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Context for Filipino community based orofacial cleft prevention interventions

Sandra Elaine Daack-Hirsch
University of Iowa

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CONTEXT FOR FILIPINO COMMUNITY BASED OROFACIAL CLEFT
PREVENTION INTERVENTIONS

by

Sandra Elaine Daack-Hirsch

An Abstract

Of a thesis submitted in partial fulfillment
of the requirements for the Doctor of
Philosophy degree in Nursing
in the Graduate College of
The University of Iowa

May 2007

Thesis Supervisor: Professor Toni Tripp-Reimer

ABSTRACT

Among Filipinos of lower SES 1/500 babies are born yearly with an orofacial cleft. This is one of the highest birth prevalence of orofacial clefting in the world. The main purpose of this study was to obtain contextual information prior to planning for community based health interventions in the Philippines regarding orofacial clefting. A descriptive ethnography was used to describe working class Filipinos' (including healthcare workers') current beliefs about the causes, prevention, and treatment of orofacial clefting, and vitamin taking practices during pregnancy. Modifications of Kleinman's explanatory models were made to include questions about people's general and personal beliefs about cause and prevention of cleft. Innovative methods were developed and used in field research and included an oral back translation method and double translation process.

Filipinos reported the following explanations for cause of cleft inheritance, falls, cravings, environmental exposures, and God's will. Beliefs about prevention of cleft included limiting their number of children, being careful not to fall, and avoiding environmental exposures. Filipinos seek surgical repair as treatment for their cleft. Iron was the supplement women reported taking most often during pregnancy. Female participants reported that feeling better, cost of multivitamin, side effects, and bad smell and taste were reasons why they quit taking micronutrients before they had completed the recommended course.

This study is the first to construct a Filipino explanatory model specifically for clefting. In constructing Filipino's explanatory model for clefting we found that people's general causal explanations for cleft were not always congruent with personal causal

explanations, and people's causal explanations for cleft were not always congruent with their prevention explanations. Modifying Kleinman's explanatory models to include questions about general and personal explanations for cause of illness and questions about prevention should be used to educe a more complete explanatory model. Results from this research can be used to inform the design of health campaigns and/or possible vitamin trials. These campaigns could include but are not limited to developing information brochures and programs about the cause and prevention of clefting, or developing public health campaigns to promote the use of prenatal vitamins in women of childbearing age.

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CERTIFICATE OF APPROVAL

PH.D. THESIS

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has been approved by the Examining Committee
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To my children, Leah and Will. To Mike, who works much harder than I do every day.

It is common sense to take a method and try it. If it fails, admit it frankly and try another.
But above all try something.

Franklin D. Roosevelt

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ABSTRACT

Among Filipinos of lower SES 1/500 babies are born yearly with an orofacial cleft. This is one of the highest birth prevalence of orofacial clefting in the world. The main purpose of this study was to obtain contextual information prior to planning for community based health interventions in the Philippines regarding orofacial clefting. A descriptive ethnography was used to describe working class Filipinos' (including healthcare workers') current beliefs about the causes, prevention, and treatment of orofacial clefting, and vitamin taking practices during pregnancy. Modifications of Kleinman's explanatory models were made to include questions about people's general and personal beliefs about cause and prevention of cleft. Innovative methods were developed and used in field research and included an oral back translation method and double translation process.

Filipinos reported the following explanations for cause of cleft inheritance, falls, cravings, environmental exposures, and God's will. Beliefs about prevention of cleft included limiting their number of children, being careful not to fall, and avoiding environmental exposures. Filipinos seek surgical repair as treatment for their cleft. Iron was the supplement women reported taking most often during pregnancy. Female participants reported that feeling better, cost of multivitamin, side effects, and bad smell and taste were reasons why they quit taking micronutrients before they had completed the recommended course.

This study is the first to construct a Filipino explanatory model specifically for clefting. In constructing Filipino's explanatory model for clefting we found that people's general causal explanations for cleft were not always congruent with personal causal

explanations, and people's causal explanations for cleft were not always congruent with their prevention explanations. Modifying Kleinman's explanatory models to include questions about general and personal explanations for cause of illness and questions about prevention should be used to educe a more complete explanatory model. Results from this research can be used to inform the design of health campaigns and/or possible vitamin trials. These campaigns could include but are not limited to developing information brochures and programs about the cause and prevention of clefting, or developing public health campaigns to promote the use of prenatal vitamins in women of childbearing age.

TABLE OF CONTENTS

LIST OF TABLES	ix
LIST OF FIGURES	x
CHAPTER 1: INTRODUCTION CONTEXT FOR FILIPINO COMMUNITY BASED OROFACIAL CLEFT PREVENTION INTERVENTIONS	1
Background.....	2
Problem.....	4
Purpose	6
Specific Aims	8
Significance	10
Summary of Introduction.....	12
CHAPTER 2: REVIEW OF THE LITERATURE	14
Biomedical Explanation of Orofacial Clefting.....	14
Genes in Orofacial Clefting.....	18
Environmental Factors and Orofacial Clefting.....	22
The Philippines	30
Biomedical Explanation of Orofacial Clefting in the Philippines.....	38
Ethnomedical Explanations of Orofacial Clefting in the Philippines.....	41
Summary of the Review of Biomedical and Ethnomedical Explanations for Orofacial Clefting	45
Conceptual Framework.....	46
Social Marketing	47
Summary of Conceptual Framework.....	53
CHAPTER 3: METHODS.....	56
Design.....	57
Informant Selection and Recruitment.....	58
Interview Schedule	63
Data Collection	69
Informant Interviews	70
Data Collection by Site.....	75
Bacolod City.....	76
Silay City.....	77
Kabankalan City.....	79
Himamaylan City.....	80
Data Management.....	81
Data Analysis.....	82
Trustworthiness and Validity.....	85
Summary of Methods	89
CHAPTER 4: RESULTS.....	90
Barangay Health Office.....	90
Informants.....	94
Healthcare workers.....	95

Noncleft Group.....	97
Cleft Group.....	99
Aim 1 Content Analysis.....	102
Label.....	103
Etiology and How Cleft Occurs	103
Prevention.....	139
Treatment.....	154
Aim 2 Content Analysis.....	154
Health Care Information Sources	155
Vitamin Use during Pregnancy	156
Perceptions about Vitamin Supplementation and Fortification.....	161
Suggestion for a Successful Vitamin Trial.....	170
Summary of Results.....	173
CHAPTER 5: DISCUSSION.....	178
Social Marketing.....	178
Audience Analysis.....	179
Channel Analysis.....	198
Market Analysis.....	199
Implications for Research Practice and Education	205
Limitations.....	220
Summary.....	223
Conclusion.....	226
REFERENCES	228
APPENDIX A: INTERVIEW GUIDE	239
APPENDIX B: INFORMED CONSENT.....	242
APPENDIX C: SOCIAL MARKETING MODEL RISK REDUCTION OF OROFACIAL CLEFTING	246

LIST OF TABLES

Table 1. Candidate genes listed by gene function.....	20
Table 2. Summary of association studies on maternal alcohol use and risk of cleft lip with or without cleft palate.....	25
Table 3. Sampling frame stratified by group and setting.....	62
Table 4. Sample stratified by gender, setting and group.....	94
Table 5. Demographics for healthcare workers stratified by setting.....	96
Table 6. Demographics for noncleft female group stratified by setting.....	97
Table 7. Demographics for noncleft male group stratified by setting.....	99
Table 8. Demographics for cleft female group stratified by setting.....	100
Table 9. Demographics for cleft male group stratified by setting.....	101
Table 10. Number of participants reporting cause of cleft by categories and by type of interview group.....	121
Table 11. Summary of discordant explanations on cause of cleft among cleft group participants.....	122
Table 12 Causes of your child’s or your cleft in the cleft group.....	125
Table 13. Attribute and values used compare responses for cause of cleft.....	129
Table 14. Number of participant responses for cravings, environmental exposure, force to the fetal face, and inheritance stratified by attributes gender, age, education, setting, and family history of clefting. (<i>n</i> = 58).....	130
Table 15. Summary of prevention categories stratified by interview group.....	140
Table 16. Modified summary of prevention categories stratified by interview group.....	142
Table 17. Number of participant responses for avoiding falls and pressure, environmental prevention, and family planning stratified by attributes gender, age, education, setting, and family history. (<i>n</i> = 58).....	144
Table 18. Summary of family planning methods used stratified by cleft and noncleft groups.....	152
Table 19. Reasons women stopped taking vitamins/iron during pregnancy stratified by cleft and noncleft group.....	161
Table 20. Perceived advantages and disadvantages of taking vitamins daily, taking vitamins monthly and fortification of rice.....	169

LIST OF FIGURES

Figure 1. Tambis.	43
Figure 2. Model of social marketing essential elements.....	48
Figure 3. Barangay health office in Patag, Silay City, Negros Occidental.....	91
Figure 4. Weekly schedule of medical services rendered at the barangay health office.	93
Figure 5. Number of participants reporting cause of cleft for cravings, environmental exposures, force to the fetal face and inheritance grouped by interview group and gender. (<i>n</i> = 58)	132
Figure 6. Number of participants reporting cause of cleft for cravings, environmental exposures, force to the fetal face and inheritance grouped by interview group and age. (<i>n</i> = 58).....	134
Figure 7. Number of participants reporting cause of cleft for cravings, environmental exposures, force to the fetal face and inheritance grouped by interview group and education level. (<i>n</i> = 58).....	136
Figure 8. Number of participants reporting cause of cleft for cravings, environmental exposures, force to the fetal face and inheritance grouped by interview group and setting. (<i>n</i> = 58).....	137
Figure 9. Number of participants reporting cause of cleft for cravings, environmental exposures, force to the fetal face and inheritance grouped by interview group and family history of cleft. (<i>n</i> = 58)	138
Figure 10. Number of participants reporting prevention of cleft for avoiding falls and pressure to the abdomen, environmental prevention and family planning grouped by interview group and gender. (<i>n</i> = 58)	146
Figure 11. Number of participants reporting prevention of cleft for avoiding falls and pressure to the abdomen, environmental prevention and family planning grouped by interview group and age. (<i>n</i> = 58).....	147
Figure 12. Number of participants reporting prevention of cleft for avoiding falls and pressure to the abdomen, environmental prevention and family planning grouped by interview group and education level. (<i>n</i> = 58).....	148
Figure 13. Number of participants reporting prevention of cleft for avoiding falls and pressure to the abdomen, environmental prevention and family planning grouped by interview group and setting. (<i>n</i> = 58)	149
Figure 14. Composite Filipino explanatory model for orofacial clefting.	183

CHAPTER 1: INTRODUCTION CONTEXT FOR FILIPINO
COMMUNITY BASED OROFACIAL CLEFT PREVENTION
INTERVENTIONS

Orofacial clefting is a global health concern and among the most common birth defects worldwide with a birth prevalence of about 1/600 (*Global strategies to reduce the health-care burden of craniofacial anomalies: Report of the WHO meetings on international collaborative research on craniofacial anomalies*, 2002). Birth prevalence ranges from 1/500 to 1/1000 depending on geographic location, ethnic background, and socioeconomic status (Schutte & Murray, 1999). Poor people of the Philippines have a 1/500 birth prevalence of orofacial clefting (Chung, Mi, & Beechert, 1987; Lasa & Manalo, 1989; Murray et al., 1997).

A social marketing framework was used to guide this study on orofacial clefting in the province of Negros Occidental in the Philippines. Social marketing is an audience or client centered planning process that offers a foundation for developing culturally innovative interventions (Lefebvre & Flora, 1988; Thackeray & Neiger, 2003; Tripp-Reimer, Choi, Skemp Kelly, & Enslein, 2001). For this study, elements of formative research from social marketing were used in a cultural assessment of Filipino's beliefs, values, and practices regarding cause and prevention of orofacial cleft, and vitamin taking practices during pregnancy. This study was designed to aid in the development of larger health campaigns. These campaigns could include, but are not limited to developing a vitamin

prevention trial, developing information brochures and programs about the cause of clefting, or developing public health campaigns to promote the use of prenatal vitamins in women of childbearing age. Knowledge of another community's beliefs and attitudes about cause and prevention of orofacial clefting and acceptable modes of treatment will best lead to culturally appropriate interventions that optimize participation.

Background

Orofacial clefting is described as a complex disorder or trait. The most widely used biomedical explanation for the cause of isolated clefting and other complex disorders is a multifactorial model. A multifactorial model of causation indicates that a condition is caused by a combination of several gene variations and environmental influences. An allele, or gene variation, is one of several alternative forms of a gene or DNA sequence. An allele or alleles of one or more genes may be inherited from one or both parents to predispose their offspring to develop a particular birth defect, disease, or trait. In order for the disorder or trait to occur, the alleles must be expressed at a critical stage of human development or lifetime and in the appropriate cells. In addition, environmental factors must be present in the required dose and at a critical stage. Genetic and environmental factors act together to influence the course of human development or wellbeing. For example, a fetus could be predisposed to develop a cleft because of the

combination of alleles present in its genome, but it will only develop a cleft if it is exposed to toxins present in the maternal environment due to maternal tobacco use (Shi et al., 2007). We are just beginning to understand how these factors act independently or interact to result in an orofacial cleft.

Several candidate genes for orofacial clefting have been identified. Candidate genes include those already known to play a role in craniofacial development as well as those genes shown to cause cleft in animal models (Christensen, 1999; Jugessur & Murray, 2005; Lidral & Murray, 2004; Murray, 2002; Schutte & Murray, 1999). Studies of environmental factors and gene environment interaction influences on clefting suggest that tobacco, alcohol, and nutritional factors may be risk factors for clefting (Christensen, 1999; Little, Cardy, & Munger, 2004; Mitchell, 1997; Munger, 2001; Munger et al., 2004; Murray, 2002; Schutte & Murray, 1999). Research also suggests that low level folic acid is a risk factor for orofacial clefting, and introducing folic acid supplements to the diets of childbearing women may prevent orofacial clefting in some populations (Czeizel, Dobo, & Vargha, 2004; Czeizel, Toth, & Rockenbauer, 1996; Rouget et al., 2005; Shaw, Lammer, Wasserman, O'Malley, & Tolarova, 1995; van Rooij et al., 2004).

Results of epidemiologic research conducted in the Philippines support multifactorial causation for orofacial clefting. Munger (2004) reported that in two areas of the Philippines, poor maternal vitamin B6 status is associated with an

increase risk for cleft lip with or without cleft palate, and that low levels of folic acid may modify the risk for clefting.

Historically prenatal micronutrient supplementation programs in the Philippines have not been successful. Several factors have been suggested as reason for the programs' limited success and include: vitamins are generally thought of as treatment/cures to sickness, pregnant women present late for prenatal care, women have low compliance because of perceived side effects, and healthcare workers lack awareness related to the programs (Paulino, Angeles-Agdeppa, Etorra, Ramos, & Cavalli-Sforza, 2005). Anthropological reports describe Filipino traditional beliefs about the cause of illness as natural and supernatural. If Filipinos hold these beliefs about orofacial clefting then interventions consistent only with a biomedical understanding of its cause and prevention, such as vitamin supplementation may not be accepted.

Problem

Under the biomedical model, orofacial clefting is said to be a complex disorder with multifactorial inheritance. It is believed that a combination of genetic and environmental factors determine the risk to develop complex disorders such as orofacial clefting. When these factors are present in the right amount and during a critical time in fetal development the combination of factors

will disrupt normal facial development and result in an orofacial cleft (Mitchell, 1997).

Early interventions such as surgical repair, speech therapy, dental management, and identification of cognitive deficits related to the cleft optimize a person's ability to lead a productive life. In developing countries, like the Philippines, large segments of the population do not have access to options for repair of the cleft defect and/or treatment of secondary effects of the cleft such as speech problems, dental problems, and cognitive and psychosocial issues. Introducing folic acid supplements to the diets of childbearing age women may prevent orofacial clefting in some populations (Czeizel et al., 2004; Czeizel et al., 1996; Rouget et al., 2005; Shaw et al., 1995; van Rooij et al., 2004). Such a project is currently under consideration in the Philippines (J.C. Murray, personal communication, June 15, 2006).

Introducing a health campaign optimally begins with a contextual assessment. Social marketing is a planning process that begins with formative research with the purpose of describing the people for whom the intervention is intended and their environment. Ethnographic reports describe widespread Asian beliefs about the cause of orofacial clefting as a natural or supernatural phenomenon such as fate, imbalance of natural forces, and demons (Cheng, 1990). Beliefs about prevention and modes of treatment may also have natural and supernatural explanations. Filipinos with these beliefs may not accept

interventions consistent with biomedical understanding of cause, prevention, and treatment. This is important because divergence between researcher/clinician and parallel participant/patient explanation of illness/disease can impede progress toward improving health outcomes. On the other hand, commonalities can be used as starting points to develop culturally innovative health campaigns and interventions to decrease the prevalence of orofacial clefting. Further, an understanding of current pregnancy vitamin taking practices can inform the development of new health promotion strategies and therapies.

Purpose

The main purpose of this study was to obtain contextual information prior to planning for community based interventions or health campaigns in the Philippines aimed at informing people about causes and prevention of orofacial clefting. The main focus of this study was to understand working class Filipinos' points of view on beliefs and practices with regard to orofacial clefting and taking prenatal vitamins. There were several reasons for choosing to focus the study on working class Filipinos. First, they are the largest population group in the Philippines. Second, they also have a greater risk for orofacial clefting compared to Filipinos from a higher socio-economic class (SES) (Cembrano et al., 1995; Chung et al., 1987; Murray et al., 1997). Third, they do not have the same access to medical treatment of their cleft compared to people from a higher SES. Fourth,

they rely on charitable medical mission to receive surgical correction of their orofacial cleft. Fifth, they do not have access to secondary services such as speech therapy, dentistry and psychological services related to care of people with orofacial clefting. Thus providing prevention services, such as vitamin programs, would be more effective and less costly.

If working class Filipinos do not acknowledge biomedical explanations of the cause, treatment, and prevention of orofacial clefting, then biomedical interventions may not be accepted. To develop culturally innovative interventions related to a possible vitamin trial, it is essential to have an understanding of current vitamin taking practices among Filipinos. For example, why do women take vitamins during pregnancy and why do they quit? Where do they get information about programs? Therefore, a descriptive ethnographic study was warranted to explore and describe current beliefs about the causes, prevention, and treatment of orofacial clefting and vitamin taking practices during pregnancy among Filipinos of lower SES (Crabtree & Miller, 1999; Giorgi, 1992; LeCompte & Schensul, 1999; Miller & Crabtree, 1999; Sandelowski, 2000). This study ascertained perceptions about vitamin supplementation and fortification of working class Filipinos and healthcare workers. A social marketing process was used to guide the study and later efforts to develop interventions and possible vitamin trials that are culturally innovative and relevant (Thackeray & Neiger, 2003; Tripp-Reimer et al., 2001). In addition, explanatory models or folk

causation offered a framework to explore patterns of disease, treatment, and prevention measures regarding orofacial clefting (Cohen, Tripp-Reimer, Smith, Sorofman, & Lively, 1994; Engelhardt, 1974; Kleinman, Eisenberg, & Good, 1978; Weiss, 1988).

The central assumption used to guide this study was that knowledge of a community's beliefs about cause and prevention of orofacial clefting and their vitamin taking practices during pregnancy is fundamental to the development of culturally appropriate prevention programs that optimize participation. Therefore, this study addressed the following specific aims:

Specific Aims

1. Describe differences and similarities in attitudes and beliefs about the cause, prevention and treatment of clefting within and across three working class Filipino groups: individuals with a cleft or individuals having a child with cleft, individuals who neither have a cleft nor have children with cleft, and local healthcare workers.
2. Describe working class Filipinos' (including local healthcare workers) perceptions about vitamin supplementation and fortification during pregnancy, and obtain suggestions from the interview subjects for the design and implementation of possible vitamin trials to prevent orofacial clefting.

Participants were asked about attitudes and beliefs regarding cause, prevention, and treatment of clefting. Participants were asked about current vitamin taking practices for pregnant women and their willingness to participate in vitamin prevention trials. Women were asked these questions because they are the target population for vitamin trials. Men were asked questions related to cause, treatment, prevention, and vitamins because they may be the spouse of someone asked to participate in vitamin trials or interventions and could influence their family member's choices (Swanson & Ward, 1995). Local healthcare workers were asked about their attitudes and beliefs regarding cause, prevention, and treatment of clefting, and for ideas on how to deliver vitamins because they are typically the primary connection between the people and health care delivery in poor areas of Philippines (Ron, 1999; Swanson & Ward, 1995). Also, healthcare workers may influence or guide people's choices among available therapies and treatments (Cohen et al., 1994). It is important to look at similarities and differences within and across the groups as all offer explanations for cleft and treatment beliefs and behaviors on a social/cultural level. This strategy should generate a range of explanatory models created by the Filipino people that are representative of their culture. Information from this study should also lead to a better understanding of current vitamin taking practices during pregnancy. In turn, this understanding can aid in the development of culturally innovative interventions related to a possible vitamin trial.

Significance

The Philippines is an archipelago in Southeast Asia. Seven thousand islands of the Philippine are home to more than 89 million people (*CIA - The World Factbook - Philippines*). Approximately 1/1000 live born Caucasians are affected with an orofacial cleft (Mitchell, 1997). While in the Philippines, 1/500 live births in indigent populations will have a cleft lip with or without a cleft palate (Lasa & Manalo, 1989; Murray et al., 1997). Of the 89 million people in the Philippines 40% live below the poverty line (*CIA - The World Factbook - Philippines, ; Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*). With a birth rate of about 26 births/1000 population (*CIA - The World Factbook - Philippines*) and cleft rate of 1/500, an estimated 4,472 children are born with a cleft lip with or without a cleft palate per year in the Philippines.

In the Philippines, few health care professionals are trained to provide services to children with orofacial deformities. Healthcare resources are not available for corrective and treatment services for the working class people who have an orofacial cleft. As a consequence, many poor Filipino people do not even have primary closure of their cleft defect. Every year non-profit organizations like Operation Smile International provide surgical treatment for children and adults who have cleft and live in developing countries such as the Philippines.

From 1981 to 2001, Operation Smile has provided over 10,000 surgeries to Filipinos who would otherwise not have had access to care (R. Van der Gaurd, Operation Smile, personal communications, December 10, 2001).

At the present time it is unlikely that genetic interventions will decrease the prevalence of clefting worldwide. However, it is reasonable to propose that environmental or behavioral intervention could decrease the prevalence of clefting in some populations. In the Philippines where the need for surgical and other treatment services far exceeds the nation's ability to provide services, prevention programs are essential. Development of comprehensive prevention programs to determine the effectiveness of interventions depends on broad participation in clinical trials and other cohort studies. If prevention/intervention trials are to take place in the Philippines and are conducted by foreign investigators (non-Filipino), it is fundamental that the investigators know and understand cultural beliefs and practices regarding the cause and prevention of orofacial cleft. This understanding is vital in order to avoid prescribing treatments that may violate or contradict the cultural norm. Violations of cultural norms could lead to limited participation in clinical trials or other strategies to decrease the prevalence of clefting (Guthrie, Guthrie, Fernandez, & Barba, 1990; Mayberry, Affonso, Shibuya, & Clemmens, 1999; Ticao & Aboud, 1998; Williams, Williams, Lopez, & Tayko, 2000).

The study was designed to generate contextual information on beliefs and attitudes of Filipinos about the cause, prevention, and treatment of orofacial clefting, and current prenatal vitamin taking practices. It is anticipated that this knowledge will lead to a community-based partnership between the investigators and health officials in the Philippines. This partnership can aid the development of health programs and research to treat and prevent cleft. Findings from this study will provide background for developing and implementing interventions to increase awareness and prevent clefting among the Filipino people both in the Philippines and the United States. Such interventions include, but are not limited to social marketing, public education programs, and possible vitamin supplementation or fortification. Future research includes testing the effectiveness of these interventions. This model will be used by the investigator to pursue a subsequent program of research to develop bio-behavioral health interventions for complex genetic disorders in minority and under served populations.

Summary of Introduction

Orofacial clefting is a global health concern and among the most common birth defects worldwide with a birth prevalence of about 1/600 (*Global strategies to reduce the health-care burden of craniofacial anomalies: Report of the WHO meetings on international collaborative research on craniofacial anomalies,*

2002). Poor people of the Philippines have a 1/500 birth prevalence of orofacial clefting (Chung et al., 1987; Lasa & Manalo, 1989; Murray et al., 1997). In the Philippines, healthcare resources are not widely available for corrective and treatment services for the working class people who have an orofacial cleft. Thus providing prevention services, such as vitamin programs, would be more effective and less costly.

Munger (2004) reported that in two areas in the Philippines, poor maternal vitamin B6 status is associated with an increase risk for cleft lip with or without cleft palate, and that low levels of folic acid may modify the risk for clefting. Prenatal micronutrient supplementation programs in the Philippines have not been successful because vitamins are generally thought of as treatment/cures to sickness, pregnant women present late for prenatal care, women have low compliance due to perceived side effects, and healthcare workers lack awareness related to the program (Paulino et al., 2005). Anthropological reports describe Filipino traditional beliefs about the cause of illness as natural and supernatural. If Filipinos hold beliefs about orofacial clefting and vitamins that differ from biomedical beliefs, then interventions such as vitamin supplementation may not be accepted. The main purpose of this study was to obtain contextual information prior to planning for community based interventions or health campaigns in the Philippines aimed at informing people about causes and preventing orofacial clefting. A social marketing framework was used to guide the study.

CHAPTER 2: REVIEW OF THE LITERATURE

The review of literature begins with a discussion of current biomedical research. This literature represents a biomedical understanding of the causes and potential prevention strategies for orofacial clefting. Next, because the study took place in the Philippines, an overview of the country is provided. This provides the reader with a context for the next two sections, review of biomedical research on orofacial cleft conducted in the Philippines and ethnomedical explanations of orofacial clefting in the Philippines. The chapter will conclude with the conceptual framework used to guide the study.

Biomedical Explanation of Orofacial Clefting

The biomedical explanation for orofacial clefting is described as a complex disorder or trait, meaning that a matrix of genetic and environmental factors contribute to the cause of clefting. Orofacial clefting can be divided into four major categories. These categories are isolated cleft lip with or without a cleft palate (cl/p), syndromic cl/p, isolated cleft palate only (cpo), and syndromic cpo. Orofacial clefting is considered isolated when the birth defect occurs alone, without other structural anomalies, or cognitive deficits. Roughly 30 % of orofacial cleft cases are classified as syndromic clefts. Causes of syndromic forms of clefting include chromosome abnormalities, single gene disorders, and

teratogens. Orofacial cleft can be found as a trait in over 400 different recognizable Mendelian syndromes (single gene disorders), teratogen complex disorders, and uncategorized syndromes (*Online Mendelian Inheritance of Man*, ; Schutte & Murray, 1999). The remainder of the discussion in this chapter regarding the etiology of orofacial cleft focuses on what is known about isolated cl/p and cpo as this type of clefting is targeted in prevention trials.

The most widely used explanation for clefting and other complex disorders in Western culture had been the multifactorial threshold model. Central to this model are three major assumptions. First, the risk to develop a given condition is determined by an underlying, continuously distributed attribute called liability. Second, liability is made up of equal, additive, and relatively small effects of a number of genetic and environmental factors that are normally distributed. Third, whether the phenotype or condition is expressed or not expressed is dependent on the accumulation of risk factors, a threshold beyond which determines the observed phenotype (Mitchell, 1997). In this model the increase in risk is proportional to the “dose” of the risk factor, but it is independent of, and added to, the background risk (E. Prescott, Hippe, Schnohr, Hein, & Vestbo, 1998). Alternative explanations to the multifactorial threshold model are that clefting is caused by a single mutation in a single gene or by exposures to environmental factors.

There is evidence to support that isolated cleft is not due to a single gene mutation. The Concordance rate for monozygotic twins is much lower than the near 100 % one would expect with a single gene disorder and it ranges from 40% to 60% (Murray, 2002). Also, the risk to relatives seems to decline nonlinearly as the degree of genetic relationship to the affected person decreases (Mitchell, 1997). While cleft malformation tends to aggregate in families, no clear-cut inheritance pattern such as autosomal dominant or autosomal recessive is observed consistently. Compared to single gene disorders, the risk for a relative to be affected with cleft is low. Another argument against cleft as a single gene disorder is the fact that clefting is more common in the general population. While single gene disorders tend to be rare in the general population and more common within families (Mitchell, 1997).

At the same time, these arguments provide evidence that genes do play a role in the development of orofacial clefting. For example, orofacial clefting aggregates in families and has a recurrence risk that is higher than the general population risk. While monozygotic twins do not have 100 % concordance rate, 40% to 60% is still a higher rate than observed in dizygotic twin, 5% (Murray, 2002), and in non twin siblings, 2% - 4% (Wyszynski, Beaty, & Maestri, 1996).

More recent models of causation for complex disorders allow for the possibility that major gene loci underlie common complex disorders and traits. The expression of these disorders or traits may be the result of major

susceptibility loci operating on a background of polygenic factors that are subject to environmental exposures. One such causation model for isolated orofacial clefting is the multiplicative model. The multiplicative model hypothesizes that one to three genes of moderate effect act on a multifactorial background that includes both genetic and environmental risk factors to result in an orofacial cleft (Mitchell, 1997; Murray, 2002). Other investigators using the multiplicative model estimate the number of genes involved in clefting to be two to infinity, meaning the upper limit cannot be determined (Schliekelman & Slatkin, 2002). Like the additive model of risk, the multiplicative model also states that risk is proportional to the dose of exposure, but unlike the additive model, risk is also proportionate to the background risk (E. Prescott et al., 1998).

A more recent analysis estimates between two and fourteen loci are likely to be involved in orofacial clefting (Schliekelman & Slatkin, 2002). Schliekelman and Slatkin believe that the multiplicative model does not give a precise enough estimate of the number of causative loci (genes) for complex diseases. In their 2002 paper they state the multiplicative model based on relative risk and affected relative pairs may be inconsistent with the prevalence rate reported for many complex disorders including orofacial clefting. They developed a new method for estimating causative loci called multiplex method. This method takes into account more than relative risk and affected relative pairs. This model estimates the number of causative loci on relative risk and the entire

distribution of affected relatives in a pedigree, not just affected relative pairs. The multiplex method also considers multiplicative interactions of loci to come up with what is believed to be a more precise estimate of causative gene loci. It is now believed there are between two and fourteen causative loci for isolated cl/p (Schliekelman & Slatkin, 2002).

As Mitchell (1997) pointed out these statistical models really only provide circumstantial evidence about the mode of inheritance for isolated forms of clefting. None of the models have been proved or disproved, but each offer a framework or theory to examine genetic and environmental factors as causes of clefting. These models are used to study the action and interactions of genetic and environmental factors and their role in causing orofacial clefting.

Genes in Orofacial Clefting

Fogh-Anderson (1942) was among the first to describe an inherited component to orofacial clefting. He concluded that genetic factors play a major role in causing orofacial birth defects based on an epidemiologic study of 703 Danes with a cleft (Fogh-Anderson, 1942). His conclusion was supported by finding a familial recurrence pattern for isolated cl/p and isolated cpo and in demonstrating a high concordance rate in twins for clefting (Christensen, 1999; Schutte & Murray, 1999). Since Fogh-Anderson's report, there have been an abundance of studies devoted to the identification of genes responsible for

orofacial clefting. Genes suspected to play a role in the development of a disorder are referred to as candidate genes.

Several genetic approaches have been used to identify candidate genes and loci responsible for orofacial clefting. The process of identifying genes is often iterative with multiple techniques used by the same investigator, or a candidate gene is discovered using one method then verified using other techniques. Some of these approaches are designed to detect linkage, and others are designed to detect association or linkage disequilibrium. Other techniques are designed to evaluate specific candidate genes such as sequencing, transgenic mice, expression assays, and gene function studies. Candidate genes and loci can also be identified by genome wide association and linkage scans (Lidral & Murray, 2004; Mitchell et al., 2002).

Several thorough review articles have been written summarizing and describing candidate genes for clefting (Jugessur & Murray, 2005; Lidral & Murray, 2004; Murray, 1995, 2002; Murray & Schutte, 2004; Schutte & Murray, 1999; Wyszynski et al., 1996). Table 1 summarizes many of the candidate genes described in these reviews; the genes are listed by function.

Inconsistent results are found among candidate gene studies aimed at associating or linking specific genes to clefting. In population studies these inconsistencies are likely due to small sample sizes, genetic heterogeneity, and lack of power to detect small or weak association between a given locus and

clefing. Inconsistencies in family based studies may be due to small family sizes and the small number of families available where there are two or more affected, living relatives.

Table 1. Candidate genes listed by gene function.

Growth factor	Signaling factor	Homeodomains*	Transcription factor	Cell adhesion molecule
TGFA ^{bd}	RARA ^{abcd}	MSX1 ^{abcd}	AP2 ^a	EDN1 ^{ab}
TGFB2 ^{abd} TGFB3 ^{abd}	MTHFR ^{bd}		BCL3 ^b	
SKIb ^{abcd}	GABRB3 ^{ab}			

*homeotic genes control the developmental fates for groups of gene, ^aanimal model, ^bassociation study, ^clinkage, ^dexpression studies

Nonetheless, these studies are tremendously useful in identify genes that may aggregate with other genetic and/or environmental factors or may by themselves lead to clefing. For example, in 2000 van den Boogaard et al. reported a mutation in the MSX1 gene that segregated with affected family members in a Dutch family (van den Boogaard, Dorland, Bemmer, & van Amstel, 2000). Based on mouse studies, MSX1 was a candidate gene thought to be associated with orofacial clefing. This study showed that mutations within the MSX1 gene can cause orofacial clefing.

It is also important to comment on research that has lead to the discovery of genes responsible for syndromic forms of orofacial clefing. Research of

syndromic clefting can provide information about isolated forms of clefting in terms of revealing developmental pathways for craniofacial development. Developmental pathways can reveal other genes or environmental factors to investigate (Chakravarti, 2004; Jugessur & Murray, 2005; Lidral & Murray, 2004; Murray, 2002; Murray & Schutte, 2004). It is also thought that variants in known syndrome genes may contribute to isolated forms of clefting as is the case with the Van der Woude (VWS) syndrome gene, IRF6. Mutations in the IRF6 gene are known to cause VWS. Cleft lip and cleft palate are two main features of VWS (Kondo et al., 2002). A common variation in the IRF6 gene, known as V274I, has been associated with isolated cl/p. Zuccherro et al (2004) reported a 12% attributable risk for the V274I allele to cleft lip with or without cleft palate. Participants from the Filipinos were part of this study (Zuccherro et al., 2004). Other known genes responsible for syndromic forms of clefting and under investigation for their role in isolated clefting include: PVRL1 in Margarita Island ectodermal dysplasia syndrome (Avila et al., 2006; Sozen et al., 2001; Suzuki et al., 2000), TBX22 in X-linked cleft palate/ankyloglossia syndrome (Braybrook et al., 2001), P63 in ectodermal ectrodactyly clefting syndrome (Barrow et al., 2002; van Bokhoven & Brunner, 2003), FOXE1, and FGFR1 in Kallmann syndrome (Dode et al., 2003).

Environmental Factors and Orofacial Clefting

In addition to genetic factors several environmental factors have been implicated as having a role in causation of orofacial clefting. As early as 1943 Warkany et al. showed that nutritional deficiencies in rats lead to congenital malformation including orofacial clefting (Warkany, Nelson, & Schraffenberger, 1943).

