Coincidental pituitary adenoma and planum sphenoidale meningioma mimicking a single tumor☆

Adenoma hipofisario y meningioma de planum coincidentes simulando un tumor único

The association of pituitary adenoma and meningioma has been well described, especially in patients previously irradiated for the pituitary tumor who develop intracranial meningiomas years after treatment.[1] However, the coincidence of both tumors in contact has been seldom reported.[2–4] We present a case of contiguous pituitary adenoma and planum sphenoidale meningioma mimicking a single tumor which could be removed through an endoscopic expanded endonasal approach (EEA).

A 61-year-old patient, without previous oncological history or radiotherapic treatment, was referred to our institution after six months of progressive visual loss and bitemporal hemianopia that was confirmed by campimetry and optical coherence tomography. Neurological examination and endocrinological testing were otherwise normal, with the following serum levels: prolactin (PRL) 8.5 ng/ml; growth hormone (GH) 0.12 ng/ml; insulin-like growth factor I (IGF-I) 122 ng/ml; adrenocorticotropic hormone (ACTH) 28 pg/ml; cortisol 26.2 μg/ml; thyroid-stimulating hormone (TSH) 2.14 μUI/ml; thyroxine (T4) 1.74 ng/dl; follicle-stimulating hormone (FSH) 4.7 mUI/ml; luteinizing hormone (LH) 2.9 mUI/ml and testosterone 5.37 ng/ml. Magnetic resonance imaging (MRI) showed a 2.0 cm × 2.2 cm × 2.2 cm homogeneously enhancing sellar lesion with suprasellar extension compressing the optic chiasm (Fig. 1A and B). The pituitary stalk was displaced posterosuperiorly, though no normal gland could be identified on the MRI. The suprasellar

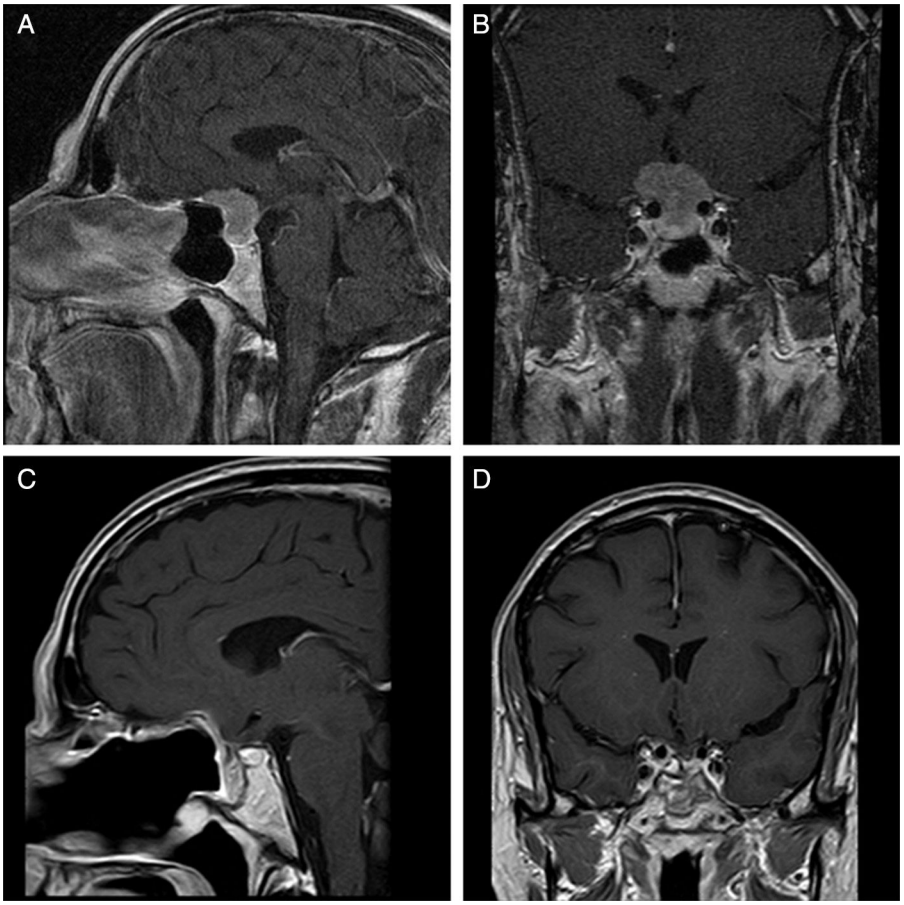

Figure 1 Preoperative magnetic resonance showing an apparently single sellar lesion with suprasellar extension to the tuberculum sellae and planum sphenoidale: (A) sagittal gadolinium enhanced T1-weighted image and (B) coronal gadolinium-enhanced T1-weighted image. Postoperative study with the same sagittal (C) and coronal (D) sequences shows a complete resection and sealing of the skull base with a nasoseptal flap.

