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Case Report

A Pediatric Case of Cauda Equina Dermoid Cyst Resected by Minimally Invasive Unilateral Hemilaminectomy

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A 3-year-old boy had difficulty sitting up and walking for several months. Magnetic resonance imaging (MRI) revealed an intradural tumor at the L3-4 level. The tumor was successfully resected by unilateral hemilaminectomy and diagnosed as dermoid cyst. The patient had an uneventful postoperative course without pain, and MRI found no recurrence after surgery. A small bone defect remained that might be favorably reconstructed with autologous and artificial bone. Hemilaminectomy allowed us to resect the cauda equina dermoid cyst with minimal invasiveness. Pediatric patients require follow-up as they are more likely to experience spinal deformity or instability after surgery.

Key words: cauda equina tumor, child, dermoid cyst, hemilaminectomy, spinal tumor

Cauda equina tumors are rare in pediatric patients [1], and the incidence rate of pediatric spinal tumors, including intramedullary and intradural extramedullary tumors, has been reported to be 1-2.6 per one million children around the world [2]. The Central Brain Tumor Registry of the United States (CBTRUS) reports that spinal cord and cauda equina tumors account for only 4.3% of all central nervous system tumors in children (age 0-14 years) [3]. Dermoid cyst is a commonly benign congenital tumor derived from ectodermal inclusions left behind after neural tube closure with subsequent separation of the cutaneous and neural ectoderm [4]. Spinal dermoid cysts account for 0.8-1.1% of all spinal tumors [5]. Surgical resection is the most common treatment for symptomatic lesions while follow-up is an option for asymptomatic lesions [6]. The standard surgical approach for the removal of a tumor in the spinal canal is single-level or multilevel laminectomy [1,7,8]. However, this traditional approach poses an increased risk of damage to the posterior supporting structure, which can lead to postoperative pain and the progression of kyphotic deformity in pediatric patients [9-11]. Importantly, spinal deformity after laminectomy is considered critical, with reported incidence rates ranging from 16% to 100% in pediatric patients [9-11]. Chou *et al.* first reported unilateral hemilaminectomy for the resection of spinal tumors [12]. This approach allows surgeons to minimize bone resection and save the spinous process, the contralateral facet joints, and the contralateral paraspinal musculature [13]. Following Chou's report, many spinal surgeons have recognized the importance of minimally invasive surgery for the removal of tumors in the spinal canal. There have been reports on hemilaminectomy for disc herniation, epidural hematoma, neu-

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romuscular scoliosis, and spinal cord tumor in pediatric cases [14-17], but there are few reports for cauda equina tumor. We describe a pediatric case of cauda equina dermoid cyst resected by minimally invasive unilateral hemilaminectomy.

Case Report

A 3-year-old boy presented with low back pain caused by increased abdominal pressure. The low back pain had progressed over several months, and he had difficulty in sitting up and walking in addition to sensory disturbance in the right lower extremity. He was referred to our department, where general examination revealed no abnormality. His past medical history was insignificant. Magnetic resonance imaging (MRI) showed a tumor at the L3-4 level with isointensity to slightly high intensity on T1-weighted imaging (T1WI), high intensity on T2-weighted imaging (T2WI) (Fig. 1A), and no enhancement on contrast-enhanced T1-weighted imaging (CE-T1WI) (Fig. 1B). Based on the above, we considered intradural extramedullary tumors (*e.g.*, schwannoma, meningioma, and epidermoid/dermoid cyst) as differential diagnoses for this tumor at this point. We concluded that the patient's pain in the low back and lower extremities was induced by compression of the cauda equina with this tumor.

We decided to remove the tumor. We planned a minimally invasive operative approach. We performed right unilateral hemilaminectomy at the L3 level and right unilateral dome hemilaminectomy in order to remove the ligamentum flavum at the L2/3 and L3/4 levels (Fig. 2A). In the event of a bloody tumor or a tumor strongly adhesive to the surrounding nerves, we had a plan to enlarge the fenestration. Intraoperative findings showed a white fragile tumor with hair on the cauda equina and the tumor did not rupture (Fig. 2B and C). According to these findings, we thought it was very likely that this tumor was an epidermoid/dermoid cyst and presumed that maximal tumor resection without neurological deterioration would be possible. We did not enlarge the laminectomy because the tumor could be gradually removed piecemeal without bleeding, although it did adhere severely to the surrounding nerves. Rapid intraoperative pathological examination revealed that the tumor was mostly composed of keratinized tissue. We also found hairs in the tumor intraoperatively. These findings strongly suggested that the tumor was a dermoid cyst. Additionally, the tumor was resected carefully with complete aspiration of the tumor contents to prevent it from seeding into the intradural space with subsequent chemical meningitis. Transcranial motor evoked potential (MEP) monitoring and nerve integrity monitoring (NIM) were performed during