There are a few, rare medication exposures known to cause orofacial clefting. These include phenytoin, valporic acid, and thalidomide (Wyszynski & Beaty, 1996). Other studies of environmental factors and gene environment interaction influences on clefting suggest that tobacco, alcohol, nutritional factors (B6, B12, vitamin A, zinc, and folic acid), altitude, and herbicides may be risk factors for non-syndromic clefting (Castilla, Lopez-Camelo, & Campana, 1999; Christensen, 1999; Garcia, Fletcher, Benavides, & Orts, 1999; Jugessur & Murray, 2005; Krapels et al., 2004; Murray, 2002; Schutte & Murray, 1999; Tamura et al., 2005; van Rooij et al., 2004; Wyszynski & Beaty, 1996). As with research of genetic factors, results from studies on environmental factors are inconsistent with one study reporting an association between a given risk factor and cleft and another not finding the association. Like genetic factors, the effect of non-genetic factors is thought to be modest, requiring large sample sizes to

have sufficient power to establish association between the risk factor and orofacial clefting.

Various methods of data collection are used in environmental exposure studies and contribute to the problems of inconsistent findings of association between environmental exposures and cleft. Exposure data can come from surveys designed to quantify exposure amounts or from chart reviews. Exposure has not been measured consistently across studies. For example, investigators may ask about amount of exposures daily, weekly, or monthly. Some investigators ask respondents to specify the amount while others use forced foil methods. Women are asked to recall exposure events months or years after they occurred. Mothers of affected babies may recall these exposures more readily compared to mothers whose babies are not affected. The etiologic heterogeneity of orofacial cleft makes it difficult to tease out the effect of any one risk factor. For these reasons studies investigating association between tobacco, alcohol or nutritional factors and orofacial clefting yield inconsistent results.

One way to examine these inconsistencies is through a meta-analysis. Maternal tobacco use is thought to increase the fetus' risk to develop an orofacial cleft. A recent meta-analysis on maternal tobacco smoking and oral clefts reviewed and re-examined evidence for and against an association between tobacco and oral cleft (Little et al., 2004). These researchers combined effect size for each reported study by re-calculating the unadjusted odds ratio (OR)/ relative

risk (RR). A study was excluded from the meta-analysis if there was insufficient data, if data was superseded by other studies, or if the use of controls was thought inappropriate. The meta-analysis found a RR of 1.34 (95% CI=1.25-1.44) for cl/p, RR of 1.22 (95% CI=1.1-1.35) for cpo, and RR of 1.26 (95% CI= 1.10-1.43) for all clefts in mothers who smoke compared to mothers who did not smoke tobacco during pregnancy. The authors also reported evidence for a modest dose response effect for cl/p. One of the weaknesses of this study was the inability to adjust for confounding factors such as sociodemographics, maternal age, education, parity, race/ethnicity, and other risk factors such as alcohol consumption. Individual studies made some of these adjustments and most studies reported similar adjusted RR compared to crude RR. The authors noted that the meta-analysis of maternal tobacco smoking and oral clefts could not investigate gene/tobacco interaction. The authors concluded that their findings are strong enough to justify an anti-smoking public health campaigns (Little et al., 2004).

Recently, the GSTT1 gene, a gene involved in detoxification pathways, was found to interact with maternal cigarette smoking to increase the risk for a fetus to develop an orofacial cleft. Pregnant women who smoked 10 -15 cigarettes or more per day and whose fetus was missing both working alleles of the GSTT1 gene had a 20 fold increase for their fetus to develop a cleft (Shi et al.,

2007). Thus, providing further evidence that maternal cigarette smoking can increase the risk of a fetus to develop an orofacial cleft.

Alcohol use has also been inconsistently associated with orofacial clefting.

A summary of a few studies of alcohol use and orofacial clefting can be found in Table 2.

Table 2. Summary of association studies on maternal alcohol use and risk of cleft lip with or without cleft palate.

Study Primary author	Association Yes/No	OR (95% CI)	Number of drinks	Reference
Khoury	No	1.07 (0.81,1.40)	Dichotomized Yes/No	(Khoury, Gomez-Farias, & Mulinare, 1989)
Werler	Yes	3.0 (1.1,8.5)	> 5/day	(Werler, Lammer, Rosenberg, & Mitchell, 1991)
Shaw	Yes	3.4 (1.1,9.7)	> 5/drinking occasion	(Shaw & Lammer, 1999)
Munger	Yes	4.7 (1.1,20.6)	> 10/month	(Munger et al., 1996)
Romitti	Yes	2.8 (1.2,6.6)	≥ 4/month	(Romitti et al., 1999)

Of the studies reporting a positive association for maternal alcohol use and orofacial clefting, the amount of alcohol consumed varies from just four drinks per month to more than five drinks per day. None of these studies found an association for cpo and maternal alcohol use. Munger et al. (1996) and Romitti et al. (1999) both found a positive association between maternal alcohol use and risk for the fetus to develop a cleft lip. Both used the same study population, Iowa newborns and their parents, yet the amount of alcohol consumed to produce the positive association differed. This difference could be due to the larger sample size used in Romitti's study compared to Munger's study. Munger's study included births from 1987 to 1991 and Romitti included births from 1987 to 1994 (Munger et al., 1996; Romitti et al., 1999). Romitti's study also found that the odds ratio increased when taking into account the fetal genotype variants in the following candidate genes: TGFA, TGFB, and MSX1. This suggests an interaction between fetal genotype and maternal exposure to alcohol (Romitti et al., 1999).

Despite inconsistent results testing for an association of orofacial cleft with maternal tobacco or alcohol use, the International Collaborative Research Project made recommendations against the use of alcohol and tobacco by women of childbearing age. The recommendation states that there is enough evidence of firm, causal relationships between alcohol and orofacial cleft, and tobacco and orofacial clefts to warrant strong worldwide, public health measures to discourage

their use near the time of conception and during pregnancy (*Global strategies to reduce the health-care burden of craniofacial anomalies: Report of the WHO meetings on international collaborative research on craniofacial anomalies*, 2002).

The role of vitamins, especially folic acid (FA) in development of birth defects has been researched for over 40 years (N. J. Prescott & Malcolm, 2002). FA has been a popular micronutrient to study in orofacial cleft research because FA has been successful in reducing neural tube defects (NTD), and tissues of the face and neural tube are derived from the neural crest cell lineage.

Periconceptional use of FA was conclusively shown to decrease the risk of having a baby with a NTD by 72% by The Medical Research Council study. The Medical Research Council study was a randomized, controlled, double blind, multicentered, trial. Women who ingested 4 mg/day of FA from before conception up until 12 weeks gestation showed a 72% decrease in recurrence of NTD (MRC, 1991). These findings have been verified in subsequent case control and prospective studies (N. J. Prescott & Malcolm, 2002).

Folate is needed for the synthesis of nucleic acids, the building blocks of DNA. Compromised synthesis of nucleic acids could lead to defects in DNA replication which in turn interferes with cell growth. Folate is also involved in the methionine cycle. This cycle produces methyl groups that are needed for DNA methylation. Methylation is fundamental for the regulation of gene expression.

Cell growth and regulation of gene expression are both central to fetal development and disruptions in these processes could lead to fetal defects. Genes that code for metabolizing enzymes, such as methylenetetrahydrofolate reductase (MTHFR) may contain variations that interfere with folate metabolism and lead to defects in cell growth and gene regulation (Cummings & Kavlock, 2004; N. J. Prescott & Malcolm, 2002; Zeiger & Beaty, 2002a). However, genetic studies have been inconclusive for evidence of involvement of metabolizing enzymes in the development of orofacial cleft (Cummings & Kavlock, 2004; N. J. Prescott & Malcolm, 2002; Zeiger & Beaty, 2002a, 2002b).

Other studies have examined the relationship between maternal use of FA, from periconceptional until the second trimester of pregnancy and fetal risk for orofacial clefting. Retrospective studies using a case control design and self report questionnaires to document use of folic acid during preconception and pregnancy have demonstrated mixed results. In these studies, mothers of children with a cleft were compared to mothers of children without a cleft for preconception and early pregnancy multivitamin and folic acid use. Results from two studies showed a protective effect for folic acid on orofacial clefting (Czeizel et al., 1996; Shaw et al., 1995). A third study found no effect (Hayes, Werler, Willett, & Mitchell, 1996).

Prospective studies investigating folic acid and cleft prevention also had mixed results (Czeizel & Hirschberg, 1997; Tolarova & Harris, 1995). Tolarova

and Harris (1995) did not use a randomized control design. They invited women to take 10 mg of folic acid per day preconception, and then compared cleft rates in offspring between takers and non-takers. This group found a significant reduction of clefting in offspring of woman who took folic acid. Czeizel and Hirschberg (1997) randomized women into two groups. Case mothers received 0.8 mg of folic acid daily and control mothers took a placebo with trace elements. No differences were found in the rate of clefting in offspring for case and control mothers (Czeizel & Hirschberg, 1997). In two recently published studies the estimated decrease in cl/p risk with supplements containing folic acid ranged from 18% (Czeizel et al., 2004) to 50% (van Rooij et al., 2004). Low dietary folate intake was reported to be associated with an increase risk for oral cleft risk by up to 70% when comparing mothers with a daily folate intake of 0.18gm to mothers with a daily intake of 0.35gm (Rouget et al., 2005).

While studies did not consistently demonstrate a role for folic acid in orofacial clefting, they suggested that low maternal levels of folic acid may be a risk factor for the fetus to develop orofacial clefting. In contrast, high dose (10 mg) of maternal supplementation of folic acid may have a protective effect for orofacial clefting. It is yet to be determined whether folic acid supplementation or food fortification can reduce the risk for orofacial defects (N. J. Prescott & Malcolm, 2002).

There is biologic plausibility for the role of FA in craniofacial development, but there is little evidence to support a link between genetic variants, FA, and cleft pathology. More research is needed to examine genetic variants and FA metabolism to clarify the relationship between FA metabolism and orofacial clefting. Results from case control and prospective studies examining the effect of FA supplementation to reduce the risk of orofacial defects have been inconclusive. Randomized, controlled, double blind, multicentered, trials of FA supplementation are needed to determine the efficacy of FA at reducing risk for orofacial clefting.

The Philippines

The following is a summary describing the Philippines from the World Factbook, Library of Congress and the researcher's personal experiences conducting research, in the Philippines, over the past 12 years (*CIA - The World Factbook - Philippines*, ; *Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*).

The Philippine archipelago lies in Southeast Asia. The archipelago is made up of some 7,100 islands. The Philippines occupies an area stretching 1,850 kilometers, located south of China, to the east of Vietnam and north of Indonesia. The total land area is almost 300,000 square kilometers just slightly larger than the state of Arizona. Approximately 1,000 of its islands are populated. Most of

the islands are very small with land areas of 2.5 square miles or less. Eleven islands make up 94 % of the Philippine landmass. Two of these larger islands, Luzon and Mindanao, and the cluster of the islands between them, called the Visayan Islands, represent the three main regions of the Philippines. The Philippines has a tropical marine climate with two seasons, a rainy season and a dry season. The summer monsoon season spans from May to October when there are heavy rains and it is hot and humid. The winter monsoon season is cooler with drier air; this season spans from December to February. The dry season spans from March to May, The temperature begins to rise during the dry season but there is less rainfall (*CIA - The World Factbook - Philippines, ; Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*).

Negritos were the earliest inhabitants of the Philippines islands, followed by Malays. The islands of the Philippines have been described as place where Malay, Chinese, Spanish and American cultures have blended over to time to become the ethnic group, Filipino (*Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*). The Philippine Islands became a Spanish colony during the 16th century. After the Spanish-American War in 1898 it was ceded to the United States. On November 15, 1935, the self-governing Commonwealth of the Philippines was established, but shortly after this World War II began and the Japanese would occupy the Philippines for

almost three years. Japanese occupation lasted from January 2, 1942 until the official surrender of Japan to the United States on September 2, 1945. The islands attained their independence from the US in 1946. However, the US maintained military bases in the Philippines until 1992 (*CIA - The World Factbook - Philippines, ; Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*).

Ethnically the Filipino people are mainly Malay (96%), followed by Chinese (1.5%), and other (3%). Eighty-three percent of people are Roman Catholic while 9% of people affiliate with other Christian religions. Five percent of people are Muslim with most of these people living in the southern region of Mindanao and the Sulu Archipelago. Buddhist and other religions make up about 3% of the population (*CIA - The World Factbook - Philippines*).

Language is the main source of internal differences between Filipino people. Comparative linguistic analysis suggests that most people may once have spoken a form of "proto-Manobo," a language of the Malay people. Over the centuries the Malay spread throughout the archipelago and each group developed a distinct vernacular that can be traced to its contact with certain groups and its isolation from others. Now eight major dialects are spoken in the Philippines: Tagalog, Cebuano, Ilocano, Ilonggo, Bicol, Waray, Pampango, and Pangasinense. Political centralization, urbanization, and extensive internal migration, are breaking the linguistic barriers down. The government has placed an emphasis on

Tagalog (Pilipino) and English at the expense of local dialects. Tagalog and English are the two official languages. English is taught in schools and is the primary language of higher education and print. Still, with eight major dialects spoken, Filipinos often communicate in English to talk with one another (*Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*). Filipinos have a high literacy rate with 92.6% of people age 15 and older able to read and write (*CIA - The World Factbook - Philippines*).

In 2006 the Philippine population numbered more than 89 million and is expected to double every twenty-nine years into the next century. The median age of Filipinos is 22.1 years, with about 36% of the population under the age of 14 years. About 60% of the population is between the ages of 15-64 and only 4% is 65 years or older. The birth rate in early 2004 was 26 per 1,000, and the death rate was 6 per 1,000. In 2004 the estimated infant mortality rate was 24.24 deaths per 1,000 live births (*CIA - The World Factbook - Philippines*).

Americans by contrast have median age of 36 years, with about 21% of our population under the age of 14 years. About 67% of the US population is between the ages of 15 and 64, and 12% of Americans are 65 or older. US birth rate in 2004 was 14 per 1,000 and infant mortality rate was only 7 deaths per 1,000 live births (*CIA - The World Factbook - USA*).

In the Philippines population density increased from 160 people per square kilometer in 1980 to 220 in 1990 (*Library of Congress - Federal Research*

Division - Country Studies - Area Handbook Series - Philippines). Population density in the State of Arizona, a state comparable in size to the Philippines is only 13.5 people per square kilometer (*United States Census Bureau*).

In 1990, nearly six out of every ten Filipinos lived in villages or barangays. A barangay is the smallest local government unit and each barangay has an elected official, the barangay captain. Each barangay consists of a number of puroks (neighborhoods). Each purok has 15 to 30 households, and most barangays have from 150 to 200 households. Barangays usually contain an elementary school, one or more small retail stores, and a small Roman Catholic chapel. Most barangays also have a health center that is staffed by at least a midwife and usually a health worker. As part of genetic follow up studies, the principal investigator has visited health centers that contain only two small rooms and others that are contained within a large office complex. (*Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*).

Municipalities and cities are usually made up of several barangays. Municipalities are what Americans may refer to as towns or small cities. Municipalities and cities typically have at least one of each of the following: a larger Catholic church, other non-Roman Catholic churches, a number of retail stores including a marketplace, a six-year elementary school and usually a high school, rice and corn mills, a pit for cockfights, and the homes of most

landowners and middle-class teachers and professionals. These urban concentrations are the administrative center for social, health, economic, educational, and recreational organizations. Municipalities that are full-scale towns and cities may also have restaurants, movie theaters, banks, specialty stores, gas stations, a health clinic, and sometimes a hospital and hotels. Electricity is found in most homes in such towns, but there are still barangays in remote areas that do not have electricity or indoor plumbing (*Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*).

In the rural Philippines the family is central to a Filipino's identity, and many puroks are composed mainly of kin. This was observed during follow-up with families for genetic studies. Large extended families would occupy a single or several puroks close by. Kin ties are said to form the basis for most friendships and extended family relationships. Filipinos also form strong ties with their neighbors who provide assistance with labor and childcare (*Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*).

Many of the people in the rural areas are subsistence farmers or tenant farmers. However, many people are landless farmers or rural squatters. These people have little chance to better their lives, as they do not qualify for opportunities created by land reform or welfare programs. These people are often

allowed to farm on the land of landowners and pay the landowner “rent” in the form of a proportion of their crop yields (*Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*).

In the provinces’ remote areas, it is not unusual to find people living in nipa houses, made of bamboo and nipa-leaf thatching with no indoor plumbing or running water. By the early 1990s many houses, except in the most remote areas, were built of stronger material such as cinder block or concrete and hard wood. These houses are equipped with electricity and indoor plumbing (*Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*). Between 1998 and 2002, during follow-up visits for genetics studies, this researcher observed people living in nipa houses that had a source of electricity for lights and a television set but no indoor plumbing.

Manila is the capital city of the Philippines. Like large metropolitan cities of the West, Manila is the headquarters to major businesses, companies, and banks. More than one-half of the residents of Metro Manila are born elsewhere. Thousands of squatters live in Manila in sharp contrast to the modern façade of the corporate world and the wealthy, gated, urban, communities. People live in makeshift shacks within slums or in single shacks along the streets. These people live without water or plumbing and make their living by salvaging material from garbage dumps, peddling, and “performing irregular day work.” Regardless of where Filipinos live, their traditional pattern of behavior is described as one of

hospitality, interdependence, patron-client bonds, and strong kinship ties (*Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*).

The Philippines has a dual health care system made up of modern (Western) and traditional medicine. The modern system is based on the germ theory of disease and there are many “scientifically” trained nurses, doctors, and therapists. Traditional views will be discussed in the upcoming section.

Tropical and communicable diseases remain common health problems in the Philippines. The provision of immunizations also remains a problem. Many hospitals still have measles and tetanus wards for children. In 1980 deaths from communicable diseases were reported to be about 26 percent. In 2004 approximately 40% of the population lived below the poverty line (*CIA - The World Factbook - Philippines, ; Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*). A high birthrate and large number of children per family tends to devour family resources and to especially compromise the health of mothers. The main general health concerns are pulmonary, cardiovascular, malnutrition, and gastrointestinal disorders. Persistent poverty and lack of access to family planning underlie many health problems of the Filipino people especially women and children (*CIA - The World Factbook - Philippines, ; Library of Congress - Federal Research Division - Country Studies - Area Handbook Series - Philippines*).

Most “modern” health care personnel and facilities are located in urban areas. There was substantial migration of physicians and especially nurses to the United States in the 1970s and 1980s and now again in the 2000s. One can only guess that the exodus of health care professionals has compromised health care advances and the general health of the people in the Philippines. It has been this researcher’s experience in the provinces and poor areas of the Philippines, that hospital equipment is often non-functional and outdated. There were not enough technicians capable of maintenance and there was no stable funding source to purchase new equipment.

Biomedical Explanation of Orofacial Clefting in the Philippines

Epidemiologic research conducted in the Philippines concurs with findings from other genetic and environmental research on the cause of isolated cleft and support a multifactorial inheritance pattern described by Mitchell (1997) and Murray (2002). Three studies report familial recurrence patterns similar to those found in Caucasian populations. These studies found a 2% recurrence risk for parents with one affected child (Cembrano et al., 1995; Lasa & Manalo, 1989; Murray et al., 1997). These studies also found a 1/500 prevalence rate of clefting in indigent people of the Philippines (Chung et al., 1987; Lasa & Manalo, 1989; Murray et al., 1997). Other studies reported a prevalence rate closer to 1/800 to

1/1000 in Filipinos living in Hawaii or those with higher SES in the Philippines (Chung et al., 1987; Lasa & Manalo, 1989; Murray et al., 1997). The discrepancy between indigent and more affluent Filipinos suggests that a change in an environmental factor such as diet may explain the differences in prevalence.

Munger and colleagues (2004) conducted a case control study in the provinces of Negros Occidental and Davao, Philippines. Cases were ascertained during an Operation Smile medical mission in February of 1997 in Negros Occidental and February, 1999 in Davao. During the respective month/year controls were ascertained from randomly selected barangays representing the same geographical location from which the cases were derived from. They found that vitamin B6 deficiency was associated with an increased risk for having a child with isolated cl/p in both case groups of women. Vitamin B6 was measured by B6 activity coefficient, the higher the value, the greater the deficiency. An activity coefficient of greater than 1.85 is defined as clinical B6 deficiency. Vitamin B6, plasma folate and red cell folate were grouped into tertiles. B6 activity coefficients in the second and third tertile (first tertile was the reference) were associated with an increased in risk of having a child with isolated cl/p (OR 2.95 95% CI 1.51, 5.73 and OR 4.98 95% CI 2.56, 9.67). Increasing tertiles of plasma folate were marginally associated with an increased risk for isolated cl/p in both groups.

However, increasing tertiles of red cell folate were associated with a decreased risk for cl/p in Negros Occidental and an increased risk in Davao. B6 had an even stronger association with isolated cl/p among mothers with the lowest plasma folate levels. These findings suggest that in this Filipino population, low levels of vitamin B6 are associated with an increased risk for having a child with cl/p and that folate levels may modify this risk (Munger et al., 2004).

Although the Philippines is categorized as a developing country, it is becoming more industrialized and the prevalence of diseases associated with industrialized nations are beginning to rise. Heart disease, diabetes mellitus, and cancer are increasing. In addition to these western diseases, subclinical vitamin A deficiency; goiter; anemia; dietary deficiencies in total calories (88% of RDA); and deficiencies in iron, calcium, thiamin, and riboflavin continue to be problems reflecting the overall poor dietary conditions of many Filipino people (Cabrera, 1998). In Cabrera's 1998 report on recent developments and future plans for Philippine recommended dietary allowances, plans specific to folic acid were not mentioned. Cabrera (1998) reports the Philippine government is looking into iron fortification programs that might include sugar, rice, bread and cooking oils.

Ethnomedical Explanations of Orofacial Clefting in
the Philippines

Filipinos use both natural and supernatural explanations for health, illness, and disabilities. The Tagalog term for disease is sakit. The term sakit also includes pain, and site of pain is a category some Filipinos traditionally use to group diseases (Frakes, 1961; Williams, 1978; Williams et al., 2000).

For some Filipinos illness or disease can be viewed as physical, emotional, spiritual, and mental discomfort and represents the negative relationship between humans and their natural and supernatural environment (McKenzie & Chrisman, 1977; Miranda, McBride, & Spangler, 1998; Williams, 1978). It is common for some Filipinos to use herbs, teas, rubs, prayers, and rituals prescribed by a traditional healer, hilot (traditional midwife), or folk doctors (Filipinos refer to folk doctors as quack doctors) to treat illnesses or symptoms before seeking care from a medical doctor or along with allopathic interventions (McKenzie & Chrisman, 1977; Williams, 1978). People can purchase medications or allopathic treatments at local pharmacies without a prescription from a physician (McKenzie & Chrisman, 1977; Williams, 1978).

Indeed while traveling to even remote areas of the Philippines over the past twelve years, we came upon privately owned and operated pharmacies. These pharmacies were usually family owned and the storefront was part of the

family home. Depending on the extent of the pharmacy's inventory one could purchase items such as syringes and needles, penicillin, aspirin, and local remedies. While participating in medical missions with Operation Smile, it was common for parents to report that the birth of their child was attended by a hilot. Families often referred to folk doctors as the person who treated their child for acute illness, chronic conditions, or who pulled infected teeth.

Natural explanations for illness include beliefs that wind and exposure to heat or cold can disturb the equilibrium between hot and cold within the body and bring about illness. Other natural explanations for illness include heredity, exposures to foods, stepping on dirt, excessive drinking, poor diet, lack of sleep, the moon, night air, and exposure to sick people. During medical missions, informal interviews of mothers revealed some of their cultural views on causes of clefting. Some mothers explained they craved and ate specific foods, chicken liver or some fruits that they thought were shaped like a cleft to cause their child's cleft. Other mothers stated that an accident such as falling or that heredity caused their child's cleft.

A traditional Filipino belief is that a pregnant woman's food cravings should be satisfied especially during the first trimester to avoid harming the baby. However, it is also believed that a baby will take on characteristics of the food craved and eaten by the mother (Miranda et al., 1998). In 2001 on the island of Cebu, Philippines, a mother of a child with a bilateral cleft lip and palate was

interviewed for genetics research. In response to questions about what cause her child's cleft, she said she craved and ate a fruit called tambis. Tambis is bright pink fruit with a spicy, sweet taste. The shape of the top of the fruit appears similar to an un-repaired bilateral cleft (Figure 1). The mother believed craving and eating tambis during the pregnancy caused her son to have a bilateral facial cleft.

Supernatural causes of illness have been attributed to witches, sorcerers, devils, dead relatives or friends, and objects as conduits of harmful influences. Talismans, amulets, a cross, or prayers may be used to ward off or drive away supernatural forces (McKenzie & Chrisman, 1977; Williams, 1978).

Figure 1. Tambis.



McKenzie and Chrisman (1977) also described three Filipino traditional concepts thought to improve health and treat diseases. These include flushing, heating, and protection. Flushing is a practice that eliminates impurities or toxins by vomiting, elimination, urinating, menstrual bleeding, coughing, or perspiration. Heating is based on the Asian concept of balancing hot and cold. In addition to food and liquids that can be ingested to correct an imbalance of temperature, a hot or cold massage can be applied to ailing body parts to reduce discomfort. Protection is taking preventative measure against either supernatural or natural elements that could invade the body or mind. Protection can be a physical barrier or can operate through actions taken to avoid the supernatural or natural elements thought to cause a particular illness (McKenzie & Chrisman, 1977; Williams, 1978). Some children coming in for exams during Operations Smile medical missions wore scapulas depicting Catholic saints to protect them from harm.

Cheng (1990) provided a general overview of Asian American cultural beliefs on birth defects with an emphasis on clefting. However, her overview did not contain research specifically documenting Filipino attitudes and beliefs about the cause and prevention of orofacial clefting. Cheng described Asian beliefs of natural and supernatural phenomena causing disease and birth defects. For example, some Chinese people believe that eating rabbit during pregnancy can lead to a “harelip” baby. People of the Buddhist faith often believe that fate or bad karma cause illness including deformity. Other Asian views on disabling

conditions included retribution for wrongdoings, spells, and the imbalances of natural forces (Cheng, 1990). Because children with disabilities and deformities are viewed as punishments for sins, as monsters, or as foreign spirits, children could be abandoned or placed in special institutions (Cheng, 1990). The most specific reference to Filipinos' views on clefting was that in Filipino and Chinese cultures children with a birth defects may be viewed as a curse and ostracize from society (Cheng, 1990).

In the past 12 years this research team has visited over 300 families whose members have cleft and have only heard of one family who has been ostracized from the community because of their clefts. Attitudes and beliefs about cause, treatment, and prevention of orofacial cleft specific to Filipino culture have not been reported.

Summary of the Review of Biomedical and Ethnomedical Explanations for Orofacial Clefting

From a biomedical perspective, orofacial clefting is most likely due to the interaction of several genetic and environmental factors. The most widely researched environmental risk factors for orofacial clefting are tobacco, alcohol, and folic acid deficiency. Filipinos have one of the highest birth prevalence of orofacial clefting worldwide, 1/500. This prevalence is lower in Filipinos of higher SES and Filipinos who live in the USA, suggesting that some nutritional

risk factor plays a role in orofacial clefting among Filipinos of lower SES.

Research findings, including one conducted in the Philippines, suggest vitamin B6 and folic acid could be nutritional factors that modify risk for orofacial clefting.

Some Asian people, including Filipinos, attribute illness and birth defects to natural and supernatural phenomenon. In the Philippines successful health interventions for families of children with clefts may require that a clinician develop interventions and communication skills that are socially and culturally anchored. These may include developing harmonious verbal and nonverbal communication strategies and becoming aware of other's personal and cultural health and illness beliefs.

Conceptual Framework

A social marketing framework was used to guide this study. Social marketing is an audience or client centered planning process that offers a foundation for culturally innovative interventions (Thackeray & Neiger, 2003; Tripp-Reimer et al., 2001). "Social marketing is the process of influencing human behavior on a large scale, using marketing principles for the purpose of societal benefit rather than commercial profit" (Smith, 2000, 2006). The product in social marketing is often nontangible such as ideas, attitudes, and life changes aimed at solving a societal problem. Marketing in social marketing is directed at developing ideas that sell themselves rather than selling the developer's idea

about what one needs in order to make money (Lefebvre & Flora, 1988; McDermott, 2000; Smith, 2000; Thackeray & Neiger, 2003).

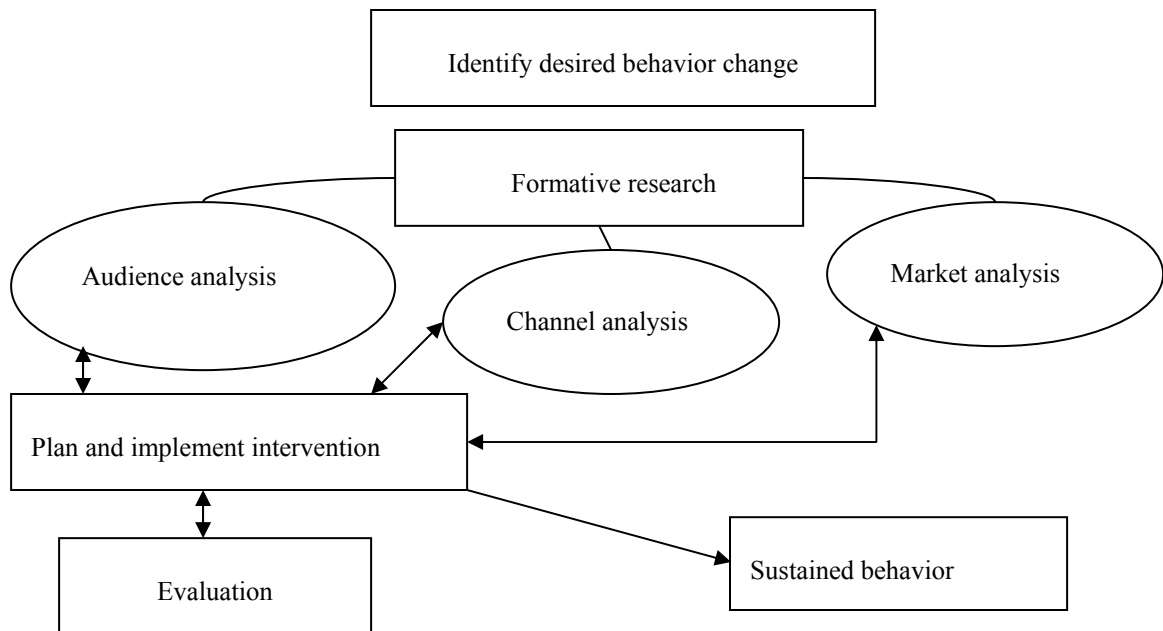
The central assumption guiding this study was that knowledge of another community's beliefs and attitudes about cause and prevention of orofacial clefting and acceptable modes of treatment will best lead to culturally appropriate prevention programs that optimize participation.

Social Marketing

Lefebvre and Flora (1988) outlined essential elements to social marketing (Figure 2). Social marketing is consumer orientated toward a voluntary exchange of goods and services between the provider and consumer. In order to begin the change process, formative research is conducted. Formative research consists of audience analysis, channel analysis, and market analysis. Findings from formative research are used to develop and implement the intervention that will affect the behavior change. An evaluation system is created to monitor the change agency and analyze problems. Evaluation planning and implementation are iterative processes. A successful social marketing campaign results in sustained new behavior.

As stated earlier, the goal of social marketing is to change consumer behavior. Business marketing, too, is concerned with consumer behavior.

Figure 2. Model of social marketing essential elements.



However, business marketing is product driven with the attitude of “we know what the consumer needs.” Certain types of health promotion campaigns also have the attitude of “we know what the consumer needs.” For example, health care professionals may believe the consumer needs to quit smoking, even though some consumers do not believe this to be true. Interventions developed using social marketing, however, will address what the consumer needs in ways that satisfies both the consumer and change agent and evolves with constant consumer input (Lefebvre & Flora, 1988). This is accomplished by utilizing exchange theory and formative research.

Marketing is about the exchange of value between two or more parties in which each party's wants are satisfied by the exchange. In social marketing an exchange example using clefting might be as follows. In order to reduce the prevalence of orofacial cleft, a goal of a social market campaign could be to increase the number of women taking prenatal vitamins before they become pregnant. However, mothers' biggest concern may be feeling less energy while pregnant, preventing them from performing daily tasks. From the mother's point of view she wants more energy. Therefore, she may take prenatal vitamins in exchange for feeling more energetic. The health care provider's concern is to reduce the prevalence of orofacial clefting. Therefore, the healthcare provider will offer a way for mothers have more energy to complete daily work in exchange for their participation in a vitamin program (Smith, 2000). The exchange principle in social marketing states that it takes more than information or forced action to change behavior; people change if they get something they value (Smith, 2006). The wants or desires of consumers are identified through formative research.

The process of social marketing involves formative research. Formative research includes audience analysis, channel analysis, and market analysis for the purpose of describing the consumer and their environment. Again the emphasis is on the consumer otherwise known as the audience.

Audience analysis identifies what the target audience currently knows, believes, and does related to the behavior change objective. This analysis may include cultural views on cause, treatment, and prevention of illness, patterns of decision making, reasons for seeking care, and readiness for change. The first aim of this study was to describe what Filipinos know and believe about cause, prevention, and treatment of orofacial clefting. One option or behavior change being considered is the possibility of prescribing vitamins before and during pregnancy as an intervention to prevent orofacial clefting. Explanatory models (EMs) were used for the audience analysis in this study.

Explanatory models appeared in health and social science literature for the first time in the early 1970s and are grounded theoretically in cognitive anthropology (Cohen et al., 1994). The explanatory model of illness was most developed by Arthur Kleinman and his colleagues to define illness within a cultural context and is based on the premise of the social construction of reality (Cohen et al., 1994; Kleinman, 1978; Kleinman et al., 1978; Weiss, 1988). Kleinman contends that illnesses are shaped by the way people perceive, experience, and cope with disease. How one perceives, experiences, and copes with illness is based in part on explanations of sickness.

Explanations are rooted in the social position people occupy as well as the meanings they give to illness events (Kleinman, 1978; Kleinman et al., 1978). People's explanations of illness are fluid and situation dependent with biologic,

psychologic, and sociocultural factors influencing the explanation they give for illness and disease. The way people define and cope with disease/illness varies across ethnic, class, and family. The researcher's or clinician's explanation of disease is also culture bound (Kleinman, 1978; Kleinman et al., 1978; Weiss, 1988). Both biomedical and ethnomedical explanations provide a clinical reality. Explanatory models provide a context for people to diagnosis and treat or seek treatments for sickness regardless of their perspective.

A second component of social marketing, channel analysis, looks at the best way to reach the target audience and their preferred methods and sources of health information. In channel analysis the program developers look at how health information is disseminated on a regular basis and how well it works. Effective use of data collected during channel analysis includes incorporating several audience preferred communication channels to promote the product. Information sources may include newspaper, radio, television, and healthcare centers. Life path points or places people are likely to encounter health messages and people in the community who are trusted sources of information are also tapped as communication channels (Lefebvre & Flora, 1988; Thackeray & Neiger, 2003).

The last component of formative research is market analysis. Market analysis has two main purposes. The first is to identify facilitators and barriers at both the individual and institutional level. Facilitators are individuals,

organizations, or behaviors that promote the achievement of the goal. One facilitator for a vitamin program may be local healthcare workers. The healthcare worker identifies women of childbearing age and monitors them for pregnancies in order to get them into prenatal clinics as early as possible. When women of childbearing age are identified, they could also be directed to the vitamin program. A barrier to a new vitamin program may be an existing iron supplementation program. Iron is known to cause gastrointestinal symptoms. Women experiencing these symptoms may stop taking their vitamins including the vitamin we want them to take.

The second purpose of market analysis is identifying the four Ps: product, price, place, and promotion (Lefebvre & Flora, 1988; Smith, 2000, 2006; Thackeray & Neiger, 2003). Identification of the four Ps goes back to exchange theory. For exchange to take place the change agent should understand the audience's preferences regarding the four Ps. In other words, will women prefer a liquid or pill form of the vitamin? How much will they pay for the vitamin?

