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Case Report

CASE REPORT

'Coexisting pituitary adenoma and suprasellar meningioma—a coincidence or causation effect: report of two cases and review of the literature'

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Abstract

Coexistence of pituitary adenoma (PA) and another type of brain tumor is a very rare clinical scenario. Even though such a presentation can be an incidental event but a thorough review of the literature will be made to elucidate the possible mechanisms and treatment options in similar cases. Two cases of concomitant sellar and suprasellar/diaphragmatic tumors are reported. A 37-year-old lady with prolactinoma and a suprasellar diaphragmatic meningioma and a 42-year-old acromegalic man with suprasellar/diaphragmatic meningioma and a PA. Both meningiomas were removed transcranially. The prolactinoma could be managed medically and the growth hormone secreting adenoma was removed trans-sphenoidally. The visual problems and hormonal imbalances of both patients improved postoperatively. The literature is reviewed on this topic and the possible pathogenesis and management protocol of similar lesions are discussed.

INTRODUCTION

Meningiomas comprise 15–25% of all intracranial neoplasms [1, 2] and pituitary adenomas (PAs) are common benign neoplasms, with a prevalence of 10–23% in unselected [3, 4]. The coexistence of PA and another type of brain tumor is a very rare clinical scenario [5, 6]. The type of PAs reported in such series varied from non-functional to functioning adenomas [2, 5, 7–10]. In cases of PA concurrent with meningioma, GH-secreting adenoma is the most predominant [8, 9]. Even though prolactinoma represent the most common type of PA in adults (70%), the association of prolactinoma other primary brain tumors is a relatively rare occurrence [4, 5, 8].

The pathogenesis of coexistence of different lesions in the sella and suprasellar region has not been elucidated [8].

We do not intend to report collision 'intrasellar' pathologies but, two cases of concomitant brain tumors (CCBTs) in the sellar region are reported. Reviewing the literature for this association, we could find only five similar reports (Table 1). 'Even though this situation can be a co-incidence', we will discuss briefly, the 'possible pathogenesis and management protocol' of similar lesions.

Case 1

A 37-year-old lady presented with 8 months history of oligomenorrhea receiving LD tablet (Ovocept-LD, Aburaihan Co.) but headache, diplopia, progressive visual impairment and persistent oligomenorrhea did not improve.

Decreased visual acuity (VA) to 80/200 with a left temporal hemianopia were detectable. The non-contrast computed tomography scan showed iso- to hypodense suprasellar lesion. Magnetic resonance imaging (MRI) revealed a well-delineated round tumor

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Table 1 Showing the reported similar cases in the literature

