Thoracic Intradural-Extramedullary Epidermoid Tumor: The Relevance for Resection of Classic Subarachnoid Space Microsurgical Anatomy in Modern Spinal Surgery. Technical Note and Review of the Literature

Giuseppe M.V. Barbagallo, Massimiliano Maione, Giuseppe Raudino, Francesco Certo

■ BACKGROUND: **Intradural epidermoid tumors of the spinal cord are commonly associated with spinal cord dysraphism or invasive procedures. We report the particular relationships between spinal subarachnoid compartments and thoracic intradural-extramedullary epidermoid tumor, highlighting the relevant anatomic changes that may influence microsurgery.**

■ METHODS: **A 40-year-old woman from compressive myelopathy owing to a thoracic epidermoid tumor extending from T3 to T4 and not associated with spina bifida, trauma, previous surgery, or lumbar spinal puncture underwent microsurgical excision. Accurate tumor membrane dissection, respecting spinal arachnoidal compartments, was performed. Reposition of a laminoplasty plateau helped in restoring thoracic spine anatomic integrity.**

■ RESULTS: **Safe gross total tumor resection was achieved. Complete neurologic recovery as well as absence of recurrent tumor was documented at 4-year follow-up. A literature review revealed only 2 other cases of "isolated" thoracic spine epidermoid tumor. However, description of the relationship between tumor membranes and spinal subarachnoid compartments was not available in either case.**

■ CONCLUSIONS: **A thorough knowledge of spinal subarachnoid space anatomy is helpful to distinguish between tumor membranes and arachnoidal planes and to achieve a safe and complete resection to avoid recurrences.**

INTRODUCTION

Spinal epidermoid tumors are benign neoplastic lesions that represent <1% of all intraspinal tumors.[1-3] They are commonly intradural and extramedullary, whereas the intramedullary location is infrequent. Spinal epidermoid cysts have been reported as acquired lesions associated with trauma and surgical or invasive spinal procedures such as lumbar puncture causing the inclusion of cutaneous epithelial tissue within the intraspinal compartment.[2] The inoculation within the intradural compartment of undifferentiated cells coming from the basal layer of the epidermis has been advocated as a pathogenetic mechanism responsible for the onset of acquired spinal epidermoid cysts.[3,4] Acquired epidermoid tumors have also been described following tubercular meningitis. Conversely, congenital epidermoid cysts originate from displaced ectoderm inclusions arising in early fetal life, between 3 and 5 weeks of gestation.[5,6] They typically occur in pediatric patients and may be associated with dysraphism[7] and/or dermal sinus.[8] Given the pathogenesis of acquired spinal epidermoid cysts, the lumbar spine represents the most common location, whereas epidermoid cysts of thoracic and cervical spine have been less frequently described.[3,8] The goal of surgical treatment of spinal epidermoid tumors should be the safe resection of both cyst content and tumor membranes. Microsurgical resection can be helped by identifying and respecting arachnoidal planes.

We report a case of thoracic intradural-extramedullary epidermoid tumor not associated with congenital spinal malformation, trauma, or previous invasive procedures. The surgical management of such lesions is highlighted, and the importance of spinal arachnoidal space anatomy is discussed. Knowledge of classic microsurgical anatomy was useful to perform safe and effective tumor dissection, and reconstruction of the laminoplasty plateau helped to restore the physiologic spine anatomy according to the principles of modern neurosurgery.

Key words
- Intradural-extramedullary tumors
- Spinal epidermoid tumors
- Spinal subarachnoid space
- Thoracic spine

Abbreviations and Acronyms
CT: Computed tomography
MRI: Magnetic resonance imaging