In full social marketing campaigns, data collected from formative research is used to create the product and messages about the product and to pre-test them. Implementation is the activation of strategies and methods developed to reach the designated goal(s) of the program. Evaluation takes place throughout the campaign to provide constant feedback. Adjustments based on feedback are made

to keep the program on target to meet its goals (Lefebvre & Flora, 1988; Smith, 2000, 2006; Thackeray & Neiger, 2003).

Summary of Conceptual Framework

A social marketing framework was used to guide this study. Social marketing is an audience or client centered planning process that offers a foundation for developing culturally innovative interventions (Thackeray & Neiger, 2003; Tripp-Reimer et al., 2001). In this study elements of formative research from social marketing were used in a cultural assessment of Filipino's beliefs, values, and practices regarding cause and prevention of orofacial cleft as well as vitamin taking practices during pregnancy. The elements of formative research included audience analysis, channel analysis, and market analysis.

To address specific aim one and as part of audience analysis, an aggregate of individual explanatory models was used to develop a cultural understanding of the cause, treatment, and prevention of orofacial clefting. Explanatory models were used to examine similarities and variations among Filipinos with different experiences of clefting, Filipino healthcare workers, and the biomedical model of cause, prevention, and treatment of clefting (Cohen et al., 1994; Kleinman et al., 1978).

Comparing models between groups can identify discrepancies. It also allows for negotiation toward a shared model as it relates to the goal of reducing

the prevalence of orofacial clefting (Caulkins & Hyatt, 1999; Garro, 1986). Explanatory models are typically constructed by asking people questions about their beliefs about the following five elements: (a) etiology, (b) onset of symptoms, (c) pathophysiology, (d) course of illness (sick role and severity of disorder), and (e) treatment (Cohen et al., 1994; Kleinman, 1978; Kleinman et al., 1978). Because this study deals with the birth defect, orofacial cleft, and associated risk factors, Kleinman's elements were modified slightly. An interview schedule was designed to draw out perceptions of clefting in these four areas: (a) label (the name people give to orofacial cleft), (b) etiology, (c) how the cleft forms (timing during pregnancy), and (d) treatment and prevention. The element, how cleft forms, was used instead of three elements in Kleinman's explanatory model, onset of symptoms, pathophysiology, and course of illness (sick role and severity of disorder). This reflects the more static state of having an orofacial cleft compared to conditions with later onset or acquired disorders.

Elements of channel analysis and market analysis were used to address specific aim 2. Questions were developed to describe people's perceptions about vitamin supplementation and fortification during pregnancy, and to obtain suggestions for the design and implementation of a possible vitamin trial. For example, participants were asked how health information is disseminated. Participants were asked to give suggestions on how to inform people about a new program such as a vitamin trial. Participants were asked to describe existing

vitamin programs and to give suggestion about how a vitamin program might be conducted. Questions were developed to explore reasons people take prenatal vitamins and reasons they stop taking them. To elicit potential barriers and facilitators for taking prenatal vitamins, questions about preference for vitamin form (pill, liquid, chewable, and fortification), dosage, how much people would pay for vitamins, and current and past prenatal vitamin taking practices were asked (see appendix A).

This research was not a full scale social marketing campaign. Instead, this research used elements of social marketing to obtain data necessary for planning for culturally innovative interventions; in this case to prepare for a vitamin intervention trial. However, this research was also designed to aid in the development of health campaigns which could include developing information brochures and programs about the cause of clefting or developing public health campaigns to promote the use of prenatal vitamins in women of childbearing age.

CHAPTER 3: METHODS

Chapter three begins with a presentation of the study design. This is followed by a description of methods and procedures used to select informants, develop the interview schedule, collect data, and analyze data. Last, rigor including efforts to establish trustworthiness and assess validity is discussed.

A descriptive focused ethnography was used to explore and describe current beliefs about the causes, prevention, and treatment of orofacial clefting and vitamin taking practices during pregnancy among Filipinos of lower SES. In addition, this study ascertained perceptions about vitamin supplementation and fortification of working class Filipinos and healthcare workers. The social marketing process provided a framework for the study. The following specific aims were addressed:

1. Describe differences and similarities in attitudes and beliefs about the cause, prevention and treatment of clefting within and across three working class Filipino groups: individuals with a cleft or individuals having a child with cleft, individuals who neither have a cleft nor have children with cleft, and local healthcare workers.
2. Describe working class Filipino's (including local healthcare workers) perceptions about vitamin supplementation and fortification during pregnancy, and obtain suggestions from the interview subjects for the design and implementation of possible vitamin trials to prevent orofacial clefting.

Design

Qualitative descriptive studies are conducted to provide a comprehensive summary of an event within the everyday context of those events. A qualitative descriptive study is especially useful for the purpose of obtaining straight forward answers to questions of special importance to practitioners. The researcher summarizes what was said or how things were described and the meanings the participant gave rather than interpreting data through an imposed theoretical lens (Giorgi, 1992; Miller & Crabtree, 1999; Sandelowski, 2000; Thorne, Kirkham, & MacDonald-Emes, 1997). The qualitative descriptive method incorporates purposeful sampling techniques and content analysis as part of the study design (Giorgi, 1992; Kuzel, 1992; Morse, 2000; Patton, 1990; Sandelowski, 2000). Each of these will be described later in more detail.

The study was a focused ethnography in that it was topic-oriented targeting on the phenomenon of orofacial clefting. Data were collected through focused informant interviews. The investigator also drew on experiences, observations, and conversations conducted in ten visits to the Philippines, over the past 12 years, to contextualize the focused data within the broader Filipino culture (Boyle, 1994; Tripp-Reimer, Enslein, Rakel, Onega, & Sorofman, 1998; Tripp-Reimer & Kelley, 1998).

The study took place in the Province of Negros Occidental, on the Island of Negros, in the Philippines Islands archipelago. Negros Occidental is part of the Eastern Visayas and has a population of just over two million. Most of the people make their living through agriculture (primarily sugar cane farming), fishing, mining, buy-and-sell trading, and working abroad (Williams et al., 2000). Focused interview data were collected between March 16, 2005 and April 20, 2005. The research team consisted of the American principal investigator (PI), two Filipino nurses from Bacolod City (BN and PG), a Filipino note taker and transcriptionists (ME), and a Filipino driver (GG). This study was approved by institutional review boards in Iowa and the Philippines.

Informant Selection and Recruitment

We used a stratified purposeful recruitment strategy to maximize heterogeneity of informants on the basis of characteristics that may differentiate the explanatory models (J. C. Johnson, 1990; Kuzel, 1992; Miles & Huberman, 1984; Morse, 2000; Patton, 1990; Rice & Ezzy, 1999; Sandelowski, 2000). In stratified purposeful recruitment specific attributes or characteristics such as income level, religion, SES, or gender can be used to stratify recruitment efforts. Qualitative sample sets are designed to make analytic generalizations (Curtis, Gesler, Smith, & Washburn, 2000; Kuzel, 1992; Morse, 2000; Patton, 1990; Sandelowski, 2000). The sample frame and size was designed to be

informationally representative. Data obtained should be from persons who can stand for other persons with similar characteristics living throughout the Philippines (Sandelowski, 1995 p. 181).

Three groups of people were invited to participate in this study: individuals with a cleft or individuals having a child with cleft (cleft group); individuals who neither have a cleft nor have children with cleft (noncleft group) and local healthcare workers. These groups were targeted for enrollment because it was thought they might differ in their ideas on cause and prevention of orofacial clefting based on their experience with orofacial clefting. These groups were further stratified for recruitment by residence and gender. These groups were included in the study to generate a range of explanatory models that were representative of the larger community. These groups were also included because interventions are likely to target women and families in general rather than only families with histories of orofacial clefting. Even if interventions are developed to target a specific group, it is still important to understand the community culture as differences and similarities can act as barriers or facilitators to the uptake of the intervention. Local health care workers were included because in poor areas of the Philippines they are typically the primary connection between the people and the health care delivery system (Ron, 1999; Swanson & Ward, 1995).

Inclusion criteria for the cleft and noncleft group were men or women between the ages of 18 and 40 and able to consent for themselves. The inclusion

criterion for the healthcare workers was the ability to give consent. This sampling strategy is a comparative method aimed at describing emerging explanatory models and hypotheses regarding Filipino's beliefs about the cause, prevention, and treatment of orofacial clefting and vitamin taking practices during pregnancy in different contexts/groups (J. C. Johnson, 1990, 1998; Kuzel, 1992; Mackenzie, 1994; Morse, 2000; Patton, 1990).

On Negros Occidental the non-government organization, H.O.P.E. Foundation Inc. (H.O.P.E.), in collaboration with investigators at the University of Iowa, maintains a database of all people who have participated in orofacial cleft studies to identify genetic risk factors for clefting. The genetic study has IRB approval from both University of Iowa and a local Philippines IRB. Over 500 Filipino families have been registered with H.O.P.E. These families come from all over the province. With approval from the H.O.P.E. foundation and the Filipino IRB (institutional review board), the database was used in the current study to identify adults for potential cleft group participants. The cleft group participants have not been asked question about their beliefs on cause or prevention of orofacial clefting by anyone from this research team. As discussed previously, there are people in this cleft registry who have informally talked with members of this research team about what they think causes cleft. However, these informal interactions took place from 1999 to 2003. The cleft group

participants for this study were enrolled in genetic studies and included in the H.O.P.E. registry after 2003.

For the purposes of this study, working class was defined as a group of people who were employed in jobs where they were poorly paid and provided little or no benefits, or were unemployed (*Elmer Social Science Dictionary*). Such occupations could include, but were not limited to manual labor, service and repair work, agriculture, street vender operators, or public transportation work. For the purposes of this study healthcare worker was defined as all persons who engage in health and health related work employed at health centers, rural health units, and barangay health stations. These people have undergone training programs in accredited government or non-government organizations and voluntarily render primary health care services ("Magna Carta of Public Health Workers," 1992).

These three groups were further stratified into urban and rural barangays as access to health care and health care information may differ in these settings. Natural science research about urban/rural ecosystems often includes only a tacit assumption about what urban/rural means. Definitions of urban and rural ecosystems are not standard and are not typically salient (Mcintyre, Knowles, & Hope, 2000). A standard definition for rural Philippines was not found. Therefore in this study, rural was defined as those Barangays that were ten or more miles away from a clinic and/or hospital staffed by physicians and nurse.

This definition was based on usage patterns of the barangay health services and access to medical doctors and nurses. People living in rural areas relied on the local barangay health office (BHO) and staff for health care. These BHOs were staffed by a trained, certified midwife and barangay health workers. Physicians and nurses typically traveled to the barangays once a week for consultation day. People attended consultation day for treatment or to be referred to a larger health center. The purpose of selecting a 10 mile radius was to operationalize the designation of rural residency consistently throughout the study.

Table 3. Sampling frame stratified by group and setting.

Group:	Urban	Rural	Total
Cleft	15	15	30
Noncleft	15	15	30
Local healthcare worker	15	15	30
Total	45	45	90

Roughly 15-30 individuals from each of the three groups was recommended as the range in which recurring themes and patterns of the data should occur (Miles & Huberman, 1984; Sandelowski, 1995). Table 3 provides an overview of the sampling frames summarizing the targeted number of

participants in each stratum. Each stratum was constructed to consist of roughly seven to eight females and seven to eight males.

Interview Schedule

A short structured interview was used to gather demographic information and included ID, group (cleft, noncleft, or healthcare worker), address, date of interview, date of birth, education, occupation, monthly income, civil status, and religion (See appendix A). The demographic data were used to make comparisons and look for response patterns in the analysis. Questions were developed to address each specific aim based on elements of social marketing's formative research.

Interview questions to address aim one, were asked as elements of social marketing's formative research, specifically audience analysis of cultural view on cause, treatment, and prevention of orofacial clefting. As described in chapter two, explanatory models (EM) were used for audience analysis in this study. An EM is typically constructed by asking people questions about their beliefs about the following five elements: (a) etiology, (b) onset of symptoms, (c) pathophysiology, (d) course of illness (sick role and severity of disorder), and (e) treatment (Cohen et al., 1994; Kleinman, 1978; Kleinman et al., 1978). This study used EM to explore a birth defect. Therefore, questions were modified to fit the static nature of birth defects. Questions related to aim one were developed

according to these four, modified elements representing explanatory models: (a) label (what name do people give to orofacial cleft), (b) etiology, (c) how the cleft forms (timing during pregnancy), and (d) treatment and prevention (see Appendix A). Questions one to three were asked to identify or label the cleft using Filipino's words or descriptions. We asked people to name the disorder with the thought that people may have different labels for cleft depending on what they believed caused it. What we as researchers believe is one disorder others may think of as multiple disorders. Questions four to six address etiology and how cleft forms. Questions seven to nine address treatment and prevention.

Based on the PI's past experiences, questions related to etiology were modified. In previous informal interviews conducted from 2000 to 2002, Filipinos who took part in family genetic studies were asked what they thought caused cleft. First, they were asked what they thought caused cleft; then, they were asked if that was the reason their child had a cleft. Of the 109 people asked these two questions, 25% were discordant in their response (Daack-Hirsch, S., unpublished data collected 2000-2001). As a consequence, for this study, persons in the cleft group were asked both what causes cleft and what caused cleft in themselves or their child. To our knowledge this sequence of questions has not been used to explore etiology in other EM studies (T. Tripp-Reimer, University of Iowa College of Nursing, personal communications, November 15, 2006).

Question development for aim two was also guided by formative social marketing research, specifically elements of channel and market analysis. Aim two was to describe working class Filipinos' (including healthcare workers) perceptions about vitamin supplementation and fortification during pregnancy and to obtain suggestions for the design and implementation of possible vitamin trials to prevent orofacial clefting.

As part of channel analysis, people were asked how they learn about new health programs or new health information. Participants were asked to provide ideas about how to utilize existing communication channels for new health campaigns related to orofacial clefting.

Market analysis included asking participants to describe current vitamin taking practices and describe potential barriers and facilitators to taking prenatal vitamins. Participants were asked about existing prenatal vitamin programs because aspects of successful programs could be used in a vitamin trial. Participants were asked to describe how and when prenatal vitamins are currently prescribed and how use is monitored. It is important to know about existing programs because existing programs could augment or compete with a new vitamin program or trial. Questions were asked to explore barriers and facilitators to taking different forms of vitamins that might be offered as part of a vitamin trial. Advantages and disadvantages of taking currently available forms of vitamins that included pills to swallow, pills to chew, or liquids were explored.

The research team was told of two dosages of prenatal vitamins on the market. There were several prenatal vitamins prescribed for daily use and there was an iron/folic acid tablet prescribed for once a month use. Because these were the only schedules known to the research team, other dosage schedules for vitamin use were not asked about. Last, people were asked to describe successful features of a prenatal vitamin program in their community.

Translation and refinement of interview schedule

Ilonggo is the Filipino dialect spoken in the area the research was conducted. The PI did not speak any of the Filipino languages. The four Filipino research team members spoke Ilonggo, Cebuano (another dialect), Tagalog (Filipino national language) and English. Before the interview schedule was used in the field, the research team practiced asking questions and verbally translating. The process of back translation was modified as part of this exercise.

Back translation is a technique used to deal with semantic problems that may occur when translating questionnaires from one language to another (Behling & Law, 2000). Back translation process involves taking a questionnaire written in one language and translating into a second language. A second, bilingual individual with no prior knowledge of the content in the questionnaire translates the contents back the original language. If substantial differences are found between the two versions in the original language another draft is made to

eliminate the differences. This is an iterative process and continues until there are no further substantial differences (Behling & Law, 2000).

The process of back translation was modified to be an oral exercise. The oral back translation process involved three people, the PI and the two Filipino nurses (BN and PG). Back translations started by having the PI ask a question from the interview schedule in English. Then, BN asked the same question out loud in Ilonggo. PG verbally translated the question back to English, immediately, without trying to interpret what was said in English. This allowed the PI's use of English words to be modified, reflecting a more typical Filipino English vernacular. This made English translation into Ilonggo easier for BN and PG during the interviews and allowed the team to clarify the meaning of questions asked in English. Also, many participants could understand English so it was important that questions were asked using a Filipino-English vernacular. Before changing the English word it was discussed whether use of a different word lost the original meaning of the question.

For example, the PI asked if people prefer to swallow or drink their vitamin. This translated back as, "Do you prefer to drink or drink your vitamin?" In the Philippines people drink even pill forms of vitamins because the pill is taken with water, therefore, one drinks pills. The question had to be modified to ask, "Do you prefer to drink your vitamin or take it as a liquid?" Similarly, there was no Ilonggo word for birth defect. Filipinos refer to birth defects as inborn

problems meaning they are born with the problem rather than developing it after birth.

Back translation was preformed as an oral exercise rather than written because the interview was entirely oral. The original plan was to have the PI ask the questions in English then have BN or PG translate the question to Ilonggo and the Ilonggo response to English. However, it was anticipated that working through an interpreter could be awkward. Oral back translation was one way to create more fluent and natural communication among the interviewer and translator and participant. To our knowledge the use of oral back translation has not been reported.

We found that questions could not be too open ended. For example, if people were asked “tell me about the practice of women taking vitamins before pregnancy?” or “what do you think about taking vitamins before you become pregnant?” people often responded with “wala.” Wala means there is nothing, I have no thought, no, or none. Instead the question was asked in two parts and more contextual, “Did you take vitamins during pregnancy?” “Do other women take vitamins during pregnancy?” These questions were followed up by asking what was prescribed, who prescribed it, how long was it taken, and why one stopped taking the vitamin.

Other questions that were difficult for people to answer were ones related to naming barriers and facilitators for different forms of vitamins. In earlier

interviews the research team asked people what was good or bad about taking one form versus another form of a vitamin. Later, the question was changed to, “in your own opinion, what do you like about taking a pill and what do you not like about taking a pill?” People would answer wala, meaning I have no opinion to both forms of the question. Eventually, the question was reframed to have a more personal context; this seemed to generate more responses. For example, to generate more responses the following explanation was giving to participants:

one way we can give you vitamins is to give you 30, 1mg tablets and ask you to take one pill every day. Another way is to give you one 30mg tablet once per month. Which way do you prefer? What is better about that? What do you not like about the other way?

Data Collection

In order to maximize interview time in the field and minimize time spent traveling, the research team spent a day or blocks of days at six different cities in Negros Occidental. The H.O.P.E. registry was used to identify areas where several cleft families lived. In this way the team could plan a day or block of days to be in that area. Families were invited to participate ahead of the time the team planned to be in their barangay. The sampling frame for the noncleft group and local healthcare workers were the various cities the cleft group came from, but not necessarily the same Barangay. Specifics about each site and participants from that site are described in more detail below.

Informant Interviews

In all groups participants had to be 18 years or older to participate. Audio taping was desired because the investigator did not speak the native language. In addition to audio taping, ME took notes during every interview. If a participant did not want to be audio taped or environmental conditions were not conducive for audio taping, extensive note taking was used to capture the interaction. No one declined audio taping. The interviews lasted between fifteen and sixty minutes.

The PI, one or both the Filipino nurses, and ME were present during each interview. The interview began with an introduction of the research team and verbal review of the consent form. The consent form was approved by the institutional review board in the Philippines (Appendix B). This was followed by a brief introduction to the interview topic and showing the participant the recording device. Once they agreed to participate, they signed an English version of the consent document and were given the choice to keep an Ilonggo and/or English version. The interview concluded once the interview schedule was complete and/or the informant felt he or she had nothing more to say. After each interview the research team members debriefed. During this time, notes were compared with what the team perceived the participant to say. If there was

disagreement among the team or if they felt more information was needed the, the participant was asked to clarify.

The first 19 cleft and noncleft interviews were conducted by the PI with PG providing English and Ilonggo translation. There were three purposes for conducting interviews this way. The first was to verify that real time translation was accurate. Therefore, translation was verified by translating Ilonggo twice. The first translation of Ilonggo/ English was during the interview; the second translation was during transcription and is described below. Double translation was done for 30 of the 69 interviews.

The second reason for double translation was to teach PG how to interview. We anticipated that the interviews may be long and the team and/or participants may experience fatigue. Also, there may be people who would not want to have the interview conducted in English. This method would teach PG the interview process in the event that there was a need to change to conducting interviews solely in Ilonggo.

The third reason to conduct interviews with a translator was to keep the PI involved in the data gathering process. This allowed the entire team to be involved in identification of categories of response, evaluation of questions and responses, and need for modification to the protocol.

All of the 11 health care person interviews were conducted in a mix of English and Ilonggo. The PI asked questions in English and responses were given

in a mix of Ilonggo and English. Many of the healthcare workers started responding in English but as they got more passionate about their response or more in depth with an explanation they spoke Ilonggo.

After the first 19 cleft and noncleft interviews, we decided to have PG conduct the interviews entirely in Ilonggo (unless the participant preferred English, none did). As anticipated the interviews seemed long; both the participant and research team were fatigued, disengaging during the interview before the questions were complete. It also seemed that people were giving short answers to questions. At this point saturation of ideas were observed, recurring themes and patterns in responses were occurring. Sometimes PG was asking questions in Ilonggo before the PI had asked the question in English. However, the team did not want to stop recruitment. One way to establish validity in the qualitative descriptive method is for data collection to go beyond saturation. This promotes accurate description of all elements of the event as it is (Sandelowski, 2000).

Therefore, the remaining 39 cleft and noncleft interviews were conducted in Ilonggo. One interview was conducted in Ilonggo but the participant responded in Cebuano (another local dialect). The interviewer, PG spoke and understood both dialects as did the participant. When interviews conducted with translation are compared to those conducted entirely in Ilonggo no change in the quality of data was observed. Length and categories of responses did not change.

Conversation flowed more freely and participants appeared more relaxed. It was observed that sometimes during interviews conducted with immediate translation the translator would provide an interpreted translation rather than a more literal translation. The following passage demonstrates this observation:

Written translation after transcribing: For me (the participant), if there will be somebody who can bring [the medicine] to me, that is much better because sometimes we tend to forget to pick it up. Some mothers would forget to pick it up or would tend to forget to take it, habits like that.

Oral translation by PG during the interview: So she wants that...she prefers that uhm...somebody would bring the medicine to her house and that somebody will supervise the mother to drink the medicine everyday.

The participant did not say anything about someone supervising her while she takes the medicine, but the translator interpreted that because the participant may be forgetful she would also want supervision. The woman did say she would like the medicine delivered to her. In another example, PG was asking a series of questions trying to establish how often this woman (C1) took her prenatal vitamins while the PI observed. P=PG speaking, T=post transcription translation by PG:

T: you started taking these vitamins on the seventh month of pregnancy?

C1: Oo.

T: Yes

P: Tapos ka magbata gid?

T: Until your last month of pregnancy?

C1: Indi man kon...may ara man kon kis-a nga inang mabakal man ko bi sang bulong na bi...

T: Not all the time. At times I also take those that I buy.

P: Multivitamins?

C1: Oo na. Indi man gid siling nga damo gid diutay man lang.

T: Yes, not too often or not too many

P: So it's like...

C1: Ma-inom ko indi man gid damo gid.

T: I take vitamins, but not too often or too much.

P: Not every day, indi adlaw-adlaw?

T: You don't take it everyday?

C1: Ga adlaw-adlaw man kay pero indi ko masiling nga maano ko bi...

T: I took it every day, (if vitamin is available) but not always.

P: Nang sige-sige.

T: Not continuously taking it.

C1: Sa kada adlaw-adlaw kon maka-inom ko indi man.

T: There are times that I take it every day and there are times that I don't.

PG then translates this interaction to the PI:

P: For her...in all three pregnancies she took vitamins but not religiously. There are times that she doesn't take any vitamins it depends on the availability of her money because she has to buy the multivitamins.

This was PG's interpretation of the why the woman did not take her prenatal vitamin daily. However, the woman does not mention lack of money as a reason why she did not take the vitamins daily. Some days she is taking vitamins that were given to her at the health clinic and other days she is taking a multivitamin she purchased. She says that vitamins were not available every day, but the reason for this was not clarified. When the interviews were coded, literal translation was coded and if needed, interpretation was used to clarify what

passages meant. For instance in this last example, lack of money would not have been coded as a barrier to taking prenatal vitamins because, when reading the literal translation it could not be determined what the participant meant. The passage could not be clarified during the interview; during the interview the PI was only aware of PG's verbal, interpreted, translation. At the time of the interview, it was assumed that this was what the participant said and therefore did not need clarification.

Data Collection by Site

Four cities within Negros Occidental were visited over a six week period. These sites included Bacolod City, Silay City, Kabankalan City, and Himmaylan City. Negros Occidental is a province in the Visayas or central Philippines. A province is subdivided into cities and municipalities (towns). In the Philippines cities and municipalities are somewhat equivalent to counties in the United States. The cities and municipalities are subdivided into Barangays. There can be both rural (as defined above) and urban barangays within a city or municipality. Bacolod City is the capital of Negros Occidental and is classified as a highly urbanized city because its population is over 200,000. It is independent of the provincial government. Most of the interviews were conducted at the H.O.P.E. foundation or in local health centers. The health centers are described in more

detail as part of participant observation in chapter four. All cleft participants were identified through H.O.P.E. as described above.

Bacolod City

One of the Filipino research team members went to the homes of the potential participants from the cleft group and made an appointment with them to come to the H.O.P.E. office to learn about this study. Healthcare workers were identified through the city health office and one of the Filipino research team members made an appointment with them to come to the H.O.P.E. office for a possible interview. The healthcare workers were asked to bring with them or identify one noncleft participant from their barangay for us to contact. All these interviews took place in the H.O.P.E. office. Participants were reimbursed for travel expenses to and from the H.O.P.E. office.

During one of the interview days in Bacolod City three healthcare workers showed up to be interviewed at the same time. There were already people waiting because people were late and interviews were taking longer than expected. Because it was late in the day and because the healthcare workers wanted to participate, they were interviewed as group rather than turning them away. The original intent was to treat each of the woman's responses as a separate interview, but the women really answered more as group so the interview was coded and

analyzed as a single interview. All participants from Bacolod City were classified as urban dwellers.

Silay City

Two cleft participants from a rural barangay of Silay City agreed to be interviewed. The interviews took place in their individual homes because the nearest clinic was about an hour travel for them by car and it was a weekend. They did not have access to an automobile. Both these participants were known to BN and PG and lived in a remote barangay of Silay City with no reasonable way to contact them prior to our visit. The research team traveled to their homes to invite them to participate in an interview. Both women agreed to interviews.

Noncleft participants were identified on consultation day at two different barangay health offices. In rural barangay health offices, consultation day is typically a weekly service where a government health team, consisting of at least a physician and nurse, come to the barangay health center to triage and treat sick people. In urban barangay health offices, consultation day may not involve a physician. Rather, people come to the barangay health center for treatment or to receive a referral to the city health office to see a physician. Noncleft participants were people who volunteered to stay and be interviewed after consultations were completed. One healthcare worker was interviewed at each of the sites. One site was a health center in a rural barangay. This was a small health center with only

one certified midwife staffing the center. The other was a barangay health center in Silay City proper. At each site, the certified midwife in charge of the clinic was contacted prior to the team's visit to ask permission to conduct the study.

It was difficult to recruit males to participate in the study. They were busy at work or too shy to be interviewed. A different type of interview method was used, natural group (Beckerleg et al., 1997; Coreil, 1995), hoping to get more men to agree to take part in the study. This type of group interview is less formal than a focus group. It takes advantage of naturally existing groups. The interview can take place in a preset location in the field or it can happen as a spontaneous event. Unlike focus groups, people in natural groups may know each other. The interview guide is typically loosely followed and the conversation is typically recorded by notes. The interviewer's role is not as directive compared to individual or traditional focus group interviews (Beckerleg et al., 1997; Coreil, 1995; Frey & Fontana, 1993). A natural group was used one other time during data collection. That group will be described in more detail below.

Three men who were friends of one the team members were invited to participate in the study. Each was reluctant to come to the health office for an interview but agreed to meet as group at a local restaurant. The research team met the three men for drinks and snacks after work in a local restaurant. The interviews were not recorded because the restaurant was open air and noisy, instead ME took notes. A less formal setting may have helped the men to feel

more comfortable during the interview and more willing to participate. However, contrary to typical nature group interviews, an interview schedule was followed (Beckerleg et al., 1997; Coreil, 1995; Frey & Fontana, 1993). This was done because the PI desired to collect similar information as that collected during individual interviews. The interviews were conducted in a mix of Ilonggo and English as the men spoke both and freely moved between English and Ilonggo. Their responses were recorded and analyzed individually. This was because the men did not answer questions collectively as the healthcare workers in the previous group did. The previous group of three healthcare workers talked to each other during the interview and gave responses as a group.

Kabankalan City

Kabankalan is the largest city outside of Bacolod City in Negros Occidental. Kabankalan is a unique city in the province because the mayor has made care of cleft children and their families a priority. Kabankalan City health center had a nurse who was in charge of tracking all individuals and families who have a person with any type of cleft. The nurse keeps demographic information on these people as well as clinical information regarding cleft type and surgical history. She is responsible for teaching parents how to care of their children with cleft and for linking them to surgical services.

The nurse identified participants both from the cleft and noncleft group and arranged for the participants to meet the research team at one of the barangay health centers located in Kabankalan City proper. All but one of the participants lived in Kabankalan City proper. This individual lived outside the city proper in a rural barangay. The nurse arranged for transportation for the participants to and from the health center. The research team interviewed two healthcare workers from two different barangay health centers and this nurse. One of the health workers was a certified midwife. Because these two healthcare workers had extensive experience working with families with cleft they were interviewed together as key informants. The interview with them focused more on how people access the health system and how a vitamin trial might be implemented in their area. Their responses were recorded together as a single interview.

Himamaylan City

Six cleft participants who lived in Himamaylan were identified. The midwives from the participants' local barangay health center were asked to locate and invite these families to come to for interviews. The interviews took place in a health center located within a large rural barangay. Most of the families accessing this center made their living in the fishing industry. The research team spent two days in Himamaylan; the first day was spent interviewing cleft participants. The next day was immunization day. This was well baby checkup

day at the health center; it was conducted by the midwives and Barangay health workers (BHW). Mothers brought their children in for weighing and immunizations. Barangay nutrition scholars (BNS) were also available to consult on babies with malnutrition and assist with immunizations. Mothers were invited to stay for interviews after the immunization clinic.

Healthcare workers staffing this health center were also interviewed. Because it was immunization day, all the healthcare workers were at the center together at the same time. This was a large Barangay with two full time, paid midwives on staff. There were two BNSs and five BHWs. The BHWs and BNSs receive stipends for their work and were considered volunteers. The center also had two volunteer midwives who helped with immunizations. All the healthcare workers wanted to be interviewed. The research team took advantage of this natural group and interviewed all but two of the BHWs together as a group. Later the two BHWs who were not present during the group interview asked to be interviewed as part of the study. These two were interviewed each separately between nonleft participant interviews.

Data Management

Interviews conducted in English and Ilonggo were transcribed verbatim into Microsoft Word. PG and the PI, independently of each other, reviewed each transcript while listening to the taped interview to check the accuracy of the

transcription. Next, the Ilonggo was translated to English by PG in Microsoft Word.

Interviews conducted in Ilonggo were transcribed in Ilonggo verbatim into Microsoft Word. PG and ME reviewed each transcript while listening to the taped interview to check the accuracy of the transcription. The Ilonggo transcript was then translated into English by PG in Microsoft Word with explanations provided for idioms, metaphors, phrases, and cultural terms that are not readily interpretable to English.

The Olympus DS 4000 digital voice recorder was used to record the interviews. The Olympus AS 4000 PC transcription kit was used to transfer digital recording to a laptop PC. The software automatically transferred the digital recordings to a file on the laptop. The transcriptionist opened each file and transcribed the digital recording to a word document. Notes taken during interviews were used to clarify or decipher parts of the audio tape that could not be heard. The Olympus AS 4000 program then automatically stores the transcribed document linked to its audio recording.

Data Analysis

Content analysis is used in qualitative descriptive studies because its dynamic nature is geared toward summarizing the informational contents of the data (Crabtree & Miller, 1999; Morgan, 1993; Rice & Ezzy, 1999; Sandelowski,

2000; Spradley, 1979). All transcripts and field notes were entered into separate files in N-Vivo. N-Vivo is a qualitative computer data analysis program that assists the investigator in content analysis of the narrative data. N-Vivo allows for coding of data by indexing text, labeling categories of text and retrieving labeled text across all cases (Richards & Richards, 1994). Each interview and each individual field note entry was considered a case.

Content analysis of the interview transcripts included the following general steps:

Specific aim 1

Each interview and set of field notes was coded for broad topical areas and descriptive sub-codes according to four areas representing explanatory models: (a) label (the name people give to orofacial cleft), (b) etiology, (c) how the cleft forms (timing during pregnancy), and (d) treatment and prevention.

Next, text related to each of the broader categories was reviewed, and more specific descriptive sub-codes were assigned to the data grouped under the broad categories (Crabtree & Miller, 1999; Rice & Ezzy, 1999; Sandelowski, 2000; Spradley, 1979). To describe differences and similarities in attitudes and beliefs about the cause and prevention of clefting across groups, data were analyzed for regularities in terms of words, categories and subcategories across groups and within each of the three groups. Categories were summarized in

narrative, then tabulated (Miles & Huberman, 1984; Morgan, 1993; Scrimshaw & Hurtado, 1987) to examine differences or similarities in the number of patterns found within and across groups as well the frequency of identified themes (Crabtree & Miller, 1999; Ford-Gilboe, Campbell, & Berman, 1995; Miles & Huberman, 1984; Morgan, 1993; Sandelowski, 2000; Scrimshaw & Hurtado, 1987; Spradley, 1979).

The use of the N-Vivo software program facilitated the analysis within and across groups. Specifically, the Boolean search function was used to construct data matrices. In addition to categorical coding, each interview was coded according to attributes of the informant (demographic information collected prior to the interview). These attributes were potential explanations for patterns of explanatory model of clefting within and between groups. These attributes included rural/urban residence, level of education, family history of clefting, age, religion and occupation. For example, the following is a description of the construction of one possible matrix: 1) merge all text data related to ideas of causation; 2) using that subset (causation), create a matrix with the vertical axis the three categories of informants and the horizontal axis the taxonomy of causation codes. In this way, patterns across the three groups were easier to compare (Richards & Richard, 1994).

Specific aim 2

Again, each interview was coded for topical areas or broad categories elicited during the interviews. Next, text related to each of the broader categories was reviewed, and more specific descriptive sub-codes were assigned to the data grouped under the broad categories (Crabtree & Miller, 1999; Rice & Ezzy, 1999; Spradley, 1979). Last, differences and similarities across groups were analyzed for regularities in terms of words, categories and subcategories across groups, and within each of the three groups. Categories were summarized in narrative, then tabulated (Miles & Huberman, 1984; Morgan, 1993; Scrimshaw & Hurtado, 1987) to examine differences or similarities in the number of patterns found within and across groups as well the frequency of identified themes (Crabtree & Miller, 1999; Ford-Gilboe et al., 1995; Miles & Huberman, 1984; Morgan, 1993; Sandelowski, 2000; Scrimshaw & Hurtado, 1987; Spradley, 1979).

Trustworthiness and Validity

Investigator triangulation was used to establish descriptive validity (an accurate description of events) (R. B. Johnson, 1993; Maxwell, 1992; Sandelowski, 2000). Investigator triangulation involves the use of multiple observers to record and describe participants' responses and behaviors (R. B. Johnson, 1993). Investigator triangulation involved the PI collaborating with

nurses, and a research assistant from the local culture to form the research team. Research team members from the local culture were viewed as active producers rather than neutral conveyors of language and information. The research team members experiences informed the study design, data collection, and data analysis (Temple, 2002).