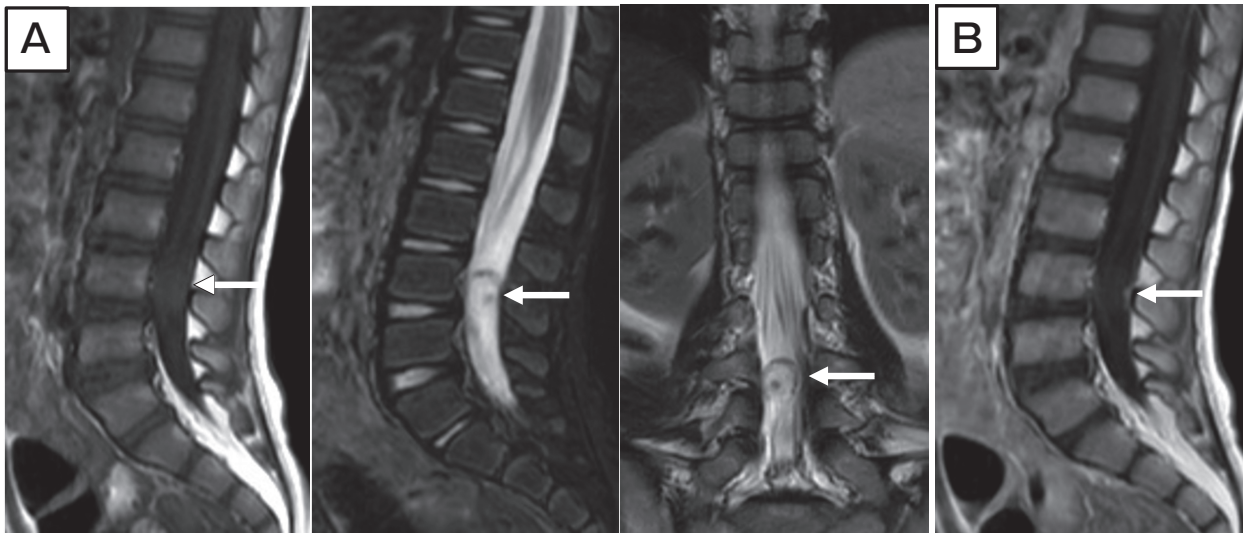

Fig. 1 Lumbar magnetic resonance imaging (MRI) findings on admission. (**A**) T1-weighted imaging (T1WI) on admission showed a tumor at the L3-4 level with isointensity to slightly high intensity (left panel, sagittal, arrow). T2-weighted imaging (T2WI) on admission showed a tumor in the right side of the spinal canal at the L3-4 level, with high intensity (middle panel, sagittal; right panel, coronal, arrow). (**B**) Contrast-enhanced T1-weighted imaging (CE-T1WI) on admission showed no enhancement of the tumor at the L3-4 level (sagittal, arrow).

surgery. MEP showed a temporary decrease in amplitude of the right quadriceps, but the amplitude had recovered to baseline by the time of closure. Gross total resection of the tumor was finally achieved, although a tiny part of the capsule was left unresected to preserve the surrounding nerves (Fig. 2D). The bone defect was filled with autologous bone and artificial bone composed of porous hydroxyapatite and collagen (HAp/Col) (Refit®; HOYA Technosurgical Co., Tokyo) (Fig. 2E). Histological examination revealed that the tumor was covered with stratified squamous epithelium and layered keratinizing material without characteristic findings such as hair follicles or sweat glands (Fig. 3A and B). Considering the intraoperative findings and the results of histological examination, the tumor was confirmed to be a dermoid cyst. Lumbar computed tomography (CT) and MRI after surgery showed the gross total resection of the tumor with a hemilaminectomy approximately 5 mm in width. The patient had an uneventful postoperative course without pain and was discharged from the hospital one week after surgery (Fig. 4A and B). Eight months after surgery, the patient's gait disturbance, low back pain, and sensory disturbance in the right lower extremity had completely disappeared. Lumbar CT showed evidence of early bone healing at eight months after surgery (Fig. 4C) and MRI found no recurrence of the dermoid cyst at 20 months after surgery (Fig. 4D). At the time of writing (29

