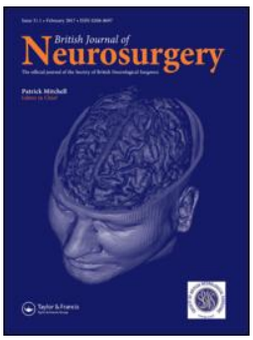

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SHORT REPORT

A case report of an intra-optic recurrent craniopharyngioma

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ABSTRACT

A 46-year old female patient presented with a left-sided superior temporal quadrantanopia due to a recurrent craniopharyngioma. The location of the recurrence was unusual. Imaging showed an enlarged left optic nerve, suggestive of a recurrent intra-optic craniopharyngioma. It was possible to remove the tumour without compromising the visual functions. In this report, we describe the case in further details.

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Craniopharyngioma; supra-sellar; intraoptic; recurrence

Introduction

Craniopharyngiomas (CP) are benign tumors of dysembryogenetic, non-glial origin with erratic growth patterns. Unfortunately, they often behave in a manner similar to aggressive tumors. Overall, they constitute 2–4% of intracranial tumors and are typically located in the infundibular region (sellar and suprasellar compartments).[1,2] CP can show high recurrence rates, based on the efficacy of the surgical excision and adjuvant therapy.[3] Most of the recurrences are located in the sellar and/or suprasellar areas.[2] They may compress the optic apparatus, hypothalamus and other adjacent structures.[2,3] Here, we describe an unusual location of recurrence. The recurrent CP was found within the boundaries of the left optic nerve.

Case presentation

A 46-year-old female patient was referred to our skull base team with a recurrent CP. The medical history was as follows; In 2004, she had undergone a right-sided pterional craniotomy for the removal of a typical CP of circa 4.5 cm on the axial plane in the infundibular region in a different Dutch centre. The presenting symptom was loss of visual fields. The location was predominantly suprasellar, and the lesion was compressing the optic apparatus and the surrounding structures (Figure 1(A)). Surgical removal was incomplete, and the patient developed an intermittent central adrenal gland insufficiency. In 2008, another surgical intervention was required due to regrowth of the residual tumour. A transnasal approach was preferred and performed in Italy (Figure 1(B)). Post-operatively, the patient developed panhypopituitarism. The transnasal surgery was complicated by CSF leakage requiring additional repair-surgery. In September 2017, the patient's vision started to deteriorate, with a left-sided loss of the temporal visual field. Perimetric examination showed a superior temporal loss in the left eye (Figure 2(A)), visual acuity was 0.4. The MRI scan showed an enlarged left optic nerve of approximately 11 mm suggestive of intra-optic recurrence extending towards the optic chiasm (Figure 1(C)). Due to progressive loss of left visual function, surgery was performed. Through a standard left-sided pterional craniotomy, we reached the optic nerve and found a clearly enlarged nerve, suggesting a mass lesion inside (Figure 1(D)). Interestingly, the nerve did not show any clear characteristics of a tumour, due to a layer of normal nerve tissue around it. Close microscopic inspection revealed a thin layer of nerve tissue on the lateral side of the nerve, close to the chiasm, through which a cyst-like appearance was visible. Microsurgical dissection through this surgical window allowed us to identify the tumour tissue. It was possible to perform a microdissection of the tumor, separating it from the optic nerve. The postoperative course was uneventful without further decline in vision. Twelve-months post-operatively, the patient indicated an improvement in vision. The visual acuity improved to 0.7. Perimetric evaluation showed also partial improvement of the visual field deficit of the left eye (Figure 2(B)). Post-operative imaging showed no signs of a residual tumour (Figure 1(E)). Pathologic examination confirmed the earlier diagnosis of a papillary CP.

Discussion

Recurrence remains one of the most important challenges in the management of CP. The risk of recurrence in partially resected tumours is reported as 25–100%, whereas in cases with gross total resection this risk is 0–26%.[4] Complete removal is not always feasible due to the adherence of tumour tissue to critical surrounding structures such as the hypothalamus. In cases where there is residual tumour, radiotherapy significantly decreases the rate of recurrence achieving a 90% progression-free survival at 10-year follow-up.[4,5] The recurrence rate is the highest in the first 3 years after surgery and most of the recurrences occur in the original area of the tumour.[1,2] Here, the location of the recurrence was unusual and complicated decision-making. Multidisciplinary discussion of the case in our skull base team, considering the advantages and disadvantages of radiosurgery vs.