	Sex, age	PA	Meningioma	Treatment
O'Connell 1956	F, 47 y	Inactive intra- extrasellar	Meningothelial suprasellar	Not stated
Probst 1971	M, 48 y	Cushing's disease intrasellar	Meningothelial suprasellar	TSS for adenoma and craniotomy for meningioma
Wild and RIP 1974	n.s.	Inactive	Fibroblastic	Not stated
	n.s.	Intrasellar	Parasellar	
Abs *et al.* 1993[1]	F, 82 y	Prolactinoma	Suprasellar	Subfrontal resection for meningioma, 3 months later resection tumor via TSS because decreased vision
	F, 47 y	Inactive-intrasellar	Parasellar	TSS approach for resection of the tumor. Partial resection had been achieved
Hainer *et al.* 1978	M, 72 y	Eosinophilic	Suprasellar	Not stated
Bunick *et al.* 1978[5]	F, 57 y	Eosinophilic	Planum sphenoid	Both tumors was removed via a right subfrontal approach
Zentner 1989	M, 46 y	Prolactinoma	Planum sphenoid	Both tumors was removed via a right sided pterional approach
Laun A *et al.* 1993	F, 61 y	GH producing	Tuberculum sellae	Both tumors was removed via a left-sided pterional approach
Jaskolski DJ 1990	M, 81 y	Inactive-intrasellar	Suprasellar meningioma	Both tumors was removed via a right subfrontal approach
Görge HH 1993	M, 53 y	Prolactinoma	Para- and suprasellar	Both tumors was removed via a left-sided frontolateral approach
Prevedello 2007	F, 52 y	Inactive-macroadenoma	Tuberculum sellae	Endoscopic transnasal resection of the pituitary tumor, then the planum sphenoidale was drilled and the suprasellar tumor was completely resected
Yu-Jen Lu *et al.* 2008	F, 52 y	Inactive-intrasellar	Tuberculum sellae	Endoscopic endonasal trans-sphenoidal intrasellar tumor resection performed then Left frontotemporal craniotomy was performed for resection of meningioma
I. Poeata 2010	F, 56 y	Inactive-intrasellar	Parasellar	An extended right temporo-pterional approach was used to reach access to both tumors
Mahvash M, *et al.*, 2014	F, 36 y	Inactive-intrasellar	Suprasellar	Endoscopic endonasal approach
Our cases	F, 37 y	Prolactinoma	Suprasellar	Craniotomy for meningioma + medication for prolactinoma Craniotomy + TSS approach
	M, 42 y	Nonfunctional	Suprasellar	

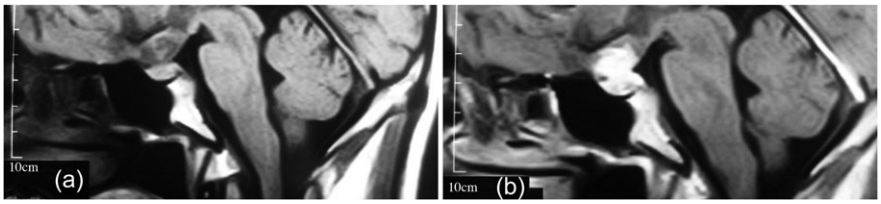

Figure 1: (**a**, **b**) MRI revealing a well-delineated round tumor 30 × 25 × 20 mm in diameter, T1W isointense and T2W hyperintense lesion located within the sella turcica and another dural-based lesion lying over the diaphragm sella extending to the planum sphenoidale. The intrasellar lesion showing a faint enhancement after contrast material injection but the suprasellar lesion has a bright enhancement.

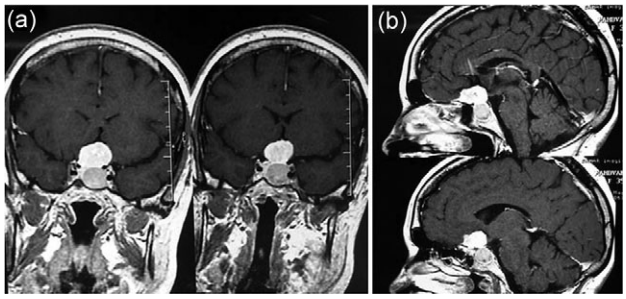

Figure 2: MRI showing (**a**) an intrasellar lesion isointense in T1W and T2W images enhancing homogenously and mildly after contrast material injection and (**b**) a suprasellar lesion 3 × 3 × 2 cm in diameter with the same intensity in T1W and T2W images which enhanced notably after contrast material injection.

30 × 25 × 20 mm in diameter, T1W isointense and T2W hyperintense located within the sella turcica and another suprasellar lesion with the same intensity. The intrasellar lesion showed a faint enhancement after contrast material injection but the suprasellar lesion had a bright enhancement with the base of the lesion creeping over the diaphragma sella and jugum sphenoidale in the meantime (Fig. 1). Serum prolactin level was 840 ng/dl (3–31 ng/dl). She was managed with Cabergoline (Dostinex tablet 0.5 mg, Pfizer Co.) during the previous 4 months without any improvement in her vision. Decompression of the optic apparatus via a right pterional craniotomy and total removal of the suprasellar tumor achieved using microsurgical technique. Diaphragm sella was intact and we did not attempt to excise the intrasellar lesion. Improvement of her vision was remarkable and further medical treatment was undertaken using 1.5 mg Dostinex weekly.

