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Clinical case

Dumbbell-shaped spinal solitary fibrous tumor: Combined approach and a review of the literature

Tumeur fibreuse solitaire en sablier de la moelle spinale : approche combinée et revue de la littérature

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ABSTRACT

Introduction. – Spinal solitary fibrous tumors are rare entities, particularly when considered in a dumbbell-shaped form.
Case description. – The authors report on a 23-year-old female patient with dorsalgia and a D11–D12 dumbbell-shaped lesion on MRI, and highly vascularized on angiography. After a biopsy-based diagnosis, an integrated approach was performed with a preoperative embolization of the feeding intercostal arteries and an en bloc resection. At 3 months postoperatively, the patient had no pain or other neurologic symptoms and a complete resection was performed and documented on MRI.
Conclusion. – To our knowledge, only 3 previous reports of dumbbell-shaped spinal solitary fibrous tumors were carried-out and this is the first case, to our knowledge, treated by pre-operatory embolization. Nevertheless, this tumor should be considered among other spinal dumbbell-shaped lesions with a differential diagnosis, i.e. meningioma and schwannoma.

Mots clés :
Tumeur fibreuse solitaire
Tumeur spinale en forme de sablier
Embolisation
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Traitement

RÉSUMÉ

Introduction. – La tumeur fibreuse solitaire est très rare, surtout si celle-ci s'étend en sablier.
Cas clinique. – Une femme de 23 ans est présentée pour une dorsalgie et une lésion D11–D12 en sablier est identifiée par l'IRM, qui se révèle très vascularisée à l'angiographie. Après une biopsie diagnostique, une approche intégrée a été effectuée par une embolisation préopératoire des artères intercostales nourrissantes et une résection en bloc. À 3 mois postopératoire, la patiente n'a pas de douleur ou d'autre symptôme neurologique et une résection complète est confirmée par l'IRM.
Conclusions. – À la connaissance des auteurs, seulement 3 cas de tumeur fibreuse solitaire spinale en sablier sont décrits, et celui-ci est le premier à être traité par une embolisation préopératoire. Cette tumeur est à considérer dans le diagnostic différentiel d'une lésion en forme de sablier, comme le méningiome et le schwannome.

1. Introduction

The first description of a solitary fibrous tumor (SFT) was in the visceral pleura in 1931 [1], but only later in 1996 that a central nervous system (CNS) case was reported [2]. This tumor of mesenchymal origin is considered to be a benign slow-growing lesion, and, in most instances, the CNS cases can be cured by total

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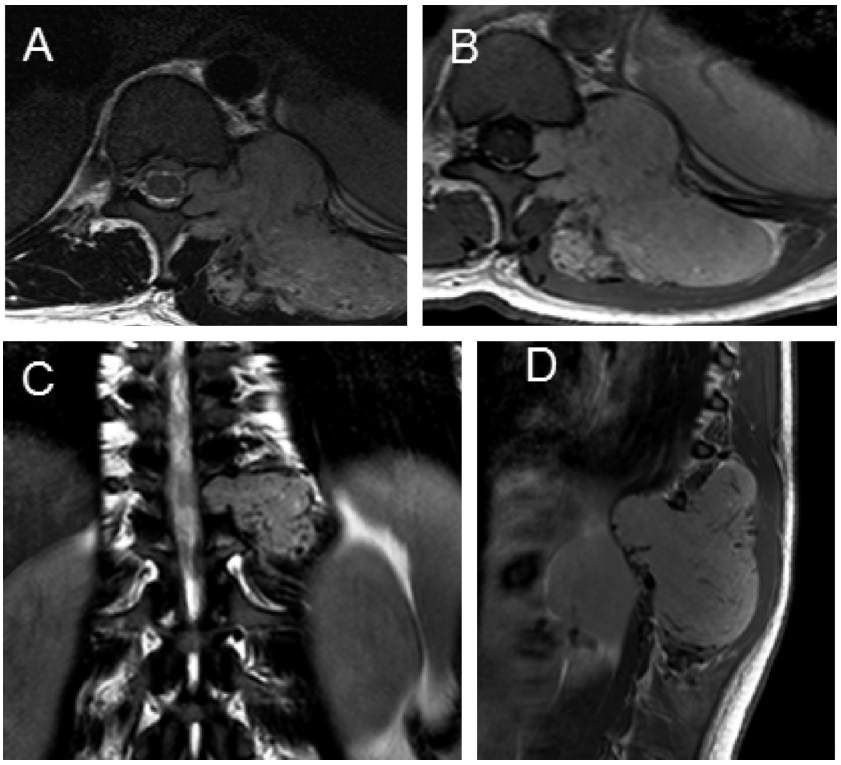

