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Clinical case

Thoracic spine intra- and extradural dumbbell-shaped meningioma: Case report and extensive review of the literature with 21 cases

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ABSTRACT

Introduction. – Spinal dumbbell-shaped meningioma is a rare condition usually mistaken preoperatively for schwannoma. The present study reported a case of dumbbell-shaped meningioma, with an extensive review of literature.

Methods. – A documented case of thoracic spine dumbbell-shaped meningioma is reported, followed by an extensive review of the literature to analyze epidemiological features, pathogenesis, histopathological diagnosis, location, Eden classification, surgical treatment and outcome in such tumors.

Results. – Case report: A 55 year-old woman was admitted with paraparesis and paresthesia of lower limbs. MRI showed a dumbbell-shaped meningioma of the thoracic spine. The tumor was totally removed via a posterolateral approach.

Review of the literature. – Twenty-one spinal dumbbell-shaped meningiomas were reported in the last twenty years (1997–2017). Mean patient age was 46.57 years, with female predominance. Mean disease progression was 23.11 months. The thoracic spine was the predominant site (38.09%), followed by the cervical spine (33.33%). Eden type-3 was the most frequent tumor type, accounting for 66.67% of cases. In all cases, meningiomas were classified as WHO grade I. Complete removal (Simpson I–II) was achieved in 75% of cases. There was recurrence in 3 patients (14%), including 1 case of malignant transformation leading to death at 12 years post-surgery.

Conclusion. – Spinal dumbbell-shaped meningioma is mainly of the benign subtype. Long-term follow-up shows low rates of morbidity and mortality.

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1. Introduction

Meningioma and schwannoma are the most common intradural extramedullary tumors. Dumbbell-shaped (or “hourglass”) tumors are usually neurogenic, such as schwannoma or neurofibroma [1].

Meningioma is a benign tumor arising from the meningotheelial cells of the arachnoid, readily identifiable as a globular fibrous solitary mass with a dural tail attachment. It accounts for 25% to 46% of primary spinal tumors [2]. Dumbbell-shaped meningioma is an unusual presentation of spinal meningioma [3]. The exact prevalence of extension through the neural foramen has not been reported [4]. The literature comprises case reports, without real series that could shed light on pathogenesis.

The authors describe a rare case of dumbbell-shaped meningioma in the thoracic spine, with an extensive analysis of reported cases at this specific location, focusing on pathogenesis, therapeutic management and outcome.

2. Method

This article reports a documented case of thoracic spine intradural and extradural dumbbell-shaped meningioma. The case description is followed by an analysis of the literature.

An extensive review of the literature of the previous 20 years (1997–2017) was performed on PubMed, Google Scholar, the Cochrane Library and ScienceDirect, using the following keywords: dumbbell-shaped tumors, dumbbell-shaped meningioma, spinal meningiomas, spinal tumors. The review covered documented spinal dumbbell-shaped meningioma in any region of the spine, including foramen magnum and sacrum.

The Eden classification of dumbbell-shaped tumors [5] was used: type 1 corresponds to intra- and extradural, type 2 to

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Table 1
Summary of reported cases of spinal dumbbell-shaped meningioma.

Case reference	Age/gender	Location	Symptomatology	Associated disease	Disease duration (months)	Eden type
1. Yoshiura et al. (1998)	16/F	C2–C4	Left cervicobrachialgia, left upper-limb weakness	NF1	3	2
2. Buchfelder et al. (2001)	72/F	C7–T2	Progressive paraparesis	–	24	3
3. Suzuki et al. (2002)	58/M	T10–T11–T12	Abnormal mass on chest X-ray	–	–	–
4. Chen et al. (2005)	16/F	C1–C2	Hearing impairment, neck mass, right upper limb weakness	NF2, bilateral acoustic schwannoma, T5–T6 schwannoma	18	2
5. Alam et al. (2005)	16/F	C4–C5	Loss of left lower limb strength	NF2, bilateral acoustic schwannoma	12	2
6. Restrepo et al. (2006)	57/M	C7–T2	Loss of lower limb strength	–	–	3
7. Santiago et al. (2009)	42/M	T2–T3	Paraparesis	–	1	3
8. Kim et al. (2011)	50/M	T6–T7	Hypoesthesia, paraparesis	–	6	2
9. Tuli et al. (2012)	42/F	T5	Weakness of lower limbs, sensitivity loss below T3	Left parietal meningioma	4	3
10. Ozaki et al. (2013)	49/F	C2–C3	Gait disturbance, muscle weakness, sensory deficit, bladder and sexual dysfunction	–	4	2
11. Yaldiz et al. (2014)	50/F	Cervicothoracic	Back pain, right arm numbness, hypoesthesia under T1, 4–/5 muscle strength in hands	–	6	3
12. Iwanami et al. (2014)	37/M	L5	Left leg pain, L5 hypoesthesia, residual urine sensation	–	8	2
13. Iwanami et al. (2014)	57/M	T12–L1	Bilateral leg pain, pollakiuria, residual urine sensation, 4/5 iliopsoas muscle strength	–	42	3
14. Iwanami et al. (2014)	59/M	L1–L5	Lower back pain	–	122	
15. Iwanami et al. (2014)	35/M	L4–sacrum	Lower back pain	–	120	
16. Sato et al. (2016)	76/F	C3–C4	Pain and weakness in the left limbs, palsy in C5 region, spastic gait	–	10	2
17. Dehcordi et al. (2016)	39/F	T3–T4	Progressive numbness and weakness in lower limbs with gait disorder, severe paraparesis	Meningiomatosis	12	3
18. Bettaswamy et al. (2016)	50/M	C2 to C4	Spastic quadriparesis, bladder disorder	–	8	3
19. Bettaswamy et al. (2016)	41/M	C3 to C7	Spastic quadriparesis, sensory loss below C6	–	4	3
20. Haranhalli et al. (2017)	61/F	T3–T4	Left upper-limb paresthesia	Cervical spondylosis	–	3
21. Present case (2017)	55/F	T2–T3	Lower limb paresthesia, allodynia, paraparesia	–	12	2
Summary	46.57 y F: 52.38% M: 47.62%	T: 38.09% C: 33.33% CTJ: 14.28 L: 14.28%	–	Neurofibromatosis: 3 Meningiomatosis: 2	23.11 m	Type 3: 66.67% Type 2: 44.37%

intra-/extradural and paravertebral, type 3 to extradural and paravertebral, and type 4 to foraminal and paravertebral tumors [5]. In the initial description by Heuer [6], dumbbell-shaped tumors were restricted to the intervertebral foramen entry point, with an hourglass shape. The present study counted types 2 to 4 as true dumbbell-shaped meningioma. Exclusion criteria comprised Eden type I tumor, which is purely intracanalicular without foraminal or paraspinal component; meningioma which was not precisely described, being included under a general description of dumbbell-shaped tumor; articles in languages other than English or French [7]; and articles unavailable in the databases [8]. The selected articles are summarized in Tables 1 and 2.

3. Results

3.1. Case report

A 55 year-old woman was admitted to our department of spine surgery with allodynia and progressive onset of lower-limb weakness. She had a 1-year history of paresthesia of the lower limbs and gradual dysesthesia in the same territory. Clinical examination showed discrete 4/5+ paraparesis, T3 hypoesthesia, urinary incontinence and hyper-reactive patellar and Achilles tendon reflexes.

The thoracic spine MRI revealed an intra-extradural mass at T2–T3 with homogenous contrast enhancement on gadolinium

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