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CASE REPORT

Giant Lumbosacral Meningioma Without Dural Attachment in an Adolescent: A Rare Diagnostic and Surgical Challenge

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Objective: To present a rare case of a huge, nondural-based atypical lumbosacral meningioma.

Background: Meningiomas are dural-based tumors that originate from arachnoid cap cells of the dural sac. Their clinical presentation is mainly due to compression of the neighboring neural structures. Management of meningiomas includes maximal surgical resection with/without radiation therapy. Herein, we present the 24th case of nondural-based, giant, lumbosacral meningioma in a young girl, a presentation that could be considered as the rarest presentation of meningiomas.

Case Presentation: The patient was a 13-year-old girl with persistent low back pain and walking difficulty for one month before our visit, without any response to conservative therapy. Imaging studies showed a huge lumbosacral meningioma at the level of L3 to S3 vertebrae.

Results: Tumor was completely resected through a posterior spinal approach. The post-operative course lacked any significant morbidity. In her one-year follow-up, complete neurological recovery without cerebrospinal fluid (CSF) leakage and any sign of regrowth of the tumor was documented. The pathologic investigation revealed meningioma grade 1 (WHO 2021 classification).

Conclusion: We presented a rare case of lumbosacral, non-dural based meningioma (WHO Grade I), which is atypical in terms of size, location, and severe adhesion to spinal nerve roots and the dura.

Keywords: atypical lumbosacral meningioma, surgical resection, non-dural tumors, cerebrospinal fluid, nerve roots

Introduction

Meningiomas are dural-based tumors that originate from arachnoid cap cells and may be intracranial or extracranial. The majority of meningiomas are slow-growing, low-grade tumors, and in some circumstances, with different grades of atypia. Ten percent of all meningiomas occur in the spinal canal.[1] Meningiomas comprise 1/4 of all the spinal canal neoplasms, with the most common site at the thoracic region, followed by the cervical and lumbar regions.[2] The location of meningioma concerning the spinal cord is mostly lateral, followed by posterior and anterior.[3] Pure sacral meningiomas have not been reported yet in the literature.[4] Meningiomas of the lumbar spine are rarely seen. Here, we present the 24th case of nondural-based spinal meningioma (L3-S3 levels) in the literature, in a young girl. It is considered "giant" due to its massive size.

Case Presentation

History and Physical Examination

A 13-year-old girl presented with a history of persistent pain in her lower back, along with bilateral radicular pain in her lower extremities for one month. No significant past medical, family, or trauma history was present. On physical examination,

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bilateral Lasegue's tests were positive, and limping was obvious during walking. On neurological examination, motor power of both lower extremities was 4/5 with bilateral hypoesthesia in L3- S1 dermatomes. No sphincter problem was complained of or detected. Deep tendon reflexes of the knees and ankles were diminished. The Babinski sign was negative on both sides.

Imaging Investigations

In a preoperative non-contrast computed tomography (CT) scan of the lumbosacral spine, scalloping of the dorsal wall of vertebral bodies, thinning of dorsal components of the vertebrae, and expansion of the spinal canal were found at L4-S3 levels, besides a hypodense mass at the same levels within the spinal canal (Figure 1). Preoperative non-contrast magnetic resonance imaging (MRI) of the lumbosacral spine showed a well-circumscribed, lobulated, intradural mass with a size of 4 × 3.5 × 8 cm, from the superior border of L3 vertebra to the inferior border of S3 vertebra, within the dural sac (Figure 2). The lesion appeared hypointense on T1-weighted MRI (T1W) and hyperintense on T2-weighted images (T2W) (Figure 2). The contrast material of the MRI could not be applied due to the patient's intolerance. The differential diagnoses of such a lesion were schwanomma. Due to a lack of bone erosion, sacral chordoma was excluded from our impression. Thus, we prepared the patient for surgical resection with an impression of lumbosacral schwanomma.

