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Treatment of active nasal fricatives substituting /s/ in young children with normal palatal function using motor-based intervention

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Abstract

Purpose: The aim was to evaluate the effect of a motor-based, hierarchically structured intervention directed at active nasal fricatives substituting /s/ in young children with normal palatal function.

Method: An experimental single-subject design was replicated across three children, aged 4–6 years, with normal palatal function, who substituted oral /s/ with active nasal fricatives. Treatment was performed weekly by a speech-language pathologist and included home training conducted by parents. Audio documented probes were registered regularly and /s/- production evaluated as oral or nasal.

Result: All children achieved 98-100% oral production of /s/ in six probed linguistic contexts at treatment end and exhibited good maintenance at follow-up. The four-year-olds showed gradual or inconsistent response and slower progress, the six-year-old direct response and faster progress.

Conclusion: The study provides preliminary evidence suggesting positive intervention effects for treating active nasal fricatives in children with normal palatal function. However, possible confounding effects such as maturation or repeated testing could not be ruled out; thus, results need to be replicated with increased experimental control. Nevertheless, the study adds to the currently meagre empirical evidence-base for the population. Individual treatment response and progress patterns were found and data suggests that the intervention may be beneficial from age 4.

Keywords: speech impairment; nasal fricative; articulation; treatment; parents; experimental single subject design

Introduction

An active nasal fricative is a nasal realisation of an oral fricative that is produced when the speech airstream is actively directed through the nose, generating turbulent airflow nasally instead of orally (Harding & Grunwell, 1998). An active nasal fricative can be perceived either as a high frequency air emission sound through the nose (anterior nasal fricative) or as "snorting" or turbulence (posterior nasal fricative) (Zajac, 2015). Such alternative articulation behaviour replacing orally produced fricatives, typically sibilants, sometimes occur in children with a normal palate and velopharyngeal function and is considered a maladaptive articulation error associated with velopharyngeal mislearning (Harding & Grunwell, 1998; Peterson-Falzone & Graham, 1990; Zajac, 2015). Active nasal fricative substitutions can compromise speech and lead to a general impression of the individual's speech being nasal and should be managed through speech therapy (Kummer, Marshall, & Wilson, 2015).

Extensive literature searches yielded only one study (Mason, Pua, & Perry, 2018) that has empirically evaluated speech treatment targeting active nasal fricatives in children without a cleft palate history. One additional study of an adult subject was also found (Ruscello, Shuster, & Sandwisch, 1991). Both studies treated active nasal fricatives as a substitute

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for oral /s/- Mason et al. (2018) in a 6-year-old boy and Ruscello et al. (1991) in a 22-year-old male. Both studies used hierarchically structured motorbased therapy approaches to establish oral production of /s/ across increasingly more complex linguistic contexts. While the treatment in Mason et al. (2018) was intensive (six 1-hour treatment sessions conducted over two weeks) and included daily home practice (5-10 minutes) conducted by the parents, the program employed by Ruscello et al. (1991) was nonintensive (nine 50-minute sessions conducted biweekly) but also utilised visual biofeedback (pneumotachograph and water manometer). Both reviewed studies showed successful articulatory outcome of established oral /s/ in all targeted linguistic contexts. Mason et al. (2018) also demonstrated good maintenance at a three-month follow-up.

The empirical research base for effects of treatments targeting active nasal fricatives in children without cleft palate is weak. The two studies found only included one subject each, whereof one adult. However, important aspects of the reviewed studies that may be clinically applicable are a motor-based treatment approach and a component of home training conducted by parents. Indeed, a motor-based treatment approach, based in the theory that a motor skill is learned through repeated actions related to the practice of that specific skill, may be both applicable and well suited for treating speech sound errors in children (e.g. Ruscello & Vallino, 2014). Further, home-training conducted by parents may be added to the speech intervention of children to increase treatment intensity in terms of dose frequency (Kummer, 2011; Mason et al., 2018) and is recommended in a motor-based treatment approach (Ruscello & Vallino, 2014). High intensity in terms of dose frequency of speech therapy sessions has been shown to yield greater gains in speech outcome (Allen, 2013) and was successfully employed by Mason et al. (2018) in the treatment of active nasal fricatives. In practice, however, achieving the high dose frequency indicated by empirical research may be very difficult (Kaderavek & Justice, 2010) due to resource demands (Keilmann, Braun, & Napiontek, 2004; Sugden, Baker, Munro, & Williams, 2016). To address the dose frequency conundrum, researchers have suggested a blended approach with speech-language pathologist (SLP) therapy sessions in combination with home training conducted by parents (Joffe & Pring, 2008; Lancaster, Keusch, Levin, Pring, & Martin, 2010; Sugden et al., 2016). Indeed, parent involvement may be both effective and practically possible (e.g. Lancaster et al., 2010; Lohmander, Henriksson, & Havstam, 2010; Sugden et al., 2016).

