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CASE REPORT

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Extensive epidural teratoma in early infancy treated by multi-stage surgery

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Abstract We report a rare case of extensive extradural teratoma successfully treated by multi-stage laminotomy and thoracotomy. A 34-day-old, dyspneic infant had a large posterior mediastinal mass identified on a chest *X-ray* radiograph. Imaging studies disclosed that the mass originated from the extradural space at the level of the lower thoracic spine, extending cephalad to C4 and caudad to L4 and severely compressing the spinal cord anteriorly, causing paraplegia. The tumor expanded bilaterally through the intraspinal foramina, coalescing to form a huge mediastinal mass. The upper half of the teratoma was removed utilizing a laminotomy from T3 through T9; 2 months later the lower half was excised via a laminotomy from T11 to L3. An additional procedure was required to resect recurrent tumor through a laminotomy from T8 to T12. The reconstructed vertebral arches were well-preserved in shape, with an almost normal spinal canal.

Key words Epidural teratoma · Infant · Laminotomy · Mediastinal teratoma

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Introduction

Extensive epidural teratomas are extremely rare at any age. We report an infant with an extensive epidural teratoma that extended bilaterally through the vertebral foramina to form bilateral mediastinal tumors. Imaging studies revealed that the mass originated from the intraspinal extradural space at the level of the lower thoracic spine, extending upward to C4 and downward to L4 and severely compressing the spinal cord anteriorly, causing paraplegia. Resection of the epidural tumor by laminectomy was thought to be undesirable because an extensive laminectomy, particularly in infancy, is known to cause late sequelae such as spinal instability, severe scoliosis, and spinal-canal stenosis, and should be avoided if possible. Staged laminotomies gave satisfactory operative fields for resection, and the reconstructed vertebral arches were well-preserved with an almost normal spinal canal.

Case report

A "normal" boy was born at 38 weeks of gestation with a birth weight of 3,032 g. On day 33, dyspnea associated with generalized cyanosis appeared after breast-feeding. His chest roentgenograph revealed large bilateral, radiolucent posterior mediastinal masses strongly suggestive of lipomas. Cyanosis associated with intercostal and hypochondral retraction was obvious on crying. Physical examination revealed complete flaccid paraplegia below the T3 level. The history disclosed that intrauterine fetal movements had been minimal and his grandmother had noticed that his lower limbs were much less active compared to the upper extremities. A computed tomographic (CT) scan showed an intraspinal epidural mass composed mainly of adipose tissue associated in part with cartilage, bone, and cysts that severely compressed the spinal cord anteriorly. Magnetic resonance imaging (MRI) showed that the mass originated in the lower thoracic spine and extended over 20 vertebrae, cephalad to C5 and caudad to L5. It also extended through multiple intraspinal foramina bilaterally to the posterior mediastinal and retroperitoneal spaces. The mediastinal masses were composed of tumor extending through each thoracic foramen. The anteriorly-compressed spinal cord was barely visible, but could not be identified in the lower thoracic area (Fig. 1).

To minimize late sequelae, staged laminotomies instead of laminectomies were performed. At 42 days of age the upper half of the teratoma was removed via a laminotomy from T3 through T9. The well-encapsulated, benign tumor could be dissected from the ligamentum flavum and dura without difficulty (Fig. 2). In the lower thoracic area, where the tumor was thought to originate, it was composed of cartilage, bone, and cysts attached tightly to the surrounding tissues, making dissection extremely difficult. The upper half of the tumor could successfuly be resected en bloc, but there were some minute residuals in the lower thoracic spinal canal and the intervertebrale foramina. After resection of the tumor, the opened vertebral arches were replaced and sutured. The postoperative course was complicated by respiratory embarrassment, which was later diagnosed to be the lazy cilia syndrome. Two months later the lower half of the tumor, extending to the retroperitoneum through the intervertebral foramina, was excised en bloc by laminotomy from T11 to L3, followed by excision of the right