Operation and Postoperative Course

The patient was put into the standard prone position after general anesthesia and application of pneumatic leg compression on both lower extremities. Intraoperative neuromonitoring was applied on both lower extremities for assessment of motor evoked potential (MEP) and somatosensory evoked potential (SSEP), prior to positioning till repositioning after the operation. Through the posterior midline approach, after bilateral L2 to S3 laminectomy, the dura was exposed. A wide laminectomy was performed without resection of any part of the facet joints. Spinal instability and need for instrumentation were out of our concern, though. The dura mater was thinned out, and following a dorsal midline incision, a large, lobulated, pink mass bulged out through the site of the dural incision. Then, with a microscopic approach, extracapsular dissection of the tumor was started at the caudal end and extended to the cranial boundary. Severe adhesions to the dura and the nerve roots of the cauda equina were observed during the exploration and dissection. The mass was not adherent to any part of the dura circumferentially. Yet the dura was significantly thinner than normal. Due to engulfment of the nerve roots, microscopic dissection of the tumor from the nerves was carefully achieved; yet, microscopic nodules were left on some of the roots to avoid neurological damage. The result of the pathologic investigation on the frozen section of the tumor during the operation was in favor of meningioma. After gross-total resection of the tumor, hemostasis and repair of the dura were performed, using a fascial graft. Post-operative

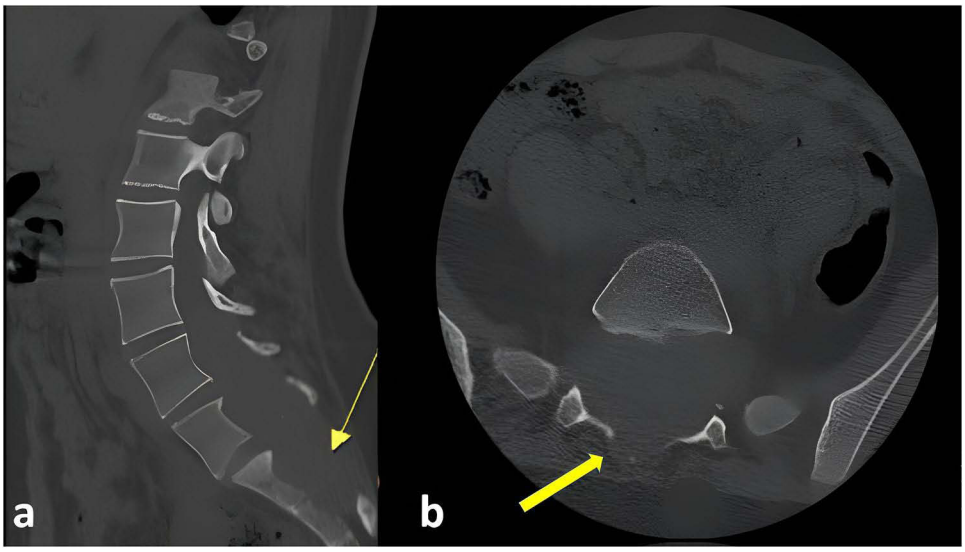

Figure 1 Pre-operative non-contrast CT-scan of the lumbosacral spine. (**a**) Mid-sagittal view, showing the scalloping of the posterior wall of L4-S3 vertebrae and a hypodense mass within the spinal column (yellow, thin arrow). (**b**) Axial view at S1-S2 level, showing expansion of the spinal canal diameter and thinning of posterior bony elements of the same level (yellow, thick arrow).