The aim of the present study was to empirically evaluate the effects of a motor-based treatment program, including home training conducted by parents, directed at active nasal fricatives substituting /s/ in children with normal palatal function. The following research questions were posed: Does a motor-based speech treatment including home training directed at active nasal fricatives improve oral articulation of /s/ in young children with normal palatal function? If so, is the improved articulation maintained over time?

Method

Experimental design

An experimental single-subject study design with A-B-FU format was replicated across individuals to evaluate the effects of articulation training using a motor-based program to treat active nasal fricatives substituting oral /s/ in children. The A phase refers to a pre-treatment baseline, the B phase to a treatment period and the FU phase to a post-treatment followup. Across all phases, articulation probes of the target behaviour, i.e. orally produced /s/, were conducted repeatedly and regularly in order to establish a pretreatment baseline, monitor treatment progress and examine maintenance of achieved effects.

Participants and initial assessment

Participants were recruited from children referred within a two-year period to the speech-language pathology clinic at a university hospital in Sweden for examination of suspected velopharyngeal insufficiency and hypernasal speech. Eligible to participate in the study were children between 4 and 6 years of age, who were substituting the oral /s/-sound with an active nasal fricative. All children who had an absent history of cleft palate, no signs of submucous cleft palate and who exhibited the particular articulation error were offered the treatment program as part of the regular clinical routine. For the participating children, the actual articulation error, active nasal fricative production substituting /s/, was diagnosed for the first time at the university clinic. Five children met the inclusion criteria and had caregivers agreeing to study participation. Subsequent to study start, two children were excluded since treatment was discontinued due to lack of motivation and cooperation in treatment, both in the clinic and at home. The three remaining participants, who took part in the study, included two boys, Peter and Tom, and one girl, Anna (fictitious names).

The initial assessment of the palate, velopharyngeal function and speech occurred at one to two separate occasions. A nasendoscopic examination was performed collaboratively by a specialist ENT medical doctor (phoniatrician) and an SLP. Speech was assessed by the SLP using the Swedish articulation and nasality test (SVANTE; Lohmander, Lundeborg, & Persson, 2017), containing single word production by picture naming and sentence repetition. A spontaneous speech sample was collected from a dialogue with the SLP around a jigsaw puzzle. In addition, speech was assessed using the 45 probe items of the study (see section *Data collection*). For detailed Table I. Detailed description of the three participants in the final study group, including results from the initial assessment at the clinic.

Participants	Peter	Tom	Anna
Age at start of therapy (years:months)	6:0	3:10	4:0
Age at termination of therapy (years:months)	6:4	4:0	4:5
Phonemes substituted by active nasal fricatives	/s/	/s/ and /f/	/s/ and /f/
Type of active nasal fricatives	Anterior	Anterior and posterior (varied)	Anterior
Hypernasality	No ^a	No ^a	No ^a
Audible nasal emission	No ^a	No ^a	No ^a
Weak consonant pressure in words without /s/	No ^a	No ^a	No ^a
Phonological development	Age appropriate ^a	Age appropriate ^a	Age appropriate ^a
Language level	Age appropriate ^b	Age appropriate ^b	Age appropriate ^b
Previous SLP contact	Yes	No	Yes
Verified VPC by nasendoscopy	Yes	_c	Yes
Normal hearing at age 3	Yes ^d	Yes ^d	Yes ^d
Typical psycho-motor development at age 3	Yes ^d	Yes ^d	Yes ^d
Attending kindergarten	Yes	Yes	Yes

SLP: speech-language pathologist; VPC: velopharyngeal competence.

According to clinical assessment at study start.

^bInformally assessed by experienced SLP at study start.

Child did not participate in nasendoscopy but VPC was verified based on clinical assessment by experienced SLP.

^dCaregiver reported results from the routine examination at age 3 offered to all Swedish children by the national child health care centres.

Table II. Detailed description of the treatment steps, corresponding linguistic contexts, examples of materials and activities at each step as well as number of probe measure items and probe item examples per each linguistic context.

