Thoracic Epidural Teratoma: Case Report and Review of the Literature

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ABSTRACT

PURPOSE: Spinal teratomas comprise a rare subset of spinal cord tumors, and here, we describe an even rarer childhood thoracic extradural–intracanalicular teratoma. The clinical presentation, management, and pathophysiology of these tumors are reviewed to promote recognition and guide treatment of these lesions.

METHODS: We report the case of a 21-month-old boy who presented with marked spasticity, as well as failure to ambulate and meet motor milestones. Additionally, we provide a literature review of spinal teratomas, including their clinical presentation, work-up, pathophysiology, and underlying genetics.

Results: An MRI of the spine revealed a large dorsal epidural tumor extending from T3 to T10 with heterogeneous contrast enhancement and severe spinal cord compression. The tumor was resected revealing a cystic mass with tissue resembling hair, muscle, as well as cartilage; pathology confirmed the diagnosis of teratoma. Gross total resection was achieved, and the child eventually gained ambulatory function.

CONCLUSIONS: Given that spinal teratomas are rare entities that can present with significant neurologic compromise, they must remain on clinicians' differentials. Unfortunately, the exact origin of these tumors remains inconclusive and requires further investigation.

KEYWORDS: teratoma, spinal teratoma, thoracic epidural teratoma, extradural–intracanalicular teratoma

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Introduction

Teratomas (Greek *teratos*: monster) are rare germ cell neoplasms composed of mixed parenchymal cells from one or more of the three germ cell layers: ectoderm, endoderm, or mesoderm.[1] Named for their disturbed growth, they are histologically classified as immature or mature depending on the degree of differentiation of their cellular constituents.[2] Because they arise from pluripotent cells, teratomas can recapitulate any diversity of tissues including skin, muscle, bone, cartilage, intestinal mucosa, fat, teeth, or hair. In children, they are usually found in midline or paraxial locations of the entire neuroaxis, but most commonly occur in the sacrococcygeal region.[3] Sacrococcygeal teratomas, in particular, are thought to arise from

pathologic, aberrantly organized remnants of the caudal end of the primitive streak.[4]

Although they can affect any age, teratomas represent approximately 3% of all childhood tumors.[5] However, teratomas of the central nervous system (CNS) are especially rare and account for only 0.1% of all CNS tumors.[6] Spinal cord teratomas are even rarer and can be present in extradural, intradural, or intramedullary locations.[7] The association between intradural spinal cord teratomas and spinal cord dysraphism[8] suggests a similar principal embryological origin. More specifically, two prominent theories exist to explain the pathogenesis underlying spinal teratomas: the dysembryogenic theory and the misplaced germ cell theory. In the former, disordered differentiation of pluripotent cells

occurs in the primitive streak,[9] whereas in the latter, there is misplacement of multipotent neural tube primordial germ cells during migration from the yolk sac.[10] While the exact pathogenesis and embryonic derivation underlying the formation of spinal teratomas remains controversial, a similar mechanism is believed to be responsible for both intradural and extradural tumor presentations.[11] In this report, we present an unusual case of an epidural–intracanalicular teratoma in a 21-month-old male. Additionally, we discuss the differential, work-up and treatment for these spinal lesions, examine other cases from the literature, and finally reflect on the prevailing understanding of the pathophysiology and genetics of spinal teratomas.

Case Report

History and examination. The patient was an otherwise healthy 21-month-old male who was the product of an induced vaginal delivery at 34 weeks gestation secondary to preeclampsia. His parents observed a 10-month history of progressive lower extremity weakness. They ultimately brought the patient to the emergency room when he was no longer able to ambulate. He had no evidence of bowel or bladder dysfunction, but his neurological examination was notable for decreased strength throughout his lower extremities bilaterally (4/5), hyperreflexia at the patellar and Achilles tendons with clonus, Babinski signs bilaterally, and significant spasticity. Magnetic resonance (MR) imaging delineated a 7.5 mm × 14 mm × 14 mm elongated cystic mass, with mixed T2 signal intensity and faint gadolinium enhancement, in the dorsal epidural space extending from the T3–T10 vertebral levels (Fig. 1A). There was severe spinal cord compression, with the cystic components of the mass extending into the T5–T6 and T6–T7 intervertebral foramina (Fig. 1A, C, D). Post-operatively, the patient required physical therapy and at six-month follow-up ambulation had been restored.