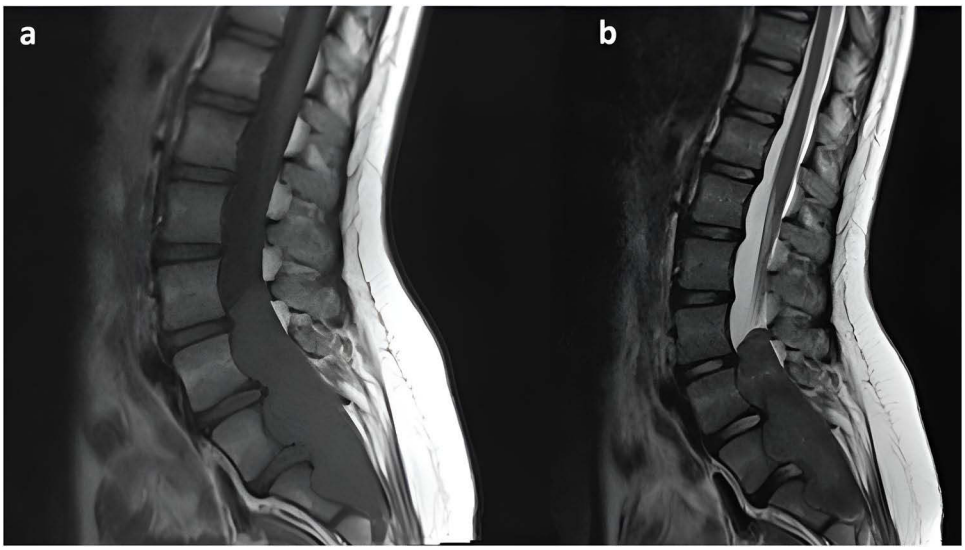

Figure 2 Preoperative non-contrast lumbosacral MRI showing an intradural space-occupying lesion that is hypointense on T1W (**a**) and hyperintense on T2W (**b**) in association with expansion of the spinal canal.

course was eventless, without any new neurologic deficit or general complications. She was discharged on the 8th postoperative day. Radicular pain was markedly improved during the first month of the postoperative status. No new neurological deficit and sign of instability were discovered in early postoperative course.

On her 9-month follow-up, no significant neurological complaint or sign was detected. MRI (with and without contrast) was performed, which showed no sign of regrowth of the tumor but a large pseudo-meningocele at the site of operation (Figures 3 and 4). On histopathological examination, the gross appearance consisted of multiple fragments of brown soft tissue, and on microscopic evaluation, the tumor comprised meningothelial cells with eosinophilic cytoplasm and an indistinct cytoplasmic border with round to oval nuclei arranged in a whorling pattern and microcalcification in a collagenous background; mitosis was about 1–2/10HPF, and necrosis was not identified. Immunohistochemistry NO: 579 in Shiraz Chamran Hospital revealed positivity for epithelial membrane antigen (EMA), with negativity for cytokeratin, S100, progesterone receptor (PR), and glial fibrillary acidic protein (GFAP) in association with a low Ki-67 proliferative index (1–2%) (Figure 5). The pathological diagnosis was meningioma grade 1 (WHO 2021 classification).

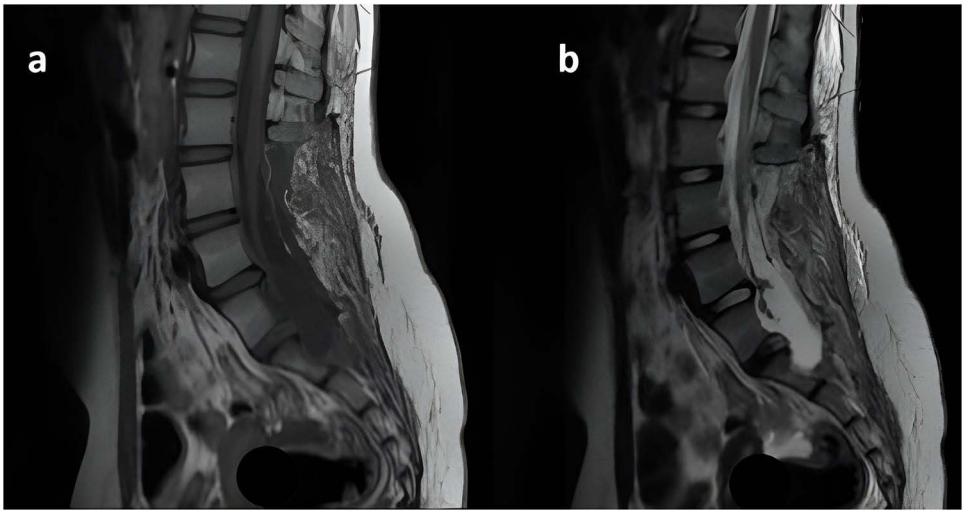

Figure 3 Early Postoperational lumbosacral MRI (T1W (**a**) and T2W (**b**) without contrast) showing the mass lesion was totally resected.