Treatment step: linguistic context	Treatment step description	Examples of materials and activities ^a	Number of probe measure items	Probe item examples	
1: Onomatopoetic expressions	onomatopoetic fricatives	Pictures depicting onomatopoetic expressions (e.g. hushing-sound, wind blowing). Imitating, gluing pictures into booklet.	_	-	
2: Phoneme	isolated /s/	Toy snakes + pictures of snakes. Play with toy snakes, imitating "the snake sound".	5	/s/	
3: Syllable	/s/+vowel	Plastic letters: $S + O$, $S + I$, etc. Post box game.	5	si, so	
4: Word initial	word initial /s/	Toy snakes + pictures of words with initial /s/. Play with toy snakes.	10 ^b	sol (sun)	
5: Word final	word final /s/	Toy snakes + pictures of words with final /s/. Memory game.	b	hus (house)	
6: Word medial	word medial /s/	Toy snakes + pictures of words with medial /s/. Bingo game.	b	läser (is reading)	
7: Cluster	word initial /s/-clusters with oral consonant(s) ^c	Small items + pictures of words with initial /s/-clusters. Memorising and recalling game.	10	skor (shoes), sprutar (is splashing)	
8: Nasal cluster	word initial /s/- clusters with nasal consonant ^d	Pictures of words with initial nasal /s/- clusters. Picking up pictures from a box and	5	snö (snow), smulor (crumbs)	
9: Sentence	sentences containing words with /s/ in different positions	naming them. Depicted story with sentences containing multiple words with /s/. Reading and retelling the depicted story.	10	Sissi och Lasse sover. (Sissi and Lasse are sleeping.)	

^aEvery step included a colour-coded booklet with pictures and instructions.

^bWord contexts of /s/ in initial, final and medial position were grouped together in the probe measure (a total of 10 items). ^cIncluded the following consonant clusters: /sp/, /st/, /sk/, /spr/, /str/ and /skr/.

^dIncluded the following consonant clusters: /sm/ and /sn/.

participant description and results from the initial assessment, see Table I.

Procedures

Data collection

Ethical considerations

The treatment procedure had been an established method at the university hospital speech-language pathology clinic for several years before study start and the evaluation was approved by the head of the relevant university clinic department. After receiving written and verbal information about the details of the study in adherence to the Helsinki Declaration, the parents gave their written informed consent.

In total, the probe measure included 45 structured opportunities, hereafter called the "probe total score", to produce the target sound /s/. Specifically, the probe measure included production of /s/ in different linguistic contexts corresponding to the steps in treatment (see Table II). A structured protocol and a material with pictures presented in a random order were used for the probe measurements. Since the children, due to age, were non-literate, they were asked to verbally repeat the depicted target sounds, syllables, words and sentences which were modelled by the SLP. The probed target words and sentences

were not the same as the ones used in treatment based on the recommendation by Maas et al. (2008). No suitable phonological or articulatory control measure could be found, since none of the participating children exhibited any other speech difficulties but active nasal fricative production substituting /s/ and, in two of the cases, /f/. Generalisation effects between the different linguistic contexts could be expected. Each probed linguistic context was thus considered to be a generalisation measure until the corresponding step had been introduced in treatment.

Baseline probes were collected at the initial assessment sessions at the clinic (see section *Participants and initial assessment*) and at the beginning of the first treatment session before the actual treatment began. Three baseline probes were aimed for in order to ensure stability of the children's attempted /s/-productions. For one participant, however, two baseline probes only were conducted to facilitate participant attendance, supported by a probe total score of 0 in both probes. Like the last baseline probe, treatment phase probes were collected at the start of each treatment session. Follow-up probes were planned to be collected approximately 1 and 10 weeks, respectively, after the last treatment session.

Audio recordings of probes were conducted in a sound proof booth at the clinic using the Soundswell WorkstationTM (Soundswell Signal Signal Workstation, 2020) (eNeovius Data och Signalsystem AB). An electret-condensed microphone (Sennheiser MKE 2) was used, positioned on a stick mounted on a head set so that the mouth-to-microphone distance of 15 cm could be constantly maintained. All recordings were stored directly in a computerised database. Directly after each therapy session, the treating SLP listened with head phones (Sennheiser HD205) to the audio recording of the probe recorded at the beginning of the session for decision on advancement to the next level in the program. The target /s/ in all probe items was assessed to be either correct (oral) or incorrect (nasal). Thus, different placements of oral articulation (interdental, palatal, lateral) were all judged as correct.