Operation, histology, and outcome. Based on the clinical presentation and associated radiographic findings, the patient was taken to the operating room for resection of the epidural mass. Spinal cord function was monitored via intraoperative recording of motor- and sensory-evoked potentials. Bilateral laminectomies were performed from T3 to T10, and the lesion was noted to be a yellow, cystic epidural mass extending from T3 to T9. A plane was identified under microscopic visualization, and the tumor was elevated with forceps and electrocautery, with no intradural component observed. On gross examination, the mass was heterogeneous in appearance, with tissue resembling muscle, cartilage, and hair, consistent with a teratoma (Fig. 2). A gross total resection was achieved and on histologic examination, the mass lesion was consistent with a mature teratoma, containing numerous well-differentiated cell types including stratified squamous epithelium, simple columnar epithelium, smooth muscle, mucous glands, as well as angiomatous differentiation (Fig. 3). The patient had motor improvements immediately
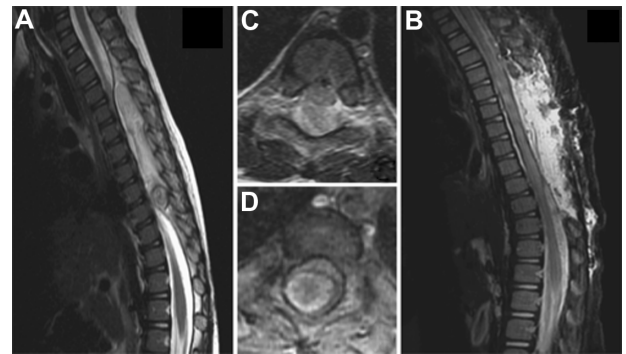

Figure 1. Pre-operative thoracolumbar spinal MR image detailing a 7.5 mm × 14 mm × 14 mm elongated cystic mass, with mixed T2 signal intensity in the dorsal epidural space extending from the T3–T10 vertebral levels (**A**). Post-operative enhancement of the dorsal epidural space in the area of the excised lesion (**B**). Pre-operatively, there is severe pre-operative spinal cord compression, with the cystic components extending into the T5–T6 and T6–T7 intervertebral foramina (**C**–**D**).

post-operatively. With physical therapy, he regained the ability to ambulate during the ensuing months.

Literature Search

Given the rarity of spinal teratomas, we performed a review of the English literature to document similar cases of extradural spinal teratomas. Only evaluating the English literature yields an inherent selection bias, but nevertheless still demonstrates the paucity of these tumors. We employed our search through PubMed using a combination of the keywords "spinal teratoma," "spinal," "epidural," "extradural," "intradural," "intraspinal," "intramedullary," "extramedullary," "teratoma," or "teratoid" as search terms. Intramedullary tumors and metastatic tumors were excluded. This search yielded five cases of epidural–intracanalicular spinal teratomas, which are discussed below. These cases illustrate the rarity of these
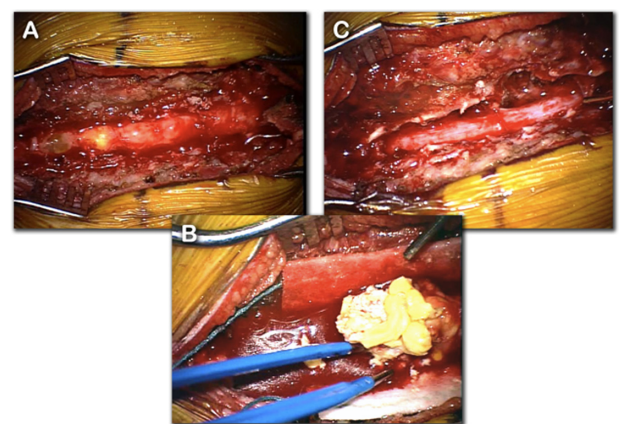