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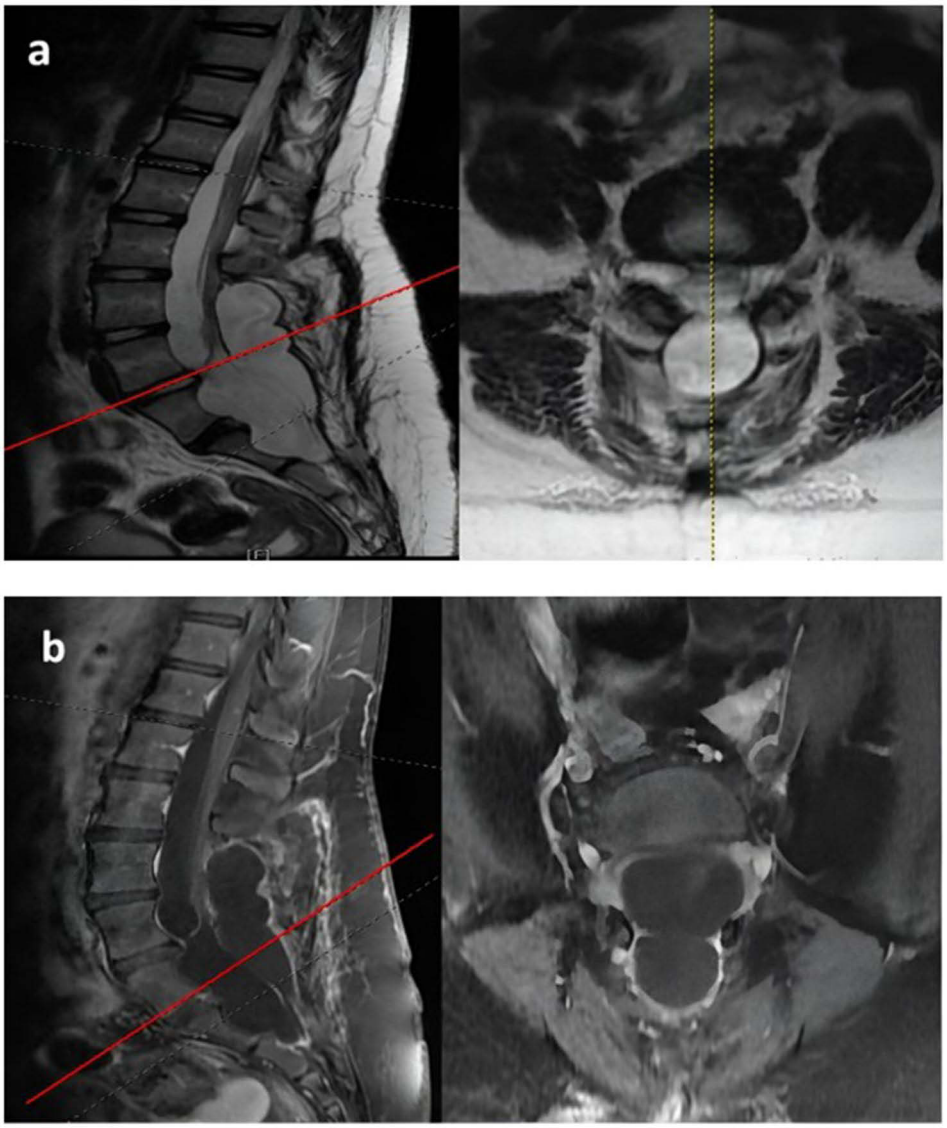

Figure 4 Late Postoperational lumbosacral MRI (1 year), showing no sign of regrowth of the tumor in contrast-enhanced T1W sequence (**b**), and with a large pseudo-meningocele at the surgical site, in T2W sequence (**a**).