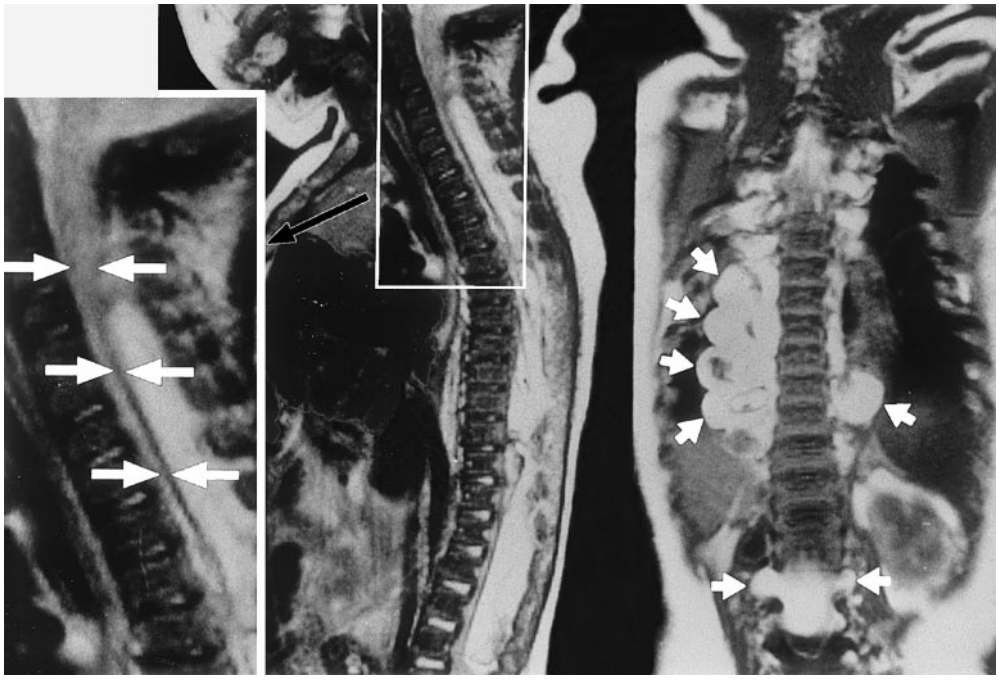


Fig. 1 Preoperative MRI study. Inset, an enlargement from left picture, shows cervical spinal cord (*arrows*) markedly shifted anteriorly by intraspinal tumor (high signal intensity due to high lipid content) extending from below. Spinal cord could not be seen in lower thoracic spine, which is shifted anteriorly by intraspinal tumor. Mass extends through multiple bilateral intraspinal foramina to form mediastinal and retroperitoneal masses with calcification (right picture)

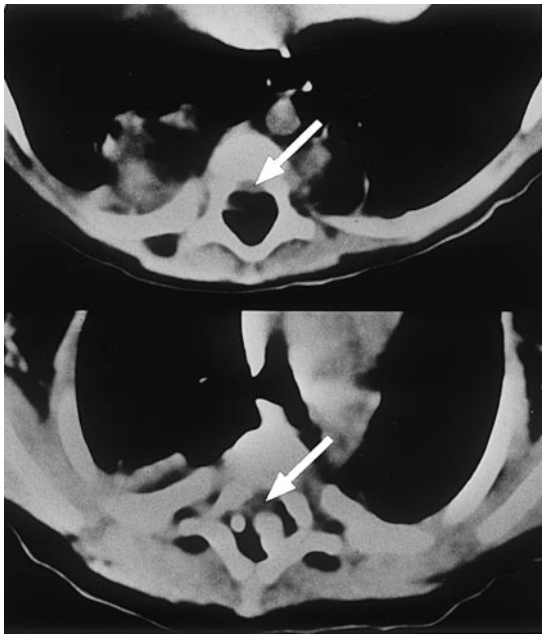


Fig. 2 CT of thoracic spine. Spinal cord (*arrows*) can hardly be seen at anterior side of spinal canal, which is occupied by tumor consisting mainly of adipose tissue (top) and osseous tissue (bottom)

