Spinal Dumbbell Meningiomas: A Systematic Review

Ann. Ital. Chir., 2024 95, 6: 1009–1025
https://doi.org/10.62713/aic.3696

Gianluca Scalia[1], Federica Paolini[2,3], Evier Andrea Giovannini[2,3], Gianluca Ferini[4,5], Alexandra Giorlandino[6], Eliana Giurato[6], Luigi Basile[3], Massimo Furnari[1], Giovanni Federico Nicoletti[1], Giuseppe Emmanuele Umana[5,7], Francesca Graziano[1]

[1]Neurosurgery Unit, Department of Head and Neck Surgery, Garibaldi Hospital, 95124 Catania, Italy
[2]Neurosurgical Clinic, AOUP "Paolo Giaccone", Post Graduate Residency Program in Neurologic Surgery, Department of Biomedicine Neurosciences and Advanced Diagnostics, School of Medicine, University of Palermo, 90127 Palermo, Italy
[3]Neurosurgery Unit, S. Elia Hospital, 93100 Caltanissetta, Italy
[4]Department of Radiation Oncology, REM Radioterapia srl, 95029 Viagrande, Italy
[5]School of Medicine and Surgery, Kore University of Enna, 94100 Enna, Italy
[6]Anatomic Pathology Unit, Garibaldi Hospital, 95124 Catania, Italy
[7]Department of Neurosurgery, Trauma Center, Gamma Knife Center, Cannizzaro Hospital, 95126 Catania, Italy

AIM: This systematic review aims to synthesize the epidemiology, clinical presentation, diagnostic approaches, treatment strategies, and outcomes of spinal dumbbell meningiomas to enhance understanding and improve patient management.
METHODS: Following Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines, four major databases (PubMed, Scopus, Web of Science, and Cochrane Library) were searched until June 2024. Studies included patients diagnosed with spinal dumbbell intradural-extradural meningiomas, focusing on patient demographics, tumor characteristics, diagnostic methods, treatment modalities, and clinical outcomes. Two independent reviewers extracted the data, with discrepancies resolved by consensus or a third reviewer. Due to heterogeneity in study designs, a narrative synthesis was conducted.
RESULTS: From 2602 studies, 22 articles comprising 32 patients met the inclusion criteria. The median patient age was 48 years (P25: 44.76, P75: 51.24), with a slight female predominance (59.38%). The cervical spine was the most common tumor location (63.63%). Symptoms included movement disorders (59.38%), sensory deficits (40.63%), and pain (34.38%). All patients underwent surgical resection, with 75% achieving gross total resection (GTR). Recurrence occurred in 15.63% of cases, and 68.75% of patients had good recovery outcomes. Histopathological analysis showed all tumors were World Health Organization (WHO) Grade I meningiomas, with meningothelial and psammomatous subtypes being most common.
CONCLUSIONS: Spinal dumbbell meningiomas are rare but challenging due to their complex anatomical features and potential for neurological deficits. Surgical resection is the primary treatment with generally favorable outcomes, but recurrence remains a concern. Long-term follow-up and research into advanced imaging and minimally invasive surgical techniques are essential for improving management and patient outcomes.

Keywords: dumbbell; tumor; meningioma; intradural-extradural; surgical resection; recurrence; imaging; systematic review

Introduction

Spinal dumbbell meningiomas are rare, typically benign tumors originating from the meningothelial cells of the arachnoid layer. Unlike more common intradural meningiomas, these tumors present a distinctive "dumbbell" shape, involving intraspinal, extradural, and paraspinal spaces, which poses unique challenges in diagnosis and treatment [1]. The extradural extension of these tumors necessitates different therapeutic approaches compared to their intradural counterparts. While meningiomas and schwannomas are the most common intradural extramedullary tumors, dumbbell-shaped (or "hourglass") tumors are usually neurogenic, such as schwannomas or neurofibromas [2]. Meningiomas, though generally benign, constitute 25% to 46% of primary spinal tumors and are typically identified by their globular, fibrous solitary mass with a dural tail attachment [3]. However, the occurrence of dumbbell-shaped meningiomas is relatively rare, and their extension through the neural foramen has not been widely documented in the literature [4, 5]. Despite their rarity, spinal dumb-

Submitted: 13 August 2024 Revised: 21 September 2024 Accepted: 29 September 2024 Published: 20 December 2024
Correspondence to: Gianluca Ferini, Department of Radiation Oncology, REM Radioterapia srl, 95029 Viagrande, Italy; School of Medicine and Surgery, Kore University of Enna, 94100 Enna, Italy (e-mail: gianluca.ferini@grupposamed.com).

bell meningiomas can cause significant morbidity by compressing neural structures, leading to various neurological deficits such as myelopathy, radiculopathy, and, in severe cases, paraplegia [5]. The complex anatomical location of these tumors often necessitates advanced surgical techniques to achieve complete resection while minimizing the risk to surrounding neural tissues. Understanding the epidemiology, clinical presentation, diagnostic methods, treatment strategies, and outcomes of spinal dumbbell meningiomas is crucial for effective management. This systematic review aims to synthesize the current literature on these tumors, providing a comprehensive analysis that can improve clinical practice and guide future research.

Materials and Methods

Search Strategy

A thorough literature search was conducted using four major databases: PubMed, Scopus, Web of Science, and the Cochrane Library. The search spanned from the inception of each database to June 2024. The search terms used were a combination of keywords and Medical Subject Headings (MeSH) terms, specifically: "spinal meningioma", "dumbbell meningioma", "extradural meningioma", "intradural meningioma", and "spinal tumor". Boolean operators (AND, OR) were utilized to ensure a comprehensive retrieval of relevant studies.

Inclusion Criteria

To be included in this review, studies had to meet the following criteria:

- **Population:** Studies involving patients diagnosed with spinal dumbbell intradural-extradural meningiomas.
- **Language:** Only studies published in English were considered to avoid language bias and ensure the accessibility of full-text articles.
- **Study Design:** Eligible studies included clinical trials, cohort studies, case-control studies, case series, and case reports.
- **Outcomes:** Studies must report on at least one of the following outcomes: patient demographics, tumor characteristics, diagnostic methods, treatment modalities, or clinical outcomes.

Exclusion Criteria

Studies were excluded based on the following criteria:

- **Non-spinal meningiomas:** Studies focusing on meningiomas located in areas other than the spinal region.
- **Intradural meningiomas without extradural extension:** Studies involving meningiomas confined to the intradural space without dumbbell or extradural components.
- **Non-English articles:** Articles not published in English were excluded due to potential translation inaccuracies and resource limitations.

Data Extraction

Data extraction was performed independently by two reviewers (GS and FP) to minimize bias and ensure accuracy. The extracted data included:

- **Authors and year of publication**
- **Patient Demographics:** Age and gender.
- **Tumor Characteristics:** Location (cervical, thoracic, lumbar), size, and extent of extradural involvement.
- **Symptoms of presentation and associated diseases**
- **Diagnostic Methods:** Imaging techniques used, including magnetic resonance imaging (MRI), CT, and any advanced imaging modalities.
- **Treatment Modalities:** Surgical approaches, and use of adjuvant therapies (chemotherapy, and/or radiotherapy).
- **Histopathology**
- **Clinical Outcomes:** Postoperative complications, outcomes, recurrence rate, follow-up data, and Simpson's grade.

Discrepancies in data extraction were resolved through discussion and consensus between the reviewers. If consensus could not be reached, a third reviewer (FG) was consulted.

Data Synthesis and Analysis

Data were synthesized through a narrative approach due to the heterogeneity in study designs, patient populations, and reported outcomes. Descriptive statistics were used to summarize patient demographics, tumor characteristics, and treatment outcomes. This comprehensive approach is designed to provide an in-depth understanding of spinal dumbbell meningiomas, offering valuable insights into their management and identifying key areas for future research. To ensure transparency and reproducibility, we followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines throughout the study selection process [6]. A PRISMA flow diagram was also created to visually represent the study selection process, detailing the number of records identified, screened, assessed for eligibility, and ultimately included in the final analysis (Fig. 1). This systematic review was conducted in accordance with the PRISMA checklist (**supplementary material**) to ensure transparency, completeness, and accuracy in the reporting of the review process and findings.

Results

A total of 2602 studies were identified through the PubMed database and reference section screening. After removing 16 duplicates, 2586 unique articles remained. The selection process began with an initial screening of titles to assess relevance to spinal dumbbell tumors, even when the term "meningioma" was not explicitly mentioned. This step resulted in the exclusion of 2390 articles, leaving 196 articles for further consideration.