Fig. 1. A. T2-weighted axial. B. T1-weighted axial. C. T2-weighted coronal showing a dumbbell-shaped form slightly hyperintense lesion with intralesional vascular voids. D. T1-weighted sagittal using gadolinium injection with intense contrast uptake.
A. Pondération T2 axial. B. Pondération T1 axial. C. Pondération T2 coronal montrant une lésion en forme de sablier légèrement hyperintense avec des voids *vascularites intralésionnelles. D. Pondération T1 sagittal avec une prise de contraste intense après injection de gadolinium.*

surgical resection [1–3]. However, reports of recurrences, malignant transformation, metastasis and CSF dissemination have been published [4]. In the CNS, they are most frequently intracranial and, due to their meningeal involvement, they should be differentiated from the fibrous meningioma and the hemangiopericytoma (HPC) [5]. Spine is an unusual location, and some MRI features has been described suggesting this diagnosis, namely a well-circumscribed and ovoid lesion, rarely a dumbbell-shaped lesion, that may have both solid and cystic components and show an isointense signal on T1-weighted images and variable on T2-weighted images with avid enhancement after gadolinium injection [6].

To our knowledge, only 55 cases of spinal STFs have been reported and only 3 of them with a dumbbell-shaped presentation [7–9]. In this report, we describe one case with this particular shape and the first one where preoperative embolization was performed as part of the surgical planning.

2. Case report

A 23-year-old female patient was referred to our neurosurgical outpatient clinic for a dorsolumbar mass diagnosed 4 years previously. She noticed it during her pregnancy, when she started complaining of a slow-growing dorsal pain. This mass remained stable after puerperium and until one year before consultation, when it significantly enlarged, increasing the pain and promoting left-sided convex scoliosis. Neurological examination, apart from the cosmetic deformity, was otherwise normal.

An MRI showed a left paravertebral, extradural, intra- and extracanalar lesion in a dumbbell-shape through D11-D12 foramen, with spinal cord compression, isointense on T1 and slightly hyperintense on T2, with intense and homogeneous contrast uptake and exuberant vascular voids (Fig. 1). Subsequently, an angiography was performed and revealed an intense tumoral blush after D10 and D11 left intercostal arteries injection (Adamkiewicz artery–leaving at left L1).

A biopsy was carried-out through a paravertebral incision. Microscopic examination (Fig. 2) revealed a tumor with a biphasic structure. It was made of irregular, round or fusiform cells, tightly arranged around a ramifying, variable in caliber, vascular network and another composed of spindle cells presenting variable arrangements with more or less extensive areas of hyalinization, with a much less impressive vascular element. Anaplastic features were absent, the proliferative index (Ki67 immunoreactivity) < 1% and the immunohistochemical evaluation showed widespread immunoreactivity for vimentin, CD34 and Bcl-2 antibodies. No bone invasion was observed. The diagnosis was a solitary fibrous tumor.

The patient was then submitted to a combined approach regarding a total resection endpoint: a tumor embolization was carried-out through left D9 and D12 intercostal arteries with SQUID 12® during two different sessions (Fig. 3); then, surgery was performed with a L1 left hemi-laminectomy and en bloc tumor resection (Fig. 4). The patient was discharged at D6 postoperative with no neurological deficits and improvement of the pain. The second histopathological examination confirmed the final diagnosis. At one-month postoperative follow-up consultation, the patient returned to her daily living activities and at 6 months the patient no longer had any pain or other neurologic symptoms. A postoperative MRI was performed with no residual lesion (Fig. 5).

3. Discussion

SFT may occur in many locations in the human body, including the CNS [1]. In our case, most SFTs were intracranial and only one-fifth were intraspinal. Regarding the intracranial location, supratentorial and infratentorial compartments, the cerebellopontine