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Case report

Cavernous sinus haemangioma with intrasellar extension mimicking non-functioning pituitary adenoma – A case report and review of literature

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ABSTRACT

Fifty-three-year-old male suffering from a presumptive non-functioning pituitary adenoma was qualified for stereotactic radiosurgery in our institution. Two attempts of surgical treatment were taken three months before radiotherapy. Excessive bleeding did not allow to remove the tumour or to take samples for histopathological examination. Diagnosis was put on the basis of radiological assessment and lack of hypersecretion of pituitary hormones. However, radiological reevaluation in our Institute revealed the presence of a well-bounded tumour invading the cavernous sinus with high contrast enhancement in FLAIR and T2-sequence. Moreover, a constriction of the normal pituitary gland with tumour mass was seen. The imaging features of the lesion finally led to diagnosis of cavernous sinus haemangioma with intrasellar expansion. The patient received radiosurgical treatment with the use of linear accelerator (LINAC). A dose of 7 Gy in one fraction was administered to achieve satisfactory local control, prevent potential further bleeding and reduce the risk of progressive neurological deficits. Stable size of the tumour and absence of any complications are confirmed in six years of follow-up.

1. Introduction

Cavernous sinus haemangioma is a rare tumour, confirmed in less than 1% cases of parasellar abnormalities.[1] Cavernous haemangiomas are thought to be hamartomas or malformations of the microcirculation.[2,3] The tumour is diagnosed mostly in middle aged women.[1,4] Headaches and cranial nerve deficits are the most common clinical presentation.[4] Interdural localisation (haemangioma is situated within the lateral wall of the cavernous sinus, between the outer dura propria and the inner membranous layer) and the presence of the pseudocapsule enables dissection of the tumour mass.[5–7] Contrary to cavernous malformations, cavernous sinus hae-

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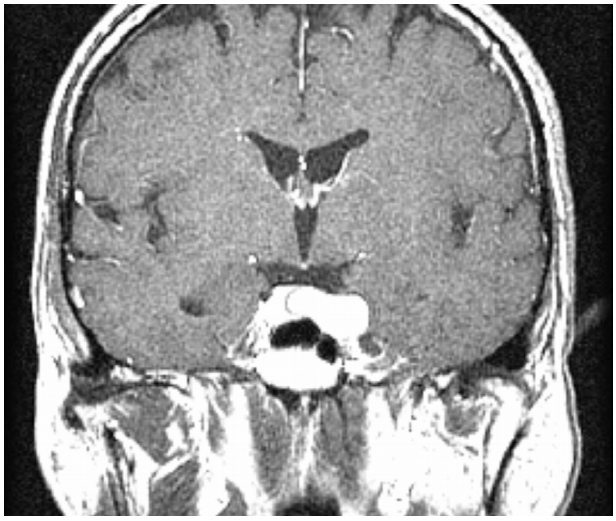

Fig. 1 – Preoperative coronal T1-weighted magnetic resonance (MR) images presenting tumour mass localised in the left cavernous sinus and sella turcica. Homogeneous contrast enhancement is seen.

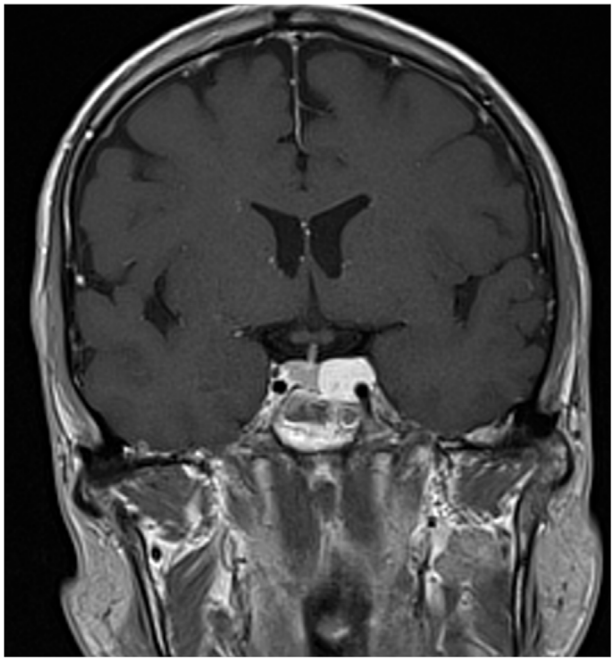

Fig. 2 – Postoperative coronal T1-weighted magnetic resonance (MR) presenting tumour mass localised mainly in the left cavernous sinus with normal pituitary gland constriction. Intense, homogeneous contrast enhancement is seen.

mangiomas grow over time and may infiltrate into the middle fossa.[8,9] Tumour is usually well bounded and grows without destruction of neurovascular structures in the cavernous sinus.[5] However, abundant vascularity makes surgical resection inconvenient.[1] Thus, radiosurgery is considered as an adjunctive or a primary therapy. In fact, optimal treatment in this rare neoplasm has not been established to date.