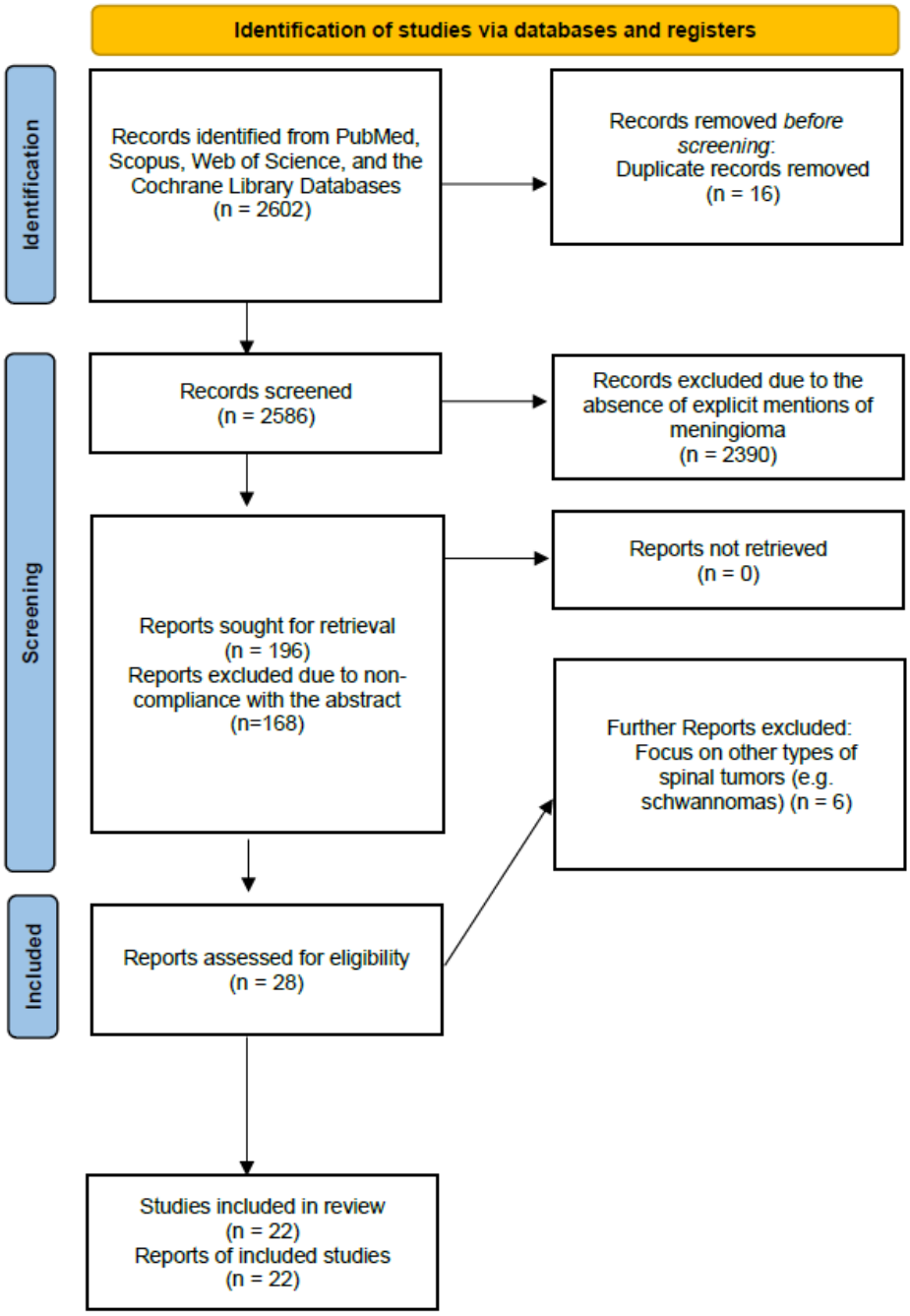

Fig. 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram of study selection process for systematic review on spinal dumbbell meningiomas. This flow diagram illustrates the systematic review process of selecting studies related to spinal dumbbell tumors. The review initially identified 2602 records through database searches. After removing 16 duplicates, 2586 records were screened for relevance. Following the exclusion of 2390 records, 196 reports were sought for retrieval, all of which were retrieved. In the eligibility assessment stage, 28 reports were evaluated, and 6 were excluded due to their focus on other types of spinal tumors, such as schwannomas. Ultimately, 22 studies were included in the final systematic review.

Next, the abstracts of these 196 articles were carefully reviewed according to predefined selection criteria. As a result, 168 articles were excluded, reducing the pool to 28 articles for full-text evaluation. Upon thorough assessment of the 28 full-text articles, 6 studies were excluded due to their focus on other types of spinal dumbbell tumors (such as schwannomas) or because they lacked sufficient patient data relevant to spinal dumbbell meningiomas. Ultimately, this systematic review includes 22 articles, encompassing a

Table 1. Summary of patient demographics, clinical features, surgical approaches, and outcomes in studies on spinal dumbbell meningiomas.

Authors/ Year	Age	Gender	Location	Symptoms	Associated disease	Disease duration (months)	Eden type	Surgery	Extent of resection	Histopatho-logy	Recurre-nce	Reopera-tion	Adjuvant therapy	Outcome	Follow-up (years, months)	Simp-son
Yoshiura *et al.* (1998) [7]	16	F	C2–C4	Left cervicobrachialgia, left upper-limb weakness	NF1	3	2	–	Complete resection	–	–	–	–	Good	0.66 y, 8 m	I
Buchfelder *et al.* (2001) [8]	72	F	C7–T2	Progressive paraparesis	–	24	3	C7, T1, T2 laminectomies, thoracotomy	Complete resection	Meningothelial	–	–	–	Good	3 y, 36 m	I
Suzuki *et al.* (2002) [9]	58	F	T10–T11 –T12	Abnormal mass on chest X-ray	–	–	–	T10, T11 laminectomies, thoracoscopy	Complete resection	Fibroblastic	–	–	–	Good	–	I
Chen *et al.* (2005) [10]	16	F	C1–C2	Hearing impairment, neck mass, right upper limb weakness	NF2, bilateral acoustic schwannoma, T5–T6 schwannoma	18	2	C1, C2 laminectomies, anterolateral transforaminal approach for paraspinal part	Subtotal resection	Fibroblastic	–	–	–	Good	0.83 y, 10 m	IV
Alam *et al.* (2005) [11]	16	F	C4–C5	Loss of left lower limb strength	NF2, bilateral acoustic schwannoma	12	2	Posterior approach	Complete resection	–	–	–	–	Good	–	–
Restrepo *et al.* (2006) [12]	57	M	C7–T2	Loss of lower limb strength	–	–	3	T1, T2 laminectomies	Complete resection	Psammomatous	–	–	Radiotherapy	Good	0.66 y, 8 m	I
Santiago *et al.* (2009) [13]	42	M	T2–T3	Paraparesis	–	1	3	T2, T3 laminectomies	Complete resection	Psammomatous	–	–	–	Good	–	I
Kim *et al.* (2011) [14]	50	M	T6–T7	Hypoesthesia, paraparesis	–	6	2	T6 hemilaminectomy, T5–T7 pedicle screw fixation	Complete resection	Meningothelial	–	–	–	Good	–	I

Table 1. Continued.

Authors/ Year	Age	Gender	Location	Symptoms	Associated disease	Disease duration (months)	Eden type	Surgery	Extent of resection	Histopatho-logy	Recurre-nce	Reopera-tion	Adjuvant therapy	Outcome	Follow-up (years, months)	Simp-son
Tuli *et al*. (2012) [15]	42	F	T5	Weakness of lower limbs, sensitivity loss below T3	Left parietal meningioma	4	3	T4, T5 laminectomies, T4–T6 pedicle screw fixation	Complete resection	–	–	–	–	Good	0.5 y, 6 m	I
Ozaki *et al*. (2013) [16]	49	F	C2–C3	Gait disturbance, muscle weakness, sensory deficit, bladder and sexual dysfunction	–	4	2	C1 to C4 hemil-aminectomies	Subtotal resection	Meningothelial	–	–	–	Good	2.5 y, 30 m	IV
Yaldiz *et al*. (2014) [17]	50	F	Cervicothoracic	Back pain, right arm numbness, hypoesthesia under T1, 4−/5 muscle strength in hands	–	6	3	T1 laminectomy	Complete resection	Psammomatous	–	–	–	Good	–	I
Iwanami *et al*. (2015) [18]	37	M	L5	Left leg pain, L5 hypoesthesia, residual urine sensation	–	8	2	L4, L5 laminectomies	Complete resection	Meningothelial	–	–	–	Good	13 y, 156 m	I
	57	M	T12–L1	Bilateral leg pain, pollakiuria, residual urine sensation, 4/5 iliopsoas muscle strength	–	42	3	T12, L1 laminectomies	Subtotal resection	Meningothelial	+	+ (1.3 years, 2.9 years)	Radiotherapy	Good, death from other disease	11 y, 132 m	IV
	59	M	L1–L5	Lower back pain	–	122	–	Posterior approach	Complete resection	Transitional	+	+ (1.8 years, 2.3 years)	–	Death from disease	12 y, 144 m	I
	35	M	L4-sacrum	Lower back pain	–	120	–	Posterior approach	Subtotal resection	Psammomatous	–	–	–	Good	2 y, 24 m	IV
Sato *et al*. (2016) [19]	76	F	C3–C4	Pain and weakness in the left limbs, palsy in C5 region, spastic gait	–	10	2	Posterior approach	Complete resection	–	–	–	–	Good	–	–

Table 1. Continued.

