Recurrent Intrasellar Meningioma After Total Excision In a Woman With Hyperprolactinaemia

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Abstract : Intrasellar meningioma is a rare disease which is difficult to diagnose preoperatively. We describe an interesting case of intrasellar meningioma in a 39–year–old woman who presented with amenorrhea, galactorrhea and severe headaches for six months. An intrasellar mass was demonstrated by computed tomography. The mass was approached by the transcranial route,an intrasellar tumour was found pushing against the diaphragma sellae.After complete removal of the tumour, the patient was discharged with resolution of visual and endocrine abnormalities and normal serum prolactin level. Histopathological examination revealed meningioma. Within six, months, the lesion reappeared with the same visual and endocrine abnormalities. Computed tomography revealed a contrast enhancing intrasellar lesion and serum prolactin level was again elevated. At reexploration, the tumour was excised. Pure subdiaphragmatic intrasellar meningiomas simulating prolactinomas are unusual lesions.Such early recurrences may be attributed to incomplete resection or to the histological character of the tumour. In spite of the rarity of intrasellar meningioma, preoperative differentiation from a pituitary adenoma is vital for the proper surgical approach.

Key Words: Intrasellar meningioma, prolactinoma, rhinoseptal route, subfrontal route.

INTRODUCTION

Meningiomas constitute approximately 15 % of all intracranial neoplasms and are the most common primary non-glial lesions (3). A suprasellar location is the fifth most common site of origin with 5 % (13). Suprasellar meningioma (tuberculum sellae,clinoid processes, diaphragma sellae) may grow downward into the sella turcica. But pure subdiaphragmatic intrasellar meningiomas are well known though uncommon lesions. These tumors seem to be exceptional and are thought to arise within the sella turcica and extend superiorly into the chiasmatic cistern, causing visual and endocrine disfunction by compressing optic pathways and pituitary stalk. There is some data in the literature referring to this uncommon location (7,8,9,11,14,15). An unusual case of intrasellar meningioma causing hyperprolactinaemia, mimicking prolactinoma and early recurrence of the tumour with endocrine abnormality is reported with a brief discussion of possible surgical routes and diagnostic criteria.

CASE REPORT

This 39 year–old woman who earlier had regular periods for years presented with a 6 months' history of headache, amenorrhea and galactorrhea, and deterioration of vision. On examination corrected visual acuity was 20/30 on the right and 20/25 on the left. A left superior temporal quadrantanopia and a right temporal hemianopia were present on formal testing of the visual fields. X–rays of the skull and tomograms of the sella turcica showed sellar enlargement with anterior and inferior erosion (Fig.1). Coronal and axial cranial CT revealed an intrasellar lesion with homogenous contrast enhancement extending superiorly into the chiasmatic cistern (Fig.2). Carotid angiography showed bilateral slight upward displacement of the A1 segments of the anterior

Fig. 1 : X–ray of the sella showing discrete enlargement of the sella turcica with anterior and inferior erosion.