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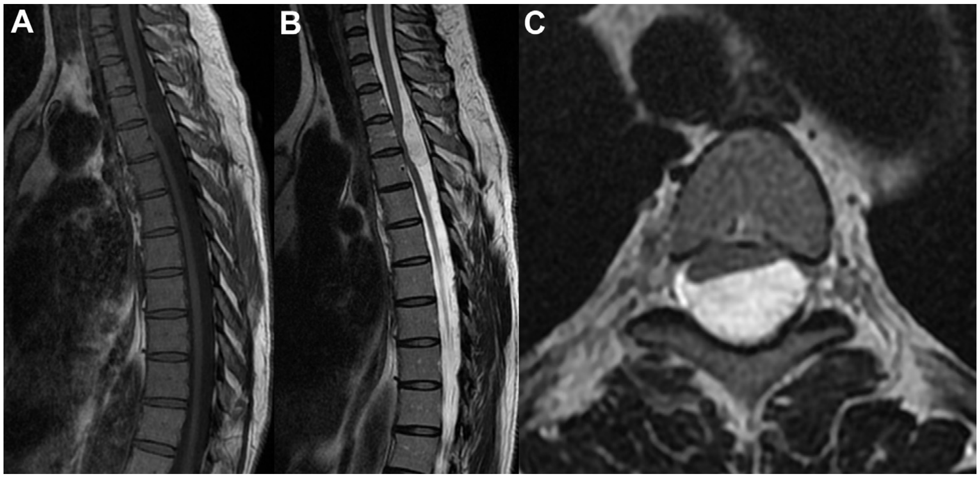

Figure 1. Preoperative thoracic spine magnetic resonance imaging. (**A** and **B**) Sagittal images show severe spinal cord compression owing to a posterior intradural-extramedullary mass at T3-T4 level. The mass appeared hypointense on (**A**) T1-weighted and hyperintense on (**B**) T2-weighted images. (**C**) Axial T2-weighted image demonstrates that the tumor mainly occupies the left posterolateral intraspinal compartment.

TECHNICAL NOTE AND SURGICAL CONSIDERATIONS

Clinical Presentation and Examinations

A 40-year-old woman presented with a 5-month history of dorsal burning pain radiating to the chest and breast. She also reported impaired sensation in her feet with numbness and freezing sensation showing a dermatomic distribution with an upper level at T3. She was referred to our institution because of worsening of neurologic deficits and recent onset of sphincter impairment with urinary retention and obstipation.

Spinal magnetic resonance imaging (MRI) documented a severe cord compression at T3-T4 level owing to the presence of a wide lesion occupying the dorsal portion of the spinal canal and anteriorly dislocating the spinal cord (Figure 1A—C). The lesion appeared hypointense on T1-weighted sequences (Figure 1A) and hyperintense on T2-weighted sequences (Figure 1B) and showed no contrast enhancement after gadolinium administration. Computed tomography (CT) scan of the thoracic spine was performed to rule out alterations of the spinal osteoligamentous structures. Neurologic examination documented tactile and thermal hypoesthesia with allodynia in the lower limbs with upper dermatomic level at T3 and lower limb hyperreflexia with the presence of bilateral suprapatellar reflexes. She had no segmental loss of strength, but mild hypotrophy of the right quadriceps muscle was detected.

Surgical Procedure

The patient signed an informed consent form, and surgical resection of the intracanalicular lesion was planned. Ethics committee approval was not required, as the proposed surgical treatment was not experimental. Surgical resection of the intracanalicular lesion was indicated. Neuromonitoring was not used in the present case, as it was not available at time of surgery. The patient was positioned prone on a radiolucent table, and a T3-T4 laminotomy plateau was elevated, creating an osteoligamentous flap including both T3 and T4 spinous processes and laminae (Figure 2A). Laminotomy was performed using a piezoelectric ultrasonic osteotome. Laminotomy was performed where the vertical segment of the lamina turned into the horizontal one, medially to the costal process, with the same surgical exposure routinely used for a standard thoracic laminectomy. The dura mater was incised in the midline and suspended to the surrounding stitches, according to standard microsurgical technique. A typical pearly, avascular lesion appeared under the dura mater, covered by the arachnoidal sheet and entirely occupying the dorsal portion of the spinal canal (Figure 2B). Following arachnoidal opening over the most prominent tumor component, microsurgical debulking of the tumor was started. After reduction in volume, careful identification and dissection of the tumor membranes began, trying to respect arachnoidal planes and avoiding coagulation. The medial dorsal arachnoidal septum was identified during dissection of tumor membranes off the right side of the spine. It was fenestrated and displaced to the right side where it was attached to the right posterolateral arachnoidal septum (Figure 2C). Both the medial dorsal septum and the right posterolateral septum had been stretched by the tumor mass and dislocated over the right dorsal rootlets. Consequently, the dissection of tumor membranes from arachnoidal planes on the right side did not cause any traction on nerve roots. Conversely, on the left side, tumor membranes were adherent to the left posterolateral arachnoidal septum (Figure 2D). This was widely fenestrated and pushed over the left dorsal nerve rootlets (Figure 2E). A gentle dissection of residual tumor membranes on the left side was started from the posterolateral arachnoidal septum toward the midline. Avoidance of inadvertent traction of neural structures was helped by the presence of the posterolateral arachnoidal septum, albeit incomplete, covering the left dorsal rootlets. After removing the tumor and its membranes (Figure 2F), dentate ligaments were visualized on both sides. The anterior subarachnoid compartment was also exposed and fenestrated at