Authors/ Year	Age	Gender	Location	Symptoms	Associated disease	Disease duration (months)	Eden type	Surgery	Extent of resection	Histopatho-logy	Recurre-nce	Reopera-tion	Adjuvant therapy	Outcome	Follow-up (years, months)	Simp-son
Dehcordi *et al*. (2016) [20]	39	F	T3–T4	Progressive numbness and weakness in lower limbs with gait disorder, severe paraparesis	Meningiomatosis	12	3	T4, T5 laminectomies	Complete resection	Both meningothe-lial	–	–	–	Good	0.5 y, 6 m	II
Bettaswamy *et al*. (2016) [21]	50	M	C2 to C4	Spastic quadriparesis, bladder disorder	–	8	3	C2 to C4 laminectomies	Subtotal resection	Meningothelial	–	–	–	Good	0.5 y, 6 m	IV
	41	M	C3 to C7	Spastic quadriparesis, sensory loss below C6	–	4	3	Posterior approach	Complete resection	–	–	–	–	Good	2 y, 24 m	I
Haranhalli *et al*. (2017) [22]	61	F	T3–T4	Left upper-limb paresthesia	Cervical spondylosis	3	–	Posterior approach	Complete resection	–	–	–	–	Good	–	I
N'da *et al*. (2018) [23]	55	F	T2–T3	Lower limb paresthesia, allodynia, paraparesia	–	12	2	T2, T3 hemil-aminectomies, T2 partial corpectomy	Complete resection	Transitional	–	–	–	Good	0.25 y, 3 m	I
Zhang *et al*. (2018) [24]	20	F	C1–C2	–	–	–	4	Posterior approach	Subtotal resection	Meningothelial	+	–	–	–	120 m	IV
	43	F	C1–C2	–	–	–	1	Posterior approach	Subtotal resection	Psammomatous	+	–	–	–	5 m	IV
	40	F	C6–C7	–	–	–	1	Posterior approach	Complete resection	Psammomatous	–	–	–	–	60 m	I
	76	F	C2–C3	–	–	–	1	Posterior approach	Subtotal resection	Psammomatous	+	–	–	–	156 m	IV
Zhan *et al*. (2019) [25]	47	F	C1–C2	Neck pain, inferior limbs numbness	–	4	2	C1–C2 hemilaminectomy	Complete resection	Concurrent Neurofi-broma and Meningothe-lial Meningioma	–	–	–	Good	0.25 y, 3 m	I

Table 1. Continued.

Authors/ Year	Age	Gender	Location	Symptoms	Associated disease	Disease duration (months)	Eden type	Surgery	Extent of resection	Histopatho-logy	Recurre-nce	Reopera-tion	Adjuvant therapy	Outcome	Follow-up (years, months)	Simp-son
Nguyen *et al*. (2021) [26]	22	F	C5–T1	weakness and numbness in both lower limbs	–	–	1	Posterior approach	Complete resection	Psammomatous	–	–	–	Good	0.5 y, 6 m	I
Zheng *et al*. (2023) [27]	53	M	C2	Left hand sensory reduction	–	12	1	Posterior approach	Complete resection	Concurrent Schwan-noma and meningioma	–	–	–	–	–	–
	47	F	C2	Left half body pain and numbness	–	72	2	Posterior approach	Complete resection	Concurrent Schwan-noma and meningioma	–	–	–	–	–	–
	55	M	C2	Right extremities motor weakness and gait disturbances	–	9	2	Posterior approach	Complete resection	Concurrent Schwan-noma and meningioma	–	–	–	–	–	–
	61	M	C2	Neck pain, gait disturbances	–	6	2	Posterior approach	Complete resection	Concurrent Schwan-noma and meningioma	–	–	–	–	–	–
Fukui *et al*. (2023) [28]	43	M	cervical	Neck pain, unsteady gait	chronic inflammatory syndrome	12	2	Posterior approach	Complete resection	Lymphoplasmacyte-rich meningioma	–	–	–	–	–	–

NF, neurofibromatosis; M, male; F, female.

total of 32 patients. These studies are summarized in Table 1 [7, 8, 9, 10, 11, 12, 13, 14, 15, 16, 17, 18, 19, 20, 21, 22, 23, 24, 25, 26, 27, 28].

This table 1 presents a comprehensive summary of 22 studies reporting on the demographics, clinical features, surgical treatments, and outcomes of patients with spinal dumbbell tumors, specifically meningiomas. It includes information on the age, gender, tumor location, symptoms, associated diseases, disease duration, type of tumor, surgical approaches, extent of resection, histopathology, recurrence, reoperation, adjuvant therapy, outcomes, follow-up periods, and Simpson grading of the extent of resection. The data provide insights into the clinical presentation and management strategies for these rare spinal tumors.

Clinical Database

All selected reports were used to construct a clinical database, highlighting the most relevant characteristics of the included patients (Table 2). The database comprises 32 patients, consisting of 13 males and 19 females, resulting in a male-to-female ratio of 0.68. The median age of the patients was 48 years (P25: 44.76, P75: 51.24) (range 16–76 years). These tumors were categorized according to the Eden's classification: Type I (5 cases), Type II (13 cases), Type III (9 cases), and Type IV (1 case), with 4 cases not reported (Fig. 2).

Movement disorders were the most common symptoms, affecting 19 out of 32 patients (59.38%). These included limb weakness, paresis, or spastic gait. Other frequent symptoms were hypoesthesia, reported in 13 patients (40.63%), pain in 11 patients (34.38%), and dysautonomia disorders, such as urinary incontinence, in 6 patients (18.75%). Although there was a higher prevalence of tumors in the cervical region (21 out of 32 cases, 63.63%), these tumors were found to affect almost all levels of the spine (Fig. 3). Median disease duration was 16.12 months (P25: 15.03, P75: 17.21) (range 1–122 months). All patients underwent surgical intervention via a posterior approach to the spinal cord for tumor resection. Specifically, 10 patients underwent laminectomy, 3 underwent hemilaminectomy, and 3 patients received posterior fixation + laminectomy/hemilaminectomy (9.38%) using transpedicular screws and rods. Most of cases (24 patients, 75%) underwent gross total resection (GTR), while a smaller portion (8 patients, 25%) underwent subtotal resection (STR). Cases of STR are related to pleural space involvement or calcified portions adherent to the spinal cord. Surgical radicality of meningiomas was classified according to Simpson's grading system, with the following distribution among the patients: 16 patients were classified as Simpson grade I, 1 patient as Simpson grade II, and 8 patients as Simpson grade IV. No patients were classified under Simpson grades III or V. Additionally, 7 patients were categorized as not reported. The treatment outcomes were favorable, with 22 out of 32 patients (68.75%) experiencing good recovery. Only one patient (3.13%) had a tumor-related death, underscoring the generally benign nature of the disease. Histopathological analyses revealed that all tumors were World Health Organization (WHO) Grade I meningiomas, with the most common subtypes being meningothelial and psammomatous meningiomas (8 cases each) (Fig. 4). The recurrence rate was 15.63% (5 out of 32 patients) with a median follow-up of 33.50 months (P25: 31.23, P75: 35.77) (range 3–156 months).

This table 2 provides a descriptive statistical summary of the clinical characteristics, surgical interventions, and outcomes in patients with spinal dumbbell meningiomas. It includes data on the sex distribution, age, presenting symptoms, surgical approaches, extent of resection, associated diseases, disease duration, Eden's classification, histopathological findings, Simpson grading of resection, recurrence rates, reoperations, and patient outcomes. Most patients underwent gross total resection (GTR) with favorable outcomes reported in most cases. The mean follow-up period was 47.15 months, highlighting the long-term monitoring of these patients.

Case Illustration 1

A 56-year-old male patient presented with a six-month history of progressive difficulty walking, diffuse lower limb pain, and subjective numbness in the lower limbs. The symptoms had progressively worsened, leading to sensations of "electric shocks" in the lower limbs, stiffness, and an inability to walk independently, necessitating bilateral support. Upon physical examination, the patient exhibited significant gait disturbances, requiring bilateral support for ambulation. Neurological examination revealed muscle strength of 3/5 in both lower limbs, hyperreflexia in the lower extremities, decreased sensation to light touch and pinprick in the lower extremities, marked ataxia, and a positive Lhermitte's sign. A cervico-thoracic spinal MRI revealed a large intra- and extra-vertebral lesion on the right side, likely extradural, showing significant homogeneous enhancement after Gadolinium administration. The lesion had maximum dimensions of approximately $50 \times 35 \times 14$ mm. This dumbbell-shaped mass caused bony remodeling of the right posterior vertebral body of Th2, widening the ipsilateral neural foramen, and extended extraforaminally to the costovertebral joint. The mass occupied a large part of the spinal canal and formed a "sleeve" around the spinal cord, displacing it to the left (Eden type 3) (Fig. 5).

The patient underwent a posterior dorsal surgical approach, including laminectomy of Th2 and partial laminectomy of Th1 and Th3, followed by subtotal microsurgical resection of the extradural mass, which extended laterally to the pleural space and anteromedially to the spinal cord. The procedure was performed under intraoperative neurophysiological monitoring, including somatosensory evoked potentials (SEPs), transcranial motor evoked potentials (TcMEPs), and free-run electromyography (EMG), alongside intensive hemodynamic monitoring.

Table 2. Descriptive statistical summary of clinical characteristics, surgical interventions, and outcomes in spinal dumbbell meningioma cases.

Indicator	Value (n%/median (P25, P75))
Sex	
Males	13 (40.63%)
Females	19 (59.38%)
Age	48 (P25: 44.76, P75: 51.24)
Symptoms	
Motor	19 (59.38%)
Sensory	13 (40.63%)
Pain	11 (34.38%)
Dysautonomia	6 (18.75%)
Surgical approach	
Posterior approach	16 (50%)
Laminectomy	10 (31.25%)
Hemilaminectomy	3 (9.38%)
Posterior fixation + laminectomy/hemilaminectomy	3 (9.38%)
Extent of resection	
GTR	24 (75%)
STR	8 (25%)
Associated disease	
NF type 1	1 (3.13%)
NF type 2	2 (6.25%)
Meningiomas in different sites	2 (6.25%)
Disease Duration	16.12 (P25: 15.03, P75: 17.21)
Eden type	
I	5 (15.63%)
II	13 (40.63%)
III	9 (28.13%)
IV	1 (3.13%)
Not reported	4 (12.5%)
Histopathology	
Meningothelial	8 (25%)
Transitional	2 (6.25%)
Psammomatous	8 (25%)
Lymphoplasmacyte-rich	1 (3.13%)
Concurrent Schwannoma and meningioma	4 (12.5%)
Concurrent Neurofibroma and Meningothelial Meningioma	2 (6.25%)
Fibroblastic	1 (3.13%)
Not reported	6 (18.75%)
Simpson grade	
I	16 (50%)
II	1 (3.13%)
III	0 (0%)
IV	8 (25%)
V	0 (0%)
Not reported	7 (21.88%)
Recurrence	5/32 (15.63%)
Re-surgery	2/5 (40%)
Outcome	
Good	22 (68.75%)
Death (tumor-related)	1 (3.13%)
Not reported	9 (28.13%)
Follow-up	33.50 (P25: 31.23, P75: 35.77)

GTR, gross total resection; STR, subtotal resection; NF, neurofibromatosis.