Treatment

The treatment included SLP therapy sessions at the university speech-language pathology clinic as well as training with parents at home. Participants received a motor-based speech treatment program, developed and clinically established at the university clinic, with the goal of achieving oral instead of active nasal fricative production of /s/. The treatment program was hierarchically structured and steps, which corresponded to different linguistic contexts, were introduced sequentially in a preset order, see Table II. The treatment was comprehensive in that repetitions of the articulatory target were conducted in several different linguistic contexts at diverse linguistic levels and multi-component in that the SLP used a variety of facilitating techniques. These, in turn, were used in an assortment of activities. Techniques included frequently demonstrating and modelling oral articulation of /s/ as well as contrasting oral versus active nasal fricative /s/-production, all while providing simple, comprehensible instructions. Auditory discrimination of oral versus nasal productions was also used. Further, the already mastered sibilantic sounds [c] and/or [J] were used to provide an experience of oral airflow in a fricative sound. The SLP also utilised the strategy of using the established consonant /t/ as a transition to oral /s/-production (/tsss/; e.g. Kummer, 2008). Moreover, extrinsic performance feedback (Ruscello & Vallino, 2014) from the SLP included comments both on the result, such as whether the child's articulation attempts were successful or not, as well as on the practice, such as specified information about the articulatory movement pattern just carried out by the child and in what ways it could be changed or refined. Intrinsic performance feedback (Ruscello & Vallino, 2014), i.e. biofeedback from the child's own sensory systems, included for example the SLP occluding the child's nose to achieve oral airflow (Golding-Kushner, 2001, chap. 6) as well as visualising oral airflow using a mirror. Finally, the SLP supported the child in developing self-monitoring of his/ her articulation by verbally reflecting on his/her attempts to produce oral /s/ in order for the child to become more aware. For materials and activities, see Table II.

The treatment sessions with the SLP lasted 45 minutes and were conducted approximately once a week at the clinic. Efforts were made to spread out the sessions evenly during the course of the treatment with the aim of moving to the next step once the probe of the corresponding linguistic context indicated 80% (Olswang & Bain, 1985) oral production of /s/ (performance-based criterion) or when a certain articulation step had been practised for two consecutive sessions (time-based criterion), whichever came first. Exceptions were the phoneme context, which was always introduced one session after the first step, and the non-cluster word contexts (initial, final and medial position), which were introduced during three consecutive sessions since those contexts were collapsed in the probe measure.

To increase dose frequency, a home training component involving the parents was included in the intervention. The parents were asked to regularly complete home assignments with the child in the time between sessions at the clinic by using the same structure and materials as the clinic SLP. They were advised to practise three to five times per week for at least five minutes, and possibly longer, per session, but only as long as the child was motivated and concentrated. Parents were asked to document the home training by marking each day that practice took place in a training log. Parents received information about

the general procedures of the treatment program, including its hierarchical structure, as well as of the facilitating techniques used. The accompanying parent always observed the session in the therapy room with the purpose of becoming familiar with the activities associated with the current treatment step. The parent received verbal instructions about the techniques to be used at home until the next session. The SLP also modelled how to carry out the suggested home assignments with the materials provided. At the following clinic session, the parent was encouraged to tell the SLP about the implementation at home and the SLP gave feedback and suggested changes as needed.

Reliability

Repeated assessments were performed on three probe sessions for each participant. Recordings of one probe session from each respective phase (A-B-FU) were randomly selected and copied onto a CD by an independent technician at the clinic, lending a total of 135 probe items (3×45) per child. Head phones (Sennheiser HD205) were used when listening to the selected reliability probes. The inter- and intra-rater reliability were measured in terms of percentage exact agreement. One external SLP, who had not been involved in the recordings or treatment of the participants, and one internal (the second author) performed the judgements independently and blindedly. Both SLPs had more than 20 years of full-time experience from the Craniofacial team at the university hospital. The probe items were judged as correct (oral) or incorrect (nasal) fricative production of /s/ but did not include judgement of articulatory placement. The mean inter-rater percentage of exact agreement (point-by-point) was 95.2% (ranging from 88.0 to 98.5%). The same evaluation was also compared to the initial probe assessments to examine intra-rater agreement for the SLP who had delivered the treatment (i.e. the second author). The mean percentage of exact (point-by-point) agreement was 91% (ranging from 85.0 to 100%).

