Clinical Neurology and Neurosurgery 115 (2013) 1180–1183

Contents lists available at SciVerse ScienceDirect

Clinical Neurology and Neurosurgery

journal homepage: www.elsevier.com/locate/clineuro

Case report

Epidural intracranial metastasis from benign leiomyoma: A case report with literature review

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ARTICLE INFO

Article history:
Received 14 May 2012
Received in revised form
11 September 2012
Accepted 28 October 2012
Available online 28 November 2012

Keywords:
Benign metastasizing leiomyoma
Smooth muscle tumors
Extra-axial brain tumors
Ovarian leiomyoma

1. Introduction

Uterine leiomyomas are benign tumors of smooth muscle origin believed to result from a clonal proliferation of uterine smooth muscle tissue [1]. They are the most common gynecologic tumors diagnosed in women of reproductive age, and the most common indication for hysterectomy. Rarely, a benign uterine leiomyoma can be found in sites of the body other than the uterus. It is called a benign metastasizing leiomyoma (BML). BML is a slow growing metastatic tumor, usually associated with a current or prior history of a uterine leiomyoma. The lung is the most common site of BML, but extrauterine locations such as the pelvic cavity, retroperitoneal space, and even the lymphatic and vascular channels have also been reported. BML of the brain is extremely rare and very few cases have been reported [2]. It has similar radiologic features as a meningioma, and consequentially has often been misdiagnosed. Ovarian leiomyomas are rare as well, and constitute less than 1% of all benign ovarian tumors. The treatment of BML is surgical resection of the mass to exclude malignancy, and the outcome is generally good. We present a case of BML of the brain and the ovary in a woman with prior hysterectomy, along with a succinct review of the disease.

2. Case report

A 43-year-old woman, gravida four, para two, presented with symptoms of headache, blurred vision, diplopia and tinnitus that had persisted for one month. The tinnitus was initially treated at a local Otorhinolaryngology and had gotten better. Visual disturbances had persisted and she was diagnosed with papilledema of the left eye at a local Ophthalmology. Regarding other past histories, the patient had received a total hysterectomy for a uterine leiomyoma 6 years ago. No other past medical history existed. On admission, ophthalmological examination was done to evaluate diplopia and blurred vision. Intraocular pressure was 14 mmHg/16 mmHg (normal range: 10–20 mmHg). The bilateral pupils showed good light reflex without RAPD (relative afferent papillary defect). The conjunctiva was normal and the cornea was clear for both eyes. However, there was swelling on the margin of the optic disc on the fundus of both eyes. The patient complained of headache as well. An intracranial pressure of 30 mmHg (normal range: supine position –7–15 mmHg, vertical position-negative (−10 to −15 mmHg)) was measured so a lumbar puncture was done to sample CSF (cerebral spinal fluid) for analysis. No abnormalities could be found. Additional evaluation of raised intracranial pressure and papilledema was needed. A magnetic resonance imaging (MRI) of the brain was performed for the possibility of

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0303-8467/$ – see front matter
http://dx.doi.org/10.1016/j.clineuro.2012.10.028

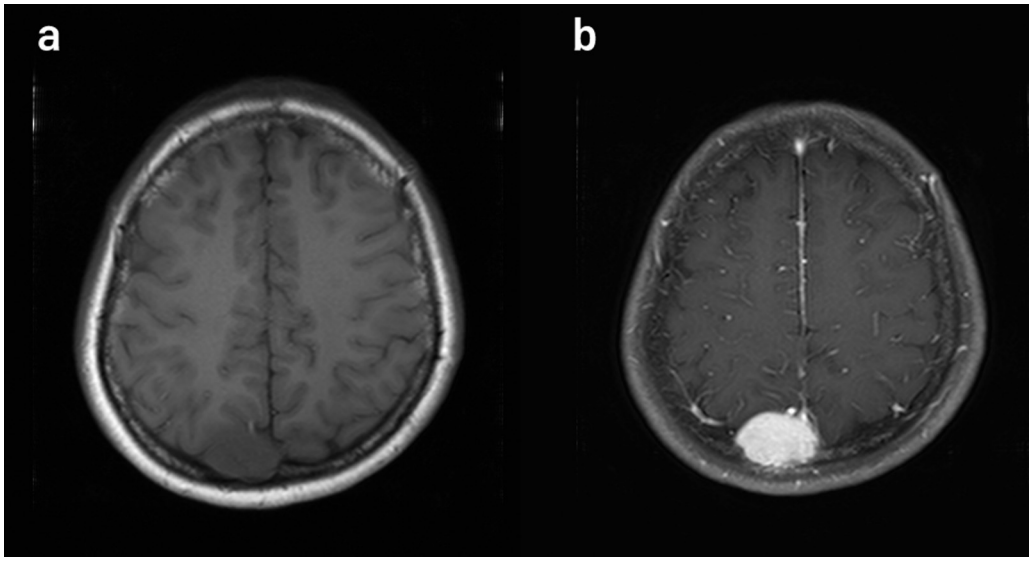

Fig. 1. Axial T1-weighted SE image showing a homogeneous lesion in the right posterior parietal area invading the calvarium (a). Axial T1-weighted enhanced image showing a well enhancing tumor in the epidura of the right posterior parietal area, obliterating the superior sagittal sinus but without brain parenchymal edema (b).