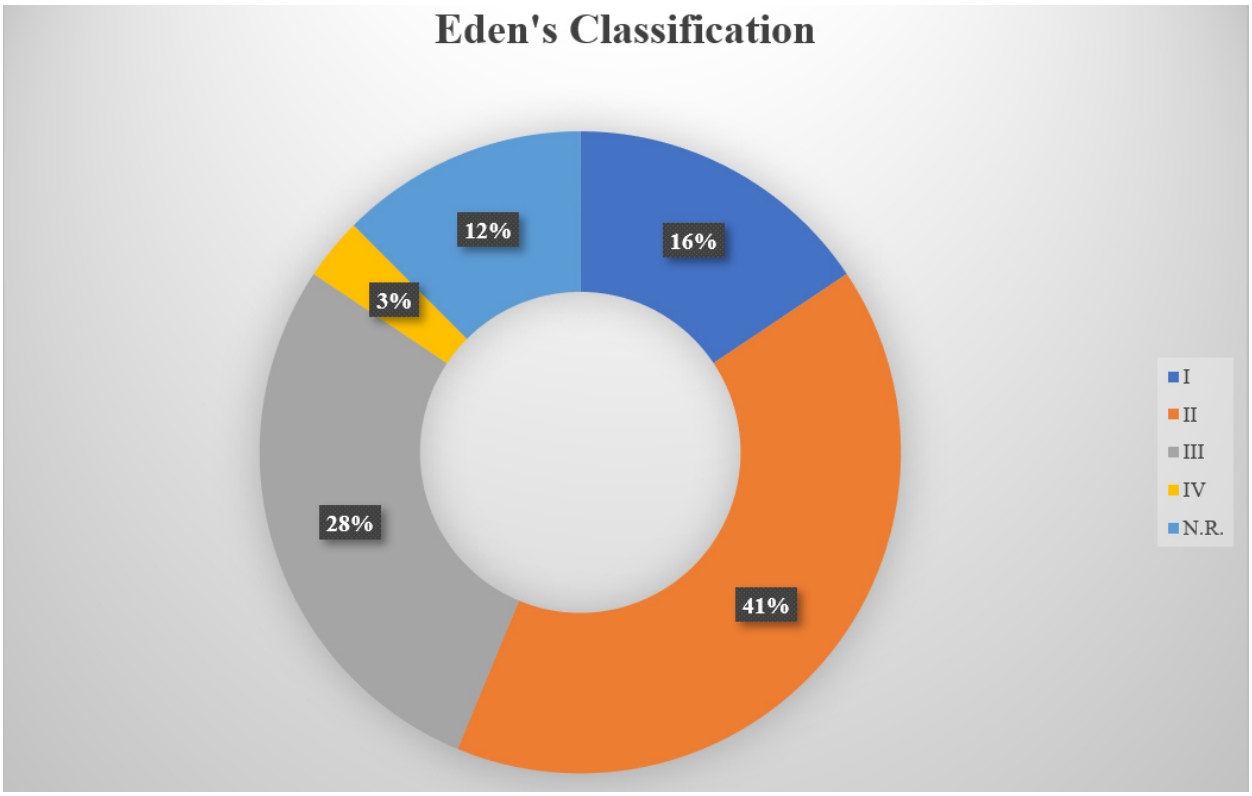

Fig. 2. Eden's Classification distribution. This donut chart presents the distribution of cases according to Eden's classification. Most cases fall under Type II (41%), followed by Type III (28%). Type I and Type IV represent 16% and 3% of the cases, respectively, while 12% are not reported (N.R.).

In the immediate postoperative period, the patient was transferred to the Intensive Care Unit (ICU) for protected awakening and multiparametric monitoring. Approximately 24 hours later, the patient was transferred to the neurosurgery department for continued care. Mobilization was initiated on the second postoperative day, and the patient exhibited progressive improvement in lower limb strength and gait. One-month follow-up thoracic spinal MRI confirmed the recent surgical resection of the intracanal, extradural expansile process at Th1 to Th3, with a small remnant in the pleural space (Fig. 6).

Histopathological examination revealed a meningothelial meningioma, classified as WHO Grade 1, with a mitotic index of 2/10 high-power fields (HPF), no necrosis, and an immunophenotypic profile positive for epithelial membrane antigen (EMA), 70% positive for progesterone receptor (PgR), negative for S100 protein, glial fibrillary acidic protein (GFAP), and pan-cytokeratin (panCK), with a ki67 proliferation index of 5% (Figs. 7,8).

The patient was discharged on the fifth postoperative day with a referral to a neurorehabilitation center, demonstrating the importance of multidisciplinary care and postoperative rehabilitation in optimizing outcomes for patients with significant neurological deficits due to spinal meningiomas. At 6-month follow-up no recurrent disease was documented on neuroimaging.

Case Illustration 2

A 51-year-old female patient with no significant medical history or comorbidities initially presented with subjective sensory disturbances, which began as "tingling" sensations localized to the sole of her left foot. Over the subsequent months, she developed lumbosacral pain radiating to the right gluteal region, along with bilateral tingling and numbness in the lower extremities, predominantly on the left side. Neurological examination revealed hyperreflexia in the left lower limb and a slight decrease in muscle strength in the left leg (grade 4/5).

Thoracic spine magnetic resonance imaging (MRI) with and without contrast enhancement revealed an intradural-extramedullary lesion at the Th9–Th10 vertebral level. The lesion exhibited a "dumbbell" shape, extending through the intervertebral foramen, and measured approximately 3.5 cm at its greatest dimension (Eden type 2). The mass demonstrated heterogeneous contrast enhancement on post-gadolinium sequences. Despite these findings, the lesion did not show the characteristic "dural tail" sign typically associated with meningiomas. Based on the lesion's morphology and location, the initial imaging findings were suggestive of a schwannoma (Fig. 9).

The patient underwent a laminectomy, and facetectomy at the Th9–Th10 level to achieve complete resection of the mass, and subsequent pedicle screw fixation. Intraopera-

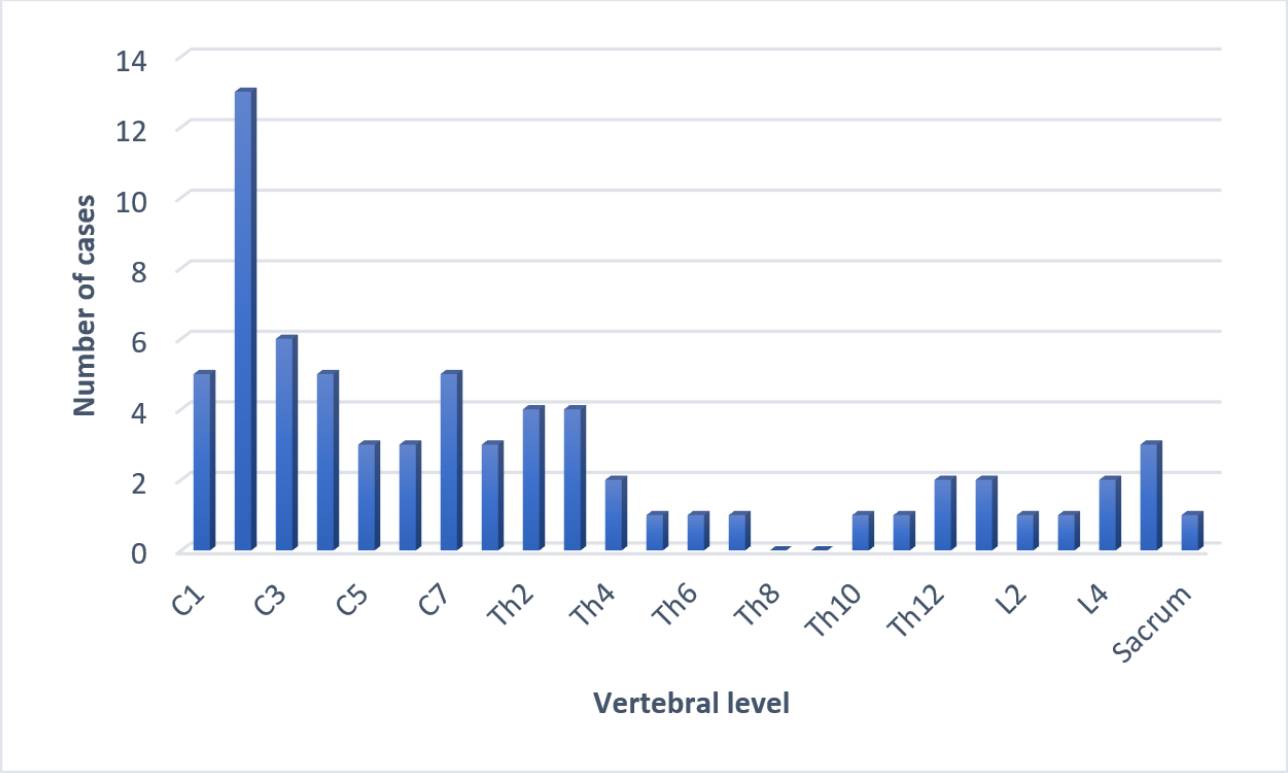

Fig. 3. Distribution of spinal dumbbell meningiomas by vertebral level. The graph illustrates the distribution of tumors across different vertebral levels. The highest peak is observed at the C2 vertebra, with 13 tumors recorded. Other levels with significant tumor presence include C1, C3, and C4.

tively, the tumor was found to be well-circumscribed and moderately adherent to the dura, without significant infiltration into adjacent neural structures. The resected specimen was sent for histopathological analysis. Microscopic examination revealed a highly cellular tumor composed of uniform spindle cells with ovoid nuclei arranged in a whorled pattern, typical of meningioma. The tumor stained positively for epithelial membrane antigen (EMA) and vimentin, while S-100 and GFAP were negative, effectively ruling out a schwannoma. These findings confirmed the diagnosis of a WHO Grade I meningioma, which was unexpected given the initial radiological impression.