Data analysis

Data analysis was conducted by calculation of mean probe total score per phase as well as of mean difference in probe total score between phases for each participant. In addition, graphs displaying probe outcome per session across all three phases for each participating child were visually inspected. Home training was descriptively evaluated by compiling the parent documentation.

Result

Mean success rates of the probe total score across study phases, mean differences in the probe total score between phases, total number of treatment sessions, total intervention duration as well as time from treatment end to FU1 and FU2 for Peter, Tom and Anna are presented in Table III. Figures 1-3 show results pertaining to the six linguistic contexts probed, as well as probe total score success rate, for each participant across all study phases.

Results showed that the participating children exhibited individual treatment response and progress patterns. Peter's response to treatment was direct and he rapidly learned as well as automatised the new articulatory motor program, reflected in a marked mean probe total score increase of 66.9 percentage points between phases A and B. Tom and Anna, on the other hand, both showed slower progress. Tom responded gradually to treatment, applying oral /s/ in the phoneme, syllable and non-cluster word contexts two to three sessions after those steps had been introduced. In contrast, Anna exhibited an inconsistent response pattern with unstable productions that fluctuated considerably between probe sessions, as reflected in the probe total score. Towards the end of phase B, progression then took off across linguistic contexts for both Tom and Anna, as indicated by steep positive slopes. Data also revealed improvements in probed linguistic contexts before introduced in treatment. This was the case for all participants, but to varying degrees. During baseline, all children showed total lack of orally produced /s/ in all linguistic

Table III. Mean probe total score across study phases, mean difference in probe total score between study phases, total number of treatment sessions, total intervention duration as well as time from treatment end to FU1 and FU2 for the three participants.

	Mean probe total score, raw score (%)		Mean difference in probe total score, raw score (%)		Total	Total	Time from treatment		
Participant	Phase A	Phase B	FU	Phase A to B	Phase B to FU	Phase A to FU	number of treatment sessions	intervention duration, weeks	end to FU1 and FU2, weeks
Peter Tom Anna	0.0 (0%) 0.0 (0%) 1.0 (2.2%)	30.1 (66.9%) 14.1 (31.4%) 14.7 (32.6%)	45.0 (100%) 45.0 (100%) 45.0 (100%)	30.1 (66.9%) 14.1 (31.4%) 13.7 (30.4%)	14.9 (33.1%) 30.9 (68.6%) 30.3 (67.4%)	45.0 (100%) 45.0 (100%) 44.0 (97.8%)	8 8 9	$11 \\ 11^{a} \\ 16^{b}$	11 and 25 ^c 1 and 13 1 and 10

Phase A: baseline; phase B: treatment phase; FU: follow-up.

Maximum probe total score = 45. ^aIncluding a three-week illness intermission with home-training only.

^bIncluding a five-week holiday with home-training only. ^cFollow-up probes were delayed due to summer holidays.

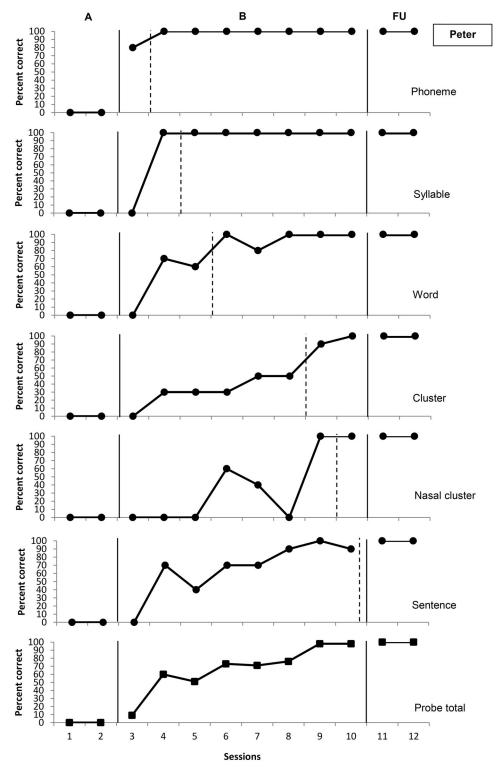


Figure 1. Percent correct (oral) /s/-production for Peter in the six linguistic contexts probed and in the corresponding probe total score across all study phases. A: baseline, B: treatment and FU: follow-up. Dotted lines indicate treatment start of a specific linguistic context.

contexts except for Anna, who exhibited a slightly rising baseline in the phoneme context. In the last probe of phase B, all children scored at, or close to, 100% in all linguistic contexts. At FU, these levels were maintained or increased. In probe session 11, Tom scored 100% in both types of wordinitial /s/-clusters as well as sentences. Hence, a decision was made to introduce both /s/-clusters with nasal consonant and sentences in the treatment session directly following.

