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Review article

Spindle cell oncocytoma: Report of two cases with massive bleeding and review of the literature

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ABSTRACT

Spindle cell oncocytoma (SCO) is a rare pituitary tumor, classified as a WHO grade I neoplasm. Due to its rarity, SCO is often preoperatively misdiagnosed as a pituitary macroadenoma. In the present study we report two recent cases of SCO, a 61-year-old male and a 65-year-old female presenting at Treviso General Hospital between March 2014 and April 2015. Tumor resection was achieved by endoscopic transsphenoidal approach but massive hemorrhagic events hampered surgery, endangering the patient's life in both cases. Both tumors featured fascicles of spindle cells with eosiniphilic cytoplasm expressing vimentin, S-100 and thyroid transcription factor-1 (TTF-1). The diagnosis of SCO was confirmed by second opinion in both cases. Extensive review of available literature, about 30 cases from 2002 to 2015, provided valuable clinical data for preoperative diagnosis and surgical removal of SCO tumors.

1. Introduction

Spindle cell oncocytoma (SCO) is a rare non endocrine neoplasm of adenohypophysis that was first described by Roncaroli et al. in 2002 [1], and was lately included in the WHO Classification of tumors of the Central Nervous System in 2007 [2]. Since then, only 28 cases have been published in English literature [1–25]. SCO is an oncocytic, non-secreting, benign neoplasm of the anterior pituitary gland that manifests in adults, with a peak incidence between the VI and VII decades of life, without any sex predilection [5]. Clinically and macroscopically it can be indistinguishable from a non-functioning pituitary macroadenoma [2]. Spindle cell oncocytoma is composed of mixed fascicles of spindle to epithelioid cells with oncocytic changes, showing an immunoreactivity for anti-mitochondrial antibody 113-I, as well as to vimentin, S-100 protein, EMA and TTF-1, with no expression of synaptophysin or pituitary hormones [4]. The genesis, progression and prognosis of SCO remain uncertain and need to be documented further [7]. Two main characteristics of SCO are the firm consistency and the high vascularization, that may prevent complete surgical resection and account for a non-negligible risk of bleeding [12,24]. We report our experience in the management of two cases of SCO with a different intraoperative and post-operative behavior. In addition, we extensively review the literature.

1.1. Case presentation

1.1.1. Case 1

A 61 year-old man presented to our Neurosurgery Department in March 2014 complaining of a headache and showing clinical signs of mild hypopituitarism. He had a medical history of ischemic heart disease treated with angioplasty, with coronary stent on right and left coronary artery, and lifetime antiplatelet medical prophylaxis. Symptoms started in November 2013 with the onset of asthenia, dyspnea and profuse sweating. MRI scans showed a sellar-sovrasellar mass with a size of 22 mm × 18 mm × 27 mm (Fig. 1a and b), involving the sphenoidal sinus and chiasmatic cistern. The neoplasm showed homogenous contrast enhancement on T1-weighted MRI scans. There was no involvement of the cavernous sinus but there were signs of severe compression of the pituitary peduncle, which could not be identified. The imaging was suggestive for pituitary macroadenoma. At the time of presentation, the patient had no evidence of neurological deficit and hormone replacement therapy was prescribed. In March 2014, he underwent endoscopic transsphenoidal surgery. The resection

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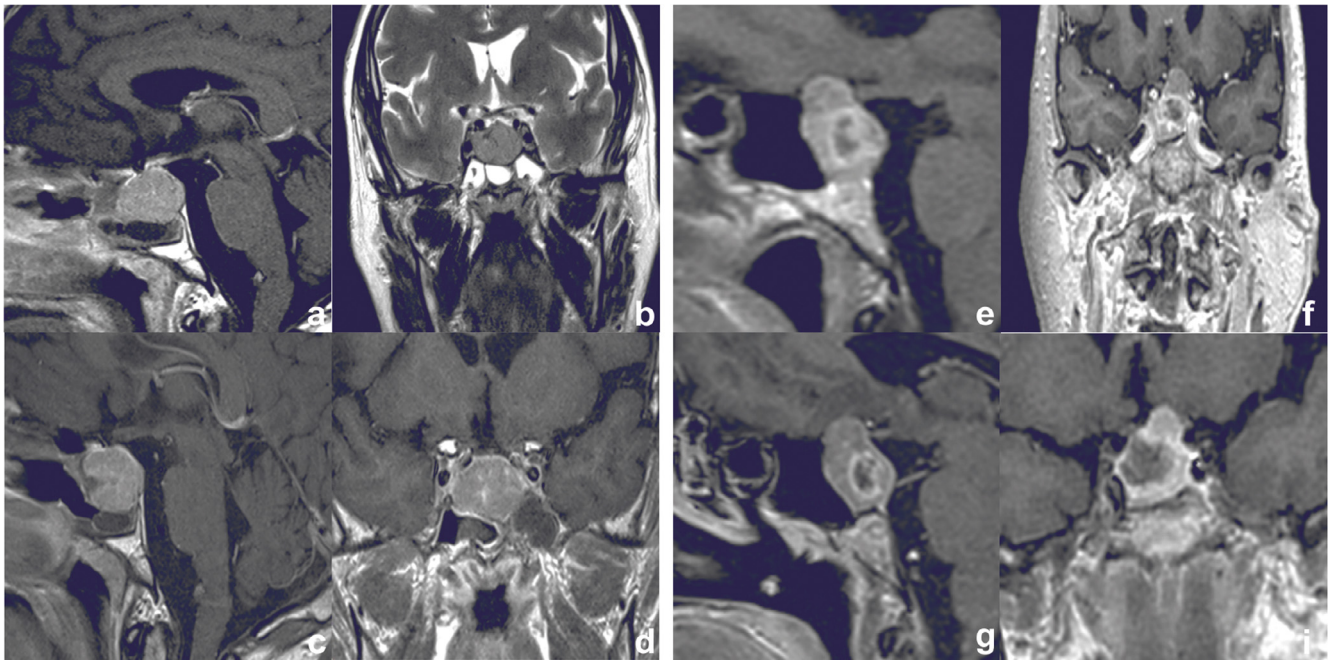