The patient's postoperative course was uneventful, with gradual improvement in her neurological symptoms and signs. She was mobilized on postoperative day one and discharged on day five with a cervical-thoracic orthosis for spinal support. At the six-month follow-up, the patient reported a significant reduction in pain and paresthesia, and neurological examination showed full strength in all extremities. One-month follow-up MRI showed no evidence of residual or recurrent disease (Fig. 10).

These reported cases underscore the diagnostic challenges in differentiating between spinal meningiomas and schwannomas or other lesions based on imaging alone. Although meningiomas commonly present with the "dural tail" sign and homogeneous enhancement, their presentation can be atypical. The dumbbell shape and heterogeneous enhancement of the lesion led to a misdiagnosis of schwannoma preoperatively. This highlights the importance of considering meningioma in the differential diagnosis of spinal intradural lesions, even in the absence of classic imaging features.

Discussion

Epidemiology and Clinical Presentation

Meningiomas represent the second most common spinal intradural tumors, accounting for about 25% of such cases [1]. Among these, dumbbell-shaped meningiomas are rare and pose significant treatment challenges due to their complex anatomy. In our review of 32 patients with spinal meningiomas, there was a slightly higher prevalence in females (19 females, 13 males), yielding a male-to-female ratio of 0.68. The mean age was 46.55 $\pm$ 16.37 years, suggesting that these tumors typically affect middle-aged adults, although younger individuals, especially those with neurofibromatosis (NF), are also at risk. Notably, three of the youngest patients in our cohort were 16-year-old females, with two diagnosed with NF2 and one with NF1. This finding aligns with existing literature, which identifies younger age and neurofibromatosis as significant risk factors for dumbbell-shaped meningiomas [10, 11].

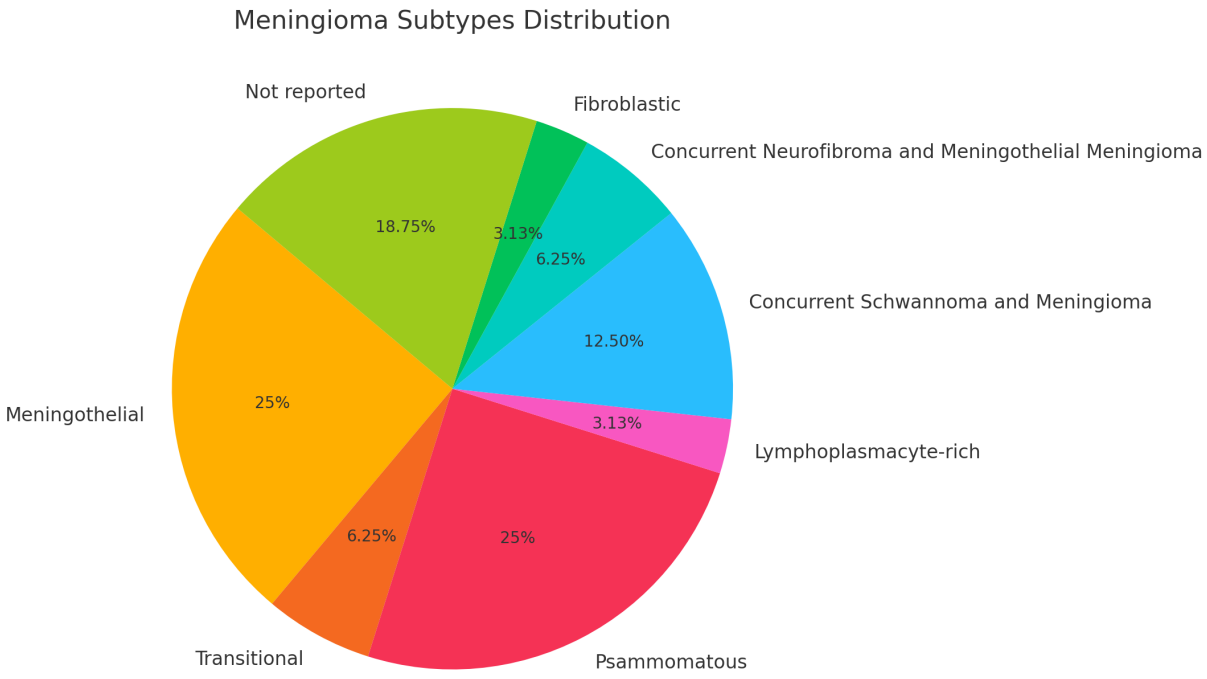

Fig. 4. Distribution of histopathological subtypes in spinal dumbbell meningioma cases. This pie chart illustrates the distribution of histopathological subtypes among spinal dumbbell meningioma cases. The most common subtypes identified were Meningothelial (25%) and Psammomatous (25%), followed by Concurrent Schwannoma and meningioma (12.5%). Other less frequent subtypes included Transitional (6.25%), Concurrent Neurofibroma and Meningothelial Meningioma (6.25%), Fibroblastic (3.13%), and Lymphoplasmacyte-rich (3.13%). In six cases, the subtype is not reported.

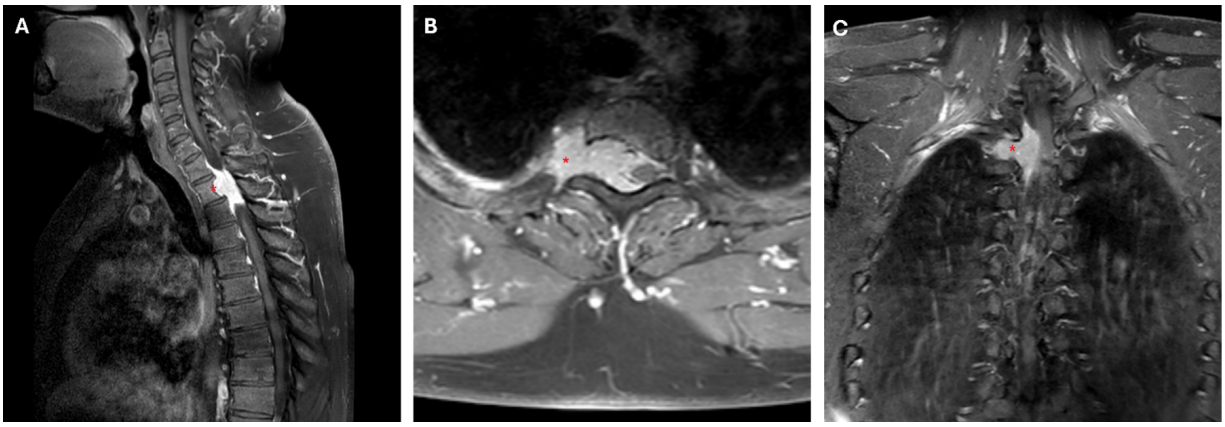

Fig. 5. Preoperative cervico-thoracic spine magnetic resonance imaging (MRI). Sagittal (A), axial (B), and coronal T1-weighted images (C) with Gadolinium contrast reveal a large intra- and extra-vertebral lesion on the right side, likely extradural, with significant homogeneous enhancement (red asterisks). The dumbbell-shaped mass, measuring approximately 50 × 35 × 14 mm, causes bony remodeling of the right posterior vertebral body of Th2, widening the ipsilateral neural foramen and extending extraforaminal to the costovertebral joint. The lesion occupies a large part of the spinal canal, forming a "sleeve" around the spinal cord and displacing it to the left (Eden type 3). This picture is from Garibaldi Hospital, Catania, Italy, and informed consent was obtained from the patient for this study.