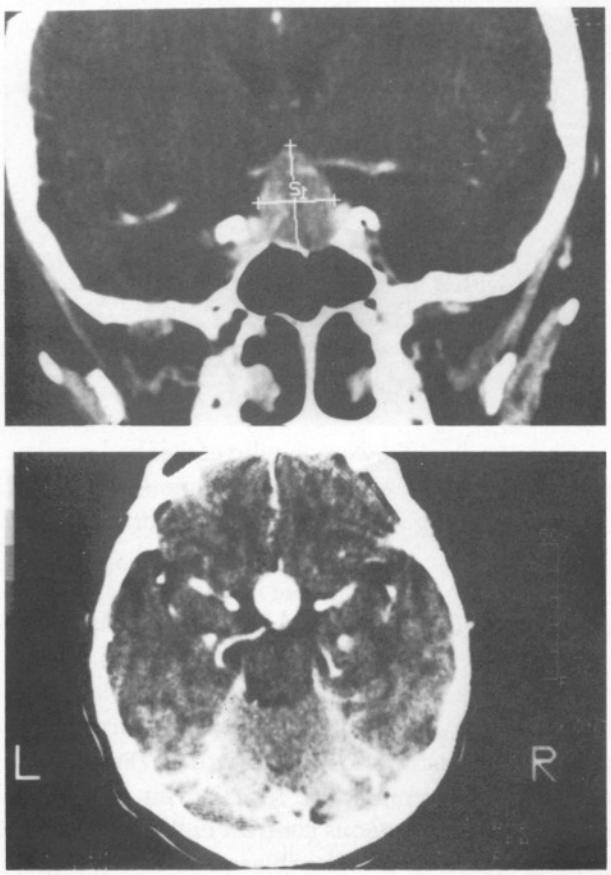
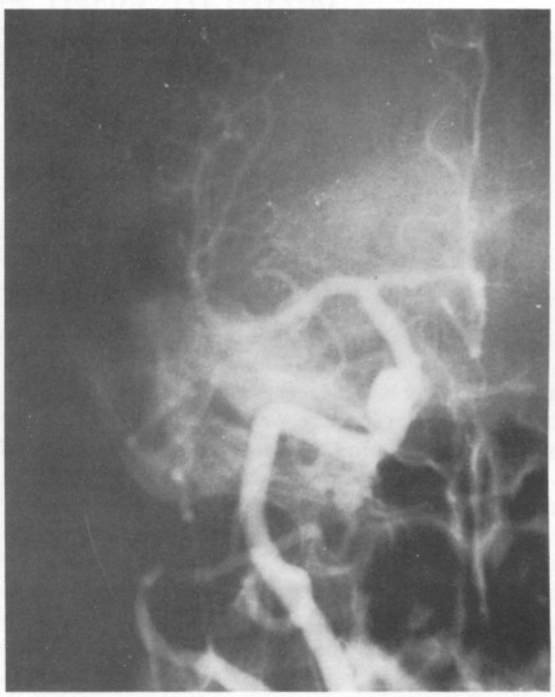


Fig. 2 : CT scans after administration of intravenous contrast medium demonstrating homogeneous intrasellar mass with suprasellar extension on coronal section (A) and mass in suprasellar cistern on axial section (B). No hyperostosis is seen.

cerebral artery without evident tumoral blushing (Fig.3). The serum prolactin level was 98 ng/ml (normal < 24 ng/ml), and other routine endocrinological tests were normal.Thus with the diagnosis of probable prolactinoma, the patient was operated upon



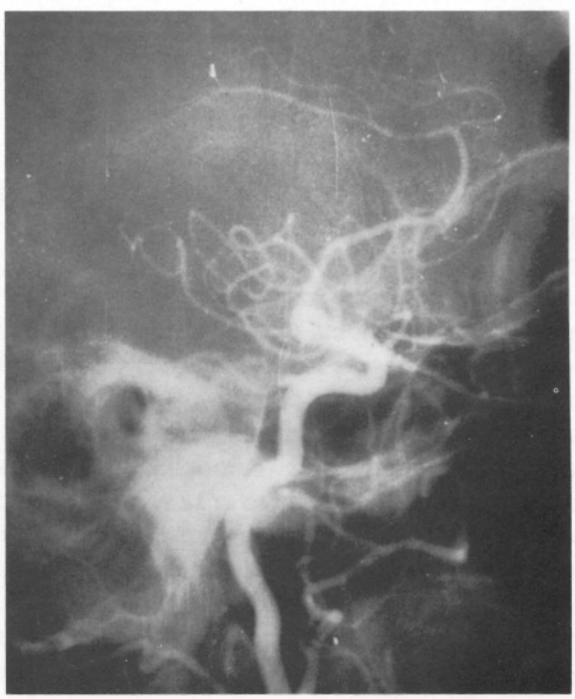


Fig. 3 : Angiography of the right carotid artery. The A1 segment of the anterior cerebral artery is elevated (A) and the lateral view shows no intrasellar or suprasellar blush (B).

by a right subfrontal approach. Surgical exploration revealed an intrasellar tumour pushing against the diaphragma sellae and the optic nerves which was removed totally sparing the pituitary gland. A surprising finding was bleeding during the operation. Macroscopically, the tumour was well circumscribed and globe shaped. The sectioned surface was gray and firm. The specimen was Stained for S-100 protein*, Vimentin** and epithelial membrane antigen (EMA)*** Microscopically, the tumour was composed of elongated cells in sheets and fascicles (Fig.4).