This research team included the PI who is a Caucasian, female, nurse from The United States. She has been a research study coordinator directing genetic and epidemiologic studies on orofacial clefting in the Philippines for over twelve years. The PI has traveled to the Philippines ten times collecting data and interviewing families for genetic studies. The PI assisted in the development of an ethics review board in the Philippines. She has been accepted by the Filipino community as culturally appropriate, such that interactions are likely to be collaborative and findings trustworthy.

Other team members included the two Filipino nurses. The Filipino nurses, knowledgeable of the goals for this research project, served as the cultural experts (Beebe, 2001). The PI and the Filipino nurses have collaborated on genetic and epidemiologic research in the Philippines for over seven years. The Filipino nurses both spoke fluent English and several Filipino dialects. The nurses' contribution to the study included: identifying participants, participating in the design of the interview schedule, and collecting and cross checking data.

Designing the interview schedule included a modified version of back translation. Back translation was performed as an oral exercise. This made English translation into Ilonggo easier for BN and PG during the interviews and allowed the team to clarify the meaning of questions asked in English. The Filipino nurses checked to make sure the questions made cultural sense to participants. The exercise also promoted consistency in asking questions during interviews regardless of which team member was the interviewer.

To increase confidence that the data was described accurately several team members were involved in cross-checking the data. The team approach of cross checking the data was to increase our confidence that there was consensus about what took place (Beebe, 2001; R. B. Johnson, 1993). Data were cross checked at several different stages during the study. Taking notes during the interview provided one way to cross check. The research assistant, ME, took notes during every interview. ME has worked on the genetics project in the Philippines for two years, is fluent in English, and a native Ilonggo speaker. The notes were used to document the interview incase the recording device malfunctioned. The notes were also used during transcription to clarify what was said when the audio recordings were not clear.

Another cross check was to debrief after each interview. Debriefing included reviewing ME's notes and a discussion among the team members present for the interview. The team discussed whether or not they agreed on

concepts presented and whether or not these concepts were consistent with what was already heard or known. The discussion also provided an opportunity to ask the participant to clarify content at the time of the interview; clarifying with participants was another way to cross check data. Debriefing also allowed the team to modify questions or the format of the interview for subsequent interviews.

The use of double translation for a portion of the interviews was another way to cross check data. Double translation involved translating Ilonggo to English on site during the interview and then again during transcription. Double translation allowed the team to check the accuracy of translation. In addition to double translation, each typed transcript was reviewed by PG and ME for accuracy in translating English and Ilonggo, and transcribing Ilonggo. Accuracy in transcribing English was also checked by the PI. The data were cross checked again during coding. The Filipino nurses were consulted on the use of labels to categorize the data to ensure that labels and categories made sense with respect to the data.

Rigor in this study was demonstrated by paying close attention to the study the design and conceptual framework and using these to guide data collection and analysis. Purposeful sampling techniques and content analysis are appropriate methods used in qualitative descriptive studies (Crabtree & Miller, 1999; Giorgi, 1992; Kuzel, 1992; Miller & Crabtree, 1999; Morse, 2000; Patton, 1990; Sandelowski, 2000). The social marketing process and explanatory models

provided the framework to develop the interview guide and to code and analyze the data.

Summary of Methods

This was a descriptive focused ethnography. Formative research from social marketing and explanatory models were used to guide the development of the interview schedule, data collection, and data analysis. Stratified purposeful sampling method was used for participant recruitment. Three groups of working class people were targeted for recruitment and included: individuals with a cleft or child with cleft, individuals who did not have cleft or a child with cleft, and local healthcare workers. The data was analyzed by content analysis. Trustworthiness was established by the long term relationship the PI has had with the research team members and through her past experiences conducting research in the Philippines. Descriptive validity (an accurate description of events) was established through investigator triangulation (team approach) and cross checking.

CHAPTER 4: RESULTS

Chapter four contains the results from this study. First, observational data used to describe the barangay health office (BHO) is presented. The BHO was described to provide context, because most of the interviews took place at a BHO. When observations explicitly pertain to a specific aim, the observational data are described with other data related to the specific aim. Second, the sample is described. Third, data for each specific aim are presented first in general and then by differences and similarities across and within groups.

Barangay Health Office

The four barangay health offices (BHO) where we spent time had the same general structure as the one pictured in figure 3. There were typically two rooms, a general waiting/intake room and a procedure room. The building on the far right of the photograph is the health office. Usually there was a basketball court on the grounds of the health office and a stage area, such as the one on the far left in the photo. The stage area was used for health teaching seminars and general community meetings or performances. The barangay health office could be used as a community center, a place for people to gather. Each BHO we visited was staffed by one or two certified midwives.

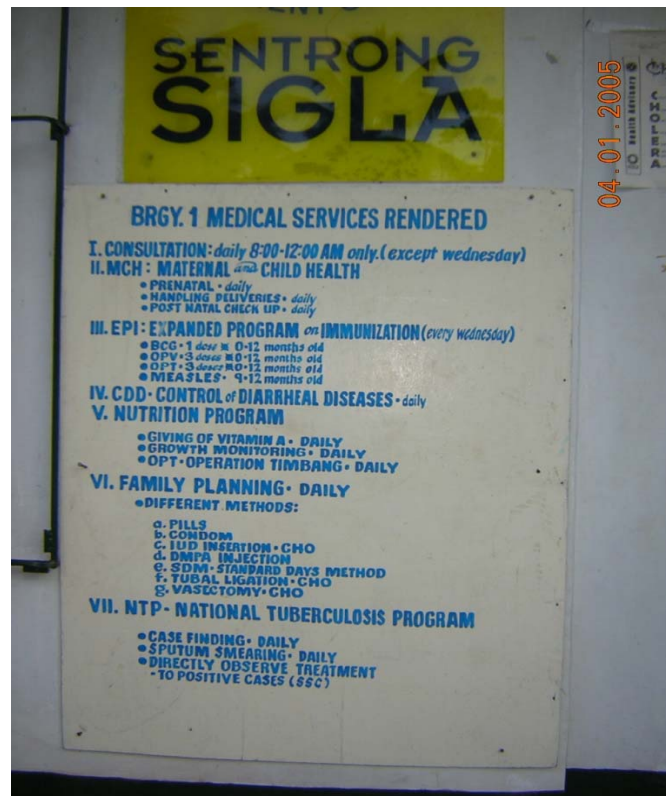
Figure 3. Barangay health office in Patag, Silay City, Negros Occidental.



Midwives are paid employees of the city/municipality and/or barangay. The midwives supervised the activities and other health workers at the health center. CMWs have a formal, two-year, post high school training program that leads to certification. The CMW is trained primarily in maternal child care; they provide prenatal care and deliver babies. They are also trained to administer immunizations and first aid. They prescribe home remedies, analgesics, and antibiotics and refer patients to the city health centers or hospitals as needed.

The CMW also supervises barangay health workers (BHWs) and barangay nutrition scholars (BNSs). The number of BHWs in a given health office depends on the number of puroks the BHO serves and funding. Some BHWs and BNSs receive small stipends, others volunteer. BHWs may receive training in preparing food safely, in hygiene and sanitation, in taking vital signs and weights, and in assisting with procedures and record keeping. They supervise patients taking oral medication, deliver medications to peoples' homes, and disseminate health information by word of mouth or house to house. The BHWs were assigned to certain puroks (neighborhoods) within the barangay. Responsibilities of the BHW include monitoring the health of the people in her puroks taking note of cases of sickness, pregnancy, and malnourished children; and encouraging people to go to the health office for care. She also informs people of new programs or seminars offered at the health office. She connects the people to the health office. All but one of the BHOs we visited had one or more BHWs. In addition, the health office may have a barangay nutrition scholar (BNS). BNSs primarily provide nutritional counseling and teaching especially for the malnutrition programs. She could also assist with other activities at the request of the midwife. There is no standardized training program for BHW or BNS. They receive training through seminars and both act as assistants to the CMW, nurse, or doctor.

Figure 4. Weekly schedule of medical services rendered at the barangay health office.



Schedules of weekly rendered services and informational posters were located outside the BHO near the front door (Figure 4). Typical services included: consultation day (described in chapter 3), immunizations, diarrhea re-hydration, prenatal services, nutrition program, family planning, and a tuberculosis program. Informational posters typically hung outside the health office or in the waiting/intake room. Many of the BHOs displayed posters for mixing hydration solutions, immunization schedules, hand washing, nutrition

guidelines, dog bites and rabies, and how to brush your teeth. Chalkboards or posters displayed outside the clinic announced new programs and seminars.

Informants

Twenty-two healthcare workers were interviewed in 11 interviews. As described above we interviewed three groups of healthcare workers and eight healthcare workers individually for a total of 11 interviews. One of the groups consisted of nine healthcare workers the other two groups had three and two members respectively. All healthcare workers were female; 13 were from rural barangays and 9 from urban barangays (Table 4).

Table 4. Sample stratified by gender, setting and group.

Groups Characteristics <i>n</i> (%)	HCW*(<i>n</i> = 22)	Noncleft (<i>n</i> = 31)	Cleft (<i>n</i> = 27)	Total N = 80
Rural male	0	1 (3)	2 (7)	3 (4)
Urban male	0	7 (23)	5 (19)	12 (15)
Rural female	13 (59)	13 (42)	8 (30)	34 (43)
Urban female	9 (41)	10 (32)	12 (44)	31 (39)

*HCW=Healthcare worker

We interviewed 58 non healthcare people. Thirty-one people from the noncleft group participated in interviews, 8 men and 23 women. Twenty-seven

people from the cleft group took part in an interview, 7 men and 20 women (Table 4).

Healthcare workers

Healthcare workers were the oldest age group of the three with an average age of 39 years. They were also the most educated with 63% having some post high school education and everyone having at least some high school education. Despite the somewhat higher education level, most healthcare workers' (77%) combined family income was still less than P6,000/month (US\$120.00). Twelve barangay health workers (BHW), three barangay nutrition scholars (BNS), six certified midwives (CMW), and one nurse took part in an interview (Table 5). Four of the five rural CMW were part of the large group interview in Himamaylan. This was a large barangay with two full-time CMW and two volunteer CMW. The two volunteers were apprenticing.

Seventy-three percent of the women in the healthcare worker group were married. This was the most diverse group in terms of religion with 64% Catholic and the remaining some other religion. Aside from age and number of CMW in rural compared to urban health centers, the rural and urban health care groups were similar in terms of demographics (Table 5).

Table 5. Demographics for healthcare workers stratified by setting.

Demographics	Urban (<i>n</i> = 9)	Rural (<i>n</i> = 13)	Combined (<i>n</i> = 22)
<u>Age</u>			
Mean	43	35	39
Range	28-53	21-50	21-53
<u>Education</u> <i>n</i> (%)			
Some high school	2 (22)	2 (15)	4 (18)
High school graduate	1 (11)	3 (23)	4 (18)
Some college	2 (22)	4 (31)	6 (27)
College graduate	4 (44)	4 (31)	8 (36)
<u>Income</u> <i>n</i> (%)			
<6,000P*	6 (67)	11 (85)	17 (77)
6,000-10,000P	1 (11)	1 (8)	2 (9)
11,000-20,000P	2 (22)	1 (8)	3 (14)
<u>Occupation</u> <i>n</i> (%)			
BHW	6 (67)	6 (46)	12 (55)
BNS	1 (11)	2 (15)	3 (14)
CMW	1 (11)	5 (38)	6 (27)
Nurse	1 (11)	0	1 (5)
<u>Civil Status</u> <i>n</i> (%)			
Married	6 (67)	10 (77)	16 (73)
Single	1 (11)	2 (15)	3 (14)
Separated	1 (11)	0	1 (5)
Widow	1 (11)	0	1 (5)
No data	0	1 (8)	1 (5)
<u>Religion</u> <i>n</i> (%)**			
Catholic	6 (67)	8 (62)	14 (64)
Baptist	0	2 (15)	2 (9)
Born Again Christian	1 (11)	1 (8)	2 (9)
Mormon	1 (11)	0	1 (5)
Protestant	1 (11)	0	1 (5)
Islam	0	1 (8)	1 (5)
Jehovah Witness	0	1 (8)	1 (5)

*P=peso; US\$1=P50, April, 2005. ** Participants self-identified their religious affiliation.

Noncleft Group

Tables six and seven summarize demographics for the noncleft group.

This was the youngest group. We interviewed 31 people in this group, 23 females and 8 males. Ten women from urban areas and 13 from rural areas participated.

Table 6. Demographics for noncleft female group stratified by setting.

Demographics	Urban (<i>n</i> =10)	Rural (<i>n</i> = 13)	Combined (<i>n</i> = 23)
<u>Age</u>			
Mean	31	28	29
Range	18-50	20-35	18-50
<u>Education</u> <i>n</i> (%)			
Elementary	2 (20)	2 (15)	4 (17)
Some high school	1 (10)	2 (15)	3 (13)
High school graduate	3 (30)	5 (38)	8 (35)
Some college	2 (20)	1 (8)	3 (13)
Vocational	0	1 (8)	1 (4)
College graduate	2 (20)	2 (15)	4 (17)
<u>Income</u> <i>n</i> (%)			
<6,000P*	8 (80)	13 (100)	21 (91)
6,000-10,000P	1 (10)	0	1 (4)
11,000-20,000P	1 (10)	0	1 (4)
<u>Occupation</u> <i>n</i> (%)			
Service	2 (20)	1 (8)	3 (13)
Domestic	7 (70)	9 (69)	16 (70)
Professional	1 (10)	1 (8)	2 (9)
None	0	2 (15)	2 (9)
<u>Civil Status</u> <i>n</i> (%)			
Married	7 (70)	9 (69)	16 (70)
Common Law	1 (10)	2 (15)	3 (13)
Single	2 (20)	2 (15)	4 (17)
<u>Religion**</u> <i>n</i> (%)			
Catholic	9 (90)	13 (100)	22 (96)
Born again Christian	1 (10)	0	1 (4)

*P=peso; US\$1=P50, April, 2005. ** Participants self-identified their religious affiliation

Unlike the health workers, women in the noncleft group were more evenly distributed in terms of education levels. Thirty percent had less than a high school education, 35% had a high school education and 34% had some post high school education (Table 6).

Again despite education level, 91% of the women lived in families whose monthly income was less than P6,000. Eighty-three percent worked in the service or domestic sectors mainly as housewives, house keepers, or street vendors. There were only two professionals, one teacher from an urban barangay and one newly trained health worker from a rural barangay. Most of the women were married and Catholic. Women from the urban and rural groups were similar in terms of demographics (Table 6).

The demographic characteristics for the eight men in the noncleft group are summarized in table 7. Of the males in this group only one came from a rural barangay. He was a sari-sari store owner. Sari-sari stores are small road side convenience stores selling individually wrapped and packaged snacks, beverages, and cigarettes. Of the eight men, four had a post high school education, two had a high school education, and two had less than a high school diploma. They had the following jobs: vendor, fishermen, accountant, engineer, custodian, and sales clerk. One person was out of work. All the men were Catholic and most were married (Table 7).

Table 7. Demographics for noncleft male group stratified by setting.

Demographics	Urban (<i>n</i> = 7)	Rural (<i>n</i> = 1)	Combined (<i>n</i> = 8)
<u>Age</u>			
Average	29	50	32
Range	22-40	N/A	22-50
<u>Education</u> <i>n</i> (%)			
Some high school	2 (29)	0	2 (25)
High school graduate	1 (14)	1 (100)	2 (25)
Some college	1 (14)	0	1 (13)
Vocational	1 (14)	0	1 (13)
College graduate	2 (29)	0	2 (25)
<u>Income</u> <i>n</i> (%)			
<6,000P*	6 (86)	1 (100)	7 (88)
11,000-20,000P	1 (14)	0	1 (13)
<u>Occupation</u> <i>n</i> (%)			
Service	2 (29)	1 (100)	3 (38)
Agriculture	2 (29)	0	2 (25)
Professional	2 (29)	0	2 (25)
None	1 (14)	0	1 (13)
<u>Civil Status</u> <i>n</i> (%)			
Married	5 (71)	0	5 (63)
Common Law	1 (14)	1 (100)	2 (25)
Single	1 (14)	0	1 (13)
<u>Religion</u> ** <i>n</i> (%)			
Catholic	7 (100)	1 (100)	8 (100)

*P=peso; US\$1=P50, April, 2005. ** Participants self-identified their religious affiliation

Cleft Group

The female and male cleft groups were similar in sizes to the noncleft groups. There were 7 men and 20 women. Unlike the women in the noncleft group, there were more women from urban barangays than rural. Table 8 summarizes demographics for women in the cleft group.

Table 8. Demographics for cleft female group stratified by setting.

Demographics	Urban (<i>n</i> = 12)	Rural (<i>n</i> = 8)	Combined (<i>n</i> = 20)
<u>Age</u>			
Average	33	33	33
Range	22-45	23-41	22-45
<u>Education <i>n</i> (%)</u>			
Elementary	2 (17)	3 (38)	5 (25)
Some high school	2 (17)	1 (13)	3 (15)
High school graduate	2 (17)	3 (38)	5 (25)
Some college	1 (8)	1 (13)	2 (10)
Vocational	3 (25)	0	3 (15)
College graduate	2 (17)	0	2 (10)
<u>Income <i>n</i> (%)</u>			
<6,000P*	10 (83)	8 (100)	18 (90)
6,000-10,000P	1 (8)	0	1 (5)
11,000-20,000P	1 (8)	0	1 (5)
<u>Occupation <i>n</i> (%)</u>			
Service	6 (50)	0	6 (30)
Domestic	5 (42)	5 (63)	10 (50)
Agriculture	0	3 (38)	3 (15)
Professional	1 (8)	0	1 (5)
<u>Civil Status <i>n</i> (%)</u>			
Married	10 (83)	8 (100)	18 (90)
Common Law	2 (17)	0	2 (10)
<u>Religion** <i>n</i> (%)</u>			
Catholic	11 (92)	6 (75)	17 (85)
Agilipay	1 (8)	0	1 (5)
Baptist	0	1 (13)	1 (5)
Mormon	0	1 (13)	1 (5)

*P=peso; US\$1=P50, April, 2005. ** Participants self-identified their religious affiliation

Like the noncleft women, education levels for women in the cleft group were distributed evenly with 40% having less than a high school education, 25% having high school educations, and 35% having some high school education. Unlike the noncleft group, there were fewer women in the cleft group with a high school education. Despite education, 90% of women in the cleft group were from families earning less than P6000/month.

Most of the women said they were housekeepers or housewives. There were three women working as farm hands, six in the service sector, and one professional. Similar to the other two groups, most of these women were married and Catholic. One woman was Agilipay which is a mix of Catholicism and an indigenous religion.

Table 9. Demographics for cleft male group stratified by setting.

Demographics	Urban (<i>n</i> = 5)	Rural (<i>n</i> = 2)	Combined (<i>n</i> = 7)
<u>Age</u>			
Average	33	40	35
Range	28-41	36-43	28-41
<u>Education</u> <i>n</i> (%)			
Elementary	0	2 (100)	2 (29)
High school graduate	3 (60)	0	3 (43)
Vocational	1 (20)	0	1 (14)
College graduate	1 (20)	0	1 (14)
<u>Income</u> <i>n</i> (%)			
<6,000P*	1 (20)	2 (100)	3 (43)
6,000-10,000P	3 (60)	0	3 (43)
11,000-20,000P	1 (20)	0	1 (14)
<u>Occupation</u> <i>n</i> (%)			
Service	2 (40)	2 (100)	4 (57)
Labor	1 (20)	0	1 (14)
None	2 (40)	0	2 (29)
<u>Civil Status</u> <i>n</i> (%)			
Married	2 (40)	2 (100)	4 (57)
Common Law	1 (20)	0	1 (14)
Single	2 (40)	0	2 (29)
<u>Religion</u> ** <i>n</i> (%)			
Catholic	4 (80)	2 (100)	6 (86)
Born again Christian	1 (20)	0	1 (14)

*P=peso; US\$1=P50, April, 2005. ** Participants self-identified their religious affiliation

Of the seven men who were part of the study two had a cleft lip and cleft palate and five were partners of women we interviewed. Income for this group of men was reported to be a little higher although their wives/partner reported lower family income. Regardless of occupation, all but one man the reported monthly incomes of less than P10,000 (US\$200.00). These men worked in the service sector, in labor, or were un-employed. Six were catholic, most had partners, and only the two affected males were single (Table 9).

Aim 1 Content Analysis

Each interview was coded for broad topical categories and descriptive sub-codes according to four elements representing explanatory models: (a) label (the name people give to orofacial cleft), (b) etiology, (c) how the cleft forms (timing during pregnancy), and (d) treatment and prevention. Each element will be described first in general, and then differences and similarities across and within groups will be presented. Interview data from healthcare workers were gathered through individual and group interviews. Their data are presented as one group because the categories of responses for cause, prevention, and treatment of orofacial clefting were similar.

Label

Interviews for cleft and noncleft participants began by showing a large photograph of a child with a cleft lip and palate. Universally cleft is called sungi among Ilonggo speakers on Negros Occidental. Sungi is used regardless of subgroup: cleft, noncleft, or health worker. The health workers referred to cleft lip as sungi or cleft and were not always asked to label the picture because the word sungi came up in conversation before they were asked to name to the defect.

Only four people offered another name for cleft lip in another dialect. In Cebuano and Tagalog dialects cleft lip is called bunggi. Only one person interviewed was a native Cebuano speaker. In addition to labeling the cleft lip as bunggi, she referred to cleft lip as bunggi throughout the interview. We did not ask people specifically to label a cleft palate because most people did not notice that the child in the picture had a cleft palate, three people did label the cleft palate as punga the Ilonggo word for cleft palate. One person labeled the cleft lip as sambil, also an Ilonggo word, meaning swelling and in this case swelling of the lips.

Etiology and How Cleft Occurs

Participants named seventeen categories of cause of cleft. The three most common causes were inheritance (33 responses), force to the fetal face (29

responses) and craving (12 responses). Each of these categories was described by participants in more detail.

Inheritance

Inheritance (heredity) was given as a cause for clefting especially when there was a clear family history of orofacial cleft. When asked how inheritance works, people did not talk about genes but often noted other family relatives who had a cleft. They also explained inheritance as something in the blood, “it came through the blood” or “it was present in a distant relative” and showed up now after a few generations where no one had a cleft. Vertical inheritance was recognized but affected sibling pairs were not always recognized as inheritance:

Interviewer (I): In your opinion, what do you believe ... why is it that this defect is appearing, what do you believe in?

First Client (C1): Inheritance. For me it is in the blood of...in the blood that flows.

I: When you were pregnant with your son were you worried that he will be cleft too?

C2: No.

I: You were not worried.

C2: No because we don't have it [cleft] in our family, just my daughter. I was just shocked when I saw her.

C3: Because on my father's side, my father's uncle, which is already my grandfather, he is a cleft.

I: Your dad's uncle or grandfather, so he is your uncle?

C3: No. It's my dad's uncle so he is already my grandfather.

I: Ah yes, it's your grandfather. Paternal side.

C3: Then my male cousin, paternal side, he is also a cleft.

I: Your cousin paternal side is also a cleft.

C3: Yes.

I: So, what do you call it?

C3: Inheritance.

I: Can you make me understand what does inheritance means?

C4: It came from their side, from their uncles, or grandparents, something like that. The sibling of their mother has a cleft.

I: What if it's normal...I mean for example I have a grandfather who is a cleft then I gave birth to a cleft child. If my grandfather is normal do you think I will have a cleft child?

C4: I don't think so.

C5: I don't believe in that [falling] because it is really in the blood. It's within the blood of a family that causes cleft.

I: Ah, so it's within the blood, do you have a term for that, what do you call that?

C5: It's like there is an inheritance of that.

I: So you mean to say it is inheritance? For example, my grandfather is cleft, and then I also have a child with cleft. Do you think I would have a child with cleft if my grandfather is not cleft?

C5: No.

I: Why?

C5: Because you don't have it in your blood line.

C6: Because my father told me that they have ancestors who were clefts. Then it never appeared on [my father's] children's generation.

I: From the great, great grandfather?

C6: Yeah, we are already the great, great grandchildren. Like that. Then he told me that I was the one who got the cleft, like that.

C7: We discussed this, our family; we were trying to discover what caused the cleft of my nephew. And we were able to trace that to the grandfather of the father, of my brother-in-law, who was a cleft also. And then you know what, there's a funny side of this because way back in 1984 I had a boyfriend who was... whose mother was cross-eyed. And then we visited his place, and then I was so shocked to see the mother was cross-eyed. And then, oh anyway I'm not going... I was telling myself anyway I'm not going to... I'm not going to marry the mother but the man. And then we get into their house and then there was I think the soap opera was Flor de Luna. My best friend was my companion, and she was already singing "Flor de Luna" like that and then my boyfriend called his niece and the

girl came to us... the girl was about eight or seven something and then he gave the pasalubong (gift you give when you are a visitor) and I was so shocked that the girl was also cross-eyed. So three months later...I broke up with the man.

C8: There is no study made wherein cleft will occur on that specific generation. There are some instances where cleft comes out if not on this generation; it might come out two generations from now.

I: For example, your uncle or your grandfather is not cleft. Do you think your two kids will be born with cleft?

C9: Because that's what they say years back [my ancestors had cleft].

I: If they were all born normal, do you think you will have cleft children?

C9: No.

I: No, why?

C9: Because they are normal so it [cannot be passed on].

One health worker described inheritance in terms of consanguinity saying she knew of a family who has cleft because the members are “related, belong to one family.” In this case the mother of the cleft child married her uncle.

Force to the fetal face

Force to the fetal face was explained as the result of fetal positioning and force or pressure applied to the pregnant abdomen. People believe that in the fetal position the fetus brings his or her thumbs or fingers to the face near the mouth or places digits inside the mouth, often referred to as thumb sucking. A sudden movement by the mother, such as slipping, falling, or being startled, causes the fetus to tear the face open. In the case of cleft of the lip only, the thumbs or fingers lie outside the mouth and are pushed into the oral cavity causing a cleft lip

and sometimes all the way through to tear the palate too. If the palate is also cleft, the fetus could have had the thumbs or fingers inside the mouth and pulled them through the palate and lip. A bilateral cleft lip is caused by two digits in or near the mouth while a unilateral cleft is caused by a single digit. The face must be formed then broken open. People believed the baby's face was formed some time later in the pregnancy, usually after the fifth month with around seven months being the most common response:

I: Why do you believe that the fall caused cleft? Did your sibling slip?

C1: Yes.

I: So why, why, did you think that was the reason for the cleft?

C1: Because she suddenly fell down.

I: So what do you think happens inside the womb to cause the cleft?

C1: I think because while the baby is inside the womb, it was already thumb sucking.

I: The hands are like this? (Thumb sucking position)

C1: Yes.

I: They thumb suck.

C1: Probably when the mother suddenly fell down it caused the tearing.

C2: If you will ask me Ma'am, as far as I know, the baby is fully developed in 5 months. So if it's only one month [into the pregnancy] falling won't affect the baby.

I: What if for example, because you said that the baby is fully, fully develop in 5 months, how about if I slipped during my 3rd month?

C2: It won't be affected.

I: Are there any other reason that you have heard people give for the cause of cleft? I've heard slipping, are there other reasons?

C3: None, just slipping.

I: Slipping. Do moms really fall that much here? I just hear this so much.

C3: There is a story behind the slipping, each has its own story. Yes.

There are times during the course of their pregnancy that they slipped or stumble. What they usually say is, based on their experience if the child has cleft lip; the baby is positioned with the fingers on the lip.

I: Because the fingers are placed right here (I put my fingers on the outside of my face)?

C3: Yes.

I: On the outside?

C3: On the outside.

I: Okay.

C3: Then the damage goes through the palate.

I: Oh, they stick their thumb in there.

C3: Yes, inside.

I: Ah, if [the cleft] is on one side, it's just one finger and if it's both sides they are like this (holding two fingers up to the mouth for bilateral cleft)?

C3: Yes.

I: Alright then.

C3: That causes the deformity of the lip.

I: What do you think cause other children to have a cleft?

C4: When a pregnant mother slipped.

I: How does slipping cause a pregnant mother to have a cleft child?

C4: Maybe because the baby inside the womb is thumb-sucking. For example, in my case maybe when I was a baby inside the womb of my mother, I was sucking my two thumbs that is why I have two slits in my lip. While in the case of my brother, he used only one finger for thumb-sucking that is why he only had one slit on his lip.

I: Why is it that your palate also breaks?

C4: It is because the palate is so soft, that is why when the thumb pushes through the lip and breaks the palate.

I: So, for example, I am pregnant then I slipped during 7-8 months, my baby will have cleft, what if I slipped around three to four months, will my baby be cleft?

C5: Not yet.

I: Not yet, why?

C5: Because he is not yet...Because he is not yet formed.

I: You said that if a woman is 7-8 months pregnant then she slips her baby will be cleft. What if she is 2-3 months pregnant then she slipped, why did you say that the baby won't be born as a cleft?

C6: Because during those early months the baby is not that formed yet. The baby is [completely] formed during the 7-9 month of pregnancy.

I: How does slipping cause the cleft?

C6: If you slipped the baby will be affected because the baby's position is like this.

I: What about the position of the baby? Describe it for me.

C6: The baby is positioned like this (showing us thumb sucking position)

I: Yes.

C6: If the fingers experienced impact it will break the skin.

I: Will it reach the palate area?

C6: Yes.

I: So the baby is already formed and slipping can cause the thumb to break the face open?

C6: Yes.

Slipping and falling was the most common type of force to the fetal face.

Similarly, attempted abortions and pressure on the mother's abdomen also put force on the fetal face causing it to tear open. Participants from both the cleft and noncleft group offered attempted abortion as a cause of cleft. However, no one from the cleft group stated that an attempted abortion caused their child's cleft. Attempted abortions were described as those that use catheter, abdominal massage, or medications. Abortion methods cause force to the face either by direct contact (a catheter) to the fetal face or by a perceived shaking of the uterus induced by medication such as ipecac that causes cramping and vomiting. Folk medicines used to induce abortions are sold at the open market and induce severe abdominal cramping and vomiting. Abortions are also thought to be induced by a vigorous massage usually performed by a traditional midwife (hilot).

In addition to falling, pressure to the mother's abdomen was believed to cause cleft. This kind of pressure is not as sudden as with falling or being startled. Pressure to the abdomen presses the baby's thumbs or fingers into the

face. This force seemed to come from a more constant type of pressure causing the face to deform rather than tear open:

C1: Others say that it [cleft] is because of medicine that they take.

Interviewer (I): What medicines?

C1: I don't know. That is what they say. Whatever form they will use to abort the baby.

I: Abortion pills.

C1: Yes, that is what they say. I did not take medicine when I was pregnant with my daughter.

C2: Sometimes pregnant women take medicines.

I: What kind of medicines do you think?

C2: Medicine for abortion...If it is not successful it affects the baby.

C3: And then the use of the catheter.

I: Used for abortions?

C3: Yes, for abortions, if the abortion attempt fails the baby could have cleft.

I: What do you think are the causes or reasons why we have cleft children?

C4: When I was pregnant with her, I can't, I went to the hilot and had my tummy massaged.

I: Hilot, was it for prenatal or abortion?

C4: Abortion.

I: How many months?

C4: I think around three.

I: Three months?

C4: Oo (yes), three to four.

I: You attempted to...?

C4: Abort.

C5: Yes, she was overworked, it was too much and it also depends on how the baby is positioned [thumb sucking position]. Especially if she just works and works, always sitting because she is washing clothes everyday, of course the baby will be cleft.

I: Why... so while the mom is working or washing clothes, the baby is positioned like this (thumb sucking position); what do you think is happening?

C5: Yes, because the baby is being squeezed in and the mom would suddenly sit.

I: So the baby is being squeezed?

C5: Yes, it's uncontrollable. Maybe it's because of that.

C6: Sometimes if you are very tired, you are not aware that your weight is on the child. So, they get stuck beneath you without you knowing it. Yes, they become cleft because you lie on their face while you are on your abdomen.

I: What do you think is the cause of your child's cleft?

C7: Prone position.

I: What, prone positioning?

C7: He was in a prone position inside the womb, then when he was born he was positioned like this, his fingers are here (thumb sucking). He was positioning like this (thumb sucking) inside the womb and his fingers are inside that is why his palate was affected too.

The woman, who talked about her attempted abortion when she was pregnant with her cleft child, went on to say her child's cleft was caused by the chickenpox she had during that pregnancy. She further noted that if she did not have chickenpox then perhaps the attempted abortion could have caused the cleft.

Cravings

The third most common reason for clefting was craving. In this response category, people described that their babies took on features or phenomenon of the environment during a specific time in pregnancy. Participants said a baby could have a cleft if the mother saw a person with a cleft or if the mother or father teased a person with a cleft during the period of craving. The period of craving is the first three months (first trimester) of pregnancy. One reason to categorize

seeing and/or teasing, and reprimanding a person with cleft as craving is that people said the cleft could only happen if seeing, teasing, and reprimanding occurred during the period of craving. Another reason these were categorized as craving is that people described them as a craving. People described seeing a person with cleft as “liking to look at the cleft child,” “falling fond of cleft,” and dwelling on the image of a cleft child. One woman told us that if a person reprimands a child with cleft, the image stays on their mind and then their child will have a cleft. Another woman said cravings caused her child to have a cleft; when we asked what she craved, she reported that her husband likes to tease a child with cleft. In the case presented in chapter 2, the mother said she craved a food (tambis) that looks like a cleft. These cravings or not being able to get the image or desire off one’s mind, are transferred to the fetus. One other reported craving explanation for cleft was if cravings were denied during the period of craving, the child could suffer consequences such as cleft:

Client 1 (C1): We do not have cleft inheritance, but my younger brother has a cleft child because his wife likes to look at, likes to look at the cleft child.

Interviewer (I): Yes, how many months?

C1: Around three months, three months cravings.

I: Three months cravings.

C1: But she really hated cleft. That is why she gave birth to a cleft child.

C2: If you are on your first trimester of pregnancy, do not reprimand a child who has a defect because it has side effect.

C3: That when a woman conceives and falls fond of cleft, the tendency of her baby/child is to be that [cleft]. There were two occasions associated to

my late father who had lost his right middle finger. Two pregnant women laughed at his hand with the missing finger and both had boys with deformed right middle hands. One, [missing] portion of the finger tip which is the nail; the other one has short fingers, also on the right hand.

C4: Like when I went to Mabinay, I saw a child, a child that looks like my son's defect.

I: What kind of a child?

C4: Female. Then I rode in the Ceres Bus, when I rode the Ceres I saw that child again. I was wondering, what was that? I have not seen that type of cleft she had.

I: But is she cleft?

C4: Yes, cleft just like this one (in the picture). I was only one month pregnant.

I: Okay, then?

C4: Yes, then I continued thinking in the corner of my mind, about that cleft. It really stuck on my mind. When I arrived in our house, I told my husband, "I saw a cleft, it looks really bad".

C4: Because this one (pointing to her lip) it has a slit.

I: Where you pregnant then with your son who has a cleft?

C4: Yes.

I: When you were pregnant with your son that was the time that you saw that cleft child?

C4: Yes, that time. Then I always thought about that and when my baby was born, he looked like that child.

I: So you thought that, that was the reason he was born cleft?

C4: Yes.

I: What do you think is the cause of a baby's cleft? What is your belief on why your child is cleft, or why other people's children have a cleft?

C5: According to the oldies it is due to cravings. Like if you always like to look at the cleft child or tease him it will have a deeper effect on your pregnancy.

I: Explain what you mean when you say cravings?

C5: When you crave for something you will give birth to a baby that has the same physical appearance.