2. Case report

Fifty-three-year-old male suffering from a pituitary tumour was qualified for radiosurgical treatment at our Institution. His general status was good (WHO 0). There were no visual field deficits in ophtalmological examination. Hormonal blood tests did not reveal any abnormalities. Neurological symptoms were not present as well.

Six months before oncological consultation, the patient was admitted to the neurology department because of a transient ischaemic attack (TIA), manifesting as a transient right hemiparesis with aphasia. MR imaging revealed a presumable pituitary macroadenoma 17.6 mm × 18 mm × 14 mm in size with cavernous sinus invasion. Homogeneous contrast enhancement of the tumour was confirmed (Fig. 1).

Two attempts of neurosurgical treatment were taken in 3 and 3.5 months before radiosurgery. Excessive bleeding did not allow for excision of the well vascularised tumour mass. However, intraoperative Doppler vascular assessment revealed the presence of a marked arterial blood flow in the tumour. Samples for histopathological evaluation were not taken. Patient was referred to our Institution for further oncologic treatment. MR performed after surgery confirmed the presence of tumour mass localised in sella turcica and cavernous sinus.

Radiological reevaluation at our Institution revealed the presence of a well-bounded tumour involving the cavernous sinus. Analysis of the radiological features led to diagnosis of a cavernous sinus haemangioma with intrasellar expansion. The majority of the tumour mass was localised in the cavernous sinus with constriction of the normal pituitary gland. Intense postcontrast enhancement of the tumour was noticed in the dynamic phase of MRI, normal pituitary gland had significantly lesser degree of enhancement (Fig. 2). Tumour was hypointense on T1-weighted images and hyperintense on T2-weighted images. Boundaries of the lesion were well defined.

Radiosurgical treatment with the use of linac was performed in one fraction of 7 Gy. Radiotherapy was administered in order to achieve a satisfactory local control, prevent potential further bleeding and reduce the risk of progressive neurological deficits (Fig. 3).

In six years of follow-up, stable size of the tumour and absence of any complications are confirmed (Fig. 4).

3. Discussion

Cavernous sinus haemangiomas are not typical vascular neoplasms, but rather hamartomas or malformations of the microcirculation.[2,3] There are two histopathological types described by Zhou et al.[10] Type A, a sponge-like tumour, is characterised by the presence of a pseudocapsule. Type B is a mulberry-like tumour without or with an incomplete pseudocapsule. In our patient, histopathological assessment was not performed due to intense bleeding at the time of surgeries. The final diagnosis was based on radiological features of the tumour.

The most common signs of the haemangioma localised in the cavernous sinus are: headache, dysfunctions of the cranial

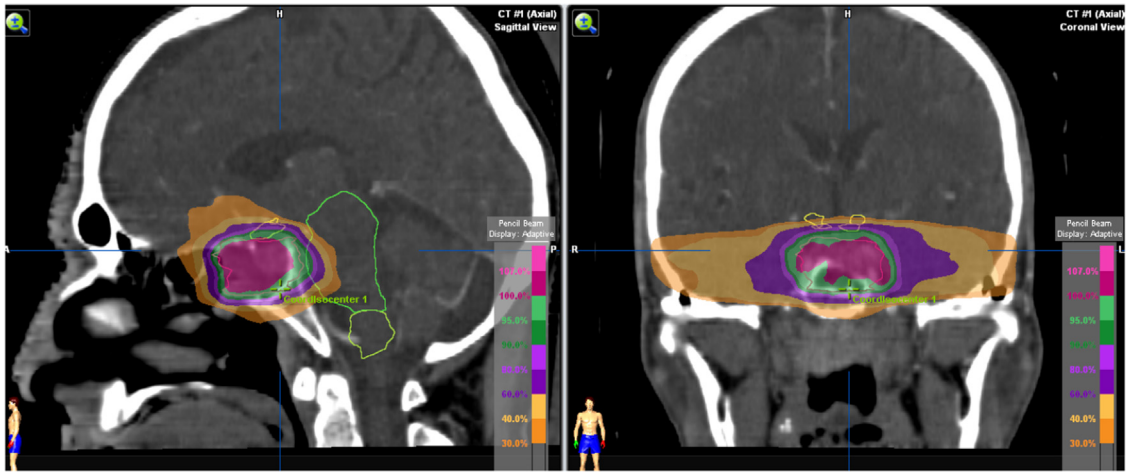

Fig. 3 – Treatment plan applied in this patient with the use of a linac equipped with an mMLC: 7 Gy in one fraction was delivered. Homogeneous dose distribution with maximum sparing of healthy tissues is seen.