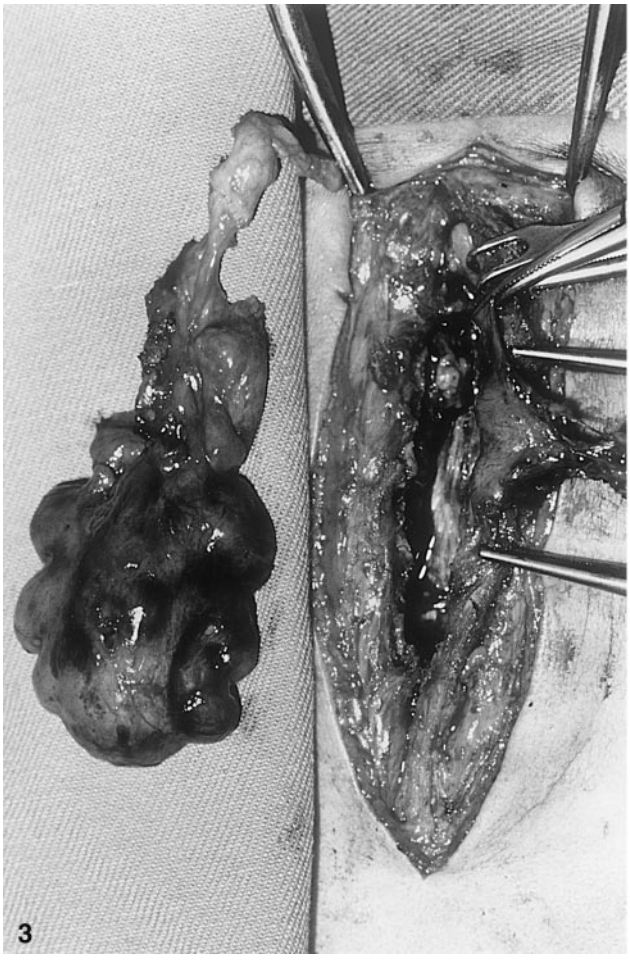
mediastinal tumor through a right thoracotomy 3 weeks later.

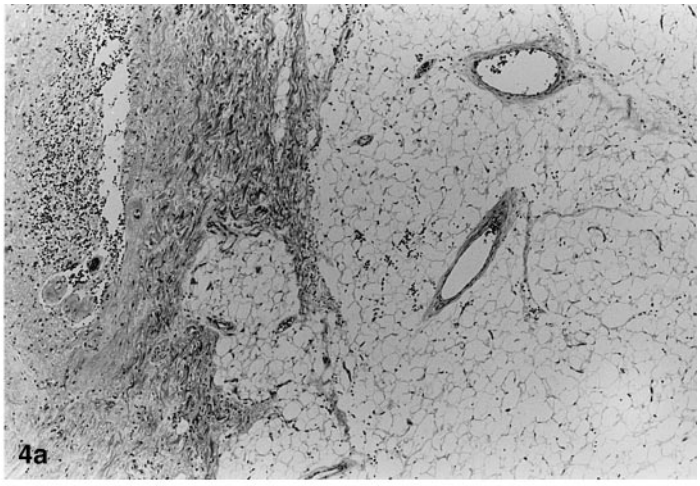
The tumor, though benign histologically, recurred rather rapidly in the thoracic epidural space and the right mediastinum, necessitating an additional intraspinal tumor resection through a laminotomy from T8 to T12. The redo laminotomy with left mediastinal resection through a left thoracotomy could be performed without much difficulty in spite of moderate adhesions and slight deformity of the spinal arches. The residual tumor which contained osseous tissue, was resected with subsequent spinal-arch reconstitution.

The postoperative spinal-canal deformity was minimal on CT; 24 months after the last laminotomy, the residual tumor in the right mediastinum, which is stable in size, can still be detected. Except for the lower thoracic spine, which has a postoperative deformity, the reconstructed vertebral arches were well-preserved with an almost normal spinal canal. However, bilateral lower-extremity paralysis below the level of the lower thoracic spine scarcely improved due to the long-standing, severe medullary compression. The paralytic scoliosis, which was not apparent on admission, has gradually progressed. He also has a progressing pigeon-chest deformity due to intercostal nerve paralysis with intact diaphragmatic movement. He cannot void urine voluntarily, but has had no episode of urinary tract infection. The latest plain roentgenogram revealed a 39° thoracic scoliosis with a 26° secondary lumbar scoliosis.