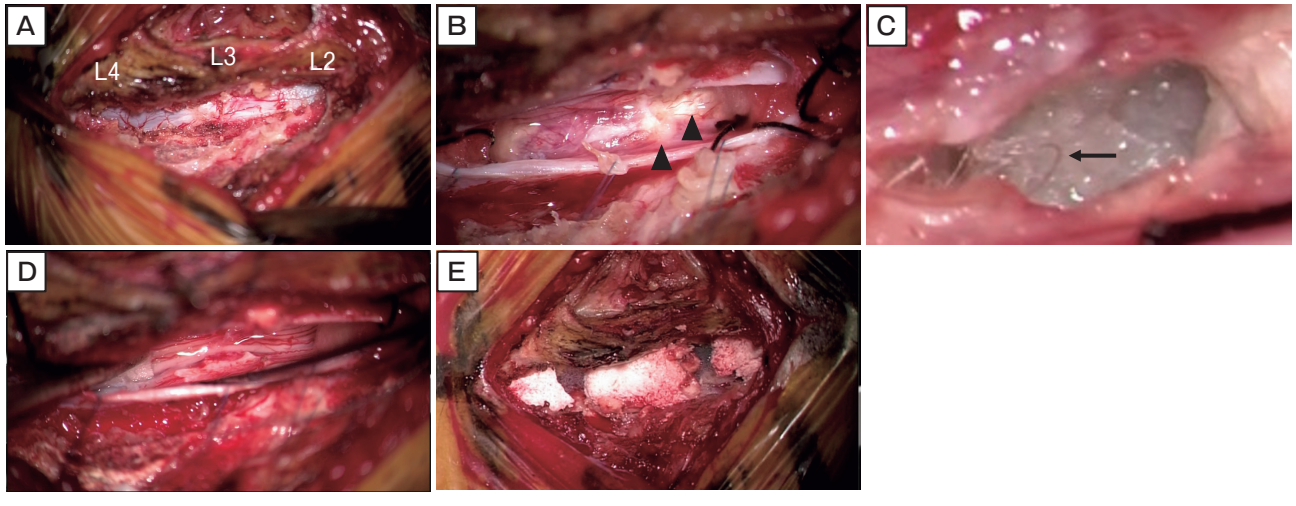

Fig. 2 Intraoperative findings. (**A**) We performed right unilateral hemilaminectomy at the L3 level and right unilateral dome hemilaminectomy at the L2/3 and L3/4 levels. (**B**, **C**) The tumor had fragile white cysts (arrowheads), and there were hairs in and around the cysts (arrow). (**D**) Gross total resection of the tumor was achieved. (**E**) The bone defect was filled with autogenous bone and artificial bone.

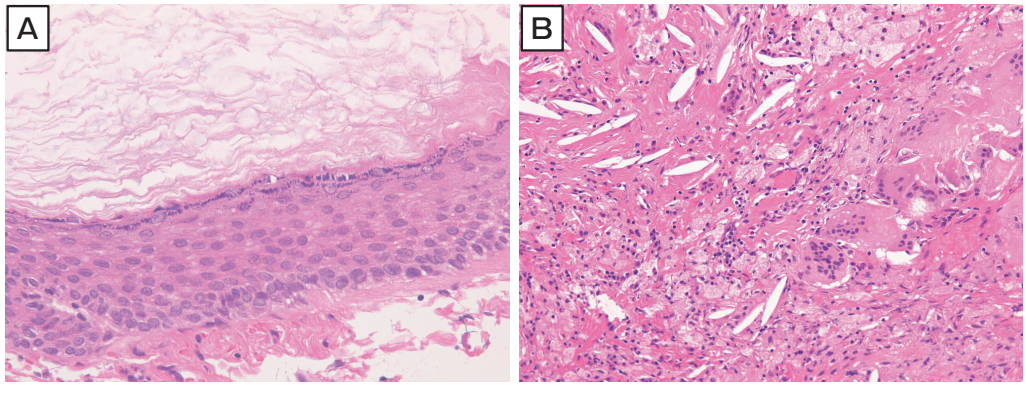

Fig. 3 Histological examination. (**A**, **B**) Hematoxylin and eosin staining showed that the tumor was covered with stratified squamous epithelium and layered keratinizing material without characteristic findings such as hair follicles or sweat glands.