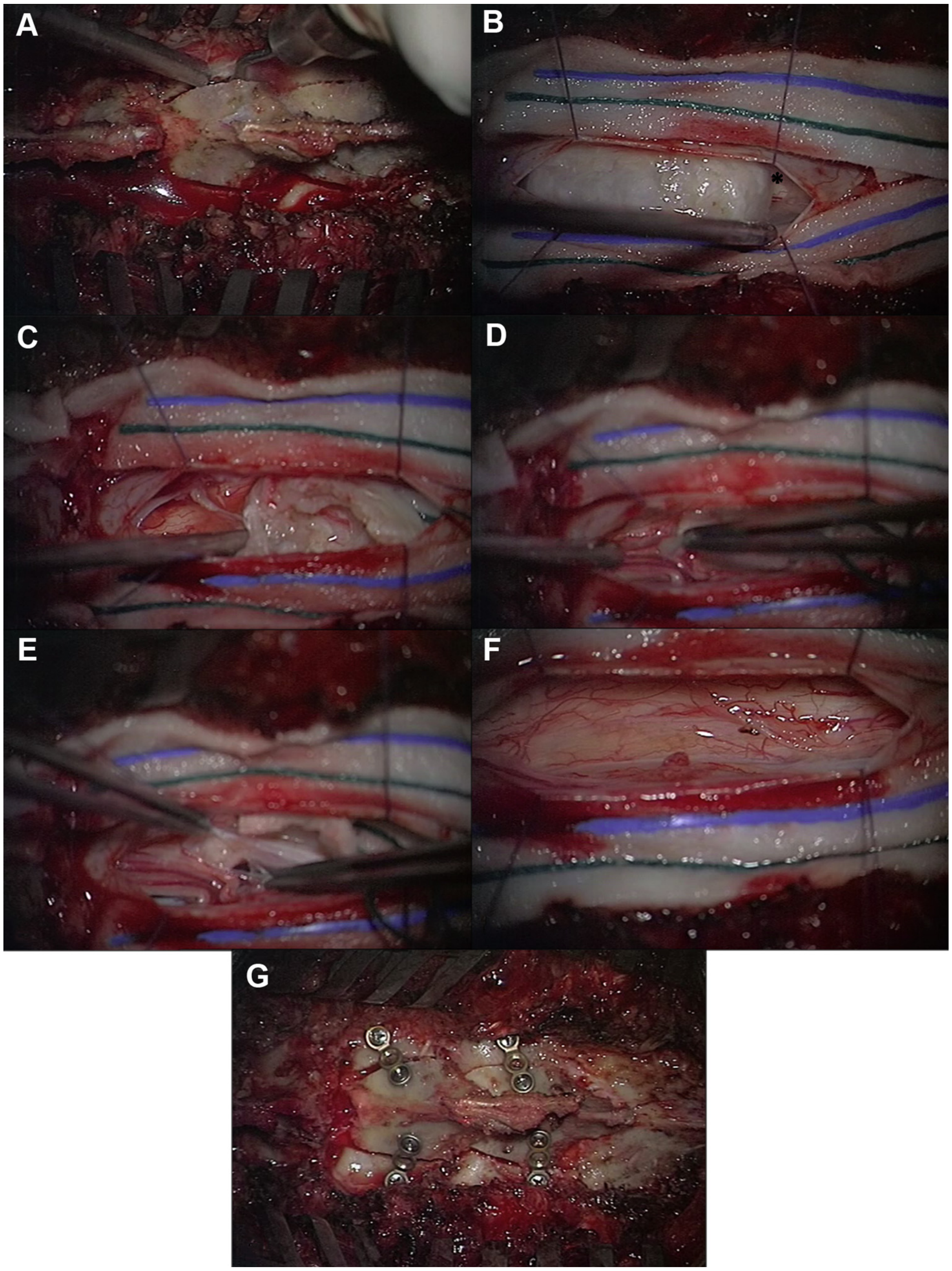

Figure 2. Intraoperative frame shots show the microsurgical removal of the intradural-extramedullary thoracic epidermoid tumor. (**A**) A laminoplasty plateau including T3 and T4 spinous processes and laminae is performed using a piezoelectric osteotome with minimal loss of bone. (**B**) After dural opening and outer arachnoidal incision, a typical pearly mass appears, displacing midline dorsal arachnoidal septum (*asterisk*). (**C**) After tumor debulking, epidermoid membranes were easily dissected and removed on the right side without exerting any traction on neural structures. (**D**) On the left side, dissection of tumor membranes was started from the arachnoidal layer covering the dorsal rootlets and proceeding toward the midline. (**E**) Left dorsolateral arachnoidal septum was widely fenestrated and difficult to recognize. (**F**) Complete resection of the epidermoid tumor including its membranes was achieved. (**G**) Osteoligamentous flap was replaced and fixed using titanium miniplates.

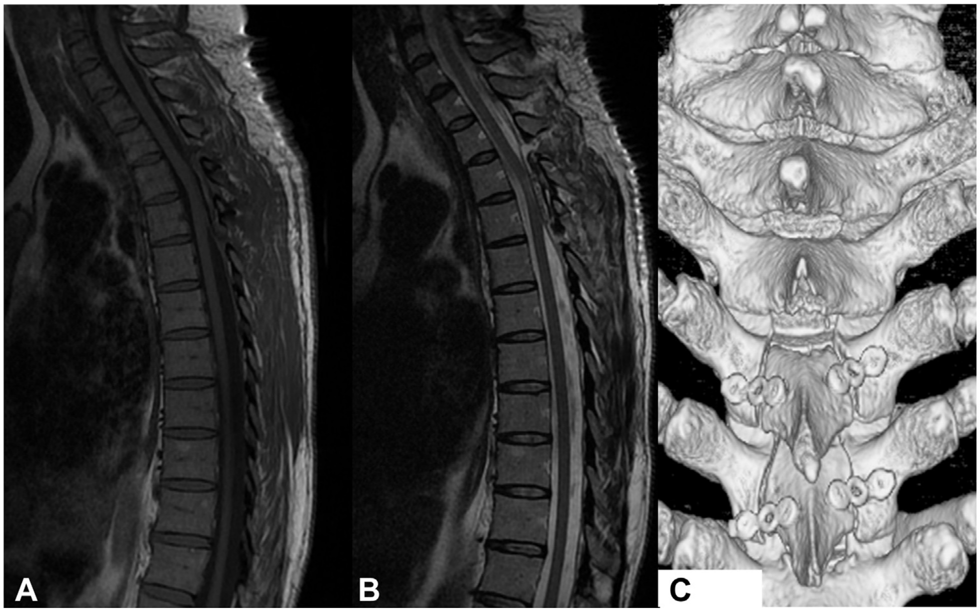

Figure 3. Postoperative (**A**) T1-weighted and (**B**) T2-weighted sagittal magnetic resonance imaging sequences show complete tumor resection without complications. A vague intramedullary hyperintense area is now visible on T2 images. (**C**) Postoperative three-dimensional rendered high-resolution computed tomography scan depicts the reconstruction of the laminoplasty plateau with titanium miniplates.

the end of the procedure. At the end of the procedure, the dura mater was sutured, and the osteoligamentous flap was repositioned and fixed with titanium miniplates (Figure 2G).

Postoperative Course and Outcome

The postoperative course was uncomplicated, and the patient was discharged home 3 days after the procedure. The patient reported improvement of lower limb sensory disturbances. Postoperative MRI and CT showed complete tumor resection (Figure 3A and B) and restoration of the physiologic thoracic spine anatomy (Figure 3C). Histology confirmed the diagnosis of epidermoid cysts (Figure 4). At the 4-year follow-up examination, full neurologic recovery was documented with no recurrence (Figure 5A–C). CT scans demonstrated ossification of the laminoplasty plateau (Figure 6A–C) and maintenance of a correct spinal alignment (Figure 6D).