Discussion

Although intraspinal lipomas and teratomas excluding the sacrococcygeal region are occasionally reported in all age groups [6, 17], epidural teratoma is quite rate [5]. Ingram and Matson [7] noted 12 cases of intraspinal teratoma in their series of 63 intraspinal tumors in children. Matson [12] later described 13 teratomas in 134 patients with intraspinal tumors in his textbook. He wrote that complete resection is usually possible when they are limited to the extradural and subdural space, but radical excision is often impossible when the lesions are attached to or located within the cord. Coulon [3] stated in a review that teratomas constitute 3% to 9% of intraspinal tumors in children and are most frequently found in the thoracolumbar and lumbar regions. He also reported that they are intradural and usually extramedullary, but can be intramedullary. The present case, however, was apparently epidural, and no intradural extension was observed.





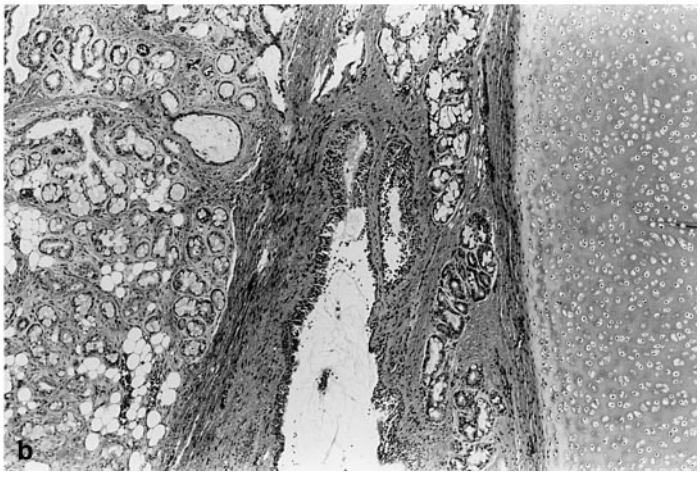


Fig. 3 Excised intraspinal teratoma from lower thoracic and lumbar spine. Well-encapsulated lower half of tumor, which extended to retroperitoneal space through intervertebral foramina, could be excised en bloc (dura is seen glistening in spinal canal). Vertebral arches are retracted by three forceps

Fig. 4a, b Photomicrograph of resected specimen. Most parts of tumor were mature lipoma and fibromatous tissue, but neural tissue, cartilage, bone, mucus glands, and alimentary and respiratory tract mucosa were also prominent. Photograph at bottom shows cartilage, mucinous gland, and ciliated epithelia

Lorenzo et al. [11] collected 40 cases of intraspinal teratoma in children and found that the incidence in the first 5 years of life is very high (84.%, with 43.8% in the 1st year). In Matson's series [12] of 134 intraspinal tumors in children, 16 (11.9%) occurred in the 1st year of life. Oi and Raimondi [13] found two cases of intradural extramedullary teratoma among 64 intraspinal neoplasms in childhood. Hendrick [8] reported three thoracolumbar teratomas with excellent results in a series of 80 intraspinal tumors. Kordas et al. [10] reported two teratomas among 80 cases of spinal tumors in childhood. In a review of 81 cases, DeSousa et al. [4] reported six intraspinal teratomas, three of which originated in the sacral region. However, an extensive intraspinal teratoma such as the present case that required multi-stage surgery is not found in the literature patients of any age.

Neurogenic tumors such as neuroblastoma are the most common cause of extradural or dumbbell tumors in childhood that require prompt resection via a laminectomy. Laminectomies, however, are known to cause late postoperative sequelae such as spinal instability and deformity. Ishida et al. [9] compared conventional extensive laminectomy and suspension laminotomy with regard to postoperative complications and found that a laminotomy causes less postoperative spinal deformity and dural constriction. Postoperative spinal instability and deformity largely depend on the age of the child and the extent of the laminectomy. To prevent these sequelae, often encountered in younger children, tumor resection via a laminotomy is recommended instead of a destructive laminectomy [1, 14–16], and reanchoring the dissected vertebral arches and ligaments has been described.

Our patient would probably have developed a greater spinal deformity and instability if a laminectomy had been used. There remained a slight shift of the midline spinous processes to the right owing to the right-sided laminotomy by an air drill. Postoperative CT from the cervical to the lumbar spine revealed a satisfactorily-preserved intraspinal space and

normal-appearing reconstructed vertebral arches. The recovery of the dural space was complete. Laminotomy can thus be strongly recommended in the treatment of intraspinal tumors in young children.

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