Clinical Symptoms and Anatomical Distribution

Movement disorders were the most common clinical presentation, observed in 59.38% of patients. These included limb weakness, paresis, or spastic gait, reflecting the significant impact of these tumors on motor function. Sensory deficits were present in 40.63%, while pain and dysautonomia were less frequent. The predominance of movement disorders may be attributed to the frequent involvement of

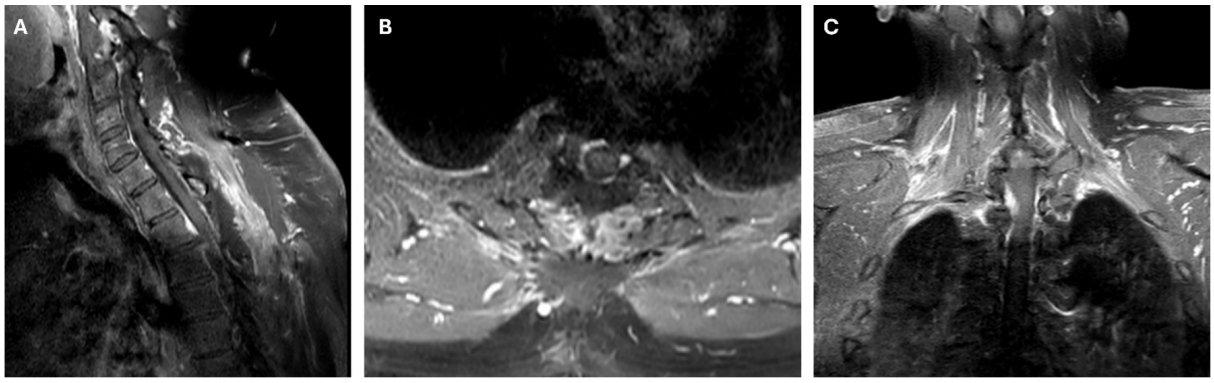

Fig. 6. Postoperative cervicothoracic spine MRI. Sagittal (A), axial (B), and coronal T1-weighted images (C) with Gadolinium contrast from the one-month follow-up thoracic spinal MRI confirm the recent surgical resection of the intracanal, extradural expansile process at Th1 to Th3. This picture is from Garibaldi Hospital, Catania, Italy, and informed consent was obtained from the patient for this study.

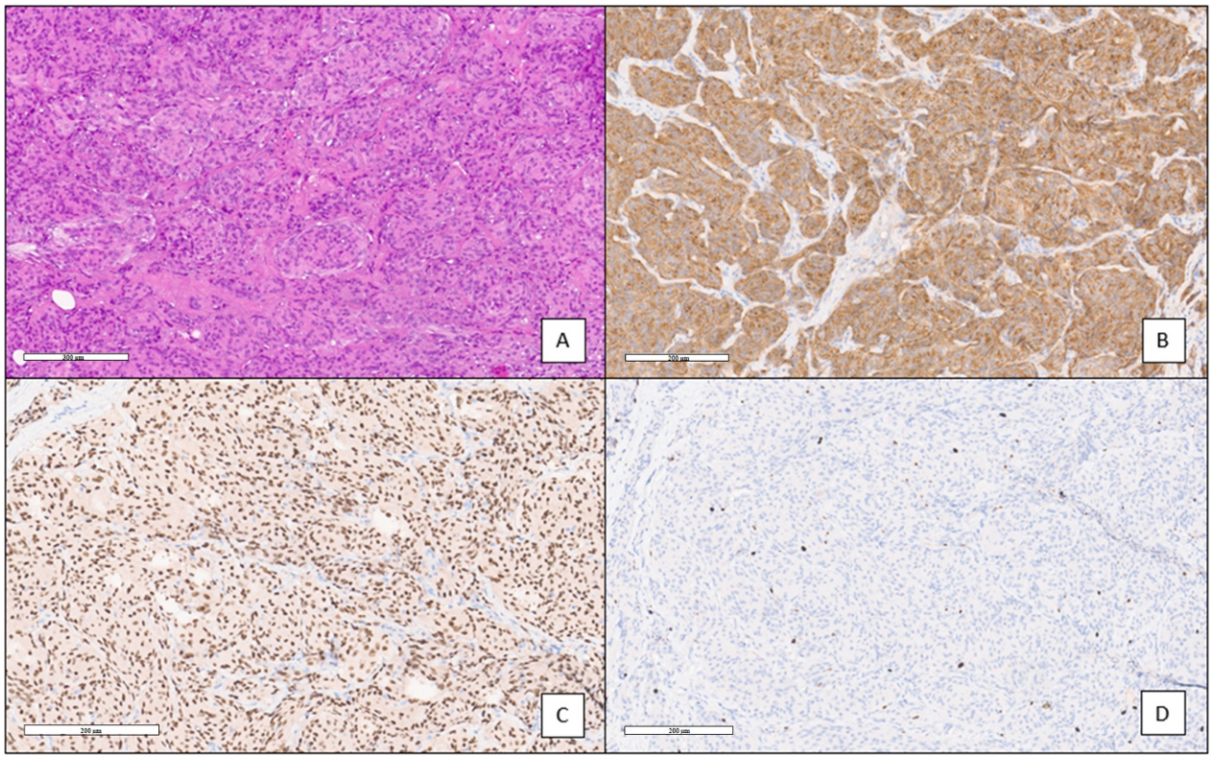

Fig. 7. Meningothelial meningioma (G1 sec. World Health Organization (WHO)). Lobulated architecture often contains meningothelial whorls; Syncytial cells with indistinct cell membranes and eosinophilic cytoplasm (A, hematoxylin-eosin (H&E) staining, scale bar, 300 µm) at 10× magnification. Immunohistochemical positivity for epithelial membrane antigen (EMA) (B) and progesterone receptor (C) at 10× magnification. Low ki67 index (D) at 10× magnification. (B–D) scale bar, 200 µm. This picture is from Garibaldi Hospital, Catania, Italy, and informed consent was obtained from the patient for this study.

the cervical spine, where 63.63% of the cases were located. This distribution is consistent with existing studies, which highlight a higher prevalence of spinal meningiomas in the cervical and thoracic regions [10, 11, 25, 26, 27, 28].

Differential Diagnosis of Spinal Dumbbell Tumors

Dumbbell-shaped spinal tumors, characterized by neural foraminal widening, present a complex diagnostic challenge due to their varied etiologies. Neoplastic tumors like meningiomas, schwannomas, and neurofibromas frequently present as dumbbell masses, often complicating diagnosis. The lack of a characteristic "dural tail" sign in spinal meningiomas, typically observed in other meningioma locations, further complicates differentiation [2]. This underscores the importance of considering a broad differential diagnosis, including other neoplastic and non-neoplastic causes such as tuberculous spondylitis, vertebral hydatid disease, and aneurysmal bone cysts [29, 30]. Ra-

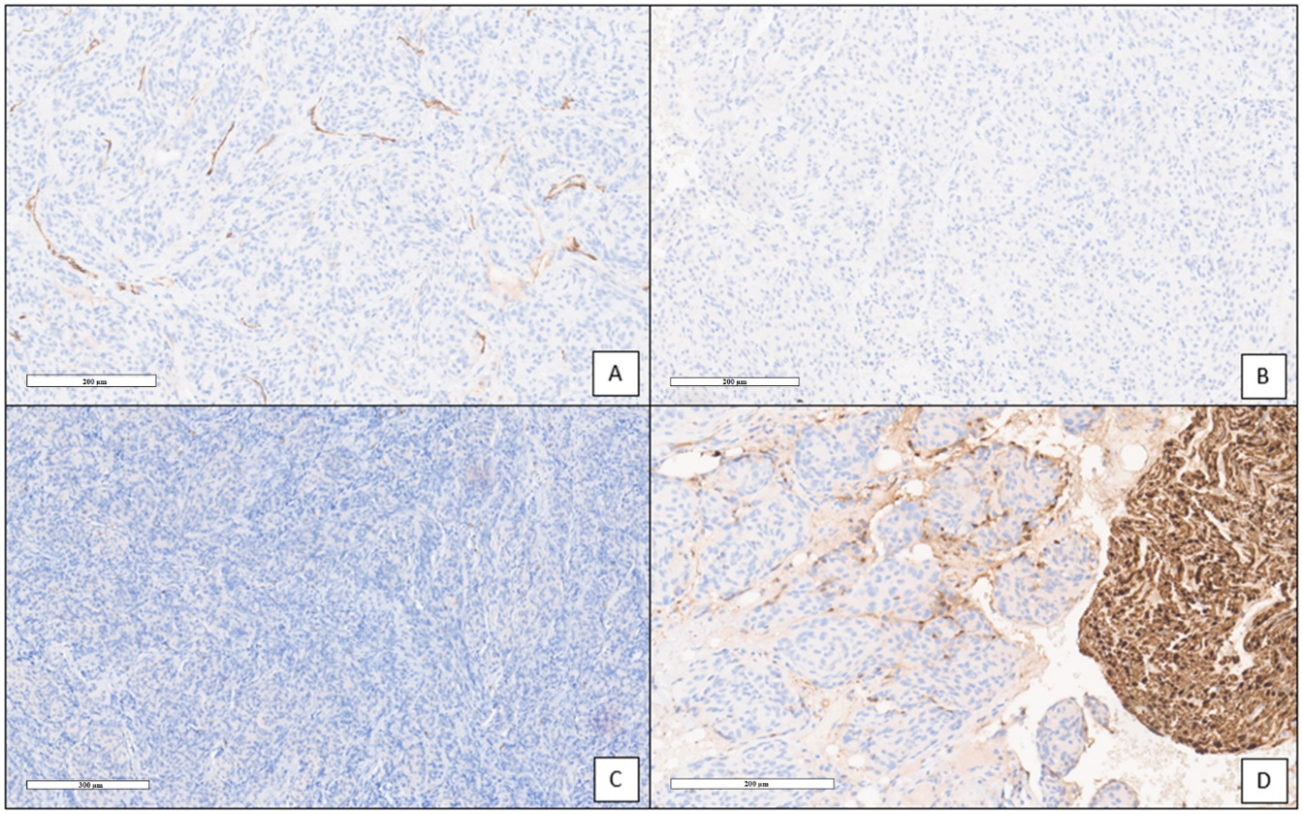

Fig. 8. Meningothelial meningioma (G1 sec. WHO). Immunohistochemical negativity for CD34 (A), glial fibrillary acidic protein (GFAP) (B) pan-cytokeratin (panCK) (C), and S100 (D) at 10× magnification. (A,B,D) scale bar, 200 µm; (C) scale bar, 300 µm. This picture is from Garibaldi Hospital, Catania, Italy, and informed consent was obtained from the patient for this study.