cerebrovascular diseases such as brain tumors. A 32 mm × 20 mm × 15 mm sized, well-enhanced solid tumor, in the right posterior parietal convexity, was located in the epidura, invading the calvarium and obliterating the superior sagittal sinus, without edema of the brain parenchyme. There were no abnormal findings around the optic nerve. The radiologic diagnosis was meningioma of the brain (Fig. 1). An F-18 FDG Torso Positron Emission Tomography – Computed Tomography (PET-CT) was done, and in addition to the right posterior parietal mass, a hypermetabolic mass was found in the pelvis (Fig. 2). No other uptake was found, including the lungs. Due to this finding, the patient underwent an enhanced abdominal-pelvic Computed Tomography (CT) with results of a well-defined pelvic mass with enhancement. Operation for the brain mass was performed. The tumor was situated in the parietooccipital area, closely attached to the calvarium and adhered to the dura and sagittal sinus. The tumor was hardly invading the parenchyme. The histopathology of the mass demonstrated to be a leiomyoma, which had the characteristic appearance of benign smooth muscle, without any apparent pleomorphism, cytologic atypia or necrosis (Fig. 3a). Immunohistochemical staining was positive for SMA (smooth muscle antibodies) and negative for Ki-67, a cell proliferation marker, and EMA (epithelial membrane antigen), an antigen that is expressed in epithelial neoplasms (Fig. 3b–d). Removal of the pelvic mass was performed 6 weeks after the neurosurgery. The histopathologic report of the pelvic mass was an ovarian leiomyoma and the immunohistochemistry was positive for smooth muscle antibodies. Postoperative course was uneventful.

3. Discussion

BML is a rare entity and about 200 cases have been reported so far. Its pathogenesis and etiology are still unclear. Theories that BML is similar to endometriosis have been proposed, as it often occurs in women of reproductive age, and is closely related to stimulation by reproductive hormones. Another suggested theory for BML is peritoneal seeding after myomectomy, or hysterectomy for uterine leiomyoma. It is possible that fragments of uterine leiomyoma implant and proliferate when accidentally left inside the peritoneum after laparotomy or within the laparoscopic trocar site, or even after morcellation. A third theory is lymphatic and vascular spread. The possibility of fragments of leiomyoma or tumor emboli from the uterine myoma entering the venous and lymphatic channels by surgical trauma and seeding to distant organs is considerable. The fourth proposed theory is coelomic metaplasia. Metaplastic transformation of the coelomic epithelium may explain BML in almost any place where mesothelial mesenchyme exists. These tumors probably originate from subcoelomic mesenchymal cells, which differentiate through the process of metaplasia into

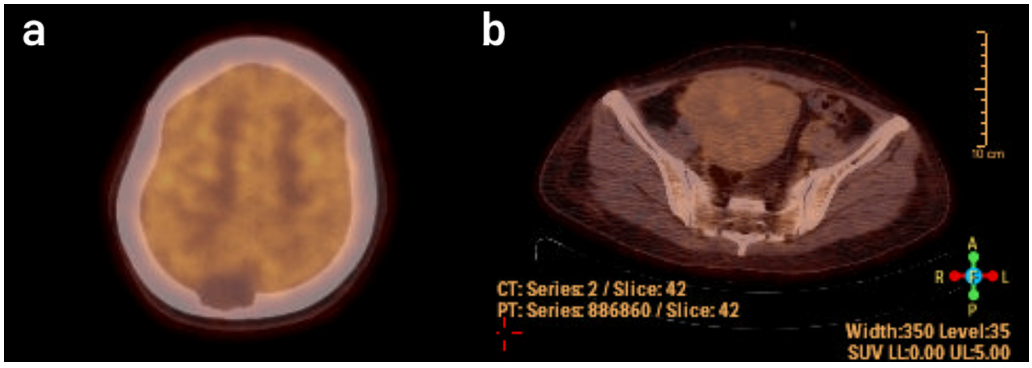

Fig. 2. F-18 FDG Torso PET-CT image of a hypometabolic lesion in the right posterior parietal area (a). A hypermetabolic lesion in the pelvis (b).