Figure 2. Intraoperative photographs detailing bilateral laminectomies from T3 to T10 (**A**), with the lesion noted to be a yellow, cystic epidural mass extending from T3 to T9 with neural foraminal extension (**B**). No intradural component was observed, and gross total resection was achieved (**C**).
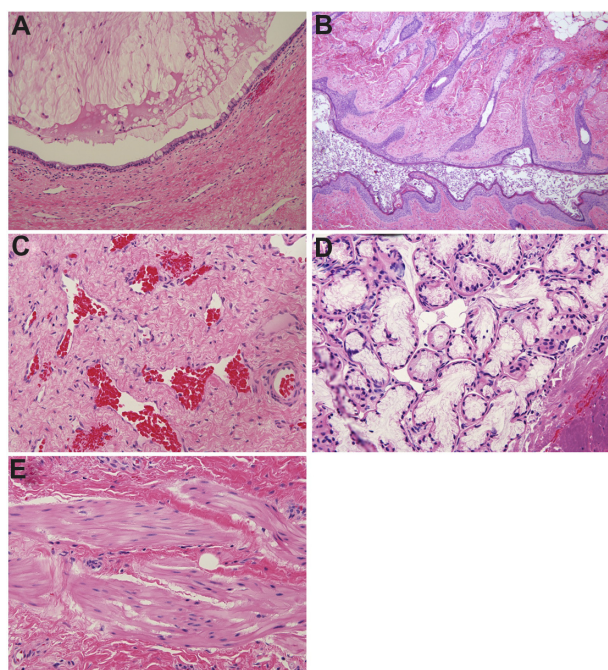
Figure 3. Histologic examination was consistent with a teratoma (hematoxylin and eosin). The teratoma contained a range of tissue types including (**A**) simple columnar epithelium with intermixed mucin secreting goblet cells resembling intestinal epithelium, original magnification ×40; (**B**) canalicular appearance of the dermoid cyst composed of multilayered stratified squamous epithelium with keratin debris and adnexal structures, original magnification ×20; (**C**) focus with angiomatous differentiation, original magnification ×40; (**D**) well-differentiated mucous glands, original magnification ×40; and (**E**) focus with smooth muscle differentiation, original magnification ×40.

spinal tumors, in particular of epidural spinal teratomas with foraminal extension.

Acar et al.[12] describe a case, very similar to our own, of a seven-month-old girl presenting with flaccid paraparesis and decreased sensitivity below the T4 level. MRI revealed a thoracic intraspinal extradural mass extending from the T6–T8 level producing severe cord compression. The lesion extended through the left T6–T7 intervertebral foramina and was found to be a benign cystic teratoma. The patient underwent laminectomies and post-operatively, the patient demonstrated gradual motor and sensory improvements.

Choi et al.[13] report the case of a full-term eight-month-old infant, with a normal birth history, presenting with severe flaccid paraparesis of both lower extremities of two months duration. MRI of the spine revealed a cystic, extradural lesion compressing the spinal cord from T2 to T4. Right-sided multilevel hemi-laminectomies from T2 to T4 were performed with attempts to preserve the facet joints to maintain spinal stability. The tumor originating from the spinal nerve root sheath was excised in its entirety, with histology revealing a mature teratoma. Post-operatively, the patient exhibited immediate improvement in strength, which continued to

gradually improve until the child could stand. Voiding and defecation difficulties were practically resolved.

Kaneko et al.[14] report the case of a full-term, 33-day-old newborn presenting with dyspnea and cyanosis following breast-feeding. A conventional radiograph of the chest revealed large bilateral, posterior mediastinal masses suggestive of lipomas with physical examination consistent with flaccid paraplegia below the T3 level. A computed tomographic (CT) scan detailed an extradural mass composed of adipose, cartilage, bone, and cysts compressing the spinal cord anteriorly. MRI delineated that the mass originated in the lower thoracic spine extending from C5 to L5, with expansion through multiple intraspinal foramina. Owing to concerns about delayed degenerative sequelae following the extensive laminectomies that would be required for such an extensive lesion, the tumor was resected through two separate surgeries (T3–T9 and T11–L3) with a two-month interval between the procedures. Three weeks later, the right mediastinal tumor was excised through a right thoracotomy. Of note, even though the tumor was benign histologically, T8–T12 laminotomies and a left thoracotomy were required when the tumor recurred at an unspecified time interval. The surgeons were able to achieve a subtotal resection, with the residual tumor maintaining a stable size via CT imaging at 24 months. The patient's bilateral lower extremity paralysis barely improved secondary to the longstanding severe cord compression before resection, leaving the patient with significant residual deficits. The patient eventually developed paralytic scoliosis and the inability to urinate voluntarily. This lesion is distinguished from our case and that reported by Acar et al by its extensive infiltration bilaterally through the intervertebral foramina to form a large mediastinal mass.