DISCUSSION

Surgical Management of Spinal Epidermoid Tumors

The rationale of surgical treatment of spinal epidermoid cysts is similar to that reported for intracranial epidermoid cysts.[9] An extended resection should be pursued, but respect of anatomic integrity of neurovascular structures is mandatory.[9] Gross total resection of the pearly content of the cyst, including external membranes, is the optimal treatment and minimizes the risk of epidermoid tumor recurrence.[3,10] All surgical maneuvers must be delicately and gently performed to avoid spreading of cyst content into the subarachnoid spaces (responsible for chemical arachnoiditis) and the inadvertent traction of neural structures during membrane dissection.[11]

Surgical management of spinal thoracic epidermoid tumors can be performed correctly only when considering and respecting the anatomy of the subarachnoid compartments.[12] The microsurgical anatomy of the spinal subarachnoid space was extensively described by Nauta et al.[13] in an article based on surgical experience and cadaveric dissection. They studied the structure of spinal subarachnoid spaces in 13 adult cadaveric specimens without premortem evidence of spinal pathology. Nauta et al.[13] highlighted the importance of knowledge of the particular anatomic features of spinal arachnoidal septa and compartments for proper surgical planning.

The normal anatomy of subarachnoid compartments and septa is illustrated in Figure 7A. According to the observations by Nauta et al.,[13] the outer spinal arachnoidal membrane is formed of a dense, continuous, and watertight layer. The longitudinal midline dorsal septum, variably fenestrated, is located between the outer layer and the middorsal superficial vein, separating 2 symmetric and specular arachnoidal posterolateral compartments. This longitudinal septum is present from the middle cervical to the superior lumbar levels. In the higher cervical region as well as at lower lumbar segments, it progressively becomes more fenestrated before disappearing. Two more laterally sited subarachnoid compartments are also present. These are separated from the posterolateral spaces by 2 further dorsolateral septa (1 on each side), which follow the oblique course of dorsal nerve rootlets and envelop the neural structures from the root entry zones to the neural foramina. These dorsolateral septa are considerably less regular and often fenestrated compared with the midline dorsal septum. Therefore, the posterior subarachnoid space is often divided into 4 compartments, 2 on each side, albeit several intraindividual and interindividual variations were described by Nauta et al.[13]

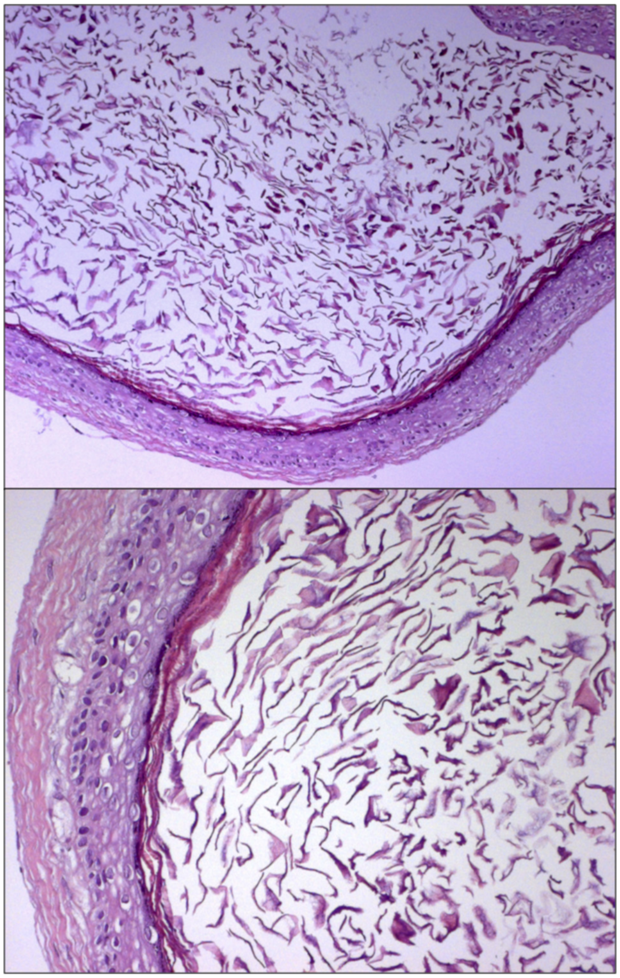

Figure 4. Histology microscope slides confirming the diagnosis of epidermoid cyst. (*Upper panel*) The squamous epithelium sheds the anucleate squames that create the bulk of the mass (hematoxylin and eosin, 100×). (*Lower panel*) No adnexal appendages, hair follicles, or sebaceous glands were observed in the fibrous wall underlying the squamous epithelium (hematoxylin and eosin, 200×).

Conversely, the anterior subarachnoid space is commonly a single compartment without septa; posterior compartments are separated from the anterior compartment by longitudinal arachnoidal membranes covering dentate ligaments (**Figure 7**A).