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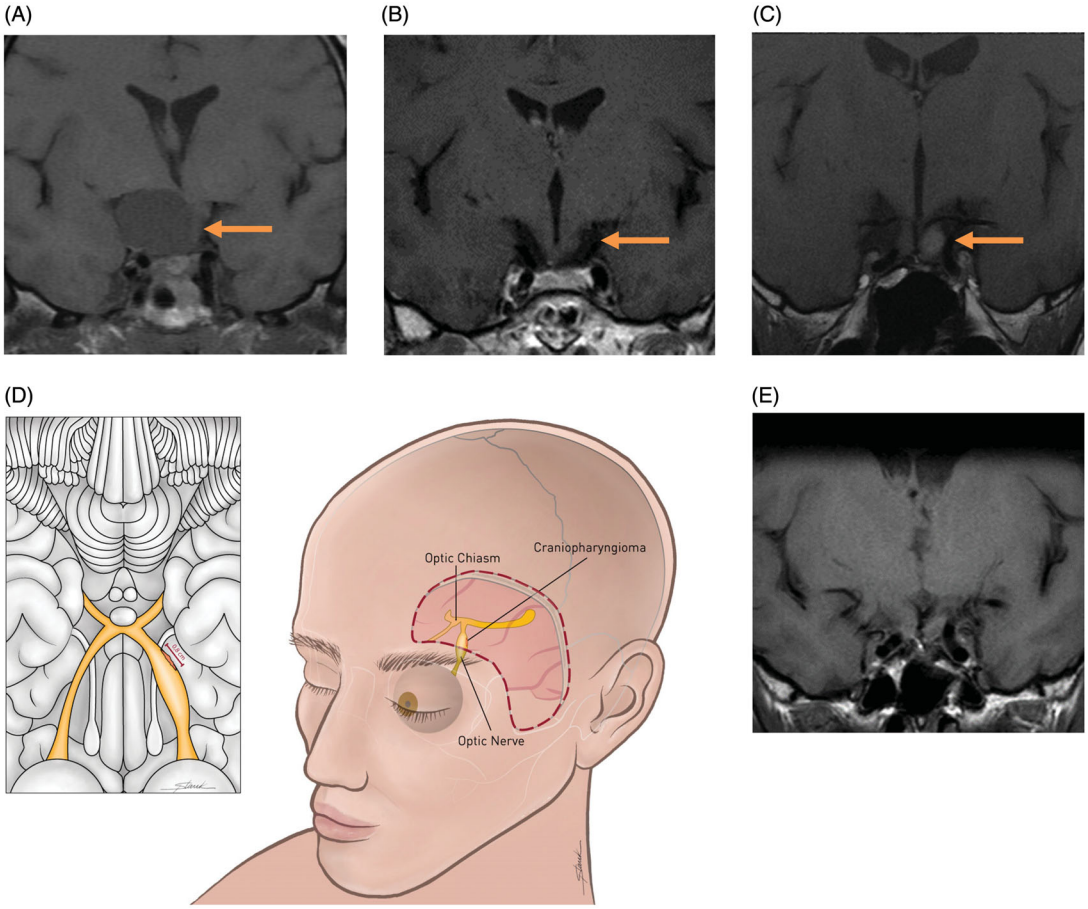

Figure 1. This figure shows the time course of the craniopharyngioma (CP) with routine MRI. Figure 1(A) is a 2004 coronal T1-W image at the level of the optic nerves in which the arrow points towards the tumour. Figure 1(B) shows a follow-up MRI from 2011 at a similar anatomical plane where there is no clear sign of recurrence. Figure 1(C) is again a similar MR-image demonstrating recurrent CP within the bounderies of the left optic nerve. Figure 1(D) is an artist drawing of this unusual recurrence. Figure 1(E) shows the MRI image 6 months after micro-surgery showing adequate removal of the tumour. Since the MRI of 2004 was without contrast-enhancement, we have inserted T1-W non-contrast enhanced images for comparison purposes.

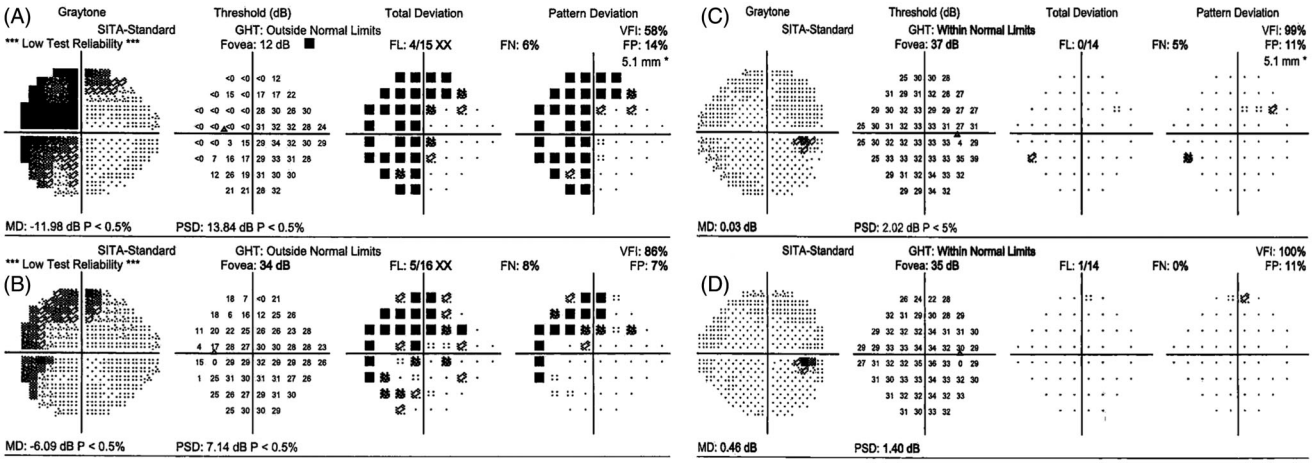

Figure 2. This figure shows the pre and post-operative perimetric examination of the left and right eye. Figure 2(A) shows the visual fields preoperatively and Figure 2(B) shows them postoperatively. Figures 2(C) and 2(D) represent the visual fields of the right eye.

surgery, considering the risk of complete visual loss, a surgical approach was preferred since this would give the highest chance of preservation of left optic nerve function.

Conclusion

In this case report, we share our experience with the management of an intra-optic recurrent CP.

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Disclosure statement

No potential conflict of interest was reported by the author(s).

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