Murovic et al.[15] report a 37-week gestational newborn boy who was found to have a firm immobile abdominal mass, with neurological examination revealing a flaccid paralysis of the left lower extremity, a right foot drop, decreased lower extremity sensation, and loss of sensation in the perianal region. A CT scan demonstrated a 6 cm × 10 cm × 10 cm retroperitoneal mass originating at the foramen of the right T11 nerve root. The patient underwent a staged resection – first, the retroperitoneal mass and then the extradural component – with histology consistent with a benign teratoma. Approximately six months later, the patient had recurrence of the retroperitoneal mass that was excised successfully, again consistent with a benign teratoma. One month later, there was recurrence again with a new extradural L1–3 mass found and resected; there was no recurrence following this.

Park et al.[11] describe a 17-month-old boy with delayed standing and walking. Spinal MRI detailed an extradural multicystic mass compressing the spinal cord at T8–T10, with extrusion through the intervertebral foramina. In the operating room, there was no dural invasion, with the tumor well encapsulated and filled with heterogeneous contents. The tumor was suspected to originate from the left T9 root, and

the patient underwent a gross total resection. Histologically, the tumor contained all three germinal layers, with no immature components found, consistent with a mature teratoma. The patient recovered some lower extremity motor function, and there was no evidence of recurrence at two years post-resection.

Discussion

Presentation and differential diagnosis of spinal cord tumors. For children failing to achieve motor milestones, compressive spinal cord tumors are an important treatable cause. In children, spinal cord tumors represent 5–10% of CNS tumors,[16] with an overall incidence of 1–2.6 per million.[17] Presenting symptoms relate to the pathological origin of the lesion, its location, the degree of compression or infiltration, as well as the associated deformity of the surrounding area. The differential for primary spinal cord lesions is best categorized by location, which broadly includes epidural tumors, intradural extramedullary tumors, and intradural intramedullary lesions. This categorization is intricately linked to symptom presentation; for example, epidural tumors most often present with back pain, which is otherwise rare in children without recent trauma,[18] but can be also present with radiculopathy, myelopathy, cauda equina syndrome, or other neurologic ailments. On the other hand, intradural extramedullary spinal cord tumors are usually of insidious onset, with the most common complaint being localized or radicular pain, in addition to gait difficulty, weakness, paresthesias, and autonomic dysfunction.[19] Finally, intramedullary tumors have a non-specific presentation and can include radiculopathy, paresthesias, dysesthesias, spasticity, profound weakness, autonomic dysfunction, and failure to achieve developmental milestones.[19]

More specifically, extradural tumors comprise 30% of all spinal cord tumors in children and include benign bony tumors (ie, osteoid osteoma, osteoblastoma, osteochondroma, giant cell tumor, aneurysmal bone cyst, and hemangioma), Langerhans cell histiocytosis, fibrous dysplasia, as well as malignant tumors such as chordomas, teratomas, metastatic lesions, and sarcomas.[7] Intradural extramedullary tumors, which again include teratomas, as well as meningiomas, paragangliomas, neurofibromas, and schwannomas, compose 25% of pediatric intraspinal tumors.[16] Seeding of primary CNS tumors can also lead to extramedullary metastases.[18] Intramedullary tumors have been reported as 25–35% of pediatric intraspinal tumors and include low- or high-grade astrocytomas, ependymomas, and gangliomas.[17] These lesions can sometimes be distinguished radiographically with CT or MRI, but histology is usually required for definitive diagnosis.

In our patient, symptoms were secondary to compression of the adjacent spinal cord with resultant myelopathy, and the differential broadly included extradural spinal lesions that respected the dura mater without associated bony malformations. MRI delineated an elongated cystic mass with mixed T2 signal intensity and faint gadolinium enhancement. The

diagnosis of teratoma was confirmed on gross examination in the OR and confirmed on histopathology.