Discussion

About 10–20% of all meningiomas occur in the spinal column.[5] The common age at diagnosis is 40–70 years. It is more frequent in females than males and can occur all along the spinal canal.[6] The thoracic spine is the most common site of spinal meningiomas, followed by the cervical and lumbar areas.[6,7] Lumbosacral meningiomas are a very rare presentation of such tumors, though. Cohen-Gadol et al, after 21 years of experience, reported that 40% of spinal meningiomas in the cervical spine occurred in individuals under 50 years.[8] Cunha et al reported a patient with a presentation like a lumbar spinal meningioma and a final diagnosis of tophaceous gout.[9] We presented a lumbosacral meningioma with persistent low back pain as the only presenting symptom, with atypical characteristics based on the size, location, and intraoperative characteristics.

Clinical manifestations of the lumbar spine meningioma are related to the tumor location and its relation to the spinal cord and nerve roots. These patients may complain of axial and/or radicular pain, weakness, sensory disturbances, and urinary or fecal incontinence as a late finding.[10,11] The time interval for diagnosis of spinal meningioma varies from person to person and is about 1 to 2 years.[12] In cases of nondural-based spinal meningiomas, as nearly all of them have been found in the lumbosacral region,

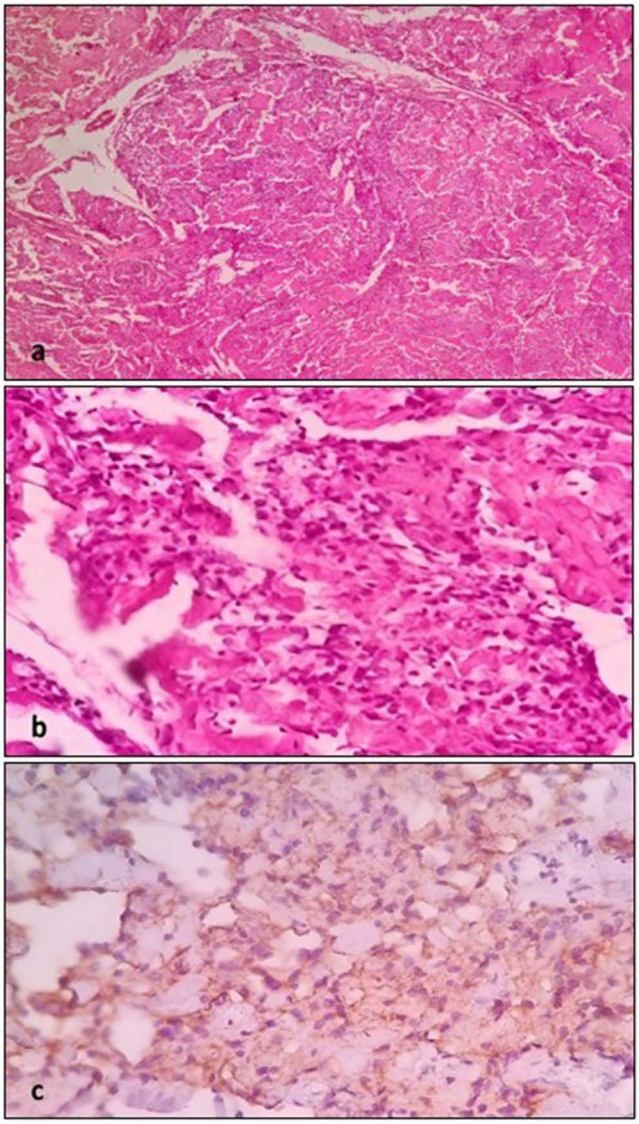

Figure 5 (**a**) A low-power view shows that the tumor has lobulated architecture and meningothelial whorls (Left, light microscopy, x40). (**b**) The high-power view shows syncytial cells with occasional whorling patterns with indistinct cell membranes, eosinophilic cytoplasm, round, uniform nuclei, and intranuclear pseudoinclusions (Middle, light microscopy, Masson Trichrome stain, x400). (**c**) High-power view shows patchy weak positive stain in the membrane of meningothelial cells (Right, light microscopy, EMA stain, x400).