Parent documentation indicated that home training was conducted relatively regularly, oftentimes on two to four days a week and usually lasting about 15–20 minutes. Unfortunately, however, parent documentation was incomplete and consequently, no systematic data can be presented.

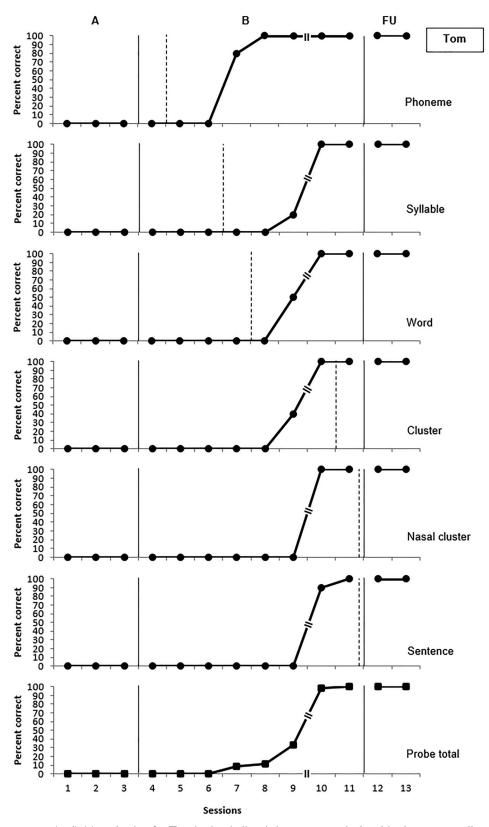


Figure 2. Percent correct (oral) /s/-production for Tom in the six linguistic contexts probed and in the corresponding probe total score across all study phases. A: baseline, B: treatment and FU: follow-up. Dotted lines indicate treatment start of a specific linguistic context. Hatched lines indicate a three-week treatment break.

Discussion

All three children in the current study successfully changed their production of active nasal fricatives substituting /s/ to oral articulation after participating in a motor-based speech treatment including home training. At baseline, Peter, Tom and Anna all showed total or near total lack of oral /s/-production. In contrast, at post-treatment follow-up, each participant produced /s/ orally in all 45 probe items,

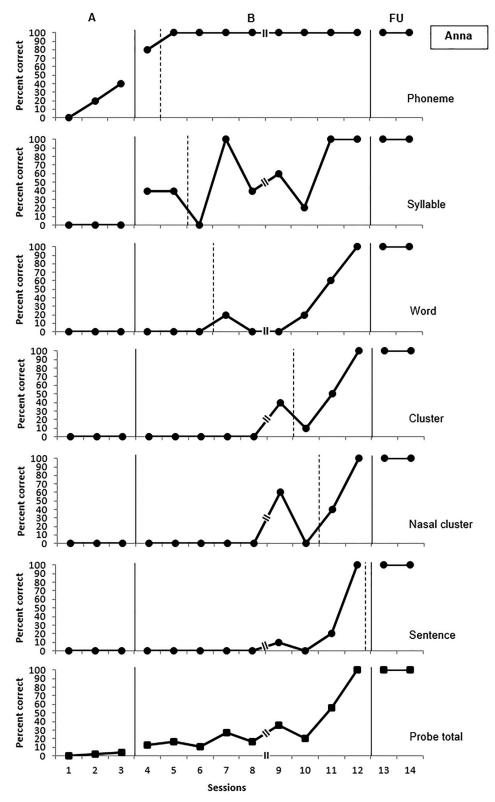


Figure 3. Percent correct (oral) /s/-production for Anna in the six linguistic contexts probed and in the corresponding probe total score across all study phases. A: baseline, B: treatment and FU: follow-up. Dotted lines indicate treatment start of a specific linguistic context. Hatched lines indicate a five-week treatment break.

spanning all linguistic contexts and production levels. This is in agreement with the, to our knowledge, two previous studies in the field (Mason et al., 2018; Ruscello et al., 1991). Like Mason et al. (2018), we also showed maintained stable oral production of /s/ in the long-term follow-up sessions. This preliminary

evidence suggests that the treatment program may have been effective, given the stable lack of correct productions before treatment onset in two of the three children, although it should be noted that we cannot rule out possible effects of confounding variables such as maturation or repeated testing (see also sections *Generalisation effects* and *Methodological strengths and limitations*).