Diagnosis of spinal teratomas. Spinal teratomas have a reported incidence of 0.15–0.18% in large spinal tumor series overall, and 5–10% in infants and children.[11] Teratomas of the sacrococcygeal region are the most common tumor in live births and have an incidence of 1 in 40,000 live births.[2] It is therefore important that clinicians consider spinal teratomas in the differential diagnosis when evaluating patients with spasticity and neurologic deficits localizing to the spinal cord. Plain radiography may show associated vertebral anomalies (though these were not present in our patient) or erosive changes in the vertebral bodies even when these anomalies are not present.[20] Neuroimaging with either CT or MRI can be used to diagnose the lesion, demonstrating a heterogeneous mass with components suggestive of various tissue types. While CT identifies calcifications, as well as fat and soft tissue densities, the ability of MRI to distinguish various soft tissue components and cystic features makes it the gold standard for diagnosing spinal teratomas.[21]

Treatment of spinal teratomas. The treatment of choice remains surgical excision, which can also serve to confirm the diagnosis, with histology allowing for further subtyping. Pure mature teratomas can be treated with total surgical resection, whereas teratomas with malignant histological features or germ cell elements may also require adjuvant chemotherapy and radiotherapy to target malignant cells.[22] Total resection is recommended, but may be difficult if there are significant adhesions surrounding neural structures. Owing to the slow-growing nature of mature teratomas, the difference in recurrence following total compared to subtotal resection is considered insignificant.[20] Even with total resection, there is a risk of leaving microscopic traces of tumor behind, and perhaps the adherent nature of the tumor increases this risk. However, in some cases of mixed teratomas, subtotal surgical resection followed by chemotherapy and radiation allows further proliferation of the remaining benign portion. This is known as "growing teratoma syndrome," for which a second surgical resection may be needed for further decompression.[22] When treating immature teratomas, the additive benefit of radiation and chemotherapy is not well defined relative to its known toxicities and should therefore be used with caution, especially in the pediatric population. Some authors advocate an aggressive surgical approach with sparing use of adjuvant therapy until evidence of recurrence.[23] Of note, using radiation therapy as the sole treatment for spinal teratomas is thought to have unsatisfactory outcomes.[22] In our patient, a plane was identified between the teratoma and the dura, and total resection was achieved despite the intraforaminal extension of the tumor.

Genetics of teratomas. Spinal teratomas may have a common genetic origin that manifests itself at different times during embryologic development (ie, before or after neural tube closure). A recent report begins to unravel the genetics

underlying sacrococcygeal teratomas, which may arise in association with local developmental errors affecting the caudal embryonic segments, which may also be related to spinal teratomas.[24] These authors describe two siblings – one presenting at birth with a sacrococcygeal teratoma and the other sibling with a lumbar lipomyelomeningocele – and propose an inherited regional propensity of developmental errors affecting the caudal embryonic segments in both siblings, predisposing each sibling to the development of their respective pathologies. Although no definite genetic loci are known, mounting evidence describes a possible association between chromosomal aberrations and teratomas. For example, sacrococcygeal teratomas have been associated with distal 10q trisomy/partial monosomy 17p[25] (unbalanced karyotype from a maternal balanced autosomal translocation present between chromosomes 10 and 17). Other reported aberrations include a mosaic trisomy of most of chromosome 1q[26] (unbalanced autosomal translocation between chromosomes 1 and 15) or a mosaic terminal 7q monosomy/distal 2p trisomy[27] (balanced de novo translocation between chromosomes 2 and 7). Similarly, the autosomal dominant Currarino anomaly – comprised of partial sacral agenesis, a presacral mass (teratomas in up to 40% of cases), and anorectal defects – has been described to result from mutations in HLXB9, a transcription factor mapping to chromosome 7q.[28] The gene for autosomal dominant sacral agenesis maps to this very same region – chromosome 7q.[29] Interestingly, a report of a teratoma from a Currarino patient that contained mature neurons suggests that the caudal notochord is important for organized secondary neurulation, shedding greater light on the underlying pathogenesis of spinal teratomas.[30]

Furthermore, chromosomal abnormalities of head and neck teratomas from seven fetuses were almost always associated with a tumor-specific amplification of genetic material: a partial trisomy or tetrasomy of chromosome 1q – that is, the presence of an extra chromosome 1, duplication of the long arm of chromosome 1, or an unbalanced autosomal translocation.[25] Though rare, malignant sacrococcygeal teratomas have also demonstrated breaks in chromosome 6 with near-haploidy.[31] Congenital sacrococcygeal teratomas have been found to express the *ras*, *fos*, and *jun* oncogenes, as well as the *NM*23 and *p*53 tumor suppressor genes, implicating the molecular pathways involved in teratoma formation and development.[32] However, future research is needed to further elucidate these pathways and the precise molecular, genetic, and underlying epigenetic mechanisms.