claudication has been the most common presentation.[13] For the diagnosis of spinal meningioma, a CT scan and an MRI of the spine are helpful. On CT scan of spine, there is spinal canal expansion with remodeling of bony elements, beside an iso- or hyperdense mass within the spinal canal in addition to occasional hyperostosis. In the cases of classic meningiomas with dural origin, hyperosteosis and calcifications could be seen in spinal CT-scan. Yet, in cases without dural adhesion and origin (nondural-based), these findings are not seen.[13,14] The differential diagnoses in such cases of bone remodeling are chordomas and schwanommas. In cases of sacral chordoma, the bone erosion is obviously seen, and the bony anatomy of the surrounding structures could be distorted completely. Besides, the neurological damage is much more significant due to the aggressive behavior of these tumors.[15] In spinal schwanommas, spinal CT-scan could demonstrate expansion of the spinal canal, scalloping of the posterior wall of the vertebral bodies, in addition to dysgenesis of the bony element.[13,16,17] An uncommon but clinically important differential diagnosis, particularly in patients with atypical presentations or risk factors, is tophaceous gout of the spine. Although rare, spinal gout can present with back pain, radiculopathy, and even myelopathy, and may mimic intradural tumors on imaging. Cunha et al reported a case initially diagnosed as lumbar spinal meningioma that ultimately proved to be tophaceous

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gout. In our patient, the absence of erosive bone changes, the purely intradural location, and the lack of clinical risk factors for gout made this diagnosis unlikely; however, it remains a consideration in the differential diagnosis of lumbosacral lesions. In our case, the expansion of the spinal column with scalloping of the vertebral bodies guided us to the impression of a lumbosacral schwannoma. MRI is currently the imaging of choice for spinal cord tumors, like meningiomas. MRI findings include a well-defined lesion with broad-based dural attachment and/or a dural tail, which appears iso- to hypointense on T1W and iso- to hyperintense on T2W images, and contrast-enhanced T1W MRI shows moderately homogeneous enhancement. Densely calcified meningiomas are sometimes hypointense on both T1W and T2W sequences of MRI.[16] In cases of nondural-based meningiomas, the most common misdiagnosed differential diagnosis has been schwanomma,[13] as occurred in our patient. The MRI appearance of our case—a well-circumscribed, lobulated, intradural mass without dural tail or calcification—completely mimicked a schwannoma, leading to a preoperative misdiagnosis. This underscores the imaging overlap between these entities and the importance of considering meningioma in the differential diagnosis of lumbosacral intradural tumors, even in the absence of typical dural features. Besides, lack of hypointensities within the tumor (calcification), absence of dural tail or wide base on the dura, are the most misinterpreted signs as spinal schwanomma.[14,17] Also, the "dural tail" of spinal meningioma may be found in metastatic tumors and lymphomas.[18] A definitive diagnosis is mostly based on the pathological findings. The most reliable IHC marker of meningiomas is EMA,[19] which was positive in our patient. Spinal meningioma is usually diagnosed with a delay of about 2 years, and the most common symptoms are axial and/or radicular pain.