Fig. 1. Imaging. *Case 1.* (a, b): preoperative sagittal and coronal T1-weighted contrast-enhanced MRI scans: sellar-suprasellar mass with homogeneous contrast enhancement. (c, d): postoperative sagittal and coronal T1-weighted MRI scans: evidence of partial resection. *Case 2.* (e, f): Preoperative sagittal and coronal T1-weighted contrast-enhanced MRI scans: large sellar-suprasellar mass with involvement of the optic chiasm and left optic nerve; inhomogeneus contrast enhancement and central area of necrosis. (g, I): postoperative sagittal and coronal T1-weighted MRI scans: signs of partial resection.

was incomplete, as a massive bleeding, resistant to platelets administration, fibrin glue, cellulose gauze (Tabotamp) and heamostatic matrix (Floseal) application, prematurely interrupted surgery. Intraoperatively the tumor appeared firm and hypervascularized. Histopathological examinations showed a proliferation of spindle cells, arranged in short fascicles with eosinophilic cytoplasm (Fig. 3A). Ki67 was expressed in 2% of the neoplastic cells. The tumor was positive for vimentin, TTF1 and S100 protein, whereas synaptophysin, GFAP, NF, Neu-N, HMB-45, CD117, PLAP, CD30, and CK-CAM5.2 where negative (Fig. 3B and D). Silver stain highlighted the presence of abundant reticuline fibers (Fig. 3C). Based on the morphological and immunophenotypical features a diagnosis of SCO was made. On June 2015 the patient underwent a new attempt of transsphenoidal resection of the residual tumor. This time antiplatelet drug was suspended 15 days before the operation, to allow a complete medication washout. As in 2014, the surgery was characterized by profuse bleeding that again prevented complete tumor resection (Fig. 1c and d). Intraoperative haemostasis was attempted with standard heamostatic agents, as described previously, with also the administration of tranexamic acid without results. Furthermore, administration of plasma expanders and blood components was necessary to restore normal haemodynamic parameters. After surgery, minding of his ischemic coronary artery disease, the patient was admitted to our intensive care unit (ICU) for intensive care follow up. Postoperatively we proceeded to perform a diagnostic brain angiography: the tumor had afferent vascularization by meningeal branches of both siphons, with a small contribution from the left internal maxillary artery. The patient was discharged with no neurological deficits. After 14 month from the second surgery no increase in residual tumor size has been documented.

1.1.2. Case 2

A 65 year-old woman presented to our Neurosurgery Department in April 2014 with severe headache and visual deficit. Her history started in November 2012, when she had experienced left third cranial nerve palsy with ipsilateral loss of visual acuity. CT scan had shown a sellar-sovrasellar mass with a size of 20 mm × 15 mm × 10 mm, with inhomogeneous contrast enhancement and erosion of dorsum sellae on T1-weighted MRI images, suggesting a pituitary macroadenoma (Fig. 2E and F). MRI scans confirmed cavernous sinus involvement and intralesional necrosis. Serum hormone and electrolytes were indicative of mild hypopituitarism. At that time we only prescribed hormone replacement therapy. Interestingly, her familial history highlighted that her brother underwent surgery for pituitary adenoma at another Neurosurgery department. In February 2014, MRI showed an increase of tumor size, with compression of the chiasm and the left optic nerve and a more extensive involvement of the right cavernous sinus compared to the previous MRI. In April 2014 the patient underwent partial endoscopic transsphenoidal resection. The tumor was firm, fibrotic and highly vascularized. Debulking was hampered by bleeding and the surgery was prematurely interrupted (Fig. 2G and H). Bleeding stopped only after several application of hemostatic agents as cellulose matrix sheets (Tabotamp), gelatine sponge (Spongostan) or fibrin glue. Histopathological examination showed long fascicles of spindle cells with eosinophilic cytoplasm. Rare mitoses were observed (Fig. 3E). Immunohistochemical analysis showed that the tumor was diffusely positive for Vimentin, S100 protein and TTF1; Ki67 was expressed in 8% of neoplastic cells (Fig. 3F and G, I). After gradual improvement of visual acuity, the patient was discharged. Nine days later, she presented at the hospital emergency room with copious epistaxis from the right nasal cavity and a remarkably low hemoglobin level (decreased from 10.1 to 8.7 gl/dl in a matter of hours). The bleeding vessel turned out to be the right spheno-palatine artery, which was promptly coagulated. Re- examination of the pre-operative imaging unveiled anastomoses between the spheno-palatine artery and the ethmoidal artery, a tumor afferent vessel. After cessation of bleeding and normal haemodynamic parameters re-establishment, the patient was discharged. We did not documented recurrences after 28 months of follow up.