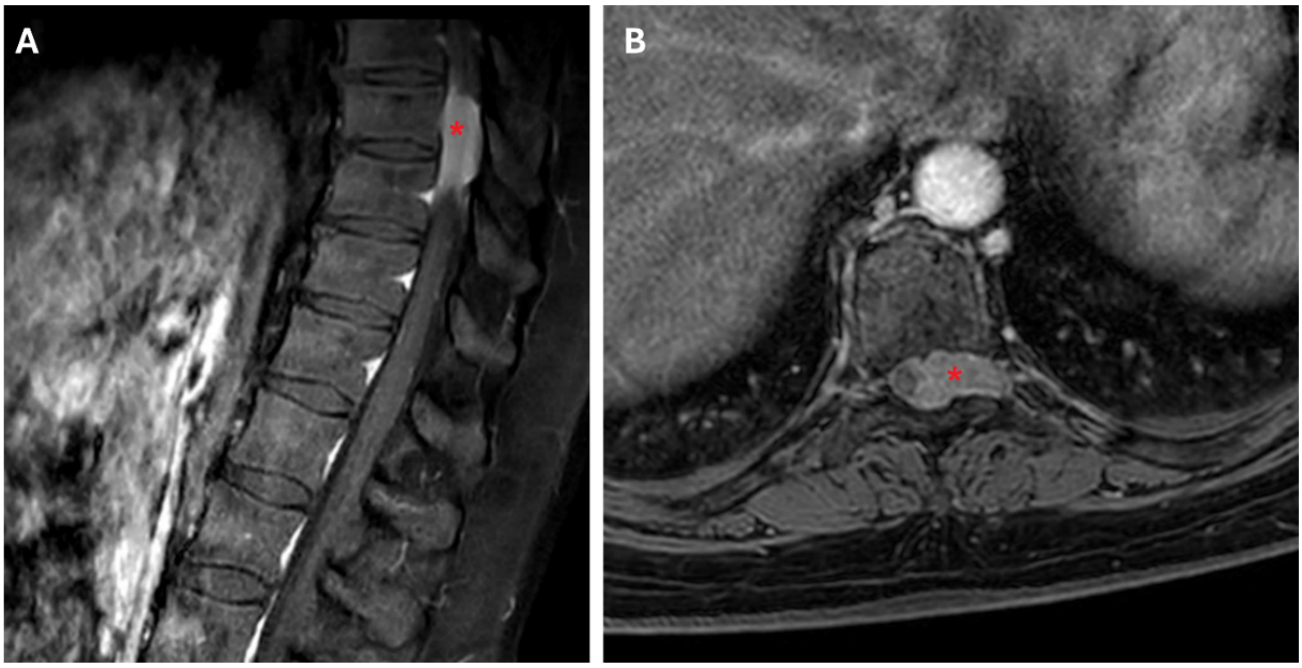

Fig. 9. Preoperative thoracic spine MRI. Sagittal (A), and axial T1-weighted image (B) with and without contrast enhancement reveal an intradural-extramedullary lesion at the Th9–Th10 vertebral level (red asterisks). The lesion displays a "dumbbell" shape, extending through the intervertebral foramen, and measures approximately 3.5 cm at its greatest dimension (Eden type 2). This picture is from S. Elia Hospital, Caltanissetta, Italy, and informed consent was obtained from the patient for this study.

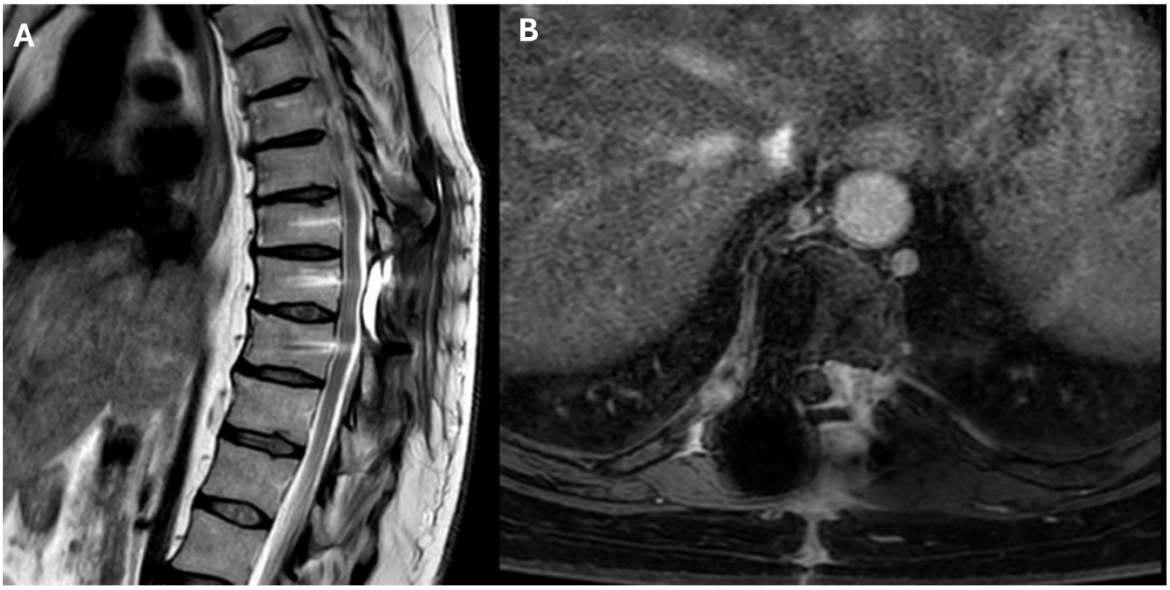

Fig. 10. Postoperative thoracic spine MRI. Sagittal T2-weighted image (A) and axial T1-weighted image (B) with Gadolinium contrast from the one-month follow-up thoracic spinal MRI confirm the recent surgical resection of the Th9–Th10 dumbbell meningioma. This picture is from S. Elia Hospital, Caltanissetta, Italy, and informed consent was obtained from the patient for this study.

diologic imaging, particularly MRI and CT, plays a pivotal role in distinguishing between these etiologies. However, the absence of typical features like the dural tail in spinal meningiomas requires careful diagnostic consideration.

Pathogenesis Theories

Several theories have been proposed to explain the pathogenesis of dumbbell-shaped meningiomas [14, 24]. One theory suggests that these tumors originate from arachnoid villi at the nerve root exit, while another posits that vestigial remnants of embryological arachnoid mater in the periradicular dura may contribute to their extradural location [21]. We categorized these tumors using the Eden classification [28], noting a predominance of Type II and III tumors, which are particularly challenging to resect due to their complex anatomical location. The progression of Type I meningiomas toward the intervertebral foramen may result in intra- or extradural extensions, while purely extradural tumors might arise from arachnoid tissue migration into the extradural space [2, 14].

Surgical Considerations and Outcomes

All patients in our study underwent surgical resection via a posterior approach. Surgical techniques varied, with laminectomy being the most common [31, 32]. Despite the technical challenges, particularly in achieving complete (Simpson I) resection, outcomes were generally favorable, with 68.75% of patients experiencing good recovery. Histopathologically, all tumors were classified as World Health Organization (WHO) Grade 1 meningiomas, with meningothelial and psammomatous subtypes being the most common. However, the recurrence rate was 15.63%, highlighting the potential for these tumors to recur despite their benign nature. Notably, one patient experienced a malignant transformation, underscoring the need for long-term follow-up [18, 31].

Challenges with Calcified and En-Plaque Meningiomas

The surgical management of calcified and en-plaque meningiomas presents significant challenges due to their adherence to surrounding tissues. These variants are associated with a higher risk of postoperative complications and recurrence, particularly in younger patients with longer life expectancies [33]. Radical surgery, including removal of the dural attachment, may be necessary to minimize recurrence risk in these patients [34, 35].

Recurrence and Long-Term Management

Recurrence remains a significant concern, as reflected by the 15.63% recurrence rate in our study. Recurrent tumors often require more extensive surgery and carry a higher risk of complications [31]. One patient experienced malignant transformation, resulting in death 12 years post-surgery despite adjuvant therapies [18]. This underscores the importance of aggressive management and long-term follow-up to prevent recurrence and malignant transformation.

Limitations

The relatively small number of cases in this study limits its statistical power, reflecting the rarity of spinal dumbbell meningiomas. Additionally, variability in reporting standards, particularly regarding surgical techniques and long-term outcomes, presents a challenge in synthesizing the data. Nevertheless, this review offers valuable insights

into the varied presentations and management strategies of these tumors, highlighting key trends that can inform clinical practice.

Future Directions

Future research should focus on multicenter studies with larger patient cohorts and standardized data collection to enhance the understanding of spinal dumbbell meningiomas. Prospective registries could capture more comprehensive data, facilitating better comparisons across studies. Long-term follow-up is crucial, particularly for cases involving subtotal resection or atypical histopathology. Additionally, investigating the molecular and genetic characteristics of these tumors could provide new insights into their pathogenesis and inform the development of novel therapies. Efforts to refine imaging criteria and explore minimally invasive surgical techniques, as well as the role of adjuvant therapies, should be prioritized to improve patient outcomes and reduce surgical risks.