As pointed out by Nauta et al.,[13] anatomic knowledge of the spinal subarachnoid spaces is essential during surgical management of intradural-extramedullary lesions. We assume that the rare "isolated" spinal epidermoid cysts have similar behavior to intracranial epidermoid cysts, as slowly growing lesions that respect arachnoidal planes. In such cases, spinal anatomy is not violated by malformative conditions or by iatrogenic injuries, and the mechanism of growth of epidermoid tumors is the same as that described for intracranial tumors. In the present case, the tumor occupied the posterior spinal canal but with a prevalent extension on the left side. Consistent with the above-mentioned anatomy, the tumor probably grew within the left posterolateral subarachnoid compartment and eventually displaced the dorsal septum to the right side (**Figure 7**B). As a consequence, during dissection maneuvers, we found the epidermoid membranes adherent to the median dorsal septum, and this formed a cleavage plane from the right-sided dorsal rootlets. We managed to identify and remove the right lateral tumor membranes from the left surface of the median dorsal septum. The septum itself appeared fenestrated in places, and we had to carefully follow and remove the tumor membranes. We hypothesized that a double arachnoidal layer derived from the conjunction of the displaced dorsal septum and the right dorsolateral one was present. This facilitated tumor membrane dissection. Conversely, on the left side, the epidermoid membranes were adherent to the left dorsolateral arachnoidal septum; however, this septum was largely fenestrated and in some areas difficult to recognize. Therefore, to achieve a radical tumor resection, we started our dissection from the arachnoidal layer covering the dorsal rootlets and proceeded toward the midline. This strategy and the awareness of the presence of the left dorsolateral arachnoidal septum over the ipsilateral rootlets allowed us to safely and entirely remove the tumor membranes (**Figure 7**C). The awareness of microsurgical anatomy and the respect of arachnoidal planes allowed us not only to achieve a radical resection of tumor membranes but also to protect neurovascular structures, avoiding damage related to the loss of a cleavage plane. The possibility to completely and safely dissect the tumor membranes from neural structures, following arachnoidal planes, was related to the surgeon feeling tissue resistance and elasticity. Arachnoidal membranes were respected when feasible, as they represent a further protection for neural structures. This concept was transferred and adapted to this case from the experience with intracranial epidermoid tumor surgery, as reported in the literature. Similarly to intracranial epidermoid tumors, we stop surgical maneuvers to remove tumor membranes if these are densely adherent to neurovascular structures to avoid new neurologic deficits. Finally, we fenestrated all posterior arachnoidal septa into the anterior septum to balance the cerebrospinal fluid pressures within the subarachnoid compartment and to reduce the risks of postoperative arachnoid cyst formation.[13]

Reconstruction of the posterior vertebral elements was obtained by fixating the laminoplasty plateau with titanium miniplates. The piezoelectric osteotome provided a precise bone cut with a minimal gap. This helped the subsequent ossification of lamina and the restoration of physiologic spinal anatomy, as documented by follow-up CT scans (**Figure 6**). Despite the fact that intradural-extramedullary tumors can be resected with a unilateral laminectomy, we preferred to perform a laminoplasty plateau in the present case to achieve wider surgical exposure and to obtain better control of the entire posterior intradural compartment. Nonetheless, repositioning of the osteoligamentous plateau and its subsequent ossification avoided postoperative spinal deformity. This occurrence is more frequent in children, but there is some evidence supporting the role of the so-called osteoplastic laminotomy in reducing the risk of long-term spinal deformity and increasing stability in the thoracolumbar spine.[15]

Literature Review of Isolated Thoracic Spine Epidermoid Tumors

Epidermoid cysts are rarely reported in the cervicothoracic spine; in particular, cysts arising in the thoracic spine account for <0.8% of all spinal epidermoid cysts.[1-3,8] Pediatric patients are more frequently affected.[8,11] Two different types and etiologies for