C5: I don't believe in other reasons; what I believe in is if you always reprimand a child who has a defect, then it will stick in your mind and eventually your baby will look like that child.

I: What do you think causes people to have a cleft?

C6: Cravings, when I was pregnant, cravings during pregnancy.

I: Did you crave something in particular?

C6: My, my husband always ran after the cleft child. He would always run after him...

I: And tease him?

C6: Yes.

I: Is that what you think causes cleft?

C6: Yes.

I: Is that true for everyone who has a cleft?

C6: That is what I believe in.

I: Like for example, if your husband will tease a cleft child, when you are about 2-3 months pregnant do you think it can make your child have a cleft?

C6: Maybe because that is the stage when you are still craving.

I: So, it could be any of those things, it could be something she saw or something she craves, or a husband was teasing. If you, what about like in the 8th or 9th month and you...if somebody was teasing or if you crave a certain food. Would the baby still be born with a cleft?

C6: No because the craving time is finished.

I: Even if your husband teased somebody in the last months of pregnancy could the baby still be born with a cleft?

C6: No, because it [the period of craving] is already finished.

Other causes

Two people thought a combination of factors added together cause cleft, much like the biomedical, multifactorial explanation:

C1: What I have heard from the old folks, the baby inside the womb is possibly floating and moving and that the stomach will be compressed. That is why it causes some abnormalities like immobilized extremities. Or, it could be that mothers are drinking medicines which can cause harm to the baby inside. We don't have enough supplement in our body, this also adds to the damage done by external forces.

C2: They say that maybe one of the reasons for clefting is that when a pregnant mother slips during pregnancy this causes the cleft, plus the fact

that she is also taking antibiotics with a bigger dosage. This is too much for the body and there is a possibility that the baby will be a cleft.

The remaining 14 explanations for cause of cleft were not described with as much detail as force, inheritance, and craving. These include medication taken during pregnancy; lack of vitamins, and in one case the wrong kind of vitamin; God's will; smoking; inborn; poor nutrition; alcohol use during pregnancy; stress; infection; do not know; pollution; the use of birth control pills; chickenpox specifically; and an accident after birth that cut the face. Medication taking during pregnancy was described as medication not approved by a doctor, incompatible with the pregnancy, or the medicine was toxic to the mother and baby and led to clefting. Eclipse was mentioned once as a potential cause of cleft, although not a personal belief of the respondent rather something she heard from a client:

I: Okay, have you heard other explanations or you have heard other people's reasons for why cleft happens?

C1: Maybe, when the mother was pregnant she took medicines that were not prescribed by the doctor.

I: They take medicine that is not recommended by the doctor?

C1: Maybe. There are some mothers who are pregnant and take medicine not prescribed by the doctor

C2: According to my friend, when she was pregnant she got sick and took some medicines, and then her child was born with a cleft. Her child has a cleft because she took medication due to her sickness after a lot of laboratory test. She took a lot of different kinds of medicines that were prescribed to her. She took them because her doctor told her to.

C3: Maybe one of the causes of clefting is the medication that mothers take when they were pregnant, medicines that didn't suit them. Maybe the

low tolerance to the medication is the reason a baby has a cleft. Some people just take medicines even if they feel better. It depends on the doctor; you follow what the doctor tells you because when you get sick you still go to the doctor. Quack doctors can't cure cleft.

C4: Maybe antibiotic that has bigger dosage for the body.

Participants did not describe how lack of vitamins and poor nutrition could cause cleft, but later people were asked how to have a healthy pregnancy and why pregnant women take vitamins. People stated that vitamins and good nutrition are needed for proper development of the baby. Likewise, people did not go into detail about how smoking or alcohol use could affect the baby except to say these affected how the baby formed. One woman read that using certain brands of vitamins, alcohol, and cigarettes could cause defects in babies. She then concluded that cleft could be one of those defects. God's will was given as a cause for cleft several times ($n = 6$):

C1: It [the birth of a child with cleft] is also a blessing of God to me. Then I am thankful that God gave it to me rather than defects like crippled and polio.

I: What do you think causes kids to have cleft?

C2: Sometimes the way they develop.

I: What do you mean by the way they develop?

C2: That they already have the defect.

I: So, why do you think that the moment they were born they are cleft? What do you think is the cause?

C2: Maybe, God gave it to them.

I: That they will be cleft?

C2: Yes.

Others explained that God's will was present when other explanations could not totally explain the cause for cleft. The participant in the first example of God's will below believed her son's cleft was caused by her fixation on a child with cleft when she was on a bus trip. Later, when we asked her if she was worried that her subsequent babies would have cleft she said no, because her other children were not affected. Even though she thought her son's cleft was because of "craving" she also states that God has something to do with the outcome of the pregnancy. Still, other participants felt that cleft was caused by inheritance, but in absence of a positive family history God's will was given as a possible cause:

C1: No. I was not worried when my 5th child was born because my daughters are just okay [they do not have a cleft]. The 6th child is him (the affected son). I thought that it is God's will because I asked for a son. I have 5 females and the 6th child is a male. I was not worried that my 7th child will be affected because my 5 daughters are normal. All I thought was, maybe it is God's will because we asked for a son.

I: Why did you say that it is inheritance?

C2: I have a sibling who also has [cleft].

I: Ah okay, you have a sibling like that. Is it open in the palate area?

C2: No, it's just the lip

I: So is it inborn?

C2: Yes, when he was born.

I: Who else?

C2: My Aunt.

I: Your Aunt?

C2: Yes, one of her children.

I: So, you do believe that it is inheritance?

C2: Yes.

I: Let me see if I have this correct, because you have a positive family history that is why there are kids born with cleft.

C2: Yes.

I: If for example you do not have affected relatives and your son is not cleft, do you think your next child will still be born with cleft lip?

C2: I believe it is a blessing from God.

I: Aside from God's blessing do you think your son will be like that if you do not have affected relatives?

C2: If it is Gods will, it will happen.

I: What if you do not have relatives who have a cleft; do you think you will still have cleft child?

C2: I am not sure, but if it's God's will then he will give you a child with a cleft.

C3: I do not believe in that [falling] because it is really in the blood. It is within the blood of a family that causes cleft.

I: Ah, so it is with in the blood, do you have a term for that, what do you call that?

C3: It is like there is an inheritance of that.

I: Inheritance. So is that what you believe in?

C3: Yes.

I: Yes, how about those mothers who do not have any inheritance traits?

C3: Ah, those who do not have inheritance. If it is God's will that he will give me a disabled child, so be it.

In addition to God's will, other explanations were given when there was no family history. In contrast, some people who believed inheritance caused cleft did not think there was risk for cleft if there was not a family history for it:

I: In your own idea, what causes children to be cleft?

C1: Maybe, because of the medicines that they are using while they were pregnant.

I: What medicines?

C1: Sometimes, it's the birth control pills that cause the abnormalities.

I: Is it also the same with cleft cases?

C1: Yes.

I: What else?

C1: Then, if they have a positive family history of clefting.

C2: We do not have cleft inheritance, but my younger brother has a cleft child because his wife likes to look at, likes to look at the cleft child.

I: Yes, how many months?

C2: Around three months, three months cravings.

I: Three months cravings.

C2: But she really hated cleft. That is why she gave birth to a cleft child.

I: In your own words, why is it that there are kids born with cleft?

C3: I do not know.

I: You do not have any idea?

C3: Maybe it is inheritance. They have a family history.

I: History, what do you mean?

C3: They have family history of cleft.

I: Can you please describe to me what you mean by inheritance?

C3: If their father or other relatives are cleft.

I: What if there's no family history and just this baby was born with a cleft?

C3: It depends, some say it is because they slipped.

I: slipping?

C3: Yes.

When asked if there were any programs or materials available addressing the reduction of risk for birth defects (inborn problems) everyone we talked with said no. People relied on hearsay and/or their own experiences to explain cause for clefting. In the quotes above, one woman said that “We don't have cleft inheritance but my younger brother has a cleft child because his wife likes to look at a cleft child.” She acknowledged that inheritance is an explanation for cleft, but based on her family's experience she said craving (her sister-in-law likes to look at a cleft child) caused cleft.

Across and within group comparison for cause of cleft

First, cause explanations will be compared across the three types of interview groups cleft, noncleft, and healthcare workers. Second, findings will be presented from within type of interview groups and across interview groups on patterns of beliefs about cause of clefting by grouping participants on the following attributes: gender, age, education, setting, and family history.

Table 10 summarizes cause of cleft categories by type of interview group, cleft, noncleft, and healthcare worker. The numbers in the table represent the number of participants who reported a particular cause, except for the healthcare workers. Numbers reported for healthcare workers represent number of interviews because we interviewed individual and groups of healthcare workers. Total response percents were not calculated because of the difference in the unit of analysis for the cleft/noncleft groups versus the healthcare worker groups.

Of the 27 people in the cleft group about half gave discordant explanations for what they thought caused cleft in general and what they thought caused their child's cleft. Of the 27 people in the cleft group, 3 were asked what caused their child's cleft; they were not asked what they thought caused cleft in general. Of the 24 people asked both questions 12 gave concordant explanations and 12 gave discordant explanations. In the concordant group there were eight females and

four males. In the discordant group there were 10 females and 2 males (Table 11).

Table 10. Number of participants reporting cause of cleft by categories and by type of interview group.*

Cause of clefting <i>Characteristics n (%)</i>	Cleft (<i>n</i> = 27)	Noncleft (<i>n</i> = 31)	Health care worker (<i>n</i> = 11)**	Total Responses
Inborn	2 (7)	1 (3)	0	3
God's will	4 (15)	2 (6)	0	6
Chickenpox	1 (4)	0	0	1
Birth control pills	0	1 (3)	0	1
Medications	1 (4)	4 (13)	2 (18)	7
Smoking during pregnancy	0	3 (10)	1 (9)	4
Vitamin (either too much or too little)	1 (4)	1 (3)	4 (36)	6
Stress	0	1 (3)	1 (9)	2
Poor nutrition	1 (4)	1 (3)	1 (9)	3
Accident as an adult	0	1 (3)	0	1
Alcohol use during pregnancy	0	1 (3)	1 (9)	2
Infection	1 (4)	0	0	1
Pollution	1 (4)	0	0	1
Do not know	0	1 (3)	0	1
Cravings	8 (30)	4 (13)	0	12
Force to the fetal face	11 (41)	13 (42)	5 (45)	29
Inheritance	10 (37)	16 (52)	7 (64)	33

* Participants could have multiple responses

** Based on 11 interviews, 3 group interviews and 8 individual interviews

One person believed that in order to prevent cleft, pregnant women should be careful not to fall during pregnancy. However, in her case she did not fall during her pregnancy, but her husband did tease a neighbor child with cleft. She believed teasing led to her own child's cleft. Similarly, participant two believed

that in general cleft was caused by cravings, specifically teasing a person with cleft. She goes on to say, “But in my case I remember falling from a carabao and felt pain.” Falling, she says, caused her daughter to have a cleft.

Table 11. Summary of discordant explanations on cause of cleft among cleft group participants.

Interview	Cause for your child's cleft	Cause of cleft in general
1	cravings, teasing	slipping/falling and cravings
2	slip fall	craving, teasing
5	slip fall	teasing inheritance
6	maternal infection	God's will, inheritance
17	not sure	inheritance or falling
32	inheritance	slip/fall, teasing
35	not sure	craving/tease, slipping/falling and inheritance
36	inheritance/God's will	slip and fall
37	chickenpox inheritance	abortion, slip chickenpox
56	inheritance	cravings and pollution
58	positioning, pressure	inheritance
79	inheritance	positioning, pressure God's will, medication

Another mother (interview 5 in Table 11) talked about a classmate of her child’s who has a cleft. The classmate’s mother says her own child’s cleft was caused by teasing a cleft child while she was pregnant with her daughter. While the mother we interviewed felt this could be true, she did not tease anyone with

cleft during her pregnancy. But she did fall and because she fell, her daughter had cleft.

Another person believed that the birth of a child with cleft is a gift from God, and some things, including cleft, happen just because it is God's will. When asked why she thought her child was born with a cleft, she said it was caused by an infection, tonsillitis, when she was three months pregnant. Three people, two women and one man, felt their child's or their own cleft was due to inheritance, but they recognized that other things may cause cleft in other people. The man (interview 32 in Table 11) said that his mother told him his cleft was because his Lola (grandmother) had a cleft. He said others were cleft because of slipping/falling and teasing. Interestingly, he has a brother who also has a cleft. His brother believed cleft is caused by inheritance. Likewise, one woman believed in general cleft was due to inheritance, but they have no family history of cleft. She recalled that when she was pregnant with her daughter who has cleft, she would let an older sibling, an infant at the time, lie on her stomach. This caused pressure on the baby's face while she was in the "thumb sucking position."

Two people (interviews 17 and 35 in Table 11) were not really sure what caused their child to have cleft. Both women thought the reasons they gave for the cause of cleft could be true, but when they examined their own pregnancies they could not find evidence to support these ideas. For example, the first woman said that while she fell during her pregnancy, and she does have a family history

of cleft, the fall was not hard. She said that the affected relative is distant causing her to doubt that these were causes her child's cleft. The other woman cited craving/teasing, slipping/falling, and inheritance all as possible explanations for cleft, but she did not experience any of those things in her pregnancy. Therefore, she could not say what may have caused her child to have cleft, and this seemed to bother her.

One woman (interview 36 in Table 11) had two children with cleft and believed that inheritance was the cause, but God's will also had something to do with their cleft. She was thankful to God for giving her children with cleft as opposed to children who were crippled or had polio. On the other hand, she worried that her subsequent children may be born with cleft if she slipped and fell.

As presented earlier, the three most common explanations for cause of cleft were inheritance, force to the fetal face, and cravings. In the cleft group the three explanations were reported with about the same frequency. However, the frequency of responses to the follow up question "what actually caused your child's or your cleft?" differed from responses given to the general question of, "what causes cleft?" Inheritance was the most common answer to "what caused your child's or your cleft?" followed by force to the fetal face and cravings (table 12).

Table 12 Causes of your child's or your cleft in the cleft group.*

Cause of cleft for your child <i>Characteristics n (%)</i>	Cleft group (<i>n</i> = 27)
God's will	4 (15)
Chicken pox	1 (4)
Poor prenatal care	1 (4)
Maternal infection	1 (4)
Not really sure	3 (11)
Cravings	5 (19)
Force to the fetal face	7 (26)
Inheritance	15 (56)

* Participants could have multiple responses

In the cleft group 24 of 27 participants reported a family history beyond themselves or their child. Still, not everyone with a family history identified inheritance as a cause of their child's or their cleft.

In the healthcare worker group no one gave cravings as an explanation although they reported that their patients believed cravings could cause cleft. Inheritance and force to the fetal face were the most frequent explanations of cause with this group.

Force to the fetal face had three sub-categories; cleft caused by falls, pressure, or attempted abortion. The healthcare workers did not report attempted abortion as a cause of cleft. Noncleft participants reported attempted abortions as much as falling as a cause of cleft (*n* = 8). In the cleft group only two people talked about attempted abortion as a cause of cleft, but neither of these people said this caused their child's cleft specifically.

Cravings had two subcategories. One was teasing, staring at, or reprimanding a person with cleft during the period of craving in pregnancy. The other was not granting a craving during that time. For example, one woman said cleft could happen because, “women craved for something to eat but then it wasn’t granted.” In general, craving was reported most frequently by participants in the cleft group.

A number of causes reported by participants were categorized as environmental exposures. Chickenpox, birth control pills, medications, smoking, vitamin use (too much or too little), stress, poor nutrition, alcohol use, infection, and pollution were all categorized as environmental exposures. The overall category of environmental exposures as a cause of cleft was created by the researcher and included in the within and across group analysis below. This category was created based on biomedical explanation for cleft and may not be recognized by Filipinos. If environmental exposures are grouped together as one group, this larger category was reported more frequently by the noncleft group and healthcare workers compared to the cleft group. If environmental exposures are grouped into one response category, environmental exposure becomes the third most common cause of cleft reported overall after inheritance and force to the fetal face. Twenty participants/interviews account for the 29 responses in the environmental category (data not shown). One or more environmental exposures were reported as cause of cleft by 5 (19%) participants from the cleft group, 9

(29%) participants from the noncleft group, and in 6 (55%) interviews from the healthcare worker group.

Environmental exposure was the third most common explanation of clefting in the noncleft group followed by inheritance and force to the fetal face. In the healthcare worker group inheritance remained the most common explanation, followed by environmental exposure, then force. In the cleft group environmental exposure becomes the fourth most common explanation; force remains the most common, followed by inheritance, and then cravings.

The explanation of inborn meant that someone had a disease or physical and/or mental problem at birth. There were only three people who said cleft was inborn. They gave no further explanation to what caused cleft; it was just simply there at birth. In order to understand how cleft fit into the category of inborn, we asked people to name other inborn problems. Other reported inborn problems included crippled, deafness, blindness, malnutrition, other deformities, paralysis, mental retardation, polio, epilepsy, Down syndrome, low birth weight, cerebral palsy, and premature birth.

Across and within cleft and noncleft group
comparison for cause of cleft grouped by attributes

The following attributes were used to group responses and look for response patterns gender, age, education, setting, and family history. Attributes

included in this analysis had at least two values per attribute with five or more responses per value in the overall study population. The healthcare worker data were not included in this analysis because that interview set included both group and individual data (Table 13).

Age was collapsed into two categories, young and old. Thirty-three years was the median age of all the participants and was used to divide the participants into the two age categories. The value young referred to participants 18 to 33 years of age, while old referred to participants older than 33 years.

For the attribute of highest level of education attained, the six attribute values elementary, some high school, high school graduate, some college, vocational school, college graduate, and post graduate education were collapsed into three values. These three attribute values were less than a high school education, high school graduate, and more than a high school education. This was done because each of these new attribute values contained about one third of the participants.

The attribute setting had two values rural and urban denoting people's residential setting. For this study rural was defined as those barangays that were ten or more miles away from a clinic and/or hospital staffed by physicians and nurse.

Family history of cleft was considered positive in the cleft group if there were additional family members affected with cleft, excluding the affected child

or participant. Family history was considered positive in the noncleft group if there were any affected relatives. Table 13 provides a summary of attributes and the values used for comparing responses for cause of cleft within and across the cleft and noncleft interview groups.

Table 13. Attribute and values used compare responses for cause of cleft.

Attribute	Attribute value
Gender	Male Female
Age	Young (18-33 years) Old (33-54 years)
Highest level of education	Less than a high school education High school graduate More than a high school education
Setting	Rural Urban
Family history of clefting	Positive Negative

Table 14 summarizes causes of cleft for inheritance, force to the fetal face, environmental factors, and cravings by attribute values for gender, age, education, setting, and family history of clefting for the cleft and noncleft participants. Other responses for cause of cleft were sporadic with too few responses to establish response patterns.

In five attribute categories, female, young, old, urban and negative family history, inheritance was the most frequent cause of cleft reported followed by

force, environment, and cravings. This was the same response pattern observed for all participants interviewed. Males reported force and environment more frequently than inheritance and cravings compared to females who reported inheritance and force most frequently.

Table 14. Number of participant responses for cravings, environmental exposure, force to the fetal face, and inheritance stratified by attributes gender, age, education, setting, and family history of clefting. ($n = 58$)*

Attribute value <i>Characteristics n (%)</i>	Cravings	Environmental exposures	Force to the fetal face	Inheritance
Male	1 (2)	6 (10)	7 (12)	2 (3)
Female	11 (19)	10(17)	19 (33)	29 (50)
Young (18-33 years)	5 (9)	9 (16)	18 (31)	16 (27)
Old (33-54 years)	7 (12)	5 (9)	7 (12)	10 (17)
Less than high school graduate	7 (12)	3 (5)	7 (12)	9 (16)
High school graduate	3 (5)	2 (3)	10 (17)	8 (14)
More than a high school graduate	2 (3)	10 (17)	8 (14)	11 (19)
Rural	8 (14)	4 (7)	9 (16)	12 (21)
Urban	4 (7)	13 (22)	17 (29)	18 (31)
Positive family history	9 (16)	7 (12)	12 (21)	11 (19)
Negative family history	3 (5)	6 (10)	12 (21)	13 (22)

* Participants could have multiple responses

People with less than a high school education had fairly similar response frequencies among inheritance, force, and cravings. People with less than a high school education reported cravings as a cause of cleft more frequently than people

who attained a high school or more education level. They also reported environmental exposures less than people with higher education levels.

People with a high school level education reported force and inheritance as cause of cleft most frequently, with fewer reporting cravings, and environment. People with more than a high school education reported inheritance and environmental exposures most frequently, but force to the fetal face remained a common explanation even by the most educated.

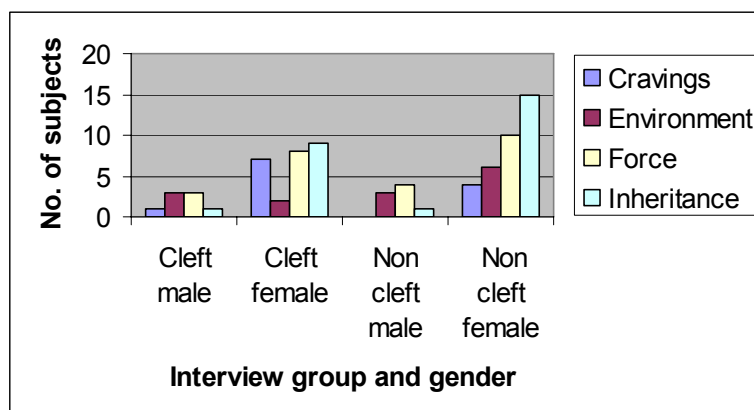
Force and inheritance were the most frequently reported causes of cleft in rural and urban groups. However, the urban group reported environmental exposures more frequently compared to the rural group. In the rural group cravings was reported as a cause of clefting more than environmental exposures and more frequently compared to the urban group. Positive and negative family histories of cleft group response patterns were similar to cleft and noncleft respectively. This was not surprising as most people in the cleft group reported a positive family history and most people in the noncleft group reported a negative family history.

Cause of cleft in the cleft and noncleft group
compared by gender

Comparing males in the cleft group to males in the noncleft group showed the same belief pattern of males in general, force to the fetal face and

environmental exposures were the most common explanations followed by all others sporadically. This is in contrast to the larger cleft and noncleft groups, females in general, and females within their respective interview groups. The response pattern for females shown in figure 5 was similar to the overall response pattern for the interview group they belonged to. The overall cleft group responses to cause were more evenly distributed among inheritance, force, environment, and cravings compared to the noncleft group. Overall, the noncleft group reported cause as force and inheritance most frequently followed by environment then cravings. This was expected as females made up the majority of participants in each interview group (Figure 5).

Figure 5. Number of participants reporting cause of cleft for cravings, environmental exposures, force to the fetal face and inheritance grouped by interview group and gender. ($n = 58$)



Males' most common explanations for cleft were force to the fetal face and environmental exposures regardless of interview group, cleft or noncleft. Females' most common explanations for cleft were inheritance then force to the fetal face. This was true in both the cleft and noncleft groups. However, females in the cleft group explained cause of cleft by cravings more often than environmental exposures while the opposite was true of females in the noncleft group.

For the attributes of age, education, setting and family history, both male and female genders were included. Response patterns without males did not change the patterns. Therefore, the following comparisons were made using both males and females.

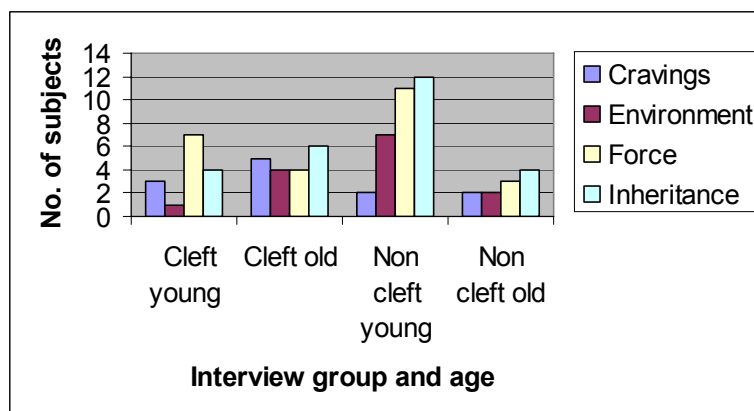
Cause of cleft in the cleft and noncleft group
compared by age

The attribute of age contained four groups, young and old, from cleft and noncleft groups respectively. In general, younger people's most common response to cause of cleft was force, followed closely by inheritance, then environmental exposure, and cravings. Older people showed a more even response pattern among these four causes with inheritance most commonly reported, followed by force, environment, and cravings (Table 14). Both old and

young noncleft groups' response patterns were similar to older people's general response pattern and to the noncleft group in general (Figure 6).

However, in the young, noncleft group, force and inheritance were more common than environment, followed by a drop to two reporting cravings as a cause for cleft (Figure 6). Younger people compared to older people in the noncleft group reported environmental exposures more frequently.

Figure 6. Number of participants reporting cause of cleft for cravings, environmental exposures, force to the fetal face and inheritance grouped by interview group and age. ($n = 58$)



In the cleft group, younger people responded most frequently that force caused cleft, followed by inheritance, and cravings, and only one participant reported environmental exposures as a cause of cleft. Older people in the cleft group reported cravings as cause of cleft most frequently compared to the other

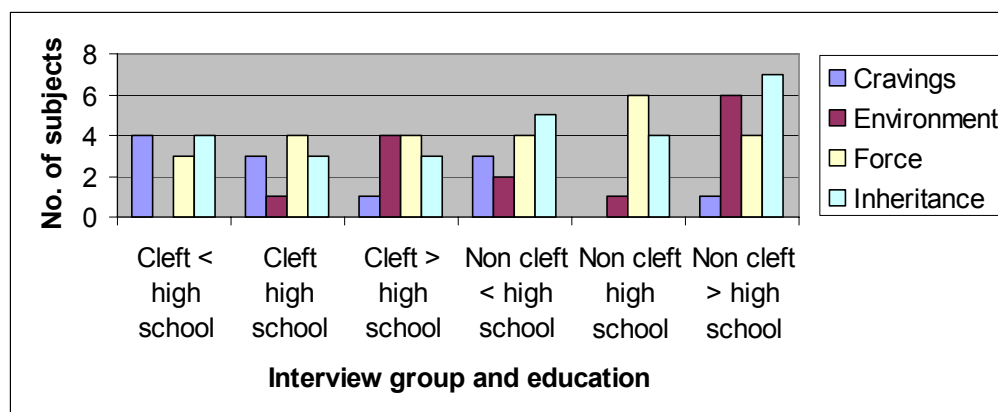
three groups. This group also had more people reporting environmental exposure as a cause of cleft compared to younger people in the cleft group and older people in the noncleft group (Figure 6). This almost even split between cravings and environment in older people in general, and within and across interview groups may reflect education levels and will be discussed later. Older people in the cleft group also accounted for the entire God's will responses found in the cleft group ($n = 4$, data not shown). In general young and old in the cleft and noncleft group explanation patterns followed the cleft and noncleft group as a whole.

Cause of cleft in the cleft and noncleft group
compared by education

Six groups for the attribute of education; those who obtained less than a high school education, just a high school education, and more than a high school education in the cleft and noncleft group respectively were compared (Figure 7). There seemed to be three trends with regard to education and cause of clefting. First, with more education there was an increase in the frequency of environmental exposure reported as cause of cleft. Second, an increase in the frequency of cravings reported as a cause of cleft was observed as education level dropped. Third, regardless of education level inheritance and force remained common explanations for cleft (Figure 7). These trends were observed within and

across interview groups as well as overall education level regardless of interview group (Table 14).

Figure 7. Number of participants reporting cause of cleft for cravings, environmental exposures, force to the fetal face and inheritance grouped by interview group and education level. ($n = 58$)

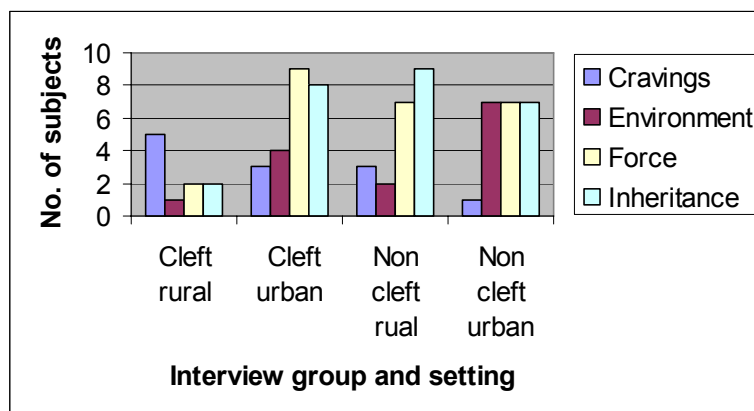


Cause of cleft in the cleft and noncleft group compared by residential setting

In terms of residential setting overall, inheritance and force were the most frequently reported cause for cleft in both rural and urban settings. Cravings were reported more frequently than environmental exposure in the rural areas where as environmental exposure was more common than cravings in the urban setting group (Table 14).

This trend was observed in both the cleft and noncleft group as well (Figure 8). The overall trend of inheritance and force as the most frequently reported cause was observed in cleft and noncleft urban groups, as well as in the rural noncleft group. However, in the urban noncleft group, inheritance, force and environmental exposure were reported with the same frequency while only one person reported cravings. In contrast, craving was the most frequently reported cause in the cleft rural group with other causes reported once or twice (Figure 8).

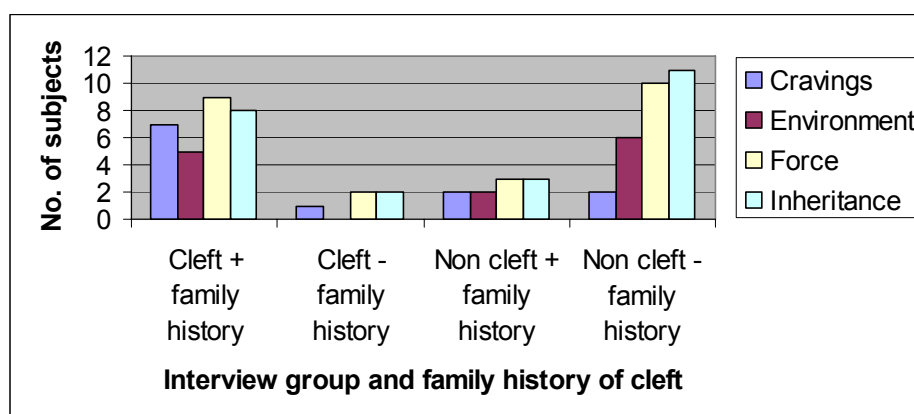
Figure 8. Number of participants reporting cause of cleft for cravings, environmental exposures, force to the fetal face and inheritance grouped by interview group and setting. ($n = 58$)



Cause of cleft in the cleft and noncleft group
compared by family history for cleft

When causes are grouped by family history and interview group, the response pattern follows the over all response pattern of cleft and noncleft group. The positive family history - cleft group reported force most frequently, followed by inheritance, then cravings, and environmental exposure. This is the same pattern observed in the cleft group. In the negative family history - noncleft group force and inheritance were reported in about equal frequencies followed by environmental exposures and craving. This is the same pattern observed for the noncleft group (Figure 9).

Figure 9. Number of participants reporting cause of cleft for cravings, environmental exposures, force to the fetal face and inheritance grouped by interview group and family history of cleft. ($n = 58$)



This is not surprising as 24 of 27 people in the cleft group reported a positive family history and 20 of 31 people in the noncleft group reported a negative family history. The groups of cleft - negative family history, and noncleft - positive family history had relatively flat response patterns because there were few participants with these attributes (Figure 9).

Prevention

Fifteen different categories of cleft prevention were named by participants. The three most frequently reported prevention methods were family planning, taking care not to fall or put pressure on the pregnant abdomen, and taking vitamins during pregnancy. Family planning was the use of some method to prevent the birth of subsequent children. It will be discussed further in a later section.

The numbers in the table 15 represent the number of participants who reported a particular prevention strategy, except for the healthcare workers. Numbers reported for healthcare worker represent number of interviews because we interviewed individual and groups of healthcare workers. Total response percents were not calculated because of the difference in the unit of analysis for the cleft/noncleft groups versus the healthcare worker groups.

The total number of responses in the health care group is less than the 11 interviews we conducted. This was a flaw with the data collection. Sometimes

the interviews with health care began by talking about the vitamin intervention trial because they had heard of the plan before we arrived. We felt this biased their response to questions about what might prevent cleft. Therefore, we did not always ask healthcare workers about prevention.

Table 15. Summary of prevention categories stratified by interview group.

Prevention categories	Cleft* (n = 27)	Noncleft* (n = 31)	Health care person (n = 11)**	Total
<i>Characteristics n (%)</i>				
Get some exercise	0	1 (3)	0	1
More research on cause is needed first	1 (4)	0	0	1
No alcohol during pregnancy	0	1 (3)	0	1
Be careful in general	1 (4)	2 (6)	0	3
No smoking during pregnancy	0	3 (10)	0	3
Healthy diet during pregnancy	0	3 (10)	1 (9)	4
Do not overwork during pregnancy	0	3 (10)	1 (9)	4
Take prescribed medicines only during pregnancy	0	3 (10)	1 (9)	4
There is nothing you can do to prevent cleft	1 (4)	3 (10)	0	4
I do not know	3 (11)	1 (3)	1 (9)	5
Do not mock reprimand or dwell on the image of a cleft person	3 (11)	2 (6)	0	5
Get regular prenatal checks	0	4 (13)	1 (9)	5
Take vitamins during pregnancy	3 (11)	3 (10)	2 (18)	8
Do not slip or fall avoid pressure to the abdomen during pregnancy	4 (15)	7 (23)	1 (9)	12
Family planning	16 (59)	9 (29)	0	25

* Participants could have multiple responses

** Based on 11 interviews, 3 group interviews and 8 individual interviews

Other health care interviews focused more on how local health care systems worked, and prevention questions were not asked (Table 15). Other

prevention strategies listed in table 15 were reported infrequently and were not included in the within and across group analysis below.

Two prevention strategies had vague meanings. One was to be careful, and only three people reported this prevention strategy; they did not elaborate on what this meant. Another prevention strategy was to avoid overworking. One healthcare worker offered this and to avoid falling as methods to prevent cleft. In her case overworking had implications for putting force on the fetal face. She said when mothers overwork they may slump over position or bend at the waist, causing cause force on the fetal face when the fetus has its hands to its face.

Three people from the noncleft also thought that to avoid overworking could prevent cleft along with an environmental prevention strategy such as taking only prescribed medications and not smoking. They did not explain how overworking could prevent or cause cleft.

As with responses to the cause of cleft, prevention methods considered to be influenced by environmental exposures were collapsed into one category, environmental prevention. The preventions included in the new category were to avoid alcohol and smoking, exercise, eat a healthy diet, take vitamins, get regular checkups, and take only prescribed medicines. Note participants talked about doing these prevention strategies during pregnancy, not before. On average, women in this study began their prenatal care at about three and one half months after they missed their period (range, one to nine months). The overall category

of environmental exposures as a cause of cleft was created by the researcher and included in the within and across group analysis below. This category was created based on biomedical explanation for cleft and may not be recognized by Filipinos.

The new distribution of participant responses by collapsing prevention strategies into one category is summarized in Table 16. Family planning remains the most common prevention strategy, followed by environmental prevention, then avoiding falls. However within interview groups this pattern did not hold true.