Definitive treatment for all kinds of meningioma is surgical resection. The outcome of spinal meningioma differs, related to the success of complete resection, preoperative neurological status, and histopathological findings, such as atypia and mitosis. In cases with recurrent spinal meningioma, adjuvant (or palliative) radiotherapy is utilized.[20] The most common postoperative neurologic complications consist of motor power decline, sensory disturbances, and sphincter/autonomic dysfunction. In our case, we planned open surgical resection with an impression of a lumbosacral massive schwanomma. To avoid instability, a wide laminectomy without manipulation and resection of facet joints was performed. After dural opening, we faced a huge intradural mass with a thick arachnoid lining. Exposing the cranial and caudal boundaries, microscopic piecemeal resection was utilized to avoid damaging nerve roots. Most of the cauda equina nerve roots were engulfed in the mass. Thus, mass was shaved from the nerves microscopically and carefully with the guidance of MER. Microscopic nodules (<1 mm) were forcibly left on the nerve roots. After the report of our pathologist on the frozen section (meningioma), circumferential exploration of dural adhesion was performed. Dura was very thin all along the mass circumferentially. After the resection, a massive collection and flow of CSF was obvious. Dura was closed water tightly, using a fascial graft and fibrin glue. Despite this, a pseudo-meningocele formed after the operation. The hypothesis for such circumstances – as seen in the similar cases in the literature[13,14] – could be the sudden negative pressure in the site of the resected tumor, vacuuming CSF, and subsequent high pressure of the fluid. Together with this, the circumferentially thin dura with microscopic pores (manipulation and sutures), could be the reason of leaking CSF into the surrounding dead spaces at the surgical site. The pseudomeningocele does not need any intervention till it is progressive and compresses the neural structures.[21] Thus, we took a watchful waiting path to manage the postoperative pseudomeningocele at the surgical site. Distant metastasis is rarely seen in spinal meningioma to the lung, pleura, and mediastinum, with a rate of about 0.1% and an unknown mechanism.[22,23] Altogether, radiation therapy for spinal meningioma, especially in cases of meningothelial meningiomas (WHO grade-1), is reserved for tumor recurrence, seeding, or metastasis.[13,14] We planned a serial neurological examination every month in the first year and contrast-enhanced MRI evaluation yearly after that.

While the outcome in this patient was favorable, several aspects of the diagnostic and surgical approach warrant reflection. First, the preoperative imaging was highly suggestive of schwannoma, and meningioma was not strongly considered due to the absence of dural attachment and calcification. In retrospect, a broader differential diagnosis including nondural-based meningioma could have been considered despite its rarity. Second, intraoperative ultrasound, though not part of our standard protocol, might have provided valuable real-time information regarding tumor margins and the relationship with nerve roots, potentially reducing the need for piecemeal resection. Third, although facet joints were preserved, the long-term risk of delayed instability following multilevel laminectomy in an adolescent remains a concern, and periodic dynamic imaging is now part of our follow-up plan. Finally, the development of a pseudomeningocele, despite watertight dural closure, highlights the challenge of dural repair in the setting of extensive dural thinning and the absence of posterior bony support. In future

similar cases, the use of dural substitutes and sealants, along with meticulous multilayer closure, may help reduce this complication.

For WHO Grade I meningiomas with gross total resection, routine adjuvant radiotherapy is not indicated. However, close clinical and radiological follow-up is essential, particularly in cases with residual microscopic disease, as in our patient. We recommend clinical evaluation every 3 months for the first year, every 6 months for the second year, and annually thereafter. Contrast-enhanced MRI should be performed annually for at least 5 years to detect early recurrence. In the event of symptomatic recurrence or progression on imaging, re-operation should be considered as the first-line option. If re-operation is not feasible or if the tumor shows atypical features on recurrence, stereotactic radiotherapy or fractionated external beam radiation may be considered. Patients should also be counseled regarding the rare possibility of late instability and the need for periodic dynamic imaging of the lumbar spine.

Conclusion

We presented the 24th case of nondural-based spinal meningioma (WHO Grade I) that is atypical with consideration of size, location, and severe adhesion to nerve roots. Giant lumbosacral nondural-based meningiomas can be considered the rarest presentation of meningioma in the literature so far. We present our unique case with diagnostic and surgical challenges.

Abbreviations

CSF, Cerebrospinal fluid; CT, Computed tomography; MRI: Magnetic resonance imaging; T2W, T2-weighted; MEP, Motor evoked potential; SSEP, Somatosensory evoked potential; EMA, Epithelial membrane antigen; PR, Progesterone receptor; GFAP, Glial fibrillary acidic protein; IHC, Immunohistochemistry.

Data Sharing Statement

The data used to support the findings of this study are included within the article.

Ethical Approval and Patient Consent Statement

Institutional approval from the Ethics Committee of Shiraz University of Medical Sciences (ethics code number IR.SUMS.MED.REC.1404.026) was obtained for the publication of this case report and any accompanying images. Informed consent was obtained from the patient's parents for publication of this case report and any accompanying images.

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors report no conflicts of interest in this work.

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