Conclusion

Failure to achieve age-appropriate gross motor developmental milestones may be observed in children less than two years of age. Spinal teratomas are a rare clinical entity that must nonetheless remain on the differential in children with signs or symptoms localizing to the spinal cord. On neuroimaging, the presence of heterogeneous masses with components

suggestive of various tissue types can facilitate the diagnosis. Finally, the diagnosis can be confirmed in the operating room, with histology allowing further subtyping and clarification. In addition to surgical excision, we recommend post-operative physical therapy to allow as much functional restoration as possible. In the end, the exact origin of these tumors remains inconclusive; however, there is strong evidence that their origin is from pluripotent cells of Hensen's node and the caudal cell mass, with the most common possible genetic aberrations from duplication of chromosome 1 or deletions in chromosome 7. Further research is needed, but stem cell transplantation for spinal cord injury may inadvertently lead to a better understanding of how spinal teratomas form given their origin from pluripotent cells.

Author Contributions

JLQ and RAG wrote the first draft of the manuscript. JLQ, RAG, AJH, and CCD contributed to the writing of the manuscript. JLQ, RAG, AJH, and CCD agreed with manuscript results and conclusions. JLQ, RAG, and CCD jointly developed the structure and arguments for the paper. JLQ, RAG, AJH, and CCD made critical revisions and approved the final version. All authors reviewed and approved the final manuscript.

REFERENCES

1. Kumar V, Abbas AK, Fausto N. *Robbins and Cotran Pathologic Basis of Disease*. 7th ed. Philadelphia, PA: Elsevier Saunders; 2005.
2. Harms D, Zahn S, Gobel U, Schneider DT. Pathology and molecular biology of teratomas in childhood and adolescence. *Klin Padiatr*. 2006;218(6):296–302.
3. Azizkhan RG, Caty MG. Teratomas in childhood. *Curr Opin Pediatr*. 1996;8(3):287–92.
4. Izant RJ Jr, Filston HC. Sacrococcygeal teratomas. Analysis of forty-three cases. *Am J Surg*. 1975;130(5):617–21.
5. Rattan KN, Malik V, Khurana P, Dhawan S, Kaushal V, Maggu S. Teratomas in infancy and childhood. *Indian J Pediatr*. 2001;68(2):117–20.
6. Isik N, Balak N, Silav G, Elmaci I. Pediatric intramedullary teratomas. *Neuropediatrics*. 2008;39(4):196–9.
7. Park T, Ghatan S, Scott R. Youmans neurological surgery. In: Winn H ed. *Youmans Neurological Surgery*. Vol 2. 6th ed. Elsevier; 2011.
8. Tsitsopoulos P, Rizos C, Isaakidis D, Liapi G, Zymaris S. Coexistence of spinal intramedullary teratoma and diastematomyelia in an adult. *Spinal Cord*. 2006;44(10):632–5.
9. Koen JL, McLendon RE, George TM. Intradural spinal teratoma: evidence for a dysembryogenic origin. Report of four cases. *J Neurosurg*. 1998;89(5):844–51.
10. Rewcastle NB, Francoeur J. Teratomatous cysts of the spinal canal; with "sex chromatin" studies. *Arch Neurol*. 1964;11:91–9.
11. Park SC, Kim KJ, Wang KC, Choe G, Kim HJ. Spinal epidural teratoma: review of spinal teratoma with consideration on the pathogenesis: case report. *Neurosurgery*. 2010;67(6):E1818–25.
12. Acar O, Guney O, Eser O. Epidural thoracal teratoma in a 7-month-old girl. *Childs Nerv Syst*. 2003;19(2):126–8.
13. Choi SJ, Choi HJ, Hong JT, et al. Intraspinal extradural teratoma mimicking neural sheath tumor in infant. *Childs Nerv Syst*. 2004;20(2):123–6.
14. Kaneko M, Ohkawa H, Iwakawa M, Ikebukuro K. Extensive epidural teratoma in early infancy treated by multi-stage surgery. *Pediatr Surg Int*. 1999;15(3–4):280–3.
15. Murovic JA, De Armond S, Nagashima T, Edwards MS, Hoshino T. Cell kinetics analysis in a case of teratoma of the thoracic spine. *J Neurosurg*. 1986;65(3):331–4.
16. Baysefer A, Akay KM, Izci Y, Kayali H, Timurkaynak E. The clinical and surgical aspects of spinal tumors in children. *Pediatr Neurol*. 2004;31(4):261–6.
17. Choi GH, Oh JK, Kim TY, et al. The clinical features and surgical outcomes of pediatric patients with primary spinal cord tumor. *Childs Nerv Syst*. 2012;28(6):897–904.