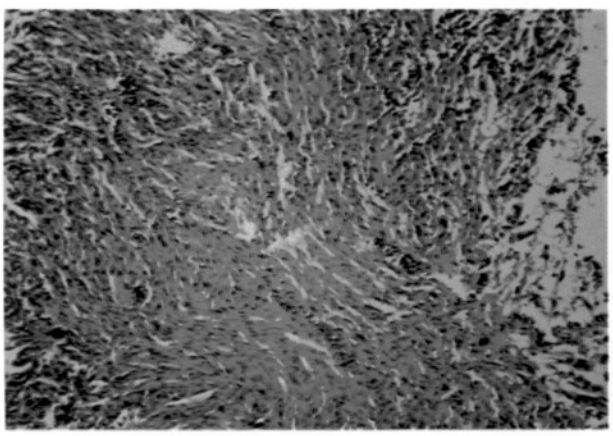


Fig. 4 : The tumour is composed of markedly elongated cells H.E x 150

The nuclei of the cells were elongated and dense in chromatin, but oval nuclei with delicate chromatin were seen. The tumour cells were positive for S–100 protein, vimentin and EMA (Fig.5,6,7). There was no mitosis or necrosis. Histopathological diagnosis was meningioma. Postoperatively, the patient's visual acuity and visual field improved gradually and the serum prolactin level declined to 24 ng/ml.Postoperative CT scan showed no residual tumour.Six months after the operation,the patient was readmitted with severe headache.

On reexamination, the patient's corrected visual acuity was 20/40 on the right and 20/60 on the left, a slight bitemporal hemianopia was present. CT scanning again revealed a heterogenous contrast enhancing intrasellar lesion with moderate suprasellar extension (Fig. 8). The serum prolactin level was elevated to 40 ng/ml. At subfrontal reexploration tumour was removed which was histologically

* S–100 (DPC, Cat. No: CKS1S, California, USA)
** Vimentin (DPC, Cat. No: CKVNS, California, USA)
*** EMA (DPC, Cat. No: CKEMS, California, USA)

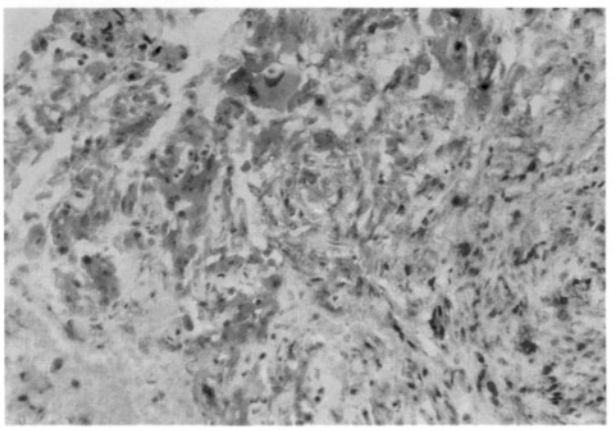


Fig. 5 : Tumour cells showing membranous and cytoplasmic EMA positivity ABC X 375

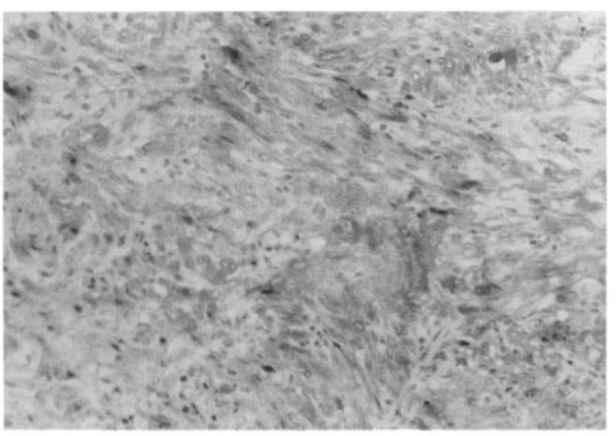


Fig. 6 : S–100 protein–positive tumour cells ABC X 375

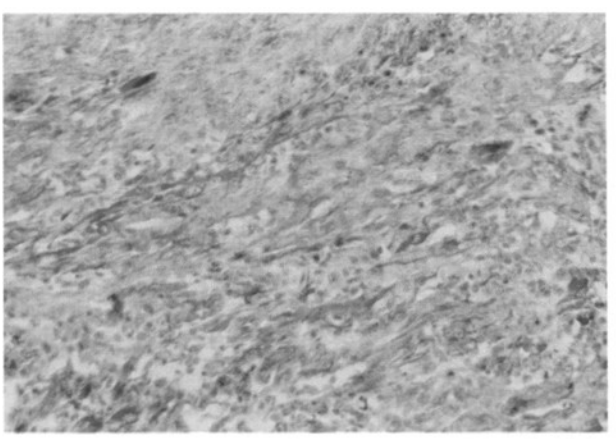


Fig. 7 : Tumor cells showing Vimentin–immunoreactivity ABC X 375

identical to the original specimen. Following the second operation, the patient's visual acuity and visual field improved and serum prolactin level declined to 20 ng/ml. Eights months after discharge, the patient was normal.