Conclusions

In conclusion, dumbbell-shaped spinal meningiomas present significant challenges in diagnosis, surgical management, and long-term care. Although these tumors are generally benign and associated with good recovery rates, the risk of recurrence and the potential for malignant transformation necessitate careful surgical planning and long-term follow-up. Achieving total resection, particularly in anatomically complex regions like the cervico-thoracic spine, is critical to reducing the risk of recurrence and improving overall patient outcomes.

Availability of Data and Materials

Data to support the findings of this study are available on reasonable request from the corresponding author.

Author Contributions

GS, FG, GF, GEU, GFN, FP, EAG, and LB conceived, designed, and performed the systematic review, and wrote the initial draft. GS, GF, EG, FG, AG, and MF analyzed and interpreted the data and provided statistical analysis. All authors revised the manuscript critically for important intellectual content. All authors read and approved the final manuscript. All authors have participated sufficiently in the work and agreed to be accountable for all aspects of the work.

Ethics Approval and Consent to Participate

Written informed consent was obtained from the patients for publication and any accompanying images.

Acknowledgment

Not applicable.

Funding

This research received no external funding.

Conflict of Interest

The authors declare no conflict of interest.

Supplementary Material

Supplementary material associated with this article can be found, in the online version, at https://doi.org/10.62713/aic.3696.

References

[1] Hohenberger C, Hau P, Schebesch KM, Kölbl O, Riemenschneider MJ, Pohl F, *et al*. Spinal meningiomas. Neuro-oncology Advances. 2023; 5: i112–i121.

[2] Ozawa H, Kokubun S, Aizawa T, Hoshikawa T, Kawahara C. Spinal dumbbell tumors: an analysis of a series of 118 cases. Journal of Neurosurgery. Spine. 2007; 7: 587–593.

[3] Ogasawara C, Philbrick BD, Adamson DC. Meningioma: A Review of Epidemiology, Pathology, Diagnosis, Treatment, and Future Directions. Biomedicines. 2021; 9: 319.

[4] Roux FX, Nataf F, Pinaudeau M, Borne G, Devaux B, Meder JF. Intraspinal meningiomas: review of 54 cases with discussion of poor prognosis factors and modern therapeutic management. Surgical Neurology. 1996; 46: 458–458–63; discussion 463–4.

[5] Solanke G, Monappa V, Kudva R. Histopathological Spectrum of Meningiomas with Emphasis on Prognostic Role of Ki67 Labelling Index. Iranian Journal of Pathology. 2020; 15: 197–204.

[6] Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, *et al*. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ (Clinical Research Ed.). 2021; 372: n71.

[7] Yoshiura T, Shrier DA, Pilcher WH, Rubio A. Cervical spinal meningioma with unusual MR contrast enhancement. AJNR. American Journal of Neuroradiology. 1998; 19: 1040–1042.

[8] Buchfelder M, Nomikos P, Paulus W, Rupprecht H. Spinal-thoracic dumbbell meningioma: a case report. Spine. 2001; 26: 1500–1504.

[9] Suzuki A, Nakamura H, Konishi S, Yamano Y. Dumbbell-shaped meningioma with cystic degeneration in the thoracic spine: a case report. Spine. 2002; 27: E193–E196.

[10] Chen JC, Tseng SH, Chen Y, Tzeng JE, Lin SM. Cervical dumbbell meningioma and thoracic dumbbell schwannoma in a patient with neurofibromatosis. Clinical Neurology and Neurosurgery. 2005; 107: 253–257.

[11] Alam AM, Shuaib IL, Ghimire R. Cervical dumbbell meningioma and bilateral acoustic schwannoma in a patient with neurofibromatosis type 2. European Journal of Radiology Extra. 2005; 55: 71–73.

[12] Restrepo CS, Herrera DA, Lemos JA. Extraforaminal meningioma with extrapleural space extension. AJR. American Journal of Roentgenology. 2006; 186: 1314–1316.
[13] Santiago BM, Rodeia P, Cunha E Sa M. Extradural thoracic spinal meningioma. Neurology India. 2009; 57: 98.
[14] Kim MS, Eun JP, Park JS. A dumbbell-shaped meningioma mimicking a schwannoma in the thoracic spine. Journal of Korean Neurosurgical Society. 2011; 50: 264–267.
[15] Tuli J, Drzymalski DM, Lidov H, Tuli S. Extradural enplaque spinal meningioma with intraneural invasion. World Neurosurgery. 2012; 77: 202.e5–e13.
[16] Ozaki M, Nakamura M, Tsuji O, Iwanami A, Toyama Y, Chiba K, *et al*. A rare case of dumbbell meningioma of the upper cervical spinal cord. Journal of Orthopaedic Science: Official Journal of the Japanese Orthopaedic Association. 2013; 18: 1042–1045.
[17] Yaldiz C, Asil K, Ceylan D, Erdem S. A dumbbell-shaped meningioma in the thoracic spine: A case report. Spine Journal. 2014; 3: 168.
[18] Iwanami A, Kobayashi Y, Takano M, Mikami S, Toyama Y, Nakamura M. Invasive dumbbell spinal meningiomas: report of four cases and a review of the literature. Journal of Orthopaedic Science: Official Journal of the Japanese Orthopaedic Association. 2015; 20: 1148–1154.
[19] Sato T, Okuda T, Saito T, Yonezawa I, Onda S, Shimamura Y, *et al*. A Rare Case of Cervical Dumbbell Meningioma with the C2 Spinous Process Preservation. Open Journal of Orthopedics. 2016; 6: 98–102.
[20] Dehcordi SR, Ricci A, Chiominto A, De Paulis D, Di Vitantonio H, Galzio RJ. Dorsal extradural meningioma: Case report and literature review. Surgical Neurology International. 2016; 7: 76.
[21] Bettaswamy G, Ambesh P, Das KK, Sahu R, Srivastava A, Mehrotra A, *et al*. Extradural spinal meningioma: Revisiting a rare entity. Journal of Craniovertebral Junction & Spine. 2016; 7: 65–68.
[22] Haranhalli N, Nakhla JP, Yassari R, Kinon MD. Radiographic Pearls in the Evaluation of an Extradural Thoracic Meningioma: A Case Report. Cureus. 2017; 9: e1031.
[23] N'da H, Dauleac C, Toquart A, Afathi M, Meyronet D, Barrey C. Thoracic spine intra- and extradural dumbbell-shaped meningioma: Case report and extensive review of the literature with 21 cases. Neuro-Chirurgie. 2018; 64: 206–210.
[24] Zhang LH, Yuan HS. Imaging Appearances and Pathologic Characteristics of Spinal Epidural Meningioma. AJNR. American Journal of Neuroradiology. 2018; 39: 199–204.
[25] Zhan Z, Yan X, Nie W, Ding Y, Xu W, Huang H. Neurofibroma and Meningioma within a Single Dumbbell-Shaped Tumor at the Same Cervical Level without Neurofibromatosis: A Case Report and Literature Review. World Neurosurgery. 2019; 130: 1–6.
[26] Nguyen BQ, Tran DDT, Dang TC, Mai TD, Pham HD, Truong VT. Cervical intra-extradural meningioma with en-plaque, dumbbell-shaped, and an unusual calcified pattern in a young patient. Surgical Neurology International. 2021; 12: 454.
[27] Zheng GB, Hong Z, Wang Z. Diagnostic value of MRI in coexistence of schwannoma and meningioma mimicking a single dumbbell-shaped tumor in high cervical level. Case series and literature review. The Journal of Spinal Cord Medicine. 2023; 46: 326–331.
[28] Fukui K, Tsuji O, Suzuki S, Nori S, Nagoshi N, Okada E, *et al*. A rare case of dumbbell-shaped lymphoplasmacyte-rich meningioma arising at the upper cervical spinal cord. Journal of Orthopaedic Science: Official Journal of the Japanese Orthopaedic Association. 2023; 28: 1439–1444.
[29] Eden K. The dumbbell tumours of the spine. British Journal of Surgery. 1941; 28: 549–750.
[30] Kivrak AS, Koc O, Emlik D, Kiresi D, Odev K, Kalkan E. Differential diagnosis of dumbbell lesions associated with spinal neural foraminal widening: imaging features. European Journal of Radiology. 2009; 71: 29–41.
[31] Umana GE, Visocchi M, Roca E, Passanisi M, Fricia M, Tranchina MG, *et al*. Thoracic Spinal Angiolipomas: A Systematic Review. Journal of Neurological Surgery. Part A, Central European Neurosurgery. 2022; 83: 161–172.
[32] Maiuri F, Del Basso De Caro M, de Divitiis O, Guadagno E, Mariniello G. Recurrence of spinal meningiomas: analysis of the risk factors. British Journal of Neurosurgery. 2020; 34: 569–574.
[33] Dang DD, Mugge LA, Awan OK, Gong AD, Fanous AA. Spinal Meningiomas: A Comprehensive Review and Update on Advancements in Molecular Characterization, Diagnostics, Surgical Approach and Technology, and Alternative Therapies. Cancers. 2024; 16: 1426.
[34] Shah DS, Reddy RV, Doğruel Y, Asfour MZ, Pour-Rashidi A, Haider AS, *et al*. Calcified spinal meningiomas: a systematic review of clinical characteristics, treatment strategies, and outcomes. Journal of Neuro-oncology. 2023; 162: 295–305.
[35] Serratrice N, Lameche I, Attieh C, Chalah MA, Faddoul J, Tarabay B, *et al*. Spinal meningiomas, from biology to management - A literature review. Frontiers in Oncology. 2023; 12: 1084404.