18. Huisman TA. Pediatric tumors of the spine. *Cancer Imaging*. 2009;9 Spec No A:S45–8.

19. Abul-Kasim K, Thurnher MM, McKeever P, Sundgren PC. Intradural spinal tumors: current classification and MRI features. *Neuroradiology*. 2008;50(4):301–14.

20. Makary R, Wolfson D, Dasilva V, Mohammadi A, Shuja S. Intramedullary mature teratoma of the cervical spinal cord at C1–2 associated with occult spinal dysraphism in an adult. *J Neurosurg Spine*. 2007;6(6):579–84.

21. Basmaci M, Hasturk A, Pak I. Cystic mature teratoma of the thoracic region in a child: an unusual case. *J Neurosci Rural Pract*. 2011;2:186–9.

22. Fujimaki T. Central nervous system germ cell tumors: classification, clinical features, and treatment with a historical overview. *J Child Neurol*. 2009;24(11):1439–45.

23. Phi JH, Kim SK, Park SH, Hong SH, Wang KC, Cho BK. Immature teratomas of the central nervous system: is adjuvant therapy mandatory? *J Neurosurg*. 2005;103(6 suppl):524–30.

24. Oliveria SF, Thompson EM, Selden NR. Lumbar lipomyelomeningocele and sacrococcygeal teratoma in siblings: support for an alternative theory of spinal teratoma formation. *J Neurosurg Pediatr*. 2010;5(6):626–9.

25. Batukan C, Ozgun MT, Basbug M, Caglayan O, Dundar M, Murat N. Sacrococcygeal teratoma in a fetus with prenatally diagnosed partial trisomy 10q (10q24.3-->qter) and partial monosomy 17p (p13.3-->pter). *Prenat Diagn*. 2007;27(4):365–8.

26. Wax JR, Benn P, Steinfeld JD, Ingardia CJ. Prenatally diagnosed sacrococcygeal teratoma: a unique expression of trisomy 1q. *Cancer Genet Cytogenet*. 2000;117(1):84–6.

27. Le Caignec C, Winer N, Boceno M, et al. Prenatal diagnosis of sacrococcygeal teratoma with constitutional partial monosomy 7q/trisomy 2p. *Prenat Diagn*. 2003;23(12):981–4.

28. Crétolle C, Pelet A, Sanlaville D, et al. Spectrum of HLXB9 gene mutations in Currarino syndrome and genotype-phenotype correlation. *Hum Mutat*. 2008;29(7):903–10.

29. Lynch SA, Bond PM, Copp AJ, et al. A gene for autosomal dominant sacral agenesis maps to the holoprosencephaly region at 7q36. *Nat Genet*. 1995;11(1):93–5.

30. Gegg CA, Vollmer DG, Tullous MW, Kagan-Hallet KS. An unusual case of the complete Currarino triad: case report, discussion of the literature and the embryogenic implications. *Neurosurgery*. 1999;44(3):658–62.

31. Noguera R, Navarro S, Carda C, Peydro-Olaya A, Llombart-Bosch A. Near-haploidy in a malignant sacrococcygeal teratoma. *Cancer Genet Cytogenet*. 1999;108(1):70–4.

32. Kruslin B, Hrascan R, Manojlovic S, Pavelic K. Oncoproteins and tumor suppressor proteins in congenital sacrococcygeal teratomas. *Pediatr Pathol Lab Med*. 1997;17(1):43–52.