Individual response and progress patterns

We found differentiated individual treatment response and progress patterns: direct, gradual or inconsistent treatment response and fast or slow treatment progress. Age might be an explanatory factor. The six-year-old (Peter) had a direct response and fast progress, comparable to the same-age boy in the study by Mason et al. (2018). In contrast, the four-year-olds exhibited gradual (Tom) and inconsistent (Anna) treatment responses and slower progress during the initial stages of treatment, but steep ascents towards the end. Lower age may, at least partly, explain Tom's and Anna's patterns, since their linguistic awareness, mental focus and self-monitoring skills - abilities important in motor-based treatment (Ruscello & Vallino, 2014) - presumably were less developed. An important question is therefore whether Tom and Anna would have progressed more quickly or recovered spontaneously had treatment been delayed. Occasional cases of spontaneously resolved active nasal fricatives have indeed been mentioned in the literature (e.g. Harding & Grunwell, 1998); however, there is little empiric data to rely on. Instead, like Zajac (2015), we argue that active nasal fricative production is deviant and important to treat early once established. These children also run the risk of being misdiagnosed as exhibiting velopharyngeal insufficiency with passive symptoms and, consequently, of being unnecessarily referred for consideration of surgical treatment (Zajac, 2015). Our data suggest that children as young as 4 years may benefit from a child centred motor-based articulation treatment, such as the current, a suggestion supported by Ruscello and Vallino (2014).

Generalisation effects

Improvements in one or more yet untreated linguistic contexts were seen for all three participants. Likely, such improvements reflect generalisation effects; however, alternative interpretations include testing effects due to repeated exposure to probes or maturation effects (Kennedy, 2005). Neither for Peter nor for Tom do testing effects seem likely since Peter showed a total lack of oral /s/-production during baseline followed by a direct and fast improvement after treatment onset, and Tom was exposed to many repetitions of probes before improvement in any context. Anna's improvements in phonemes and syllables could possibly be due to testing effects. However, since syllables started to improve after actual treatment onset, it seems more likely to be a generalisation effect assisted by structured guidance (Ruscello & Vallino, 2014) to produce an oral airstream during the first step (onomatopoetic fricatives). Maturation

effects do not seem probable for Peter because of his direct response and quick progress, whereas for Tom and Anna maturation cannot be ruled out. Tom's gradual response and slow progress could reflect maturation; though, the improvements in some untreated contexts towards the end of phase B may as well reflect generalisation. Further, maturation seems unlikely to have caused Anna's rising baseline in phonemes and improvement in syllables, since no other contexts advanced until later in phase B and the syllable progress was unstable. Yet, maturation may have occurred towards the end of phase B since it took Anna long to achieve stable improvement and when the progress rate finally increased, after an extended treatment phase, multiple contexts improved simultaneously. However, it could also be the case that Anna had grown older and thus had increased linguistic awareness, mental focus and selfmonitoring skills, which in turn resulted in a better treatment response. Finally, it is also worth mentioning that notes in Tom's and Anna's medical records indicated that at the end of their respective treatment periods, both had generalised oral production to their untreated active nasal fricatives substituting /f/; both children then produced oral /f/ in the structured treatment situation and Anna in spontaneous speech as well. In sum, due to generalisation effects, and possible confounds such as maturation and testing effects, we cannot be sure that the reported improvements were due to the treatment program. Thus, further research is needed with more controlled designs.

Home training

The home training may have contributed to the children in our study reaching the target behaviour more quickly by increasing the intensity in terms of dose frequency. Similarly, Mason et al. (2018) reasoned that parental involvement probably played an important role in the rapid articulation change for the boy studied. Indeed, using a blended approach, mixing clinician and parent training, might be a factor for success in articulation treatment (e.g. Lohmander, Henriksson, et al., 2010; Sugden et al., 2016) and might also be encouraged based on the oftentimes limited clinical resources available (Joffe & Pring, 2008; Lancaster et al., 2010).