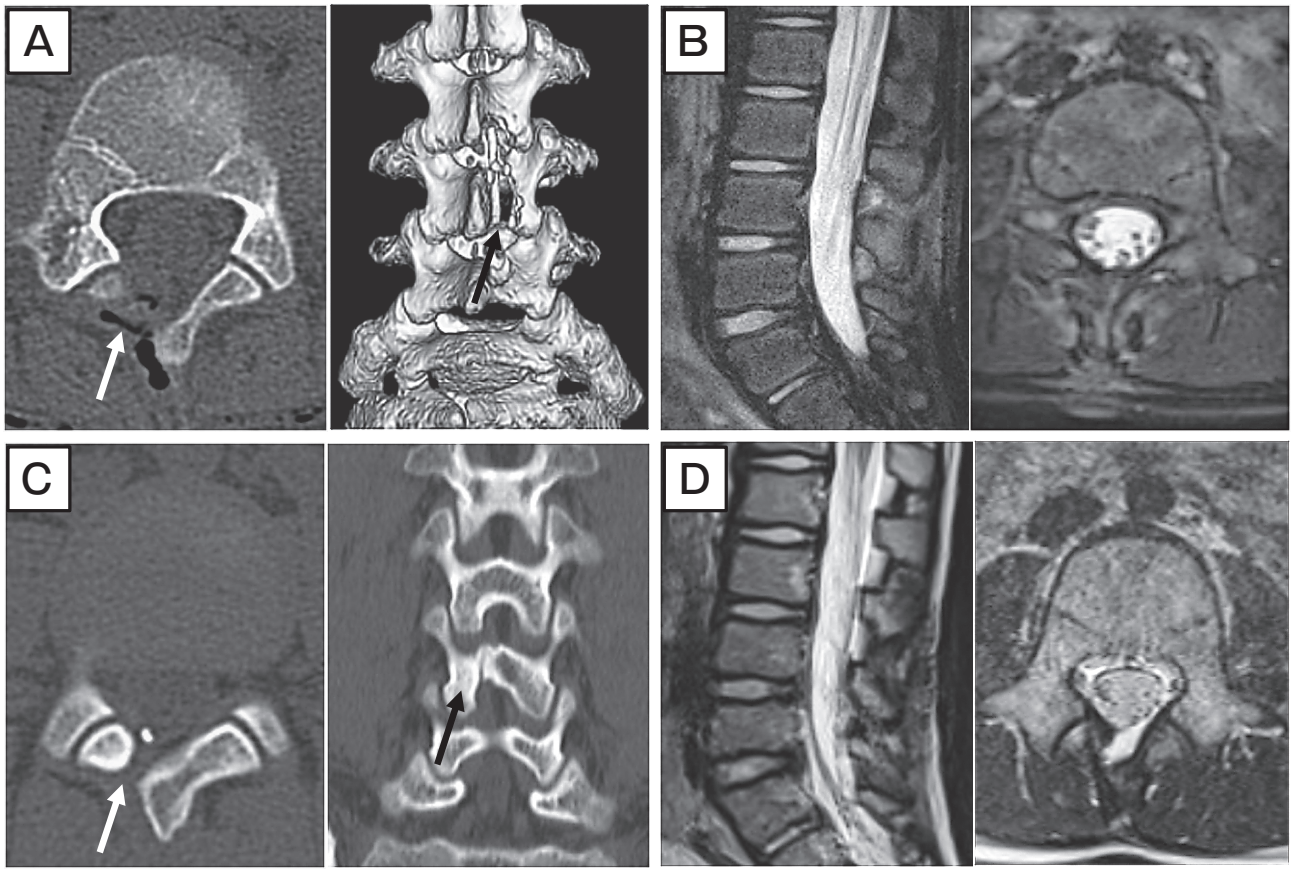

Fig. 4 Lumbar computed tomography (CT) and MRI findings after surgery. (**A**) Lumbar CT after surgery showed a minimal window opening of approximately 5 mm at the L3 level (arrow). (**B**) MRI at six days after surgery showed gross total resection of the tumor with reduced compression to the surrounding cauda equina. (**C**) Lumbar CT at eight months after surgery showed evidence of early bone healing (arrow) at the L3 level. (**D**) MRI at 20 months after surgery found no recurrence.

months after surgery), the patient has been followed up every year as an outpatient without any neurological symptoms.

Ethical approval.

All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee (IRB#1911-023), and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

Discussion

Spinal tumors account for approximately 5-10% of all central nervous system tumors [18], and intradural extramedullary tumors account for 40% of spinal tumors [18]. The distribution and incidence of cauda equina tumors are 4% and 0.3 per one million, respectively [19]. The most common histological types of intradural tumors in the lumbar spinal canal are meningiomas (29%), schwannomas (24%) and ependymomas (23%) [19]. Although we included these tumors in the differential diagnosis, the ultimate diagnosis of the present tumor was dermoid cyst, which was an unlikely candidate among the differential diagnoses.

Dermoid cysts are rare and benign congenital tumors that may locate at any level of the midline craniospinal axis [20]. It has been reported that the incidence of intramedullary dermoid cyst is higher than that of extramedullary dermoid cyst in children [21]. Dermoid cyst predominantly occurs in the lumbosacral segment, including the cauda equina and the conus medullaris (60%), followed by the upper thoracic (10%) and cervical segments (5%), with or without a concom-

itant anomaly such as spina bifida [22].