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extension was broad based, reaching above the tuberculum sellae and planum sphenoidale. A non-functioning pituitary macroadenoma was suspected and the patient was offered surgery. An endoscopic transphenoidal approach with 0° and 30° rigid nasal endoscope was performed. The rostrum sellae was completely removed up to the margins of both carotid arteries. Cranially only a limited transtubercular and transplanum exposure was deemed necessary since the soft consistency of most pituitary adenomas allows a complete dissection and removal of the whole tumor after internal debulking if the suprasellar component is not exceedingly big. After successful resection of the sellar tumor, leaving glandular tissue attached to the pituitary stalk posteriorly, the diaphragm was opened and the suprasellar component was dissected from the optic chiasm and removed. Intraoperatively the suprasellar component appeared to have a stiffer consistency than the sellar tumor. After surgery the patient showed slight visual improvement. He developed hypopituitarism and persistent diabetes insipidus that were managed medically. The postoperative computed tomography (CT) suggested a tumor remnant at the planum sphenoidale. Meanwhile, histological study demonstrated the coincidence of two contiguous tumors: a pituitary adenoma with positive immunohistochemical staining for GH and LH, and a meningothelial meningioma (Fig. 2). Moreover the patient developed an intermittent CSF rhinorrhea when standing up. Therefore a reoperation was planned to remove the residual tumor and seal the CSF fistula. The patient was re-operated on day 8 through an expanded endoscopic transplanum approach that allowed the excision of the meningioma with decompression of both optic nerves. Effective sealing of the skull base was achieved through a multilayer closure with intradural and epidural fascia lata and a pedicled nasoseptal flap (Fig. 1C and D). The patient experienced a significant and stable visual recovery that allowed him to resume a normal life and return to work.

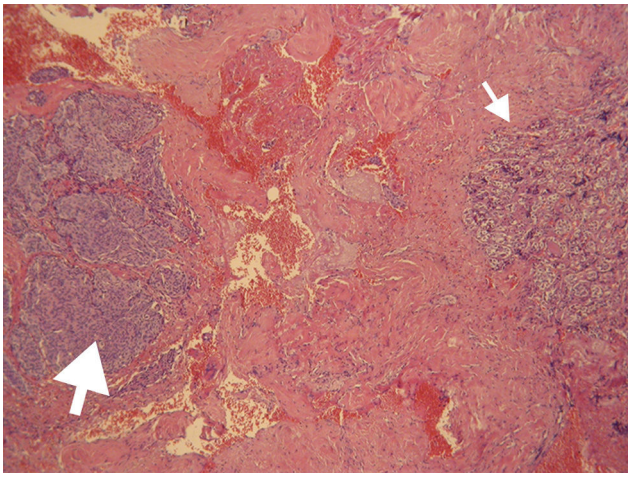

Figure 2 Photomicrograph of a section of the lesion that shows two different neoplastic tissues in contact (hematoxylin and eosin, 100×): clusters of densely packed cells of a meningothelial meningioma (big arrow) and a distorted acinar pattern of chromophobe cells corresponding to a pituitary adenoma (small arrow).

Radiotherapy is a well known cause of development of meningioma; several cases have been reported after irradiation of pituitary adenomas. On the other hand, coincidental meningiomas in patients diagnosed of pituitary adenoma and without previous irradiation are uncommon, with only 33 cases reported in the world literature.[3] The reported cases of coincidental pituitary adenoma and meningioma show a clear preference of the meningioma for a perisellar location at the planum sphenoidale, tuberculum sellae and the sphenoid wing.[1,3,5] Contiguity of the meningioma with the pituitary adenoma mimicking a single mass is extremely rare.[2,4] Coincidental menigiomas have been reported in patients with non-secreting adenoma, prolactinoma, acromegaly and Cushing disease. Although prolactinomas are the most frequent secreting adenomas, the higher prevalence of acromegaly among patients with coexisting meningioma has led some authors to propose an association between GH-secreting adenomas and the development of meningioma.[5,6] However, although meningiomas may express GH and IGF-I receptors and may show growth response to its stimulation in vitro, this effect has yet to be proved in vivo.[7]