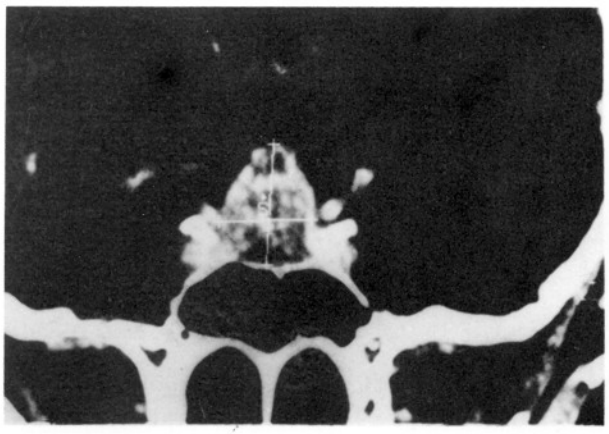


Fig. 8 : CT scan after administration of intravenous contrast medium showing heterogenous intrasellar mass with suprasellar extension on coronal section.

DISCUSSION

Intrasellar meningioma may develop from the lateral or inferior wall of the sella turcica or from the diaphragma sella (8).Although intrasellar meningioma is never reported in the large series of meningiomas of the sellar region (1,10,12), several case reports have appeared in recent literature on the successful surgical treatment of intrasellar meningioma by subfrontal or transsphenoidal route (7,8,9,11,14,15). Many authors described satisfactory resolution of the preoperative visual and endocrinological deficits, without any major complications and recurrence. Postoperative follow-up averaged one year.To the best of our knowledge no recurrence, in such a short period,has previously been reported following excision of an intrasellar meningioma. The cause of the early recurrence in this case is unknown and may be related either to the histological character of the tumour or to microscopic residual tissue.

A rational surgical approach to intrasellar meningioma is complicated by a general inability to correctly diagnose these uncommon lesions prior to operation. The most sensitive imaging technique for the diagnosis of intrasellar meningioma is MR.

Endocrinological findings of intrasellar meningioma may be similar to those of the non–functioning pituitary adenoma (8,15) or prolactinoma, as in our case.Increased prolactin secretion level was rarely reported with intrasellar meningiomas.

It is very difficult to differentiate intrasellar meningioma from other intrasellar neoplasms because the X–ray findings resemble to those of other neoplasms.But a meningioma should be suspected if bony hyperostosis is evident on plain X–ray.

CT scan is not specific in the differential diagnosis of intrasellar meningioma, adenoma or other intra lesions because pituitary adenoma can grow in any direction in the sellar region and can not always be differentiated from other intra–parasellar lesions based on morphology.Although intrasellar calcification favors a diagnosis of meningioma,it may rarely be seen with adenomas.

Although angiography may indicate an intrasellar meningioma by showing an intrasellar tumour blush,Cophignon et al (4) also reported tumour blush in pituitary adenoma.But angiography did not show a marked blush in our case as in the case of Grisoli et al. (8).

Magnetic resonance (MR) imaging is an effective modality in the differential diagnosis intrasellar region.Identification of the diaphragma sella with MR can be used to differentiate a pituitary tumor from a suprasellar mass,elevated in the former but depressed in the latter (5). But it is not useful for differentiation of a pituitary tumour from an intrasellar meningioma since the diaphragma sella elevated in both lesions. Meningiomas have shown significant enhancement on Gd–DTPA–enhanced MR studies but pituitary adenomas usually demonstrate only slight and inconsistent enhancement (2,6). Thus enhanced MR imaging may be the best way to differentiate intrasellar meningioma from pituitary adenoma.

A few reports of treatment of an intrasellar meningioma without complication by the transsphenoidal route appear in the literature (8,15).Grisoli et al (8) recommended the transsphenoidal route for biopsy or removal of every intrasellar tumour with or without suprasellar expansion.Since there are few reports of treatment of this lesion by this route, the actual incidence of recurrence and its reliability is unknown.

We believe that subfrontal route should be the preferred surgical approach since the tumor is adherent to the meninges. The transsphenoidal route has complications as CSF leak, massive hemorrhage and incomplete excision. These drawbacks are

avoided by the subfrontal route along with better visualization of the suprasellar component of the tumor allowing complete excision.

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