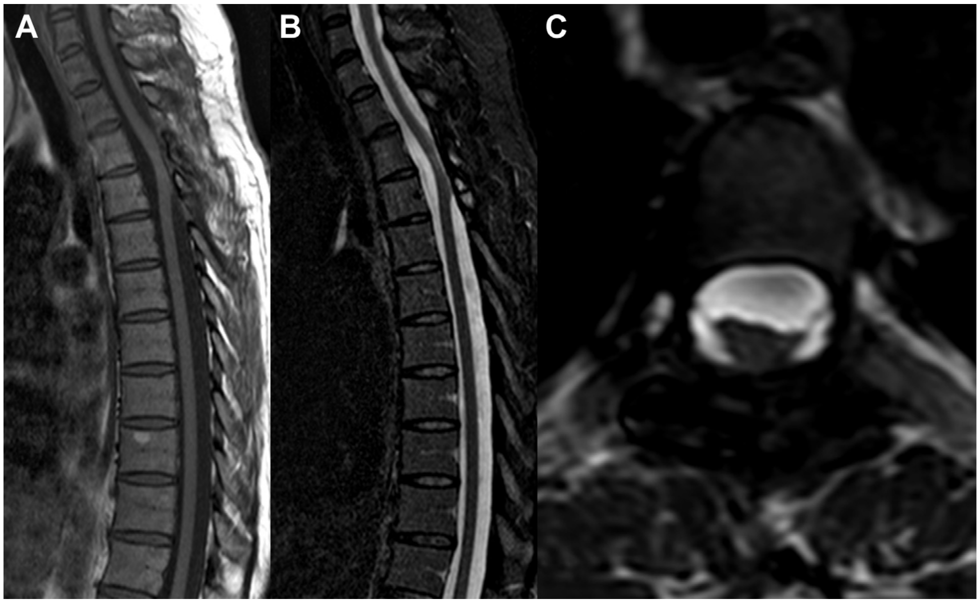

Figure 5. Magnetic resonance imaging obtained at 4-year follow-up. (**A** and **B**) Sagittal and (**C**) axial images document the absence of recurrent tumor and the reduction of hyperintense intramedullary area.

Figure 6. (**A** and **B**) Axial, (**C**) coronal, and (**D**) sagittal reconstructed computed tomography images obtained at 1-year follow-up document ossification of the laminoplasty plateau and maintenance of a correct spinal alignment.

spinal epidermoid cysts have been described: congenital malformations associated with cutaneous and osseous abnormalities (i.e., spina bifida, localized hypertrichosis, dermal sinus) and acquired lesions following lumbar puncture.[14,16,17] An abnormal inclusion of ectopic ectodermal cells during closure of the neural tube and previous serial lumbar punctures have been advocated as pathogenetic mechanisms responsible for the occurrence of spinal epidermoid tumors.[14]

Another, less common, type includes "isolated" spinal epidermoid cysts, which are not associated with dysraphisms, invasive procedures (i.e., lumbar punctures), or trauma.[17] A literature review revealed that only 8 cases of isolated spinal intradural-extramedullary epidermoid cyst have been reported.[3,6,8,10,16,18] A thorough analysis of these reports demonstrated only 3 cases of spinal epidermoid tumors occurring in adult patients, 2 in the thoracic spine[3,8] and 1 in the lumbar spine[16]; the remaining cases occurred in pediatric patients.[6,10,18]

The differential diagnosis of an "isolated" spinal epidermoid cyst can be difficult. The MRI features of such lesions can be similar to features described for an arachnoid cyst on T1-weighted and T2-weighted sequences, and postcontrast images may be inconclusive.[3,6] Moreover, spinal epidermoid cysts should be differentiated preoperatively from spinal dermoid cysts and spinal neurenteric cysts. Similar to intracranial epidermoid cysts, the differential diagnosis can be helped by diffusion-weighted imaging, fluid attenuated inversion recovery sequences of MRI, and, in some cases, CT scan.[5,19] Spinal arachnoid cysts do not show restriction on diffusion-weighted imaging, and their signal is often suppressed by fluid attenuated inversion recovery; spinal dermoid cysts usually contain fatty elements, and CT scan may show intratumoral calcifications; spinal neurenteric cysts are usually located in the ventral part of the spinal cord or within the spine and are frequently associated with vertebral anomalies. In the present case, diffusion-weighted imaging and fluid attenuated inversion recovery sequences were not performed.