Table 16. Modified summary of prevention categories stratified by interview group.*

Prevention categories	Cleft (n = 27)	Noncleft (n = 31)	Health care person (n = 11)**	Total
<i>Characteristics n (%)</i>				
More research on cause is needed first	1 (4)	0	0	1
Be careful in general	1 (4)	2 (6)	0	3
Do not overwork during pregnancy	0	3 (10)	1 (9)	4
There is nothing you can do to prevent cleft	1 (4)	3 (10)	0	4
I do not know	3 (11)	1 (3)	1 (9)	5
Do not mock reprimand or dwell on the image of a cleft person	3 (11)	2 (6)	0	5
Do not slip or fall avoid pressure to the abdomen during pregnancy	4 (15)	7 (26)	1 (9)	12
Environmental prevention	3 (11)	11 (35)	3 (27)	17
Family planning	16 (59)	9 (29)	0	25

* Participants could have multiple responses

** Based on 11 interviews, 3 group interviews and 8 individual interviews

In the cleft group family planning was the most common prevention strategy reported. This was followed by avoiding accidents such as falling and putting pressure on one's abdomen. Environmental prevention methods, to avoid mocking/teasing those with cleft, and I do not know how to prevent cleft were reported three times each.

In the noncleft group prevention strategies were more evenly distributed among environment ($n = 11$), family planning ($n = 9$) and avoiding falls and pressure to the abdomen ($n = 7$) compared to the cleft group. Prevention response patterns also fit the noncleft group's beliefs on what caused cleft; inheritance was the most common cause ($n = 16$), followed by force the fetal face ($n = 13$) and finally environmental exposures ($n = 9$).

Across and within group comparison for prevention of cleft grouped by attributes

Next, participant's responses to what can prevent cleft for the categories of avoiding falls and pressure to the abdomen, environmental prevention, and family planning were grouped according to attributes, gender, age, education level, setting, and family history of clefting (Table 17). Other prevention categories were not included because there were too few responses in those categories overall. As in the above analysis of cause, healthcare workers were not included in this analysis.

As with the previous comparisons for cause of cleft, it is difficult to compare males' responses to females' responses because there were few males participating in the study. However, family planning was the most common prevention strategy reported by males and females.

Table 17. Number of participant responses for avoiding falls and pressure, environmental prevention, and family planning stratified by attributes gender, age, education, setting, and family history. ($n = 58$)*

Attribute value	Avoid falls and pressure to the abdomen	Environmental prevention	Family planning
Male	2 (3)	2 (3)	8 (14)
Female	9 (16)	12 (21)	17 (29)
Young (18-33 years)	7 (12)	9 (16)	12 (21)
Old (33-54 years)	5 (9)	5 (9)	13 (22)
Less than high school graduate	5 (9)	2 (3)	8 (14)
High school graduate	5 (9)	5 (9)	9 (16)
More than a high school graduate	1 (2)	7 (12)	8 (14)
Rural	2 (3)	8 (14)	12 (21)
Urban	9 (16)	6 (10)	13 (22)
Positive family history	6 (10)	4 (7)	15 (26)
Negative family history	4 (7)	8 (14)	6 (10)

* Participants could have multiple responses

Age did not appear to have influenced the distribution responses to prevention of cleft. Both old and young reported family planning most, followed by environmental prevention, and then avoiding falls and pressure (Table 17). Likewise, residential setting did not seem to influence prevention responses except for avoiding falls and pressure. Only two people from the rural areas

compared to 9 from the urban areas reported avoiding falls and/or pressure to the abdomen during pregnancy to prevent cleft (Table 17). The family history of clefting response pattern once again reflected cleft and noncleft groups. As explained previously, in the cleft group more people reported a positive family history than a negative family history. In the noncleft group more people reported a negative family history than a positive family history. Family history was not useful to compare groups (Table 17).

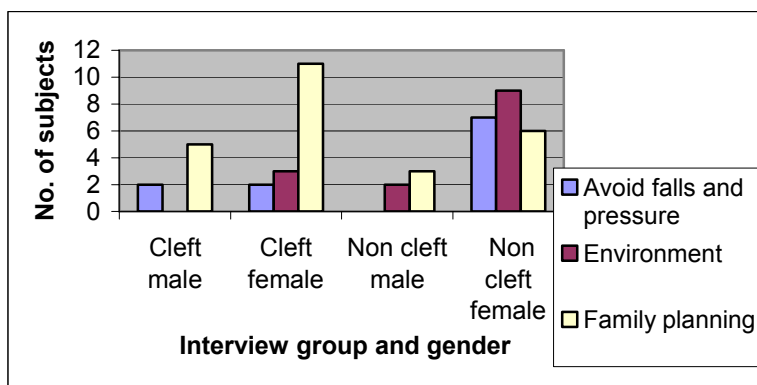
Education did appear to influence people's beliefs about how to prevent cleft. As the education level increased the number of people reporting environmental prevention strategies also increased. Only one person with a more than a high school education reported to avoid falling or pressure to the abdomen as a means to prevent cleft. Family planning was common across all three education levels (Table 17).

Prevention of cleft in the cleft and noncleft group
compared by gender

Males from both the cleft and noncleft group reported family planning most frequently as a means to prevent cleft. Only two males from the cleft group reported avoiding falls and pressure during pregnancy, and only males from the noncleft group reported environmental prevention. Females from the cleft group also reported family planning most frequently. Females from the noncleft group

showed a more even distribution of responses among avoiding falls/pressure, environmental prevention, and family planning (Figure 10).

Figure 10. Number of participants reporting prevention of cleft for avoiding falls and pressure to the abdomen, environmental prevention and family planning grouped by interview group and gender. ($n = 58$)

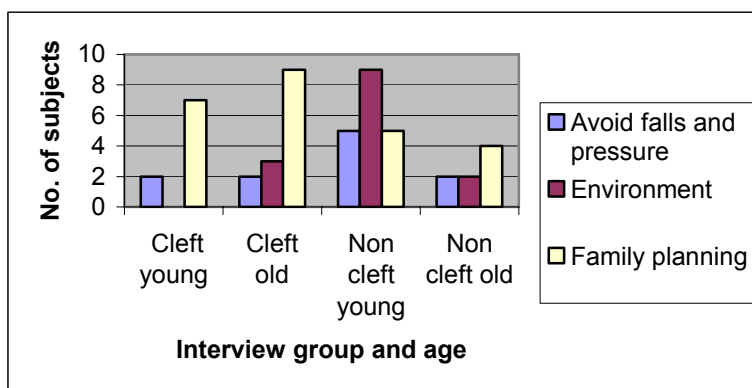


Prevention of cleft in the cleft and noncleft group compared by age

Every age group, except the noncleft young, reported family planning as a way to prevent cleft most frequently. The noncleft young group reported environmental prevention strategies most frequently. All age groups, except the noncleft young, reported family planning most frequently; avoiding falls and environmental prevention strategies were reported less but with just about the

same frequency. The noncleft young reported environmental prevention strategies most frequently; family planning and avoiding falls were reported less but with just about the same frequency (Figure 11).

Figure 11. Number of participants reporting prevention of cleft for avoiding falls and pressure to the abdomen, environmental prevention and family planning grouped by interview group and age. ($n = 58$)

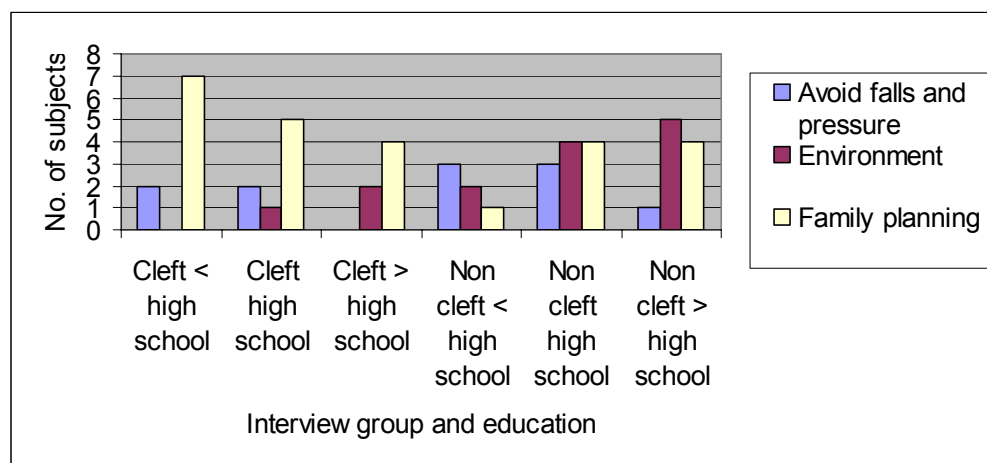


Prevention of cleft in the cleft and noncleft group compared by education

Avoidance of falls and pressure to the abdomen tended to be more common prevention strategies for those with a high school education or lower in both the cleft and noncleft group. The number of participants reporting

environmental preventions increased as education levels increased. This was also true in both the cleft and noncleft group (Figure 12).

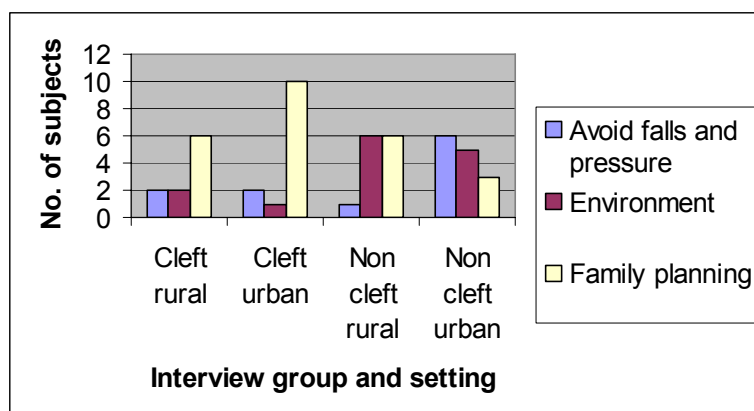
Figure 12. Number of participants reporting prevention of cleft for avoiding falls and pressure to the abdomen, environmental prevention and family planning grouped by interview group and education level. ($n = 58$)



Overall the noncleft group reported environmental prevention methods more than the cleft groups. In the cleft group no one with less than a high school education reported any environmental prevention methods. In the cleft group as education level increased the number of people reporting family planning decreased. In the noncleft group the opposite trend was observed, as education level increased the number of people reporting family planning increased (Figure 12).

Prevention of cleft in the cleft and noncleft group
compared by residential setting

Figure 13. Number of participants reporting prevention of cleft for avoiding falls and pressure to the abdomen, environmental prevention and family planning grouped by interview group and setting. ($n = 58$)



Cleft urban and rural groups followed the overall response pattern of the cleft group in general. Cleft urban and rural groups clearly reported family planning as the most common prevention method. Few people in either group reported environmental prevention or avoidance of falls/pressure to the abdomen during pregnancy as strategies to prevent cleft. In the noncleft rural and urban groups environmental prevention methods was common as was true of the noncleft group in general. However, compared to the noncleft urban residents

noncleft rural residents reported family planning more than they reported avoidance of slipping and falling (Figure 13).

Family planning prevention method

Family planning was the most frequent prevention method given by people in both the cleft and noncleft groups, but never mentioned by the healthcare workers we interviewed. Of all the prevention methods reported by participants, family planning especially the use of birth control could have a big impact on a vitamin trial. If people are not willing to become pregnant then a vitamin trial to examine the impact of vitamin use to reduce recurrence of cleft may not be feasible. Therefore, it will be described in more detail.

Most people who thought family planning was a way to prevent cleft said that in order to prevent clefts we must prevent the birth of more children. There was not a distinction made between occurrence and recurrence of cleft. We simply asked people, “How could we prevent cleft?” Also, family planning as way to prevent cleft was not limited to only those who believed cleft was caused by inheritance:

I: What do you think are ways to prevent having babies born with cleft?

C1: Not to have a lot of children, because abnormality cannot be avoided if you bear lot of kids.

I: So you have use control (birth control)

C1: Yes, control, (birth control), that’s it.

I: What do you think can be done to prevent cleft from happening?

C2: For me all I will do so that I will not have another cleft child, I will not have another pregnancy.

I: So, not having children will prevent babies being born with cleft or not having children at all because you are too poor to feed the children?

C2: Not having children so we can avoid having babies born with cleft (she takes hormone injections as family planning method and falling caused her child's cleft).

I: What do you think we can be done to prevent having cleft children?

C3: Don't get pregnant again.

I: Don't get pregnant. Why did you say that we can prevent having cleft children when we stop being pregnant?

C3: You can't prevent having cleft children. Because there will be times wherein you really can't guess what will happen.

I: You can't guess what?

C3: That your child will be cleft. You won't know that because he is inside the womb.

We added questions about family planning when we realized that people were often giving it as a prevention method. We asked if people used family planning in all but three of the interviews from the cleft group, and 10 in the noncleft group. Overall, people reported the use of birth control pills most often followed by no use of family planning. Patterns of type of family planning use did not differ between cleft and noncleft group (Table 18).

However, uptake of family planning did differ between the groups, with more people in the cleft group reporting use of family planning. Of the 24 people in the cleft group asked about family planning, 83% reported use of a family planning method and 17% used no family planning. In the noncleft group, 71% of the 21 people asked reported use of family planning, while 19% reported no use of family planning.

Table 18. Summary of family planning methods used stratified by cleft and noncleft groups.

<i>Characteristics n (%)</i>	Cleft (<i>n</i> = 27)	Noncleft (<i>n</i> = 31)	Total (<i>n</i> = 58)
Not asked	3 (11)	10 (32)	13 (22)
Birth control pill	9 (33)	8 (26)	17 (29)
Condom	3 (11)	1 (3)	4 (7)
Hormone injection	1 (4)	2 (6)	3 (5)
IUD	2 (7)	0	2 (3)
None	4 (15)	6 (19)	10 (17)
Rhythm	1 (4)	1 (3)	2 (3)
Tubal ligation	1 (4)	1 (3)	2 (3)
Withdrawal	3 (11)	1 (3)	4 (7)
Abstinence	0	1 (3)	1 (2)

No pattern of family planning method was observed when comparing rural and urban areas except that the IUD was found only in the urban areas and hormone injection was found only in the rural areas. However, overall only two people reported using an IUD and three people reported hormone injection as their family planning methods.

The use of birth control pill may be influenced by age. As described above, the cohort of participants was grouped into two age groups, young and old. There were 33 people in the young group and 25 in the old group. Eight people from the young and five from the old were not asked about family planning. In both groups more people reported use of some form of family planning rather than none at all. In the young group most (13 of 25) reported using birth control pills, and four reported no use of family planning. In the older group 4 of 20

reported some use of birth control pills, while 6 reported no use of family planning. Other methods (condom, hormone injection, IUD tubal ligation, abstinence, rhythm, and withdrawal) were reported more than the pill alone.

The use of birth control pills may also be influenced by education with its reported use more common in people with a high school education or less. In all three education groups, more people than not reported use of some form of family planning. Eighteen people who received less than high school education reported use of family planning compared to one person who reported none. Eight of the high school educated reported use of family planning compared to four who reported none. Eleven people who received more than high school education reported use of family planning compared to five who reported none.

The use of birth control pills specifically was reported by eight people in the less than high school group, six in the high school group, and 3 in the more than high school group. There were 10 people who were older than 33 and 11 who were 33 or less in this highest education level. We did not investigate family planning any further as the study was not about family planning. However, we did ask people if they planned to have another pregnancy. In the cleft group ($n = 27$) 5 said yes, 14 said no, and 3 were not sure (5 had no response recorded). In the noncleft group ($n = 31$) 13 said yes, 13 said no, and 1 was undecided (4 had no response recorded).

Treatment

No one described any “special” treatment when asked how to treat and care for children with cleft. When asked how to treat children with cleft people responded, “Surgery.” Some of the mothers talked about taking extra time to feed their children compared to their children who do not have cleft.

Aim 2 Content Analysis

Aim two was to describe working class Filipinos’ (including healthcare workers) perceptions about vitamin supplementation and fortification during pregnancy and to obtain suggestions for the design and implementation of possible vitamin trials to prevent orofacial clefting. Participants were asked to provide ideas about how to utilize existing communication channels for new health campaigns related to orofacial clefting. We asked questions to explore how and why people take vitamins during pregnancy. We also asked questions to explore participants’ perceptions about vitamin supplementation. This was done by asking about barriers and facilitators to taking different forms of vitamins that could be offered as part of a vitamin trial. Last, we asked people to give us ideas on how we might make a prenatal vitamin trial more successful. Interview data from healthcare workers were gathered through individual and group interviews.

The data are presented as one group because the categories of responses specific to aim two were similar between the two types of interviews.

Each interview was coded for broad topical categories and descriptive sub-codes. Responses to questions pertaining to each of these elements: vitamin use in pregnancy, health care information delivery, and perceptions of vitamin supplementation will be described. Then, differences and similarities across groups will be presented.

Health Care Information Sources

Participants in this study reported obtaining most of their health information from the local health office. As described above, the weekly schedule of available medical services are posted outside of the barangay health (Figure 4). Informational posters hung outside the health office or in the waiting/intake room. Announcements for new programs or seminars are written on chalkboards or paper posters and displayed outside the clinic. In addition to the postings, we asked participants how they were informed of new programs offered at the health office.

More than half (55%) of the non healthcare participants ($n = 58$) reported that they found out about new and existing health programs from the barangay health worker (BHW) or other health office staff going house to house. The BHW was described as going house to house and personally inviting targeted

audiences to the health office for a given event. About 20% responded of non healthcare participants ($n = 58$) reported they heard of new programs by word of mouth, one neighbor telling another and so forth to spread the word of a health program or event. Fourteen percent ($n = 58$) of non healthcare workers reported finding out about new health programs through postings outside the health office. Three or fewer non healthcare participants reported one or more of the following methods of finding health information: radio, newspaper, public announcements, personal letter, routine visit to the health center, through their barangay captain, and seeking information on one's own. This response pattern remained consistent regardless of how the data were grouped.

Vitamin Use during Pregnancy

Only one group of healthcare workers reported dispensing folic acid. They said they used folic acid primarily to treat and prevent anemia, and not to prevent birth defects. Another group of health workers reported that they gave multivitamins until their supply ran out, and then people had to purchase their own.

Healthcare workers from health offices in Kabankalan City and Himamaylan city reported that their usual practice regarding prenatal vitamins is to dispense one bubble pack of 30 tablets at a time. Healthcare workers give iron at the first prenatal visit. The women were instructed to come back each month

for a prenatal checkup and new supply of iron. The checkup entailed a weight check, blood pressure monitoring, palpation of the abdomen, and dispensing another 30 tablets. These activities are recorded and kept in a patient record maintained in the health office.

Based on the healthcare workers description, iron and other free medications or vitamins are typically dispensed from the barangay or city health office, not private physician clinics. For example, women may consult a physician on her first or second prenatal visit. The physician may prescribe iron for anemia. The patient would then most likely go to the barangay health office to obtain iron for free rather than pay for it at a pharmacy. One group of health workers reported that sometimes their supply only allowed them to give a one month supply of iron for free, and then the women had to purchase their supply for the remainder of the pregnancy. A different group of healthcare workers said they began iron in the third or fourth month of pregnancy after the period of craving when women are not nauseated:

C1: So we start them (on iron) on the fourth month the third and the fourth month of pregnancy....Because that's the time that they stop craving.

I: Craving. They are giving the ferrous sulfate in the fourth month of pregnancy because during that time the mothers are not...

S: not as nauseated.

I: Yeah, nauseated.

While iron was dispensed without a prescription by the midwives, multivitamins were prescribed by a physician and usually purchased at the

pharmacy. Some of the healthcare workers said that multivitamins were routinely prescribed by physicians, but it was up to the women to decide to take it. Most of the women we talked to did not take multivitamins because they said they could not afford it. One woman took said she took multivitamins during her pregnancy because she could not tolerate the iron and could not afford to pay for the vitamins. If a woman wanted to switch to a different brand of iron (different from the one given for free at the health center) she would have to purchase it from the pharmacy. Some women would alternate between iron and a multivitamin. Most of the women we talked to only took iron during their pregnancies.

We asked participants to tell us why women take vitamins during pregnancy. The health of the baby and the mother were stated reasons for vitamins use. People stated that vitamins helped the baby “develop good inside” without abnormalities. Vitamins were also thought to give the baby resistance to sickness and give the baby energy so the he would be active inside the mother. One person reported that vitamins promoted brain growth. Two other reasons to take vitamins were so the baby would not be malnourished and would gain weight. The health of the baby was reported more frequently ($n = 21$) compared to the health of the mother ($n = 10$) in the noncleft group only. In the cleft group and healthcare worker groups, mother and baby health were reported in about equal frequencies. Within the noncleft group all but two people who reported vitamins were for the health of the mother also said vitamins were for the health

of the baby. In the healthcare worker group the same participants reported health of the baby and mother. In the cleft group however, only four people reported both health of the mother and baby as reasons to take vitamins during pregnancy. Within the cleft and noncleft groups no distinct pattern of responses (mother versus baby) stratified by age, education, gender or setting was appreciated:

C1: The child can get vitamins from the mother and he will be free from sickness.

C2: To avoid abnormalities, so the child will develop well and to avoid abnormality while pregnant.

C3: For pregnant women, to keep their baby strong and healthy. Vitamins taken by mothers are for their babies to make it healthy, and for brain development, and for moms too.

C4: So that by the time the baby is born he will have healthy body.

C5: So that, it [the baby] will be energetic inside.

C6: So that when their baby grows it won't be malnourish.

Vitamins were reported to give energy, promote sleep, give resistance to sickness, be a dietary supplement, and treat anemia for mothers taking them during pregnancy:

C1: To add more energy.

I: To add energy, for the baby or the mother?

C1: For the mother.

C2: So that the mother will have enough strength to do work and to sleep.

C3: It acts as a food supplement especially if the mother does not have the appetite to eat more. Also for pregnant women who can't sleep.

C4: Vitamins just give resistance to you and the baby. (Vitamins give resistance against sickness for the mother and the baby)

I: What did they say about the purpose of ferrous?

C5: To add blood.

C6: Like when you're anemic, you need to take vitamins.

It did not occur to us during the time we were interviewing to distinguish between the purpose of iron and purpose of a multivitamin. In our conversations with participants, iron was referred to as a vitamin as well. About half the time participants talked about anemia they made reference to iron specifically, and half the time they said vitamins were used to treat or prevent anemia. There were times, too, when iron was reported to be for the health of the baby.

Women reported that they began taking vitamins/iron at their first prenatal visit; this could be anywhere from one to nine months with 3.5 months the average and the most commonly reported time to begin prenatal among women in this study. We also asked women why they quit taking their vitamins/iron during pregnancy (Table 19).

Women reported they stopped taking vitamins/iron during pregnancy because of side effects and cost. Complaints of side effects had to do with iron while costing too much money was in reference to multivitamins. The following complaints were reported about iron and caused women to discontinue use: it made them feel sick, nauseated, and/or dizzy; tasted bad; and smelled bad (like fish). One person stopped taking a multivitamin because it was not available, and

another stopped taking iron because the health center did not have any more available. One woman said she did not take her multivitamin because she was afraid her baby would be too big. Two women from the cleft group stopped taking their iron because they felt “better” and found no need to continue. Response patterns were not influenced by age, education or setting.

Table 19. Reasons women stopped taking vitamins/iron during pregnancy stratified by cleft and noncleft group.*

Reason to stop taking vitamin/iron <i>Characteristics n (%)</i>	Cleft (<i>n</i> = 20)	Noncleft (<i>n</i> = 23)
Complaints	12 (60)	9 (39)
Delivered baby	7 (35)	11 (48)
Felt better	2 (10)	0
Cost too much money	2 (20)	4 (17)

* Participants could have multiple responses

Perceptions about Vitamin Supplementation and Fortification

Perceptions about vitamin supplementation and fortification were explored by asking participants to list advantages and disadvantages of taking vitamins as a liquid, chewable tablet, pill to swallow and in fortified grains. We did not ask which form people preferred although a few people talked about preference as they gave ideas on advantage and disadvantages. We asked about advantages and

disadvantages with the idea that these data could be useful in developing a more widely distributed survey on preferences for vitamin supplementation.

These questions seemed difficult for participants to answer. We tried reframing the question to ask what do you like or not like about taking a given form of vitamin. We used the words advantage and disadvantage and good and bad, but our word choice did not seem to matter. We tried asking people, what would be better for you, chewing vitamins or swallowing vitamins. Then we asked what is better about X, and what do you not like about Y. We put the scenarios into personal context. We explained that we are trying to come up with the best way to give vitamins to people, and that we would like their help to develop a plan. We explained that we may conduct a vitamin trial to see if vitamins could reduce the overall number of babies born with cleft. Still, more than half the people answered “wala” (I have no thoughts about this).

One concern voiced about vitamins in a chewable form was that it might taste bad, even if flavored. Other concerns were that children may take it and overdose and that chewing the vitamin was too much work compared to just swallowing it. One woman thought that because the vitamin is chewed, people may not get the full dose. Pieces of the vitamin could get caught in one’s teeth. Another concern was that adults just are not familiar with this form of vitamin so they may not want to take it.

On the other hand, convenience was offered as an advantage of chewable vitamins. A chewable form would not need water and could be put in one's purse and ingested on the way to work or anywhere. Other advantages were that chewable vitamins could be shared with children and the flavor would hide the bad taste of vitamins. Two people liked the idea of a chewable vitamin because it would be something "new" to try.

We described liquid forms of vitamins as a powder to be mixed with water to drink. Participants thought this would be inconvenient and too much work. Another common concern was that a liquid vitamin might taste bad. Others said they are not familiar with this type of vitamin. In addition, one person felt that he may not get a full dose of vitamin because the liquid sticks to the cup. Faster absorption, slower uptake in the body, easier to swallow, and the added benefit of drinking water were reported as advantages to taking a liquid vitamin.

We asked people what they thought were disadvantages and advantages to fortifying rice with vitamins. Several possible concerns about fortification were raised by participants. One was that the vitamin might be harmful to those who ate the rice but did not need the vitamins. People could be allergic to the vitamin. Another concern was dosage, some were skeptical that they would get the correct dose if the vitamin was placed in rice or bread. Other concerns were that the fortified food could be expensive, people do not always prepare food at home, the vitamin could get washed out during preparation, and they may not buy the brand

of rice being fortified. One other concern was fairness. If the fortified food was meant for only women, for example, it would not be fair to others in the home who could not take advantage of the rice.

The main benefit of fortification was that in addition to vitamins, people also got food. Another advantage reported by participants was that fortified rice could benefit more people not just pregnant women. They thought fortification could cost less because they would only need to purchase rice, not both rice and vitamins. Another benefit was that rice is eaten practically every day so one would forget their vitamins if it was in their rice. One person said that rice already had vitamins so adding more to it made it even better. Another person was more confident in the “purity” of the vitamins in rice versus in a tablet. She said that tablets include other ingredients that are not vitamins.

Most people did not report disadvantages for the pill form of vitamins. Two people said they have problems swallowing pills, but reasons women stopped taking iron were bad taste and smell, and GI disturbances. Advantages for the pill form included convenience, “it goes directly to the body” and “it has a quick effect” compared to other forms.

We asked participants to list advantages and disadvantages to taking a daily dose of vitamins and a monthly dose. People were familiar with taking their vitamins on a daily basis; no one reported experience taking it weekly or monthly. A drawback to taking a daily pill was the daily burden of remembering to take it.

One person said it is extra work to take vitamins daily and others said they have a tendency to forget to take daily doses of medicines and vitamins. Others said that taking vitamins daily meant that you would have daily side effects. If you take vitamins less often, then maybe you would not have to deal with the side effects as often. The most common negative aspect of taking daily vitamins was the notion that if one forgot a dose it would be more difficult to make it up. Some people felt if they forgot a daily dose, they might not receive the entire monthly dosage. Some people were concerned that if they skipped doses and made it up by taking two doses in one day, they would get too much. They were concerned that taking two pills in one day may not be tolerated and cause side effects:

CI: I prefer the once a month because I won't tend to forget that tablet. The disadvantage of taking the pill once a day is that, what if I forget my dosage for this day? I won't be able to complete the whole one month of taking the pill.

C2: She thinks that taking it once a month would be a lot better than taking the everyday pill... because [when she forgets] sometimes she may double the dose and that might not be good for her (because of side effects).

On the other hand, taking vitamins daily had the benefit of taking a more constant dose, and some felt the body would tolerate that better. Receiving the correct dose daily was associated with receiving daily benefits of the vitamin. In addition, daily dose was the pattern people were most familiar with:

C1: [I prefer daily vitamins] So that you can continuously take it and you will be energetic every day that you do your obligations at home (I'll have energy when I do my household chores).

C2: It will help a lot if we take vitamins every day because we work every day. If we take vitamins every day it will help us a lot because we are working every day. It can give me energy.

C3: Because our body needs it for daily work. Especially when I wash clothes, I want to take vitamins every day so that I will feel lighter when I work. It's nice to work when you take it every day.

C4: So that you will know the effect is daily. If you will take it all at once you might fly (because if you take the bigger dosage in one intake you might get high).

C5: For me, it will be better to take the vitamins every day because I will have a better sleep, then good appetite, in that case the more food I eat the more nutrients the baby will have.

C6: I prefer a small dose every day, because the doctor prescribed it. Based from what we have experienced and encountered at the center, that's just it, except it is taken twice a day.

C7: A usually here, the doctor prescribes [it] once a day. That's all only once a day, every day, daily. I never experienced once a month.

C 8: I prefer once a day, we are use to taking vitamins once a day.

C9: The good thing about taking the vitamins every day is that your body will just receive the minimal amount every day.

C10: My doctor gave me a prescription of Natalence N once a day. I am also a mother and when my doctor prescribed me Natalence N to be taken once a day I followed it because he is a doctor and he knows what he is doing.

C11: The doctor would always prescribe them one capsule per day for three months because the daily requirement should be met.

One common concern about taking a larger monthly does of vitamin was the possibility that the body would not tolerate it, and it could lead to an overdose:

C1: All in one dose your body can't take it. If you take it every day, you take it little by little and you will be able to tell if your body can take it or not.

C2: She was concerned that the body could not tolerate a large dose or at least that is what the general person would think. I asked her, even if a larger dose was free of side effects did she think that people would attribute or perceive any physical abnormal feeling like dizziness to the high dose and likely discontinue it, and she said yes.

C3: Because it will be the first time that I will take vitamins [monthly], I might not like it, and I fear that it will increase my blood pressure.

C4: Because you can take it slowly (that is better). You might have an overdose on one tablet a month... your body may not be able to take it.

C5: Of course pregnant women might be afraid to have an overdose because something might happen to their baby inside; the one that the doctor suggests (the daily dose) is the one that they follow.

I5: So most mothers like you are afraid to take large doses of vitamins?

C5: Of course, because we might have an overdose and it won't be good for our pregnancy.

People were also concerned that taking a vitamin only once a month

would not provide daily benefits:

I: Okay, so if you will take the (vitamin) once a month, you feel like you don't have resistance or energy because you will only take it once a month?

C1: Yes.

C2: Because there is no effect when you take it once a month. But if it is every day then it has an effect on your body.

I: I think what you're getting at is that the baby gets, the mom and baby get the right amount every day. So every day there is a consistent amount of vitamin going to the mom and the baby instead of one big dose all at once. I think that is what she said. Can you ask her if that is what she said?

C3 (through the interpreter) She is telling us that this is how she understands your statement. Women would love to take vitamins every day because it can help them have a better sleep. Then the nutrients will go directly to the baby every day.

Other concerns were that the larger monthly dose would require a larger pill and would be difficult to swallow. Larger dosage pills may also be more expensive. Some people felt once a month dosage would be easier to forget and have greater consequences. Some were concerned that if they forgot a pill that was to be taken only once a month they would miss an entire month's supply, not just one day.

Perceived advantages of a monthly pill were that there may be less of a burden to remember, it is easier to make up the dose if you forget, more convenient, less expensive, and there may not be daily side effects:

C1: You can say that it's only once in a month (I only have to take the vitamin once a month) so it's okay. But if you take it every day...I even get lazy to brush my teeth every day.

C2: Because it's just once that you will drink the medicine then you won't have to worry that you will forget it [every day].

C3: She prefers to take it once per month because if you forget you can make it up easier than if you take it every day. Plus there is less to remember when taking it once per month.

C4: Because if you will take it every day it will be expensive. Imagine how many tablets, one month is thirty. So I choose the tablet that is taken once a month.

C5: Yes, it will be effective because it won't be expensive because it will be once a month.

C6: [I choose] The once a month.

I6: Once a month, why?

C6: Because if I choose the other one (daily dose), every night I take it I would feel nauseated.

Table 20. Perceived advantages and disadvantages of taking vitamins daily, taking vitamins monthly and fortification of rice.

Method of vitamin intake	Disadvantage	Advantage
Fortified rice	<ul style="list-style-type: none"> -People who do not need vitamins will get them -Allergy to vitamin -Dose is not individualized -Fortified food will be expensive -Vitamin gets washed out during food preparation -Do not always prepare food at home -It will not be fair if the rice is not for everyone 	<ul style="list-style-type: none"> -You get food plus rice -More people could benefit not just pregnant women -Eat rice every day so you won't forget your vitamins -Rice is already good for you -More pure form of vitamins are found in fortified rice
Daily vitamin pill	<ul style="list-style-type: none"> -Daily burden to remember -Daily side effects -Extra work to take vitamins -Difficult to make up missed dose 	<ul style="list-style-type: none"> -Receive a consistent dose, you get what you need daily -Daily benefits (energy) -Familiar with this form
Monthly vitamin pill	<ul style="list-style-type: none"> -Overdose -Body cannot tolerate -Do not receive daily benefits -Not familiar with this dose -More expensive -Greater consequence if you miss a dose -Easier to forget to take it 	<ul style="list-style-type: none"> -Experience side effects less frequently -Less expensive -Easier to make up missed dose -Convenient -Less burden to remember

Table 20 summarizes advantages and disadvantages perceived by participants for taking vitamins once a day, taking vitamins once a month, and

fortification of rice. Response pattern remained consistent regardless of how the data were grouped.

Suggestion for a Successful Vitamin Trial

Finally, we asked people to give suggestions on how to make a vitamin trial more successful. People gave three categories of ideas: (1) who should endorse the vitamin trial, (2) how to educate people about the trial, and (3) how to conduct the trial. Very few people gave suggestions for how to make a vitamin trial more successful. Therefore, response pattern comparisons were not made within and across groups.

Endorsement

Endorsement of the vitamin trial by trusted medical personal, such as doctors, nurses, midwives and barangay health workers from their own community was one suggestion. Another was to seek endorsement from the barangay captain. These people were viewed as people who could influence the decision of others to participate in the trial, and they are trusted as people who are looking out for the welfare of the people in their community.

Education strategies

The most frequent suggestion for the vitamin program was the need to educate people before the program began. Mother's class was suggested as a possible venue to educate the public about vitamins. Mother's classes are seminars for mothers. Topics for mother's classes may include: immunization, cooking healthy foods, infectious disease, sanitation, and health related topic. People said that if they understand why they are taking vitamins, then they would be more likely to follow the program. The need to advertise using methods previously discussed, house to house, radio, television, newspaper, and public announcements were also suggested.

Suggestions for how to conduct a vitamin trial

A few people suggested using role models from the community as spokespersons for a vitamin program. Two people offered to be that spokesperson. The idea is that the spokesperson forms a trusting relationship with people who conduct the study; so that the spokesperson can testify that the people conducting the study are good people. One woman said she would convince others that the vitamins really helped, and that she is proof that the vitamins can work. She said because people know and trust her, they will listen to her. We asked if the researcher were the one who went house to house to convince people

to take vitamins, would people participate. People said no, because they do not know her:

C1 If a collector or vendor would knock on our door, we would say nobody is home or the boss is not here. That's the style. So, it is really nice if people from the community will be the one to convince others. To that end, community involvement was also suggested by participants as

way promote a successful vitamin trial. For example, one person suggested we use the barangay captain to get the word out about the vitamin program. One group of health workers suggested tying into existing programs through the barangay health office such as prenatal visits and appointment cards. Another suggestion was to form a cooperative with members from the barangays and puroks to help introduce the project to the community.