Methodological strengths and limitations

A major limitation of the study design, and thus internal validity, was the lack of any suitable, untreated phonological or articulatory control target. Therefore, experimental control could unfortunately not be demonstrated per se. However, functional relations, i.e. systematic changes in the target behaviour due to intervention (Kennedy, 2005), were first observed after the first treatment step (onomatopoetic expressions) had been introduced (with the one exception of phonemes for Anna). It is also worth noting that even though the program was step-based and probes presented per corresponding linguistic contexts, the treatment was a given as a "package", which perhaps is best reflected in the probe total score. Moving to the next treatment step was based on the child managing 80% oral /s/-production in a linguistic context or having practised a step for two consecutive sessions. Clinically, however, some individual adjustments had to be made to sustain child interest or because of fast progress. Moreover, we used probe data with untrained items to make decisions about moving to the next step as recommended by Maas et al. (2008), who argued that regular transfer tests, rather than treatment practice data, reflect actual motor learning.

Additional limitations include the lack of data on number of target item repetitions and rate of correct child responses during clinical sessions. Accordingly, SLP treatment fidelity was not evaluated as such. Due to the clinical nature of the study, the exact number of target repetitions varied somewhat based on child interest and motivation. A summary of each treatment session documented in the child's medical record showed that the right treatment steps had been followed, which is also an aspect of treatment fidelity (Kaderavek & Justice, 2010). Documentation from the parents unfortunately was incomplete. Therefore, no systematic home training data could be presented. Nonetheless, the documentation indicated that training at home had been carried through on a regular basis. In addition, fidelity was enhanced by training parents through observation, verbal instructions, modelling and by encouraging parent reflection, supported by for example Kaderavek and Justice (2010) and Sugden et al. (2016).

The listening assessments only included judging oral versus (non-oral) active nasal fricative production and not articulatory placement of the /s/. Although results showed emerging and then established oral production of /s/, the consonant was produced at varying places of articulation and thus not correctly produced as such. However, to the ear, incorrectly produced oral /s/-production sounds much more correct compared to active nasal fricative production substituting /s/ and the change from nasal to oral was therefore the main purpose of the treatment. Judging /s/-productions as oral or nasal should be a straightforward task. Surprisingly, however, our study did not reveal excellent reliability. Deviant place of articulation of /s/ might have hampered the assessment and the use of phonetic transcription, including place of articulation, could have facilitated the task (Heselwood & Howard, 2008). Moreover, the treating SLP was the primary analyst, scoring probes after each session. The analyst was therefore not blinded in the first round of scoring the probes and there may have been perceptual drift that coincided with potential treatment progress, thus creating a potential confound when examining treatment effects. However,

when inter- and intrarater reliability was examined, both the primary analyst and the external analyst were blinded (see section *Method*).

Including place of articulation for /s/ as a treatment goal would have required normal hearing of the speakers. The hearing status of the participating children was not formally assessed. This was another limitation to the study, as was the fact that the children's middle ear effusion history could not be securely verified. Training of correct place of articulation would also have required reasonable dental occlusion. Reference data on typically developing Swedishspeaking children age 5 years reveal that approximately 30% produce /s/ with deviant place of articulation (Lohmander, Lundeborg, et al., 2017). Thus, correct place of articulation could not have been expected to occur in the participating children.

In spite of the listed methodological limitations, this single-subject study with its replication across three participants add to the very limited evidence base for treatments targeting active nasal fricatives in children with normal palate and velopharyngeal function. Still, due to the preliminary nature of this study, there is a need for further replication. To exert greater experimental control, future studies might use a multiple baseline across subjects design. Although, since the incidence of the particular articulation error is rare, a non-concurrent multiple baseline design (see e.g. Kennedy, 2005) might be a more feasible alternative, or else a multi-clinic study. Finally, to further strengthen the evaluation of treatment effects, a measure of intelligibility before and after treatment should be added.

Conclusion

In conclusion, the current study provides preliminary evidence suggesting that a motor-based, hierarchically structured intervention including home-based training may be effective in remediating active nasal fricatives substituting /s/ in children with normal palatal functioning. However, possible confounding effects such as maturation or repeated testing could not be ruled out; thus, results need to be replicated with increased experimental control and internal validity. Nevertheless, the study adds to the currently meagre empirical evidence-base for the population. Individual treatment response and progress patterns were found. Further, the data suggest that the intervention may be beneficial from 4 years of age.

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Declaration of interest

The authors report no declarations of interest.

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