CONCLUSIONS

Isolated epidermoid cysts have to be considered in the differential diagnosis of intradural-extramedullary lesions of the thoracic spine. A thorough knowledge of classic spinal subarachnoid space anatomy is essential for correct surgical planning and helps to achieve safe and complete dissection of membranes; this is crucial to prevent tumor recurrence. Respect of

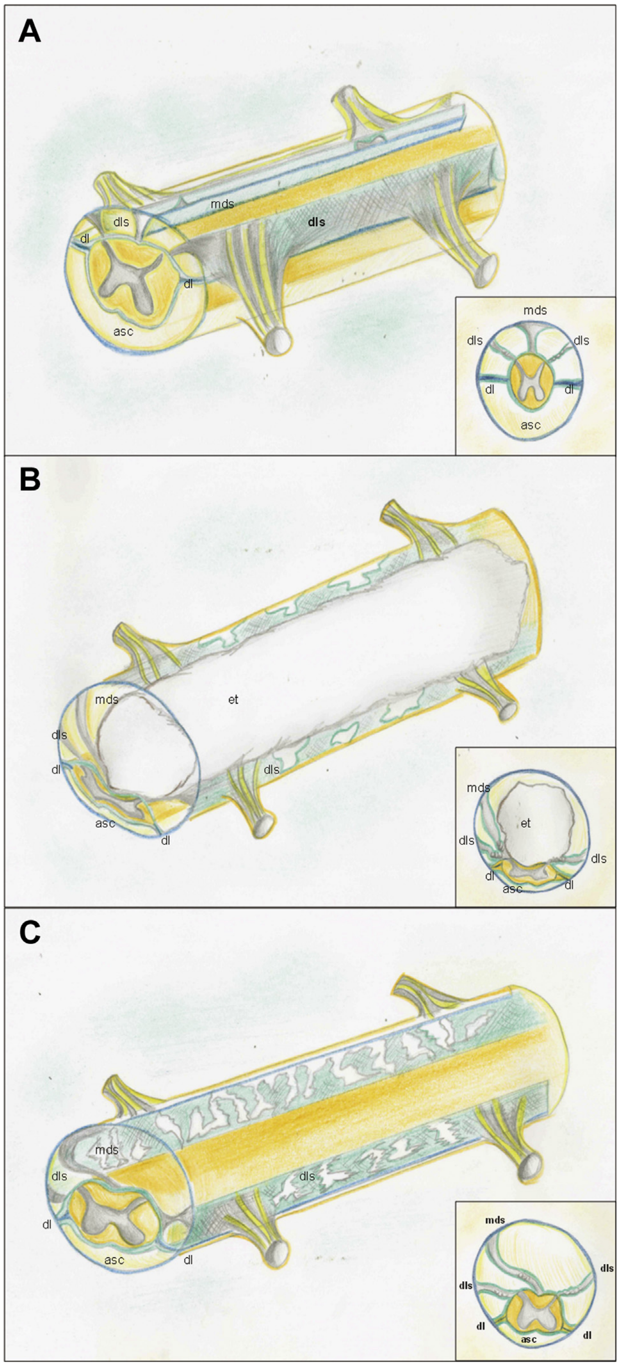

Figure 7. Anatomic relationship between epidermoid tumor and arachnoidal planes is schematically shown. The outer spinal arachnoidal membrane is formed by a dense, continuous, and watertight layer.[14] The longitudinal midline dorsal septum, variably fenestrated, is located between the outer layer and the middorsal superficial vein, separating 2 symmetric and specular arachnoidal posterolateral compartments (Figure 3A). This longitudinal septum is present from the middle cervical to the superior lumbar levels. In the higher cervical region as well as at lower lumbar segments, it progressively becomes more fenestrated before disappearing. Two more laterally sited subarachnoid compartments are also present. These are separated from the posterolateral spaces by 2 further dorsolateral septa (1 on each side) that follow the oblique course of dorsal nerve rootlets and envelop the neural structures from the root entry zones to the neural foramina. These dorsolateral septa are considerably less regular and often fenestrated compared with the midline dorsal septum. Therefore, the posterior subarachnoid space is often divided into 4 compartments, 2 on each side, albeit several intraindividual and interindividual variations have been described. Conversely, the anterior subarachnoid space is commonly a unique compartment without septa; posterior compartments are separated by the anterior compartment by longitudinal arachnoidal membranes covering dentate ligaments. (**A**) Normal subarachnoid space anatomy was distorted by the presence of (**B**) tumor arising from the left subarachnoid compartment and displacing the midline dorsal septum to the right side. (**C**) Midline dorsal septum as well as dorsolateral septa was highly fenestrated, as seen after completing tumor membrane dissection. asc, anterior subarachnoid compartment; dl, dentate ligament; dls, dorsolateral septum; mds, midline dorsal septum; et, epidermoid tumor.

classic microneurosurgical anatomy combined with the use of modern neurosurgical techniques and the application of spinal stability concepts leads to the best result.

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