Other suggestions for how to make a vitamin trial successful included giving vitamins for free, home delivery of vitamins by the BHW, and the use of giveaways. Giveaways are small gifts usually food or household items such as plastic containers and clothes baskets, used to entice people to take part in an activity or as gifts of appreciation for their attendance. One person suggested we target women before their first pregnancy and another suggested we target women who already had a child with cleft. She felt if we targeted mothers of children who did not have cleft we would not find anything out, because they are normal.

Several healthcare workers talked to us about an existing program called treatment partners. This program is intended for use with patients who have

tested positive for tuberculosis (TB). These patients are placed on a drug treatment regimen that requires strict adherence. To ensure adherence patients partner with a health worker, typically a BHW. The BHW makes sure the medications are dispensed and monitors the patient for compliance until the regimen is completed. Healthcare workers suggested a treatment partner for participants in a vitamin trial.

Summary of Results

We interviewed 80 people in 69 interviews. All healthcare workers were female; 13 were from rural barangays and 9 from urban barangays. There were a total of 11 healthcare worker interviews. We conducted three group interviews; the remaining eight healthcare workers interviews were individual interviews. A total of 31 people participated in the noncleft group, 8 men and 23 women. Twenty-seven people from the cleft group were interviewed, 7 men and 20 women. Most people were married or in common law relationships. Participants were equally divided by education levels. One third of participants had less than a high school education, one third had graduated from high school graduate, and one third had more than a high school education. Most of the people were working class having jobs in service, labor, or agriculture with monthly family incomes of less than P6,000. The majority of participants reported Catholic religious affiliation.

Specific aim 1

Sungi is the Ilonggo word for cleft lip, but there were a two people who used the Cebuano word, bungi to name cleft lip. Inheritance, force to the fetal face, environmental exposures, and craving were the most common explanations given for cause of orofacial clefting. In summary, inheritance and force to the fetal face were common explanations for cleft regardless of participants attributes (characteristics), except for males. Males reported environmental exposures and force to the fetal face most often. However, there were few males in this study. Cravings as a cause of cleft was more often reported by people who had a child with cleft, were less educated, or lived in more rural settings. In contrast, environmental exposures as an explanation of cause of cleft was reported more frequently by people who did not have a child with cleft, were more educated, or lived in a more urban setting.

Family planning was the most common prevention method reported by members of the cleft group regardless of attributes gender, age, education or setting. The number of people from the cleft and noncleft group reporting family planning was inversely related to education levels. The number of people from the cleft group reporting environmental prevention increased as the level of education increased.

In the noncleft group as the education level increased, the number of people reporting family planning or environmental prevention increased. Environmental prevention was the most commonly reported prevention method in the noncleft group. However, the number of people reporting environment, family planning, and avoiding falls/pressure as prevention strategies was more evenly distributed in the noncleft group compared to the cleft group. Within the noncleft group, environment remained the most common or second most commonly reported prevention method except among those with less than a high school education and the urban group. For these latter two groups avoidance of falls/pressure was the most commonly reported prevention.

Specific aim 2

Iron was the micronutrient supplement most women reported taking during pregnancy. Reasons women reported for not taking iron and vitamins during pregnancy were cost of vitamins and side effects related to iron. No one reported taking any kind of vitamin/mineral before pregnancy for the health of the baby or mother. Most women began taking their vitamin/iron after their first prenatal visit around 3.5 months (after they missed a menstrual period). Women received their iron through the local health center for free. People recognized some health benefits of taking vitamins/iron for mothers and babies.

Most participants in this study obtain their information about new health programs from the local health office by a BHW going house to house to deliver a message, word of mouth, or by postings at the health center. Other sources of information mentioned by three or fewer participants included: radio, newspaper, public announcements, personal letter, contact with healthcare workers at routine visits, their barangay captain, and on their own.

Regardless of vitamin form (liquid, chewable, or pill to swallow) people did not want the vitamin to taste or smell bad. They wanted it to be convenient. They were also concerned about receiving the correct dose in order to obtain daily benefits regardless of vitamin form. People voiced concern that a larger monthly dose of vitamin would have side effects and not be tolerated compared to a daily dose. Some people thought that side effects from vitamins would be less frequent if they took a monthly dose compared to a daily dose. Participants expressed concerns about the cost and the burden of remember in to take vitamins regardless of form and dosage. Reported benefits of fortification compared to supplementation included: feeling full, receiving vitamins and food at the same time, and lessening the burden of remembering to take a vitamin.

Several suggestions for making a vitamin trial more successful were given. People reported that we should get endorsements from government and health officials and seek support from the community. A role model as a spokesperson from the community was another suggested form of endorsement.

Educating the community before the program begins was the most frequent suggestion given for how to make a trial more successful. Several health workers suggested using a treatment partner for dispensing and monitoring vitamin use during a trial.

CHAPTER 5: DISCUSSION

The focus of this chapter is to discuss the main findings described in the previous chapter as they relate to formative social marketing research. Results highlighted in this chapter include: (1) Filipino explanatory models for the cause, prevention, and treatment of orofacial clefting, (2) sources of health information (3) current practices in taking prenatal vitamin, and (4) potential barriers and facilitators for a vitamin trial. These findings, as presented in Chapter 4, are examined in relation to implications for practice, future research, and education. Study limitations inherent in this research will also be discussed.

Social Marketing

A social marketing framework was used to guide this study. Social marketing is an audience or client centered planning process that offers a foundation for developing culturally innovative interventions (Thackeray & Neiger, 2003; Tripp-Reimer et al., 2001). In this study elements of formative research from social marketing were used in a cultural assessment of Filipino's beliefs, values, and practices regarding cause, and prevention of orofacial cleft as well as vitamin taking practices during pregnancy. The elements of formative research included audience analysis, channel analysis, and market analysis.

Aim one was to describe differences and similarities in attitudes and beliefs about the cause, prevention, and treatment of clefting within and across three working class Filipino groups: individuals with a cleft or individuals having a child with cleft; individuals who neither have a cleft nor have children with cleft; and local healthcare workers. Questions asked as part of aim one were asked as elements of social marketing's formative research, specifically audience analysis of cultural view on cause, treatment, and prevention of orofacial clefting.

Elements from social marketing formative research, specifically components of channel and market analysis were used to describe working class Filipinos', including local healthcare workers' perceptions about vitamin supplementation and fortification. We also sought suggestions for the design and implementation of possible vitamin trials to prevent orofacial clefting. To meet the aims of Specific Aim 2 we asked participants about (1) sources of health information, (2) current prenatal vitamin taking practice, and (3) potential barriers and facilitators for a vitamin trial. The main findings from this study are summarized in a social marketing model of the reduction of risk for orofacial clefting (Appendix C) and are discussed in more detail in the following sections.

Audience Analysis

For some Filipinos illness or disease can be viewed as physical, emotional, spiritual, and mental discomfort and represents the negative relationship between

humans and their natural and supernatural environment (McKenzie & Chrisman, 1977; Miranda et al., 1998; Williams, 1978). Natural explanations for illness among Filipinos include an imbalance between hot and cold within the body, heredity, accidents, exposures to foods, contact with dirt, alcohol, poor diet, lack of sleep, the moon, night air, and exposure to sick people. Supernatural causes of illness have been attributed to witches, sorcerers, devils, deceased relatives or friends, and non-humans acting as conduits of harmful influences. Talismans, amulets, a cross, or prayers may be used to ward off or drive away supernatural forces (McKenzie & Chrisman, 1977; Miranda et al., 1998; Williams, 1978). Traditional Asian beliefs about cause of orofacial clefting have been reported to include retribution for wrong doings, spells, curses, and imbalance of natural forces (Cheng, 1990). The findings from this study support the reports of others on some Filipino traditional beliefs about the causes of illness. Participants in the study reported heredity, accidents, poor diet, and alcohol use as causes for orofacial clefting.

Beginning in the late 1930s, George Peter Murdock and colleagues started the Human Relations Area Files (HRAF) project. The goal of this project was to organize ethnographic studies according to a standard format. This resource organizes more than 700 different aspects of culture in terms of etic cultural categories. The HRAF domain of sickness provides easily accessible information on hundreds of societies related to health categories such as preventive medicine,

theory of disease, medical care, sorcery, and bodily injury. Based on reviews of hundreds of ethnographic studies, the HRAF categorizes natural and supernatural causation of illness. The HRAF project took emic concepts of illness and translated them to etic categories. This resource provides a means for researchers to compare their results and engage in cultural comparison of illness explanations. The HRAF has five distinct ways to categorize natural causation if emic concepts are translated to etic terms and include: infection, stress, organic deterioration, accident, and overt human aggression. Supernatural causations are classified as mystical, animistic, and magical. Mystical causation is further subdivided into fate, ominous sensation (dreams, sightings, and feelings that cause illness), and mystical retribution. Animistic causation is subdivided into soul loss (by thievery) and spirit aggression. Magical causation is subdivided into sorcery and witchcraft (Anderson, 1996). This study found explanation for orofacial cleft that would fit in the HRFA categories of infection, stress, accidents, and mystical retribution.

We found causal explanations for orofacial clefting among Filipinos to be similar to the general explanation of illness describe by others. Participants gave the following “natural” causal explanations for clefting: heredity; force to the fetal face caused by falling, pressure to the pregnant abdomen, or abortions; and environmental exposures. Environmental exposures included birth control pills, medications, smoking, alcohol, stress, poor nutrition, infection, pollution, and

vitamins (too much or too little). Supernatural explanations for orofacial cleft included God's will and craving.

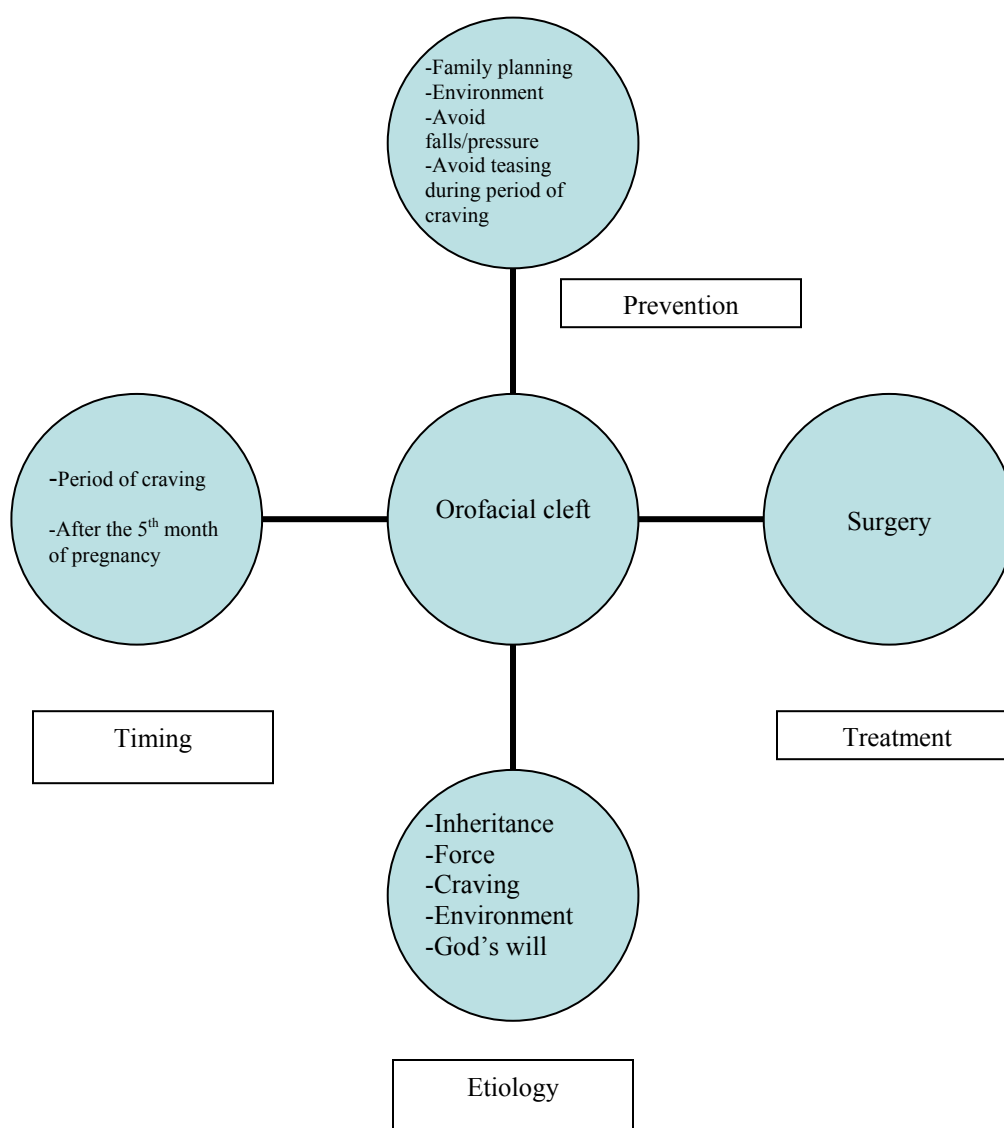
Filipino Explanatory Model for Orofacial Clefting

People's individual explanations of illness are fluid and situation dependent with biologic, psychologic, and sociocultural factors influencing the explanation they give for illness and disease. (Kleinman, 1978; Kleinman et al., 1978). An aggregate of individual explanatory models for working class Filipinos in Negros Occidental is shown in figure 14. Filipinos' explanations for orofacial cleft includes inheritance, force to the fetal face, environmental exposures, craving (seeing, teasing, and/or reprimanding a cleft person during the period of craving), and God's will. The explanatory models for orofacial clefting did not differ between the cleft and noncleft groups. The frequency of responses differed between groups, but not the explanations. Healthcare workers' explanations for orofacial cleft differed slightly from the cleft and noncleft groups. Healthcare workers did not report craving as a cause for cleft, nor did they give family planning as a method to prevent cleft.

Inheritance and force to the fetal face were pervasive explanations for cleft regardless of how participants were grouped. Observed exceptions to this were men (both cleft and noncleft) and in the cleft group from rural areas. Men favored

explanations of force to the fetal face and environmental exposures. People in the rural cleft group favored explanation of craving as cause of cleft. Cleft can occur

Figure 14. Composite Filipino explanatory model for orofacial clefting.



early in the pregnancy during the period of craving or later in the pregnancy depending on the specific cause.

Family planning, environmental prevention, avoiding trauma to the pregnant abdomen such as falls or pressure, and not teasing or reprimanding a person with cleft during the period of craving are ways orofacial clefting can be prevented. The number of participants reporting environmental preventions strategies increased as the level of education increased in both the cleft and noncleft groups. In the noncleft group the number of participants reporting family planning increased as education levels increased. In the cleft group the opposite was observed; the number of people reporting family planning decreased as education levels increased. This finding may reflect the small number of people in each education subgroup because overall family planning was the most common prevention strategy at each level of education within the cleft group. Orofacial cleft is treated by surgical repair regardless of its cause and/or prevention.

Causal and preventative explanations were compared across the three types of interview groups: cleft, noncleft, and healthcare workers. Causal and preventative explanations were compared on the following attributes gender, age, education, setting, and family history as well. These comparisons were offered as a heuristic exercise and do not reflect intracultural variability or ambiguity

(Nichter & Nichter, 1996). Correlational studies based on random sampling strategies are needed to make these associations.

In addition to cultural descriptions of Filipinos' explanations of illness as both natural and supernatural (McKenzie & Chrisman, 1977; Miranda et al., 1998; Williams, 1978), several studies have reported Filipinos' explanations for illness. Other's reported Filipinos' explanations for illness to include: an imbalance between hot and cold within the body; heredity; accidents; exposures to alcohol, cigarettes, and contagion; poor diet; and lack of sleep (Guthrie et al., 1990; Nichter & Nichter, 1996; Yamada, Caballero, Matsunaga, Agustin, & Magana, 1999). The finding from other studies regarding cause of illness will be compared to the Filipino explanatory model for cleft that was constructed as a result of this study.

Sungi

The Ilonggo word for cleft lip is sungi and for cleft palate is punga. Another word for cleft lip in this area of the Philippines is bungi; this means cleft lip in the Cebuano dialect. Nichter and Nichter (1996) conducted a focused ethnography on Filipino mother's knowledge of acute respiratory illness (ARI) in Oriental Mindoro, Philippines. They asked mothers to name and explain causes for acute respiratory illness in their children. Mothers gave several names for ARIs. The names given to ARIs were associated with causes, signs and

symptoms, and treatment courses. We heard of only one name for orofacial clefting, *sungi* (*bungi* in the Cebuano dialect), and all causes, preventions, and treatments were related to the single name for cleft. This suggests that among Filipinos orofacial clefting is the same disorder regardless of cause, prevention, or treatment.

Inheritance

Inheritance (heredity) was reported most often as an explanation for cleft by participants in this study. When the question of what causes cleft was modified and asked again for the participants in the cleft group to “what caused your child’s cleft?” inheritance was reported by 56% of participants as the cause. Fifty-two percent of noncleft participants thought inheritance caused cleft, and 64% of healthcare interviews contained inheritance as a cause of cleft. Many people understood that heredity plays a role in orofacial clefting when there was positive family history, but there were people who said that heredity was not a risk factor if there was no family history. No one talked about genes. They described heredity in terms of family history and blood lines. People also did not necessarily recognize inheritance if there was not a vertical pattern of inheritance. For example, one woman was not worried that subsequent offspring could have a cleft because her daughter was the only one in their family with cleft, no previous ancestors were affected. For others, when they could not recall an affected

relative they assumed that an ancestor from generations back had cleft, thus explaining the cleft in their child.

Others have reported that Filipinos attribute heredity as a cause for illness. Mothers in the study of acute respiratory illness (ARI) in Oriental Mindoro, reported heredity as one cause of ARI (Nichter & Nichter, 1996). Researchers conducted focus groups with Filipino immigrants to learn about their beliefs related to cause and treatment of tuberculosis (Yamada et al., 1999). Filipino immigrants to the United States identified heredity as a cause of tuberculosis. Guthrie et al. (1990) conducted longitudinal studies of breast feeding and infant growth in a rural community in the Philippines. They found that mothers believed illness, stunting of growth, and death in their children could be caused by hereditary factors (Guthrie et al., 1990).

Force to the fetal face

Like inheritance, force to the fetal face caused by falls, attempted abortions, or pressure on the pregnant abdomen was a pervasive explanation of clefting. Forty-one percent of people in the cleft group and 42% of people from the noncleft group reported that some type of force to the fetal face could cause cleft. Forty-five percent of interviews from the healthcare workers contained force to the fetal face as a cause for clefting. People who reported that force to the fetal face caused orofacial clefting generally did not believe this could cause

cleft until later in the pregnancy. There is linear and concrete logic to believing falls and force cause clefting. The idea of force to the fetal face as cause of cleft is reinforced when a women falls during pregnancy and then subsequently delivers a baby who has a cleft. Those who reported that force to the fetal face led to clefting believed the baby's face must be fully formed and then broken open. They reported that the face is fully formed after the fifth month of pregnancy.

Nichter and Nichter (1996) also reported that Filipinos believe falls can cause ARI. Filipinos in their study believed that falls or blocked flow in the body called pilay hangin could lead to one form of ARI. Pilay was described as falls or being held too tight that lead to fracture or dislocations within the body and disrupted natural "flows." ARI thought to be caused by pilay was treated by traditional healers (folk doctors). ARIs caused by pilay hangin were characterized as recurring and/or persisting. The traditional healer located the disrupted flow and massaged the body where the block was located to restore natural "flow" (Nichter & Nichter, 1996). In Ilonggo pilay hangin is called kibit. During this researcher's two months stay, the topic of kibit came up in several informal conversations with Filipinos from various backgrounds. Kibit was described and treated just as Nichter and Nichter described pilay hangin (1996). Participants in this study also reported falls as a cause of illness, specifically cleft. However, unlike ARI, participants reported only one treatment for cleft regardless of cause.

Other cultures also believe accidents cause of illness. The HRAF lists accidents as one of the five distinct ways to categorize natural causation of illness. Accident as a category of illness in the HRAF is based on reviews of hundreds of ethnographic studies (Anderson, 1996).

Craving

The third most common cause of orofacial cleft reported by participants was craving. Cravings are a supernatural explanation for cause of cleft. Participants in this study described craving as (1) seeing a person with a cleft and then thinking about them during the period of craving and (2) retribution for staring at, teasing or reprimanding someone with a cleft during the period of craving. People in this study described the period of craving as the first three months of pregnancy. Participants believed that during this time the fetus is vulnerable to take on characteristics of an object and/or person the mother or father saw and thought about. The object may be a food craving such as the example of the mother who craved tambis. Participants also described craving as “fondness for looking at a cleft person”, unable to get the image of a cleft person off their mind, or the urge to tease a person with cleft. People thought cleft was side effect of staring at, teasing, or reprimanding a person with cleft during the period of craving. The woman who reported craving tambis caused her child’s

cleft was an observation made by the researcher on a previous visit to the Philippines.

Craving as an explanation for cleft was more common in the cleft group. Its reported frequency decreased as education levels increased. Craving was more common among those from rural residential settings than urban in both cleft and noncleft groups. It could be that cravings as an explanation was more common among the cleft group because it was experienced and validated by the birth of a baby with a cleft. However, when we asked people what caused their child's cleft, inheritance was reported three times more often than craving.

A traditional Filipino belief is that a pregnant woman's food cravings should be satisfied especially during the first trimester to avoid harming the baby. Another traditional belief is that a baby will take on characteristics of the food the mother craved and ate (Miranda et al., 1998). Yet, only one person reported that cleft could be caused when a pregnant woman does not satisfy her food craving during the period of craving.

In addition to heredity, Guthrie et al. (1990) noted that Filipino mothers believed illness, stunting of growth, and death in their children could be caused by sorcery (Guthrie et al., 1990). On a prior visit to the Philippines we heard of one family who describe a curse that caused their family to have cleft. Craving, as explained by participants in this is similar to sorcery in that they are both supernatural explanations for illness. Other cultures also believe in supernatural

explanation for illness. The HRAF lists supernatural causations as mystical, animistic, and magical. Mystical causation has two subdivisions fate/ominous sensation (dreams, sightings, and feelings that cause illness), and mystical retribution (Anderson, 1996). Mystical causes of clefting reported by participants in this study include God's will (fate) and craving (seeing a person with cleft and retribution for teasing/reprimanding during the period of craving) as causes of orofacial clefting.

Environmental exposures

Participants reported environmental exposures as causes of cleft as well. Environmental exposure was not as pervasive an explanation for cleft as inheritance and force to the fetal face. Environmental exposures explanations were more commonly reported by the noncleft group and healthcare workers. It could be that the mothers and fathers we interviewed from the cleft group did not experience environmental exposure during their pregnancy with the affected child, and therefore did not offer them as explanations. The frequency of reported environmental explanations increased as education levels increased. Environmental explanations were more common in the urban group compared to the rural residential setting group.

People from the noncleft group talked about environmental preventions (avoid drinking and/or smoking; take vitamins; eat healthy and receive regular

prenatal checkups) more frequently than people from the cleft group. People in the noncleft group reported the general category of environmental prevention as frequently as they reported family planning. This again could be due to people's experience in that people in the cleft group did not say environmental exposures led to cleft in their child as often as inheritance. Therefore, it stands to reason they did not report environmental prevention strategies as often compared to noncleft and healthcare workers. A link between family history and environmental exposures as risk factors was not part of Filipino's explanations for cause and prevention.

The overall category of environmental exposures as a cause and prevention for cleft was created by the researcher and included in the within and across group analyses. This category was created based on biomedical explanation for cleft and may not be recognized as a category by Filipinos. However, the individual explanations making up this category were explanations given by the participants and represent possible explanatory models for orofacial clefting by people from the Philippines.

Nichter and Nichter (1996) reported that Filipinos in their study of ARI thought contagion (germs) could cause ARI. We also observed that some Filipinos thought that infections such as chickenpox could cause orofacial clefting.

Filipino immigrants to the United States reported environmental exposures such as bacteria and viruses, cigarettes, alcohol, dirty conditions, and wet clothing could lead to tuberculosis (Yamada et al., 1999). Imbalances of the body thought to cause tuberculosis included overwork; poor nutrition; untreated colds, cough, and fevers; and worries about problems (stress). We did not use the category of imbalances, but categorized similar causes as environmental exposures. Filipinos in this study reported that maternal exposure to cigarettes, alcohol, infections, poor nutrition, fatigue/overwork, and stress could cause cleft. These causes of cleft are similar to the environmental and imbalance causes of tuberculosis reported by Filipino immigrants.

Filipino immigrants also reported that changing the environment and exposures, and correcting imbalances were methods to treat tuberculosis. Changes in the environment included getting out of the city and into the fresh air and making work and home environment cleaner. Avoiding cigarettes was another way to improve the environment and treat tuberculosis. Correcting imbalances in the body to treat tuberculosis included activities such as exercising, eating balanced diet, getting rest, avoiding problems, and bathing when one is not tired or sweating (Yamada et al., 1999). Participants in our study reported that by avoiding environmental exposures and overworking, along with eating well and getting exercise and rest, orofacial clefting could be prevented. These prevention

explanations are similar to treatments for tuberculosis reported by Filipino immigrants (Yamada et al., 1999).

Family planning

Family planning was the most frequent prevention method given overall by people in both the cleft and noncleft groups, but it was never mentioned by the healthcare workers we interviewed. This was an unexpected finding. Only 4 of 109 people who took part in the previous informal interviews said that limiting the number of children or family planning could prevent cleft lip and palate (Daack-Hirsch, S., unpublished data collected 2000-2001). Additionally, the majority of the people interviewed for this study were Catholic, and birth control, a form of family planning, is against the doctrine of the Catholic Church.

Family planning may have been mentioned more frequently as a method of prevention because it was an active Department of Health (DOH) public health campaign, *Ligtas Buntis*, during the time of this study. *Ligtas Buntis* literally means safe pregnancy. The DOH program goals were to reduce the number of unwanted pregnancies and abortions, premature births, and birth injuries and/or deaths to mothers and infants. To reach these goals the program advocated for the freedom and responsibility of couples to raise happy and healthy families. *Ligtas Buntis* programs taught maternal child nutrition, promoted early and often prenatal care, and offered family planning options. Family planning options were

given to couples as a means to space or limit the number of children, taking into account the couple's conscience, religious beliefs, and cultural values. In addition to informed choice for family planning, the program also advocated the use of trained healthcare professions for prenatal care and delivery. As we started data collection, local health offices were just completing the *Ligtas Buntis* campaign. During the campaign the programs listed above were offered. In addition, statistics were gathered on the family planning methods currently used per household per barangay. These activities and statistics were then reported to the national DOH.

Family planning as prevention could have been a reaction to the *Ligtas Buntis* campaign that was taking place when this study was conducted. It could also reflect the general national trend in the Philippines to limit family size (*Improving reproductive health in the Philippines, 2003*).

The previous birth of a child with cleft may have influenced people's choice to use family planning as 51% of people in the cleft group said they did not plan to have more children compared to 42% of people in the noncleft group. It could be that people who had a child with cleft are more likely to limit their number of offspring because they did not want to have another affected child. This may partially explain why people in the cleft group reported family planning more frequently compared to the noncleft group.

The idea that family planning (birth control) could reduce the occurrence and recurrence of orofacial clefting is interesting. Vieira and Orioli (2002) published a metaanalysis of studies on birth order and oral clefts and found that children with higher birth order are more likely to have a cleft (Vieira & Orioli, 2002). Vieira and Orioli's metaanalysis showed that the odds of clefting increases with higher birth order peaking at an odds ratio of 3.0 for four or more children. While birth order was not associated with clefting in all studies, a combined analysis of selected studies did provide evidence to support birth order as a risk factor for clefting in some groups of people.

The relationship of birth order to orofacial clefting is not well understood. One hypothesis is that homocysteine has a teratogenic effect on the developing fetus. As folic acid levels decrease, homocysteine levels increase. Folic acid is needed to counter the rise of homocysteine. Pregnancy affects homocysteine and folate levels. With each trimester homocysteine levels naturally increase while folate naturally decreases unless the mother supplements her diet with folic acid. Folate levels continue to decrease after pregnancy (Zeiger & Beaty, 2002b).

Women who have many pregnancies in short intervals may not recover their folic acid storage level. Hence, the odds of clefting may increase with higher birth order, especially for women who have short intervals between pregnancies. A constant state of pregnancy may deplete a woman's folate stores leading to increased risk for negative outcomes for the fetus such as clefting. At

least one study of lower socioeconomic status, Filipino, women found that low red cell folate and vitamin B6 deficiency were associated with an increased risk for orofacial clefting (Munger et al., 2004). Perhaps the constant state of pregnancy depletes women of nutrients such as folic acid and is manifested in accumulation of risk factors that lead to an increase risk for clefting with increasing birth order. If higher birth order was shown to be associated with an increased risk for orofacial clefting among Filipinos, then family planning to prevent future pregnancies could be pursued as one prevention strategy.

Treatment

Participants reported surgery as the only treatment for cleft. We thought there may be special treatments, rituals, or healers that people with cleft may need, but there were none reported. If there had been, it would have been important to understand the traditional healer's views on cause and treatment/prevention of orofacial clefting. For example, researchers found that some traditional healers from South African believe orofacial cleft can be caused by failure to pay necessary homage to ancestral spirits, evil spirits, curses from jealous people, bewitching, eating poisoned rabbit meat, and mothers' failed abortion attempts. South African healer's tend to express more magical explanations (sorcery and witchcraft) (Anderson, 1996) for orofacial cleft compared to Filipinos in this study.

South African healer's approaches to treating orofacial clefting focused on treating the whole being and making things right to the ancestors or evil spirits that caused the cleft. The healers said that they treated the problems of the mind and spirit caused by the cleft, but the medical doctors did not (Dagher & Ross, 2004). Traditional healers acknowledged that they could not cure the cleft and that people need to be referred to medical doctors to repair the face. The healers felt that surgical repair was important in order for the person with cleft to be accepted in his or her community and not be mistaken for a witch (Dagher & Ross, 2004). Immigrant Filipinos reported several treatment modalities for tuberculosis as did Filipino mothers in Oriental Mindoro for acute respiratory illness (ARI). For both tuberculosis and ARI treatment type depended on signs, symptoms, and causes of the illness (Nichter & Nichter, 1996; Yamada et al., 1999). Surgery was the only treatment for cleft reported by Filipino participants in this study.

Channel Analysis

Participants in this study obtain their health information from the local health office by a barangay health worker (BHW) going house to house to deliver a message, word of mouth, or by postings at the health center. Other sources of information mentioned by three or fewer participants included radio and

newspaper advertisement, public announcements, personal letter, contact with healthcare provider during a routine visit, and their barangay captain.

Market Analysis

In summary, iron was the micronutrient supplement women reported taking most often during pregnancy. Female participants reported that feeling better, cost of multivitamin (MTV), side effects, bad smell and bad taste, and GI upset from iron were reasons why they quit taking vitamin/iron before they had completed the recommended course. Women in this study also said that sometimes the vitamin/iron was not available at the health center. No one reported taking any kind of vitamin/mineral before pregnancy specifically for the health of the baby. Women began taking their vitamin/iron after their first prenatal visit around 3.5 months (after they missed a menstrual period). Women received their iron through the local health center for free, but they had to pay for MTV.

Historically, iron supplementation programs in the Philippines have had little success because several interacting factors: vitamins are generally thought of as treatment/cures to sickness, pregnant women present late for prenatal care, women demonstrate low compliance related to side effects, and healthcare workers lack of awareness related to the program (Paulino et al., 2005). These ideas were substantiated in this study as well.

Regardless of the micronutrient selected for orofacial clefting prevention trials, there are important developments in the Philippines with women of reproductive age (WRA) and iron/folic acid supplementation. The Philippines and other countries of South East Asia are in the midst of a paradigm shift of their views on how best to treat iron deficiency and subsequent anemia.

Iron deficiency is the most common form of micronutrient malnutrition and cause of anemia in the Philippines (Paulino et al., 2005). A national study conducted in 1998 reported that the prevalence of anemia in pregnant and lactating women was 50.7% and 45.6% respectively. In Negros Occidental where this study was conducted, the prevalence of anemia for pregnant and lactating women were higher than the national average, 63.4% and 47.6% respectively (*Philippine nutrition facts and figures*, 2001). Daily iron with folic acid is the treatment of choice for iron deficiency. Folic acid is given to reduce the risk that folic acid deficiency contributes to anemia (Paulino et al., 2005). We now know that folic acid also reduces the risk of neural tube defects. In 1977 the Philippines Department of Health (DOH) made the recommendation that beginning in the fourth month of pregnancy and during lactation women should take 60 mg of elemental iron and 2 mg of folic acid twice daily. In 2003 that recommendation was modified.

In 2003 the DOH stated that pregnant women should take a supplement containing 60 mg of elemental iron and 400 μ g of folic acid daily as soon as

pregnancy is diagnosed and continue for three months after delivery (Paulino et al., 2005). We did not find this to be true for women in our study. No one reported taking folic acid and most women did not begin taking iron until they were into the second trimester.

Maternal child health complications associated with maternal anemia are well documented. Maternal anemia leads to infant anemia and is associated with stillbirth, infant death and brain damage (Cavalli-Sforza, Berger, Smitasiri, & Viteri, 2005). Low hemoglobin levels during the first half of pregnancy are consistently associated with low birth weight and preterm delivery (Viteri & Berger, 2005). An estimated 20% of maternal deaths are related to the effects of anemia. It is generally recognized that worldwide many women do not enter pregnancy with at least the 300 mg of iron reserves needed to meet pregnancy demands. Aside from maternal child consequences of anemia, it is also associated with problems of human productivity, educational success, and infection (Viteri & Berger, 2005).

These health consequences and the limited success of daily iron supplementation in pregnant women are causing a paradigm shift from iron and folic acid as a therapeutic intervention to one of prevention. For women of reproductive age (WRA) this means that satisfactory iron and folic acid levels should be maintained throughout their reproductive life. After careful review of the literature on studies of the efficacy and effectiveness of daily versus weekly

iron/folic acid supplementation, the World Health Organization (WHO) Regional Office for the Western Pacific (WPRO) decided to support pilot effectiveness projects from 1998 to 2002. The Philippines, Vietnam and Cambodia were selected to examine the effectiveness of weekly iron/folic acid supplementation of WRA in unsupervised program conditions (Cavalli-Sforza, 2005).

Weekly iron dosage has been associated with at least the same, if not improved, therapeutic effects as daily iron dosage in preventing iron deficiency and consequential anemia (Smitasiri & Solon, 2005; Viteri & Berger, 2005). In addition, once weekly folic acid supplement of 2.8 mg for 12 weeks was reported to achieve red blood cell folate levels associated with a reduction of neural tube defect risk (Cavalli-Sforza, 2005). Weekly dosage is thought to enhance compliance by reducing frequency and severity of side effects associated with iron. Weekly dosage is also thought to be operationally easier to manage (less contact hours spent dispensing and monitoring vitamins and less expensive) (Underwood, 2005).

In the Philippines the pilot project took place in three municipalities in Pangasinan, a province in northern Luzon. Using social marketing strategies the Philippines DOH and the United Laboratories (UNILAB) of the Philippines partnered to develop an iron/folic acid pill that would be appealing in terms of color, smell, size, packaging, and cost to WRA. The supplement is called Femina. It was sold to nonpregnant women at a low cost in drug stores, local

variety stores (sari-sari stores), schools, and local barangay health clinics.