Case 2

A 42-year-old man was referred with acromegaly. Serum GH level was 65 IU (up to 5 IU) and IGF1 890 IU (up to 370 IU). His VA decreased during the previous 6 months to 40/200 with bitemporal hemianopia. MRI revealed an intrasellar lesion isointense in T1W and T2W images, enhancing homogenously and mildly after contrast injection. There was also a suprasellar/diaphragma lesion 3 × 3 × 2cm in diameter with the same intensity in T1W and T2W images, which enhanced notably after contrast injection (Fig. 2). It could not be verified whether the lesions were of the same nature or 'double coexisting lesions'. A trans-sphenoidal microscopic adenomectomy was performed. The normal looking pituitary gland and intact diaphragm prohibited us from extending our search beyond the diaphragm. The overnight serum GH level was 10 IU.

Considering no improvement in his VA, a right pterional craniotomy was performed and a purplish, lobulated, meaty tumor uplifting the chiasm and optic nerves could be excised gross totally. The diaphragm sella and jugum sphenoidale were the areas coagulated and curetted to achieve Simpson I tumor excision followed by remarkable improvement of vision. There has been no recurrence of any of the tumors after 9 y.

DISCUSSION

Coexisting PA and suprasellar meningioma are a rare occurrence. Our search in PubMed and Google Scholar and handhold identification of the references of each article revealed 15 similar cases reported in the available literature including the five supasellar/diaphragmatic cases included in Table 1.

Considering MESH terminology, there have been different descriptions used to define coexistence of more than one tumor; 'collision tumors' are those with infiltration of a tumor by another type of tumor while 'coincidental tumors' are synchronous tumors of different histogenesis in contiguous or far from each other [7, 9]. We would like to suggest that the coexisting tumors in cases like ours are most probably of coincidental type rather collision tumors and not of the neurocutaneous disorders such as Neurofibromatosis Types I and II.

Several mechanisms have been suggested for triggering development of multiple primary brain tumors of different histology in a single patient, still the etiology remains unknown [2, 7, 9]. Some believed that their immunohisto-chemical analysis showed that concurrent adjacent double tumors occur because of activation of the signaling pathways of receptor tyrosine kinases or, one tumor may secrete a growth factor that initiates growth of another lesion [2, 7, 9]. It is yet to be investigated whether in GH-producing adenoma, might induce arachnoid cap cell transformation to meningioma [8, 9]. The other mechanisms hypothesized for the development of multiple tumors are exposure to the offending biochemical substances, genetic factors, prior trauma and surgery [7, 9].

In our first case, the serum prolactin level was 840 ng/dl and the stalk effect was almost excluded.

It is important to distinguish between an adenoma with suprasellar extension and an adenoma coexisting with a suprasellar meningioma because the treatment strategy for these tumors are different, even though no pathognomonic radiological characteristic is seen in imaging of some of these cases such as ours.

Several authors suggest that of both tumors should be removed in one session [10]. If removal of the tumors is not possible in one session, it is important to decide which tumor should be operated on first. Some suggested that avoiding the complications associated with the transcranial approaches, extended trans-sphenoidal surgery might be a good alternative choice [10].

'The ideal surgical approach' for treatment of such concomitant tumors is not well supported by clear evidences in the literature [1, 6, 10].

CONCLUSION

High quality imaging with good resolution and specified techniques can preclude loosing golden time for preservation of vision in these CCBT and surgical approach should be tailored according to the patient's symptom, the anatomical characteristics of the mid skull base region and the feasibility of resection of two tumors in one session.

CONFLICT OF INTEREST STATEMENT

None declared.

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