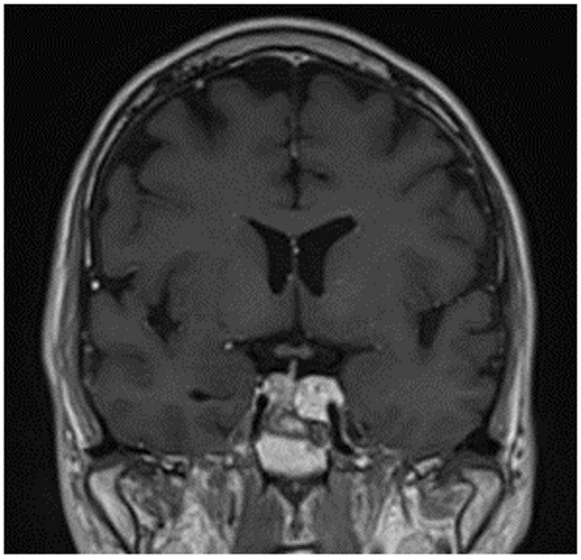

Fig. 4 – Postradiosurgical coronal T1-weighted magnetic resonance (MR) presenting haemangioma localised in the left cavernous sinus and sella turcica. Stable size of the tumour is confirmed six years after the treatment.

nerves (placed in the cavernous sinus) and ocular pain. Haemorrhage manifesting as a sudden neurological deterioration is uncommon.[10–12] However, in our patient a TIA was the first clinical manifestation of the tumour.

Not surprisingly, radiological evaluation was crucial in our patient. Especially, in view of the fact that cavernous sinus haemangiomas are difficult to distinguish from other lesions situated in this region (neurinoma, pituitary adenoma, meningioma). The rate of preoperative misdiagnosis ranges from 66.7% to 87.5%.[10–12] In our patient, haemangioma was mistaken for pituitary adenoma. Typical presentation of pituitary adenoma on MR is hypo- to isointensity on T1-weighted images and variable intensity on T2-weighted images.[11] In contrary, haaemangiomas have a marked signal hyperintensity on T2-weighted MR imaging. Sharp, well bounded margins and dumbbell-like appearance are seen.[1,4,13] The aforementioned distinguishable marks with homogeneous postcontrast enhancement made the diagnosis unambiguous in our case.

Neurosurgery is performed as a primary treatment in almost all cases of cavernous sinus haemangiomas. If the complete resection is possible, surgical treatment is sufficient to cure the patient.[14] However, the threat of excessive bleeding and the presence of critical anatomical structures are important issues in qualification of patients for neurosurgery.[11,12,15] Thus, radiosurgical treatment is usually considered as a valuable adjunctive option, especially after nonradical neurosurgery.[11] Radiosurgery is also used as an effective primary therapy.[5,15] Surgical excision is considered in young patients and in case of large tumours. Radiosurgery is usually performed in patients suffering from comorbidities for small lesions presenting indisputable radiological characteristics, and as an adjuvant therapy.[5,16] Optimal treatment paradigm is not defined to date.[5]

Radiosurgical treatment of this rare entity is usually performed with the use of GammaKnife (GK).[1,2] The mean marginal doses applied in GK SRS range from 10 to 19 Gy.[1,16–19] A similar case of cavernous sinus haemangioma mimicking pituitary adenoma was described by Hori et al.[11] In that case, CyberKnife radiosurgery was conducted, central dose of 21 Gy in 3 fractions was delivered in order to prevent further bleeding. Authors confirmed decreased size of the tumour in the first year after radiotherapy. Similar treatment (CyberKnife radiosurgery) was performed by Wang et al.[17] Applying 21 Gy in 3 fractions lead to the shrinkage of the tumour in all irradiated patients. According to the studies describing outcomes of Gamma Knife radiosurgery, satisfactory local control with decrease in risk of haemorrhage (especially after the first symptomatic haemorrhage) is noticed.[11,20] Noteworthy, many investigators revealed substantial regression of the tumour[1,16–19] induced by vessel obliteration as a consequence of irradiation.[16] Hayashi et al. confirmed significant volumetric tumour response. In their study, 83% of tumours had more than 50% of volume reduction without any complications.[1] Furthermore, Wang et al. noticed a significant shrinkage of

67.8% of the irradiated tumours.[17] Similarly, Bansal et al. achieved remarkable tumour volume reduction in almost all cases.[16] Despite the device used for radiosurgery, the goal of the treatment remains the same - satisfactory tumour control and prevention of bleeding, especially in cases of haemangioma disqualified from surgery. In our patient, radiosurgical treatment was conducted with one fraction of 7 Gy. The dose was lower than doses applied in the cited investigations. Nevertheless, stabilisation of the tumour was achieved. Noteworthy, dose of 7 Gy was efficient not only for radiological control. There was no evidence of bleeding in our case. In six years of follow-up, no late toxicity of the treatment was observed.

4. Conclusions

Treatment paradigms in the case of cavernous sinus haemangiomas are not precisely defined. However, radiosurgery is worth considering especially in cases of inoperable or partially resected tumours.

Conflict of interest

None declared.

Financial disclosure

None declared.

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