Pregnant and lactating women up to three months after delivery were given the supplement for free. Project teams at national, regional, provincial, and local levels were trained in social marketing and interpersonal communication skills (Paulino et al., 2005).

The Philippines trial was successful at improving knowledge of the benefits of weekly iron/folic acid supplementation as it relates to iron deficiency and anemia. The benefits of folic acid were not marketed (except on the package of Femina) and were not part of the reported pre and post trial knowledge assessment. Sales of Femina increased over the trial period, and self reported compliance improved with time. Adverse side effects were reported, but because there was no placebo group, frequency or severity of side effects could not be assessed (Paulino et al., 2005). Compliance was greater for those taking weekly versus daily supplements. Ferritin levels in pregnant and nonpregnant women were significantly higher in those taking weekly supplement compared to those taking it daily. However, there was not an analogous finding for hemoglobin. This may be due to concurrent vitamin A deficiency, also common in Filipinos (Angeles-Agdeppa et al., 2005).

A nationwide campaign of weekly iron/folic acid supplementation of Femina was launched in the Philippines in 2004. However, it did not appear to have penetrated the area of the Philippines where this study was conducted. One

group of healthcare workers reported the use of iron and folic acid, but they were the group who said their supply ran out, and this interrupted their ability to supplement pregnant women in their barangay. When we asked the healthcare workers who participated in this study if they had heard of giving vitamins before pregnancy specifically to prevent birth defects, they said no.

Potential Barriers and Facilitators for a Vitamin

Trial

Regardless of vitamin form (liquid, chewable or pill to swallow) people did not want the vitamin to taste or smell bad. They wanted it to be convenient. Some people were concerned that if they took a vitamin only once per month they would not receive daily benefits and this would lead to low energy. Some people were concerned that they would experience side effects and low tolerance if they took a larger monthly dose. However, some felt that while there may be side effects with a larger, monthly dose, the frequency of side effects may be reduced. The expense of vitamins and the burden of remembering to take it were concerns regardless of form and dosage. Benefits of fortification over supplementation included: more people would benefit from the vitamins in rice because people already eat rice every day and there would be no burden of remembering to take a daily vitamin.

The use of an iron/folic acid supplement such as Femina could be both a barrier and a facilitator. If women continue to experience side effects associated with iron use they may not be willing to remain on iron/folic acid supplementation long term. On the other hand, if the side effects of iron can be minimized then the benefits of iron and folic acid can be emphasized in one health campaign and with one pill.

Participants offered several suggestions for making a vitamin trial more successful. People felt we should get endorsement from government and health officials and seek support from the community. A role model as a spokesperson from the community was another suggested form of endorsement. Educating the community before the program begins was the most frequent suggestion given for how to make a trial more successful. Several health workers suggested using a treatment partner for dispensing and monitoring vitamin use during a trial.

Implications for Research Practice and Education

The two main contributions of this study were (1) construction of a Filipino explanatory model for orofacial clefting and (2) use and development of innovative interviewing methods. Each will be discussed in more detail.

Filipino explanatory models for orofacial clefting

This study is the first to describe explanatory models specifically for clefting among Filipinos. To elude working class Filipino's explanatory models of orofacial cleft, Kleinman's elements of explanatory models were slightly modified. An interview schedule was designed to draw out perceptions of clefting in these four areas: (a) label (the name people give to orofacial cleft), (b) etiology, (c) how the cleft forms (timing during pregnancy), and (d) treatment and prevention. This was done because the modified elements reflect the more static state of having an orofacial cleft compared to acquired illness and/or episodic illness. Two main conclusions are drawn from Filipino's explanatory model for orofacial clefting and include: (1) people's general causal explanation for cleft was not always congruent with their personal causal explanation for cleft and (2) people's causal explanation for cleft was not always congruent with their prevention explanation.

Based on past experiences in the Philippines, we modified questions about etiology. In previous informal interviews, Filipinos who took part in family genetic studies were asked what they thought caused cleft. First, we asked what they thought caused cleft. Second, we asked if that was the reason their child had a cleft; 25% were discordant in their response (Daack-Hirsch, S., unpublished

data collected 2000-2002). For this study 50% were discordant when asked these two questions.

Family planning, environmental prevention, and avoiding pressure to the abdomen were the most frequent prevention strategies reported by participants. Reported beliefs regarding the prevention of cleft among participants added to the understanding of what people believed caused cleft. Some of the prevention strategies were anticipated based on what people reported caused orofacial clefting. For example, if people think falling causes cleft, it seems reasonable that avoiding a fall would be a way to prevent cleft.

However, we found that people's explanations for cause and prevention were not necessarily congruent. Twenty-one of 69 interviews contained ideas about prevention that did not necessarily fit with the cause the participant had reported. For example, three people said that to prevent orofacial clefting women should eat healthy diets during pregnancy, but these three people gave different causes for cleft: (1) attempted abortion, (2) God's will, (3) slipping and falling, and (4) inheritance. In addition, attempted abortion, God's will, and inheritance were given as causes of orofacial clefting by people who reported clefting could be prevented if women avoided slipping and falling. Attempted abortion, God's will, slipping and falling, inheritance, pressure on the mother's abdomen, and teasing were all causes given by people who reported family planning could prevent orofacial clefting.

To our knowledge asking people what they think cause illness in general and then on a personal level, and asking people what can prevent illness has not been used in explanatory model research (T. Tripp-Reimer, University of Iowa College of Nursing, personal communications, November 15, 2006). Kleinman's original elements of explanatory model do not include prevention. Modifying Kleinman's elements of explanatory models to include questions about general and personal explanations for cause of illness and questions about prevention of illness should be used in explanatory model research to bring forth a more complete and accurate explanatory model.

People from all three groups, cleft, noncleft, and health workers, reported that heredity caused orofacial clefting. Inheritance is a common causal explanation of orofacial clefting for Filipinos in this study and in the biomedical literature, and it is a starting place to begin to develop Filipino patient and practitioner education programs. The concepts of genomic risk factors can be developed and introduced to Filipinos by anchoring on the shared concepts of family history and heredity. Participants were able to recognize that a family history of orofacial clefting put them at risk to have a baby with cleft. This has implications for practice.

The National Council of La Raza's Institute for Hispanic Health (NCLR/IHH) developed a genomic health program with Latino Americans using formative research (social marketing) methods. They identified that Latino

participants found the topics of risk factors, genetics, rare diseases, and family history as relevant to their own health. Culturally innovative education tools based on family history were developed to introduce genomic health concepts such as risk identification and screening/testing for use with US Latino populations. NCLR/IHH also developed methods to train local health care providers (A. Gepp, Project Director, NCLR/IHH, personal communication, October 9, 2006). This could be applied to a Filipino population as education for health care providers and patient education tools in clinical practice.

Future research could involve a more in-depth inquiry into how Filipinos understand inheritance. People did not talk about genes, and we did not ask about genes. They described that the trait of cleft lip was inherited or passed along in a blood line.

Like inheritance, force to the fetal face caused by falls and pressure to the pregnant abdomen was a pervasive explanation for clefting. However, unlike inheritance, fall/pressure is not an explanation for clefting found in Western biomedical literature. Falls and pressure to abdomen poses another challenge in that people who believe this is a cause generally did not believe this could cause cleft until later in the pregnancy. This explanation places the fetus at risk at the end of pregnancy and is not congruent with biomedical understanding that orofacial cleft formation takes place in early stages of pregnancy (N. J. Prescott & Malcolm, 2002; Zeiger & Beaty, 2002b). This represents an area of disparity

between researcher/clinician and parallel participant/patient explanations of orofacial clefting. People who believe in this explanation may be reluctant to take on prevention strategies such as vitamins and reductions of environmental exposures. They may also be less likely to join research efforts testing these prevention strategies. These ideas may be reinforced by healthcare workers who shared the belief that force to the fetal face causes cleft. Health education campaigns aimed at dispelling the belief that falls, accidents, and pressure on the abdomen cause cleft can be developed independently of vitamin trials. Health education campaigns should take into account Filipino cultural views that falling has for illness and health.

Filipinos described craving as seeing, teasing, and/or reprimanding a person with cleft. Central to these acts as cause for clefting is their timing during pregnancy. These acts cause cleft only when carried out during the period of craving (first trimester of pregnancy). Others reported that Filipinos believe that babies can take on characteristics of foods craved and eaten by the mother during pregnancy, or that the baby can develop problems if the pregnant mother does not satisfy a food craving (McKenzie & Chrisman, 1977; Miranda et al., 1998). The HRAF classifies ominous sensations and mystical retribution as mystical explanations for illness (Anderson, 1996). We did not determine that people in this study felt seeing a person with cleft was an ominous sensation. Instead, people talked about seeing, teasing and reprimanding as cravings. This

description of craving may be unique to Filipino people. Further research in the Philippines is needed to understand the nature of cravings and its connections to cause of birth defects.

Craving, like inheritance, offers a common concept to frame education materials about cause of cleft. Those who believed cleft was caused by craving explained that craving took place during the first three months of pregnancy (period of craving). The fetus seems susceptible to take on characteristics of the object or person the mother or father fixates on. The period of craving is a susceptible time for the fetus to develop cleft from a biomedical point of view also (N. J. Prescott & Malcolm, 2002). The period of craving is recognized by both those believing in folk causes of cleft and those from the medical community as a time the fetus is susceptible to develop a cleft. It can be used to frame discussions to correct misconceptions as to what the fetus is actually susceptible to. For example, in patient education materials used in the Philippines perhaps framing exposures to tobacco, alcohol, vitamins, good nutrition during the period of craving and before pregnancy would make it easier to understand that these factors could play a role in the cause and prevention of orofacial clefting. Thus, they may be more accepted as explanations for cleft and as prevention methods.

Of all the prevention methods reported by participants, family planning and use of birth control could have the biggest impact on a vitamin trial. A follow-up case control study looking at reproductive behavior related to orofacial

clefting among Filipinos may be needed before a vitamin trial is planned. It would be important to examine reproductive behaviors especially if a vitamin trial was designed to determine the effectiveness of vitamins at reducing recurrence of orofacial clefting. If women are not willing to become pregnant after the birth of an affected child, a recurrence study may not be feasible.

Birth order as a risk factor for orofacial clefting has not been determined in the Philippines and would be another appropriate follow-up study. If birth order were found to be a risk factor, then one practice implications is to partner with a program such as *Ligtas Buntis* (safe pregnancy) that addresses family planning issues such as spacing, birth control, and planned pregnancies.

Planned pregnancies may be one way to reduce the number of babies born with orofacial clefting by reducing the number of children born per sib ship and increasing spacing between pregnancies. In addition, planned pregnancies allow women who may not be able to afford daily vitamins throughout their entire reproductive years to be selective about when to take vitamins. Women who are planning a pregnancy could begin taking vitamins well before conception in order to prepare her body for the pregnancy.

Regardless of why people gave the responses to cause and prevention of cleft they did, each should be considered and addressed in any education materials developed to address risk factors of the cause of cleft. This is true of education materials used in practice or as part of research. Divergence between

researcher/clinician and participant/patient explanations of disease/illness can impede progress toward improving health outcomes. When the explanation for sickness differ between provider and patient the result can be patient noncompliance, dissatisfaction with care, missed diagnosis, inappropriate or poor care (Cohen et al., 1994; Guthrie et al., 1990; Kleinman, 1978; Kleinman et al., 1978; Mayberry et al., 1999; Ticao & Aboud, 1998; Williams et al., 2000).

From the 1960s well into the 1980s Helen and George Guthrie conducted research in the rural Philippines on breast-feeding and infant growth. In developing strategies to improve infant growth they found that Filipino mothers' belief systems and practices differed from theirs. The Guthries and their colleagues observed that long held beliefs were maintained with infrequent or intermittent confirmation while newly introduced practices needed frequent confirmation and were weakened by even minimal failures (Guthrie et al., 1990). These observations are relevant to interventions like smoking cessation or micronutrient supplementation aimed at reducing orofacial clefts. The effects of such interventions are not immediate and may have a greater impact on the population as a whole rather than individuals. In order to achieve sustained behavior changes, benefits of new interventions or ideas on how to prevent cleft will likely need constant reinforcement from all levels of health care. This means health care professionals will need training and education in these areas as well.

Methods

We used innovative methods to (1) compensate for language differences between the principal investigator and study participants and (2) interview groups of people. We developed a modified back translation process to compensate for the language differences. This process was an oral exercise that facilitated the development of the interview schedule and the interview process. Back translation is a technique used to deal with semantic problems that may occur when translating questionnaires from one language to another (Behling & Law, 2000). Oral back translation process involved three people one person who did not speak the local (native) language and two native speakers. Back translations started by having the nonnative language speaker ask a question from the interview schedule in English. Then, one of the native language speakers asked the same question out loud in the local dialect. The second native speaker verbally translated the question back to English, immediately, without trying to interpret what was said in English. Using this process we could modify the nonnative languages speaker's use of English words, reflecting a more typical Filipino English vernacular. This made English translation into Ilonggo easier for the translators during the interviews and allowed the team to clarify the meaning of questions asked in English before the interviews began. Also, many participants could understand English so it was important that questions were

asked using a Filipino-English vernacular. Before changing the English word, it was discussed whether or not the use of a different word lost the original question's meaning.

Oral back translation can be applied to research where the principal investigator does not speak the native language fluently or where there are differences in the English vernacular. This exercise should increase the nonnative language speaker's confidence when asking questions and allow for more consistent and natural interactions among the interviewer, translator and participants.

Because the principal investigator did not speak the local language, we also used a double translation process. The first 19 cleft and noncleft interviews were conducted by the principal investigator with one of the Filipino nurses providing English and Ilonggo translation. Ilonggo was translated a second time during transcription. We had three purposes for conducting interviews this way. The first reason was to verify that real time translation was accurate. The second reason for double translation was to teach other team members how to interview. The third reason to conduct interviews with a translator was to keep the principal investigator involved in the data gathering process.

Double translation provided an accurate method to verify translation. We observed that sometimes during interviews conducted with immediate translation the translator would provide an interpreted translation rather than a more literal

translation. Recording the interview and translating the interaction a second time provided the research team with an opportunity to decide which of the translations (literal or interpreted) was more appropriate to code. Double translation was also an effective strategy to train other research team members to interview and interview consistently, especially where there are differences in language among team members. Double translation permitted involvement of the entire team in identification of categories of responses, evaluation of questions and responses, and modification to the protocol.

Natural group interviews reported in this study was one method used to elicit participant's causal models of cleft and ideas about vitamin use during pregnancy. Natural group interviews were a means of inquiry which invited people to express their ideas in more casual atmosphere. A more casual atmosphere can put people at ease; some people will be more open and willing to elaborate on ideas because they feel less threatened among their peers than when talking with the researcher alone (Beckerleg et al., 1997; Coreil, 1995; Frey & Fontana, 1993). Two different types of natural group interviews were conducted in this study. The first type was a spontaneous gathering of healthcare workers. On three separate occasions healthcare workers gathered spontaneously as we were interviewing one of their peers. We did not observe differences in the quality of data generated by healthcare workers in groups versus individual interviews.

The second type of natural group interview we used was a preset meeting with a group of men who were friends. Because we were having problems finding men who were willing to participate in the study and the natural group interviews with healthcare workers were successful, we decided to arrange for a group of men to meet the research team in a preset location. There were few men in the study overall and we had only one group interview with men, but data from both types of interviews was comparable. Natural groups could be used in future research as an effective way to interview people who may be shy or find individual interviewing intimidating (Beckerleg et al., 1997; Coreil, 1995). Natural group interviews in the Philippines may be especially culturally sensitive and effective because Filipinos have a more collective (group oriented) culture and use indirect communications styles (Miranda et al., 1998) compared to Americans.

Secondary findings

If folic acid were the micronutrient proven to reduce orofacial clefting, the Femina campaign could be an ideal partner. Social marketing strategies aimed to promote maternal child health are already in place with industry and government support. Folic acid is already part of the dosage. The Femina campaign addresses several issues that were brought up by women in this study. For example, women in this study want a vitamin that has few side effects, does not smell or taste bad,

is not expensive, is easy to obtain, and has less burden for remembering to take it.

Women also wanted assurance that they are getting daily benefits from the vitamin; perhaps a weekly dose such as the one in Femina would meet that need better than a monthly dose. A weekly dose may reduce the daily burden to remember, but still provide flexibility to make up doses when one forgets.

One change to the existing Femina campaign/program would be to add information that folic acid reduces the risk of specific birth defects. As it is, the benefit of reducing neural tube defects does not appear to be part of the Femina marketing message except on the back of the package. Outcomes of the study reported by Angeles-Agdeppa et al. (2005) did not include maternal folic acid levels. Birth outcomes related to iron and folic acid were not reported. One research implication could be to consider Femina as one arm of a prevention trial aimed at reducing orofacial clefting.

On the other hand, to conduct a successful micronutrient prevention trial the intervention will need to be free and side effects minimal. Given the specific micronutrient or combination of micronutrient to reduce orofacial cleft is not known, it is difficult to make specific recommendations for the reduction of side effects. The purpose of the micronutrient intervention trial is to test efficacy or effectiveness of a micronutrient or combination of micronutrients at reducing orofacial clefting. One recommendation is to offer the trial micronutrient independent of iron because of the side effects associated with iron. Another

would be to educate participants about the side effects of iron and those of the trial micronutrient and monitor women closely for side effects, especially women who are taking iron either on their own or as part of the study. Women in this study wanted to receive a daily benefit of energy and resistance to illness but did not want to experience side effects related to iron.

Women in this study voiced concern over remembering to take vitamins whether it was daily or monthly as well as what to do when one forgets. If a micronutrient trial were developed with the goal of reducing recurrence risk of orofacial cleft, a treatment partner may be a useful strategy. Participants could be partnered with a BHW who would dispense the micronutrient and monitor the participant for compliance and side effects.

The use of a well trained and informed treatment partner has several advantages. First, they could help to reduce participant's burden of remembering to take the micronutrient. Second, involving the BHW and health centers involves the community. If the trial were successful, there would already be some community involvement with the project. Local healthcare workers could be instrumental in translating research findings into public health campaigns aimed toward all women of childbearing age. Third, properly trained BHWs could assist to correct misconceptions about side effects, motivate, and provide counseling to increase participation rates. Government and local health officials, mayors, and

barangay captains are also people who could be used to endorse a vitamin trial or program because they are trusted figures in the local community.

Limitations

The main limitation to the study was the principal investigator's (PI) inability to speak Ilonggo. This prevented spontaneous conversation with some participants in order to probe further on topics or clarify people's responses without a mediating interpretation. It removed the PI from the participant and the data. This also led to not asking people to clarify the differences between vitamin and iron use. One way to minimize this limitation is to conduct a portion of interviews then translate and interpret before conducting more interviews. This would have allowed the team to make adjustments to the questions we were asking.

However, after twelve years of experience working in the Philippines the PI was proficient at understanding Filipino English and body language. A close, collegial relationship with the Filipino research team also helped to compensate for this limitation. Data were cross checked at several different stages during the study, and this provided another way to compensate for the language barrier. Taking notes during the interview was another method used to cross checked the data. The notes were also used during transcription to clarify what was said when the audio recordings were not clear. Another cross check was to debrief after

each interview. Debriefing included a discussion on whether or not the team agreed on concepts presented and whether or not these concepts were consistent with what was already heard or known. This also provided an opportunity to ask the participant to clarify content at the time of the interview. The use of double translation for a portion of the interviews was another way to cross check data. Double translation involved translating Ilonggo to English both on site during the interview and then again during transcription. Double translation allowed the team to check the accuracy of translation.

Another limitation was that we inconsistently asked healthcare workers about prevention strategies. In the future more data collection with healthcare workers may be needed to reassure that the data are informationally representative of this group. However, there was saturation of responses for cause, treatment, and prevention of orofacial clefting within the group of healthcare workers.

We did not interview people of higher socioeconomic status (SES), medical doctors, or nurses. People from all SES will be included in vitamin trials and education campaigns. The medical/health community's explanatory models are an important part of the culture as well. As stated previously, divergence between clinician and patient explanations of illness can impede progress toward improving health outcomes, and people in the Philippines are treated and cared for by doctors and nurses. This study does not include data that represents their explanatory model of orofacial clefting.

There were few men enrolled into the study. Men said they were too busy or shy to be interviewed. We switched from a formal individual interview to the use of a natural group to interview three men in this study. We did not take advantage of this interview method until late in the study. If we had used this earlier we may have been able to interview more men.

Participants for this study were identified from the registry of cleft families held at the H.O.P.E. Foundation and referred to us by community members such as nurses or healthcare workers. At other times we took advantage of consultation day at the barangay health office knowing this would be a time people would be out and available to participate in an interview. This could have introduced sampling bias and that leads to an unrepresentative sample. Some types of people may have been over represented in this study while other types were not included.

However, most often, the sampling of respondents for open-ended interviews is based on purposeful criteria rather than random chance (Barton, Borrini-Feyerabend, de Sherbinin, & Warren, 1997). Purposeful sampling is different from the random sampling where the intent is to draw statistical probabilities in quantitative research. Statistical sampling is based on randomness so that researchers can confidently generalize results from a small sample to a larger population (Barton et al., 1997). The power of purposeful sampling for qualitative studies is in selecting information-rich cases for in-depth analysis

related to the central issues being studied (Barton et al., 1997; Kuzel, 1992; Morse, 2000; Patton, 1990; Sandelowski, 2000). The sample frame and size was designed to be informationally representative. Three groups of people were invited to participate in this study: individuals with a cleft or individuals having a child with cleft (cleft group); individuals who neither have a cleft nor have children with cleft (noncleft group) and local healthcare workers. These groups were targeted for enrollment because it was thought they might differ in their ideas on cause and prevention of orofacial clefting based on their experience with orofacial clefting. These groups were further stratified for recruitment by residence and gender. These groups were included in the study to generate a range of explanatory models that were representative of the larger community. Given we reached saturation of concepts related to each specific aim within each of the interview groups, data generated during this study should be representative of persons with similar characteristics living throughout the Philippines (Sandelowski, 1995 p. 181).

Summary

The main purpose of this study was to obtain contextual information prior to planning for community based interventions or health campaigns in the Philippines aimed at informing people about causes and preventing orofacial clefting. A descriptive focused ethnography was conducted to explore and

describe current beliefs about the causes, prevention, and treatment of orofacial clefting among Filipinos of the lower socio-economic class. Informant's perceptions about vitamin supplementation and fortification were also ascertained. Elements of social marketing, specifically formative research, and explanatory models provided the conceptual framework to conduct the study.

Social marketing formative research conducted in this study consisted of audience analysis, channel analysis and market analysis. Audience analysis was conducted by constructing an explanatory model of orofacial cleft from working class Filipinos in Negros Occidental, Philippines. Filipinos' explanations for orofacial cleft includes inheritance, force to the fetal face, environmental exposures, craving (seeing, teasing, and reprimanding a cleft person during the period of craving), and God's will. Cleft can occur early in the pregnancy during the period of craving or later in the pregnancy depending on the specific cause. Family planning, environmental prevention, avoiding trauma to the pregnant abdomen such as falls or pressure, and not teasing or reprimanding a person with cleft during the period of craving are ways orofacial clefting can be prevented. Orofacial cleft is treated by surgical repair.

Explanatory models for orofacial clefting among Filipinos in this study are in agreement with cultural descriptions of Filipino's ideas about illness as well as the finding from other research studies. However, this study is the first to describe explanatory models specifically for clefting among Filipinos.

In this study we modified Kleinman's explanatory model and included questions to draw out people's general and personal beliefs about cause of cleft. We also included questions about what people think can be done to prevent orofacial cleft. This modification of methods to obtain explanatory models should be used to capture a more complete and more accurate explanation for causal models. Even when people do not think a cause is relevant to their case they may recognize explanations given by others as valid explanations in their culture. People's ideas about prevention of illness also give information as to what they think causes the illness.

A brief channel analysis of where people receive health information specifically related to new programs revealed that most people in this study obtain their health information from the local health office. Other sources of information included the radio, newspaper, public announcements, personal letter, contact with healthcare workers during routine visits, and their barangay captain.

Market analysis of vitamin supplementation and fortification showed that regardless of vitamin form (liquid, pill to chew or swallow, and fortification) people do not want the vitamin or food to taste or smell bad. They want taking vitamins to be convenient. People voiced concern of side effects and low tolerance if they should take a larger monthly dose of vitamins. However, some felt that while there may be side effects with a larger, monthly dose the frequency of side effects may be reduced. The expense of vitamins and the burden of

remembering to take it were concerns regardless of form. Benefits of fortification over supplementation were that more people would benefit from the vitamins in rice because people already eat rice every day and no burden of remembering to take a daily vitamin.

We used innovative methods to compensate for language differences between the principal investigator and study participants such as oral back translation and double translations. Natural group interviews were successfully incorporated into data collection.

Conclusion

This research used elements of social marketing in order to begin to plan for culturally innovative interventions, specifically preparing for a vitamin intervention trial. Filipino's explanatory model of orofacial cleft constructed as a result of this study can be used in public health education campaigns to correct misconceptions about what causes cleft, direct future studies regarding specific elements of the explanatory model, and provide context for health campaigns and vitamin trials. Results from this research can be used to inform the design of health campaigns regardless of whether or not a vitamin trial actually takes place. These campaigns could include but are not limited to developing information brochures and programs about the cause and prevention of clefting or developing public health campaigns to promote the use of prenatal vitamins in women of

childbearing age. Health campaigns would be useful starting points for preparing the community for anticipated micronutrient trials.

The use of folic acid or any other micronutrient to prevent orofacial clefting is still in question. First the efficacy and effectiveness of any micronutrient needs to be established. Then these data along with findings such as the ones from this study can be useful in developing health education campaigns or to inform study designs aimed at the use of micronutrients to prevent orofacial clefting.

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APPENDIX A: INTERVIEW GUIDE

ID _____ cleft non-cleft Health care person _____ Female MaleAddress _____ rural urban

Date of interview _____

Date of Birth _____

Education (indicate highest level of education):

 none some high school some college
 Elementary high school graduate college graduate post graduate

Occupation _____

Monthly Family income:

 6,000 or below 11,000- 20,000
 6,000 to 10,000 over 20,000

Civil status:

 single married separated

Religion _____

Aim 1: Questions about birth defects and cleft:

1. What kinds of inborn problems are babies born with?

What are the causes of these abnormalities. How can these be prevented?

Show a picture a child with cleft and ask people tell us what this is called.

2 .Do you know anyone with sungi (cleft lip)? (if yes who) Have you heard other names for this?

3. How common is (cleft) in your Barangay?

Have you seen anyone with a cleft outside your Barangay? How many people have you seen with cleft?

4. What do you believe causes (cleft)? Have you heard of other causes; what are these causes?

5. What do you think caused your child to have a cleft?

Probe clarify timing for example if she said she fell in the 6 mo ask if she fell earlier would that have caused the cleft. If the cleft is caused by a curse can that happen any time?

Probe Are there things that put a woman at more or less risk for this to happen?

6. Do you plan to have another pregnancy? What is the chance that this can happen again?

7. What can be done or has been done to treat you or your child's cleft?

8. What do you think can be done to prevent (cleft)?

Probes :What can you do?

What can others do?

9. Are there programs in your barangay to prevent birth defects such as (cleft)?

Probes :How does this program work/ do you participate/why or why not?

Aim 2: Vitamin questions

11. Tell me about the practice of women for taking vitamins during pregnancy?

Did you take vitamins during pregnancy? If yes what kind

When did you start? Who prescribed it? How long did you take the vitamin? What made you stop taking your prenatal vitamin?

12. Pregnancy history of child with cleft, or in the non-cleft group history of most recent pregnancy:

Who provided you with prenatal care? Where did you take or avail your prenatal care?

When did the prenatal care begin?

What can a woman do to have a healthy pregnancy?

What are the foods you ate during your pregnancy? What are good foods for pregnant women to eat? Are there foods that you believe are not good for you or the baby that you should avoid during pregnancy?

13. How do you hear about new medical programs for example new immunizations or vitamin programs or other types of medical care programs?

14. Perceived characteristics of vitamin supplementation.

In the future we would like to conduct a study giving mothers vitamins before they become pregnant to continue until they either have a baby or the study ends. We want to do this study because there is research saying that vitamins taken before and during pregnancy can help to decrease the number of babies born with cleft lip and palate. To help us come up with a good way to deliver the vitamin we would like to ask your opinion about different ways to give the vitamin.

Note: the purposes of the following questions are to explore barriers and facilitators to taking vitamins.

For example, one way we can give you vitamins is to give you 30, 1mg tablets and ask you to take one every day (1mg every day for 30 days equals 30mg). Another way is to give you one 30mg tablet once per month. Which way do you prefer? What is better about that? What do you not like about the other way?

Another way we can give vitamins is to have you stir it in liquid and drink it? Do you prefer to take it as a liquid or drink it? What is better about _____? What do you not like about _____?

We can also give you tablets that you can chew. Which do you prefer; chewing, drinking or taking vitamins as a liquid? What is better about _____? What do you not like about _____? If you like to chew your vitamin what flavor would you like?

Another way we can give vitamins is to fortify your food, like rice. Have you read about fortification of noodles with vitamin A? What would be good about fortification?

Do you have other ideas about how to give vitamins?

Would you pay for vitamins, how much?

15. Share your ideas on how to make a successful vitamin program for women, what can we do to make this work?

APPENDIX B: INFORMED CONSENT

INFORMED CONSENT DOCUMENT

Project Title: **Context for Filipino Community Based (CB) Cleft Prevention Interventions**
 Research Team: **Sandra Daack-Hirsch, MSN, RN; Henrietta Gamboa, BSN, RN; May Ann Estosos, BS**

WHAT IS THE PURPOSE OF THIS STUDY?

This is a research study. We are inviting you to participate in this research study because you are Filipino and live in the Province of Negros Occidental.

The purpose of this study is to learn about what Filipinos think cause a birth defect of the face called cleft lip and cleft palate. We want to learn whether people's beliefs about cause and prevention may be similar or different depending on their experience with a person who has a cleft. We also want to see if there are similar or different ideas among people who have a medical background and people who do not. The main theme guiding the study is that knowing people's beliefs and attitudes about cause and prevention of clefting is vital to be able to develop culturally appropriate and acceptable prevention programs that optimize participation. This is not a study about interventions. We will not be looking at a specific method to decrease anyone's risk to have a child with a cleft. Participation in this study does not ensure or exclude you from participation in any other study having to do with a specific prevention intervention.

HOW MANY PEOPLE WILL PARTICIPATE?

Approximately 90 people will take part in this study.

HOW LONG WILL I BE IN THIS STUDY?

If you agree to take part in this study, your involvement will last for about one hour. We only have one interview planned. However some times later when the researchers review the things we talked about the meaning of what was said is not clear to us. In this case we may come back and ask you some more questions to help us understand.

WHAT WILL HAPPEN DURING THIS STUDY?

If you agree to participate we will be ask to schedule and interview with you. The interview will take place and time and location convenient for you. We would like audio record the interview. However, you have the option to participate but not be recorded. The interview should take about 1 hour and we should only need to talk with you one time. We will ask you questions about what think casuses clefting and about what might be done to prevent it from happening. We will also talk to you about how and

why people go to the doctor or health clinic. At any time during the interview you have option to skip a question or ask questions.

WHAT ARE THE RISKS OF THIS STUDY?

There may be some risks from being in this study but the possibility of risk is minimal. The primary risk is that some questions may make you feel uneasy or uncomfortable. However, you have the right to refuse to answer any questions or withdraw from the study at any time. The other main risk is that others may come to learn that you participated in this study. However, we have set up ways to protect your privacy (confidentiality) and minimize this risk. We will talk more about this under the section called, *What about confidentiality*.

WHAT ARE THE BENEFITS OF THIS STUDY?

There are no personal benefits from participating in this research. However, we hope that in the future, other people might benefit from this study because of what we learn from this study about people's beliefs about the cause and prevention of clefting. We hope that what we learn leads to the development of effective and acceptable education and prevention programs for clefting.

WILL IT COST ME ANYTHING TO BE IN THIS STUDY?

You will not have any costs for being in this research study.

WILL I BE PAID FOR PARTICIPATING?

You will not be paid for being in this research study

WHO IS FUNDING THIS STUDY?

The National Institutes of Health (NIH) and the National Institute Nursing Research (NINR) in the United States of America is funding this research study. This means that the University of Iowa is receiving payments from NIH and NINR to support the activities that are required to conduct the study. No one on the research team will receive a direct payment or increase in salary from the NIH or NINR for conducting this study.

WHAT ABOUT CONFIDENTIALITY?

We will keep your participation in this research study confidential to the extent permitted by law. However, it is possible that other people may become aware of your participation in this study. For example, the H.O.P.E. Foundation Inc. Institutional Review Board and the University of Iowa Institutional Review Board (a committees that reviews and approves research studies) or federal

government regulatory agencies may inspect and copy records pertaining to this research. Some of these records could contain information that personally identifies you.

To help protect your confidentiality, we will do the following:

All information will be kept in locked files when not in use. The computer used to keep the data is password protected. Reports and manuscripts will be written in a manner as not to identify any individuals. All persons associated with the study with access to the data will obtain human subject certification that teaches them about ways to maintain your privacy.

Audio/ Photographs

One aspect of this study involves making audio recordings of you. These tapes are made as a record of our time talking. What was said will be typed out and any names or other information that could identify you will be changed. Once the interview is typed out we will look for common and different ideas among people with respect to how cleft happens and what they think can prevent it. After your interview has been typed the audiotape will be destroyed. The only people who have access to the recordings are the people in the research team and the person who will be typing the interviews. You can participate in this study without being audio taped. In this case I will take notes about what was said during our talk together.

Yes No I give you permission to make audio recordings of me during this study.

You can participate in the study without having your picture taken. Pictures will be taken to use in presenting the results of the study. Your picture will only be used with your permission.

Yes No I give you permission to take pictures of me during this study.

If we write a report or article about this study or share the study data set with others, we will do so in such a way that you cannot be directly identified.

IS BEING IN THIS STUDY VOLUNTARY?

Taking part in this research study is completely voluntary. You may choose not to take part at all. If you decide to be in this study, you may stop participating at any time. If you decide not to be in this study, or if you stop participating at any time, you won't be penalized or lose any benefits for which you otherwise qualify.

WHAT IF I HAVE QUESTIONS?

We encourage you to ask questions. If you have any questions about the research study itself, in the USA please contact: Sandra Daack-Hirsch 1-319-335-9967 or Toni Tripp-Reimer, 1-319-335-7135. In the Philippines please contact Ms. Henrietta Gamboa 63-34-434-6965.

If you have questions about the rights of research subjects or research related injury, please contact the the H.O.P.E. Foundation Inc. at Rooms 301-302 3rd floor Northpoint Bldg, B.S. Aquino Drive, Bacolod

City, Negros Occidental, email address: hope_foundation_inc@yahoo.com. You may also contact the Human Subjects Office at 300 College of Medicine Administration Building, The University of Iowa, Iowa City, Iowa, 52242, (319) 335-6564, or e-mail irb@uiowa.edu. General information about being a research subject can be found by clicking "Info for Public" on the Human Subjects Office web site, <http://research.uiowa.edu/hso>.

Your signature indicates that this research study has been explained to you, that your questions have been answered, and that you agree to take part in this study. You will receive a copy of this form in either English or Illongo.

Subject's Name (printed): _____

(Signature of Subject)

(Date)

Statement of Person Who Obtained Consent

I have discussed the above points with the subject or, where appropriate, with the subject's legally authorized representative. It is my opinion that the subject understands the risks, benefits, and procedures involved with participation in this research study.

(Signature of Person who Obtained Consent)

(Date)

APPENDIX C: SOCIAL MARKETING MODEL RISK REDUCTION OF OROFACIAL CLEFTING

Reduce Risk of Orofacial Clefting

Audience Analysis Explanatory Model