The diagnosis of a meningioma in a close location to a pituitary adenoma poses a therapeutic challenge. In most reported cases the coexisting tumors were managed independently, usually addressing the pituitary adenoma first with either medical therapy or a transphenoidal approach, and treating the menigioma separately with conservative measures or a specific surgical approach.[1] Some authors used a single pterional approach for both tumors.[2,4,8] Any of these strategies implies a significant increase of the risk of morbidity as compared to the surgical treatment of a single pituitary adenoma. In the present case a solitary pituitary adenoma with suprasellar extension was suspected on the preoperative MRI; therefore an endoscopic endonasal approach was performed. Because of the presence of residual tumor on the postoperative CT and the pathological diagnosis of a coexisting meningioma, a reoperation through an endoscopic transplanum EEA was decided. This approach offers a direct view of suprasellar meningiomas without frontal lobe retraction and allows resection with minimal bleeding and good control of the optic apparatus and the anterior cerebral arteries.[9,10] Only one previous case of contiguous pituitary adenoma and suprasellar meningioma successfully resected through an endoscopic EEA has been reported.[3] The present case adds to the evidence that in the rare situation of contiguous sellar and suprasellar tumors located in the same sagittal plane, an endoscopic EEA allows an adequate exposure and safe removal of both tumors.

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Exclusively based parenteral fish-oil emulsion (Omegaven®) in the treatment of parenteral nutrition associated liver disease: Report of a case☆

Emulsión lipídica exclusiva de ácidos grasos omega-3 (Omegaven®) en el tratamiento de la hepatopatía asociada a nutrición parenteral: a propósito de un caso

Lipid emulsions incorporated into parenteral nutrition (PN) make it possible to provide essential fatty acids and to cover energy requirements without excessive carbohydrate content. On the other hand, both the composition of these emulsions and the dose supplied are related to the development of liver disease.[1,2]

Several articles on the effect of intravenous lipid emulsions based on omega-3 fatty acids (W3FA) alone in the treatment of liver disease in adults have recently been published.[3–7]

We report the case of an adult patient with the longest treatment period reported in Spain who showed evidence of laboratory and functional improvement with this type of lipid emulsion.

This was a 44-year-old female patient on PN since 2010 after massive bowel resection for a desmoid tumor which left a residual bowel consisting of 12 cm of duodenum, half the transverse colon, and the left colon. In addition to cyclic nutritional support, she was given from the start treatment with ursodeoxycholic acid and antibiotics for bacterial overgrowth to prevent the development of liver disease. The length of the remaining small bowel precluded enteral nutrition.

During 2010 and 2011, the lipid formula administered consisted of medium-chain triglycerides (MCTs), soybean oil, and W3FA (5:4:1 ratio) at doses of 1.1–1.4 g/kg/day, carbohydrate:lipids ratio 60:40, and a nitrogen solution with a high concentration of branched amino acids (16 g nitrogen/day). In view of liver enzyme elevation, trace elements were replaced in October 2011 by another product with a lower manganese content, which resulted in liver function improvement at the end of 2011.

After work-up at a reference hospital for bowel transplant, a liver biopsy was performed in April 2012. It revealed parenchymal and canalicular cholestasis in the liver parenchyma consistent with the changes induced by PN. The bilirubin level increased from 5.1 mg/dL to 15.4 mg/dL after the liver biopsy (complicated by a hematoma and hemoperitoneum). Because of this sudden liver function impairment, lipid administration was suspended for 3 weeks, but was restarted in June using a different lipid emulsion (a physical mixture of soybean oil, MCTs, olive oil, and W3FA in a 3:3:2.5:1.5 ratio) at a dose of 25 g/day, which was gradually increased in the following months up to 50 g/48 h. Partial improvement of the bilirubin level was found by the end of the year. When lipid provision decreased, the patient required supplementation with vitamins K and A.

During 2013, because of liver function improvement and progressive weight loss (lowest weight achieved in April, 44.5 kg; BMI 16.6 kg/m^2), lipid provision was increased up to a maximum of 65 g/day for 4 days per week and 40 g/day on the other days (Fig. 1).

Regular ultrasound examinations performed during follow-up showed a liver of normal size, morphology and echogenicity, splenomegaly, and biliary sludge. In October 2013, ultrasound examination showed evidence of portal hypertension (with no evidence of esophageal varices in gastroscopy) with a slightly enlarged portal vein with hepatofugal flow. Fibroscan® tests performed in 2012 and 2013

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