MRI is an effective imaging modality for the diagnosis of dermoid cysts. Dermoid cysts typically show high-intensity on T1WI and inconstant intensity on T2WI because of their heterogeneity, including the fat and fluid content of the cyst [6, 23]. Lipomas, teratomas, and intramedullary tumors such as ependymomas, astrocytomas, and hemangioblastomas are considered in the differential diagnosis of dermoid cyst [24]. Dermoid cysts are unlikely to be mistaken for intramedullary tumors because intramedullary tumors usually show hypointensity to isointensity on T1WI and are enhanced by contrast medium. In our case, however, the tumor showed isointensity to slightly high intensity on T1WI, and no enhancement on CE-T1WI. Thus, it was unusual to consider dermoid cyst in the differential diagnosis of this lumbar intradural tumor.

When a dermoid cyst ruptures, the patient may experience headache, nausea, vomiting, motor dysfunction, seizure, paraparesis and/or quadriparesis relating to chemical or aseptic meningitis [25]. In cases of rupture, surgical resection is the standard treatment [26]. Although the tumor had not ruptured in our case, the patient complained of severe low back pain and sensory disturbance in the right lower extremity, so we decided to perform surgery. This tumor is more likely to recur within months or years when there is residual tumorous material, including the stratified epithelial membrane [27]. This membrane adheres closely to the surrounding tissue and is very difficult to resect safely [6]. Guo *et al.* report that in three out of five cases who underwent surgery for spinal dermoid cyst, a small amount of cyst wall remained, and all three cases had recurrence within 18 to 113 months after surgery [27]. Therefore, we need to follow this patient for a long time.

The essential principles of spinal benign tumor resection are minimizing the intraoperative risk of cord/nerve injuries, removing the tumor completely, and avoiding spinal column deformity or instability after surgery [28]. Recently, it has been recognized that minimally invasive approaches are important for preserving structures that may be essential for spinal stability [29]. Several studies have addressed positive clinical outcomes regarding postoperative pain, complications, and deformity or instability in hemilaminectomy [7, 30-32]. In contrast, laminectomy is more likely to cause postoperative spinal column deformity or instability because the interspinous ligament is sectioned and the ligamentum flavum is bilaterally disrupted [33, 34]. Wei *et al.* report that pediatric patients (< 18 years of age) who underwent laminectomy for spinal tumor resection had a high incidence of progressive spinal column deformity (60%: 6 of 10 patients) [35], and preoperative spinal column deformity is also associated with risk factors [35]. We selected hemilaminectomy in our case to avoid interference with the growing spine of our pediatric patient. We needed to remove the full tumor because we had to consider dermoid cyst as a differential diagnosis in the intraoperative findings. Even though hemilaminectomy is difficult for the surgeon due to its narrow operative field, we were able to complete the gross total resection using meticulous surgical techniques. Because of this minimally invasive surgery, our patient was able to leave the hospital quite soon after surgery without any postoperative pain or complications.

The artificial bone used in this surgery was composed of porous HAp/Col. The sponge-like elasticity of HAp/Col is a significant characteristic of this graft material, which fits into the space of the bone matrix without gaps, allowing bone formation to extend continuously from the matrix bone [36, 37]. Kawasaki *et al.* report that in *in vitro* tests, porous HAp/Col was almost completely absorbed and replaced by bone tissue in skeletal sites within eight weeks, while muscular sites required significantly more time [37]. Additionally, Sotome *et al.* report that porous HAp/Col has a higher capacity for bone healing than standard bone substitutes such as β-tricalcium phosphate (β-TCP) [36]. Our case showed evidence of bone healing on lumbar CT at eight months after surgery. Murata *et al.* used CT analysis after spinal surgery to assess the effect of refilling bone defects with porous HAp/Col, revealing a high rate of success over a one-year period [38]. Therefore, the use of the porous HAp/Col in bone defects might be useful to promote early bone healing even in children.

It is important that we follow up our patient for the possibility of tumor recurrence and/or postoperative spinal column deformity or instability. At the time of writing, our patient has had an uneventful postoperative course without recurrence or spinal deformity or instability for 29 months after surgery.

Conclusion

We have presented a pediatric case of cauda equina dermoid cyst resected by unilateral hemilaminectomy. This approach might make it possible to avoid spinal instability or deformity in pediatric patients. Although dermoid cysts are rare, we should keep this possibility in mind as a differential diagnosis of lumbar intradural tumor.

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