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W R Smoker, J Biller, S A Moore, D W Beck and M N Hart

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Intradural Spinal Teratoma: Case Report and Review of the Literature

Wendy R. K. Smoker,[1] Jose Biller,[2] Steven A. Moore,[3] David W. Beck,[4] and Michael N. Hart[3]

Primary intradural teratomas are very rare tumors, only 20 cases have been reported [1–16]. Intraspinal teratomas are most often located in the dorsal aspect of the cervical or lower thoracic-upper lumbar regions. We report a case of an upper thoracic, ventrally located intraspinal teratoma associated with multiple spinal anomalies. The patient was evaluated by conventional myelography, CT metrizamide myelography, and MRI.

Case Report

A 26-year-old man was admitted with a chief complaint of progressive inability to walk. In addition to congenital absence of the pectoralis muscles, he had a long history of "occasional back problems," presumably related to a congenital thoracic scoliosis. Two months before admission, he had the onset of interscapular pain after twisting his back. He did not seek medical attention. Although the pain persisted, he was able to continue to work. Five days before admission, he noted difficulty walking, which became progressively worse to the point that he could not stand without assistance. He had fallen on a number of occasions. On the day of admission he noted the onset of urinary hesitancy without incontinence. Neurologic examination revealed decreased muscle strength in both lower extremities, bilateral lower-extremity hyperreflexia with ankle clonus, and bilateral Babinski signs. Superficial abdominal and cremasteric reflexes were absent. Sensory examination disclosed hypalgesia and thermoanesthesia below the T2–T3 level bilaterally, greater on the right. There was markedly decreased proprioception and vibratory sensation in the lower extremities bilaterally.

Thoracic spine radiographs revealed nonsegmentation of the upper five thoracic vertebral bodies and spina bifida occulta of T1 (Fig. 1). A lumbar puncture was performed at the L2–L3 interspace and 2 ml of pantopaque instilled into the subarachnoid space. In a 90° prone, head-down position, contrast material would not advance past the T5 level. Five cm³ of air were then injected, but the pantopaque could not be advanced (Fig. 2A). A lateral C1–C2 puncture was then performed, and 5 ml of metrizamide placed into the subarachnoid space (Fig. 2B). CT examination was then carried out from C6 to T8 (Fig. 3). The cord was markedly compressed from C7–T1 to T5–T6 by a ventrally located mass (Fig. 3). Pantopaque, previously trapped within the cleft in the prone position, was no longer present in the supine position. MR images were then obtained, showing the lesion to be primarily cystic in nature, with signal intensity similar to that of cerebrospinal fluid (Fig. 4). A small solid component was seen at the caudal end of the cyst. Within the spinal canal, the air partially outlined the caudal extent of the tumor, separate from the compressed spinal cord (Fig. 4).

A laminectomy was carried out from C7 to T5. The spinal cord at the level of T2–T3 was extremely thinned as to appear translucent. A 25-gauge needle was placed through the cord and 8 ml of mucoid material were aspirated from a large cyst lying ventral to the spinal cord. Following cyst aspiration, the cyst, which extended from T1 to T3, was easily shelled out and totally removed. The smaller, solid portion of the tumor, located at the T4 level, was then removed by laser and coagulation.

Histologic examination of the cyst showed it to be lined by epithelium varying from pseudostratified ciliated columnar to simple cuboidal. No squamous epithelium was identified. A thin layer of fibrous tissue underlay the epithelium and contained focal small islands of neuroglial tissue. Histologic examination of the nodule primarily revealed fibrous tissue but also included smooth muscle bundles, hyaline cartilage, and fat (Fig. 5). No immature or malignant cells were present. Sex chromatin studies were not performed.

Postoperatively, the patient did extremely well. MR performed 1 week after surgery showed no evidence of residual tumor, and the spinal cord appeared normal in size (Fig. 6). At the time of discharge, 18 days after surgery, he had full strength in his lower extremities, was ambulating with the aid of a walker, and was able to void spontaneously. With the exception of proprioceptive loss, which did not return, other sensory modalities were intact. He made a satisfactory recovery, and 2 months after surgery he was walking with only the aid of a cane.

Discussion

Attempts to extract reasonably accurate data from the literature concerning intraspinal germ cell tumors are replete with frustration owing to the variety of terms under which they have been reported. Sachs and Horrax [17] noted that, in many of these tumors, derivatives of one or two germ
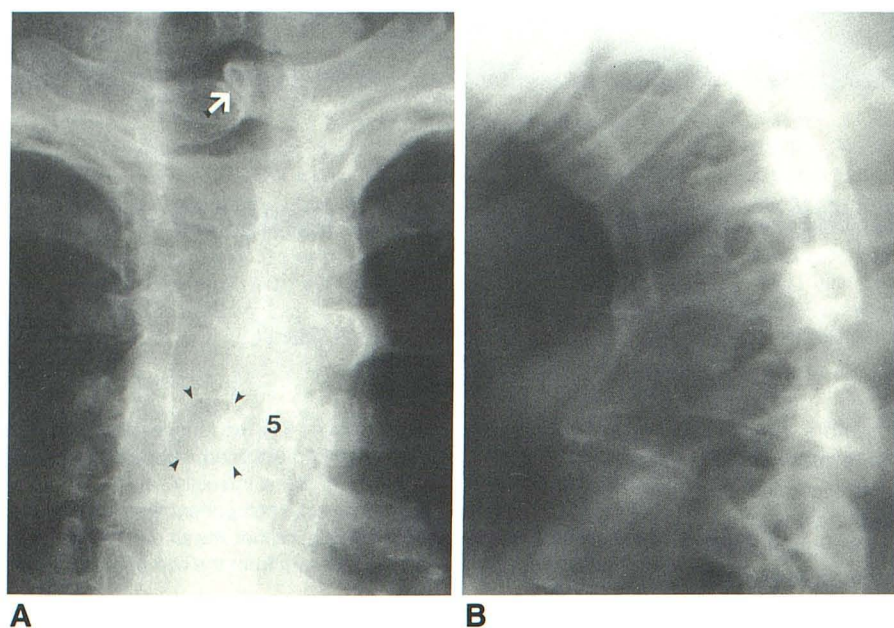
Fig. 1.—Anteroposterior (**A**) and lateral (**B**) plain radiographs of upper thoracic spine reveal nonsegmentation of bodies of T2–T6 and spina bifida occulta of T1 (*arrow* in **A**). An unusual crescent-shaped radiolucency is seen at T5 level on anteroposterior radiograph (*arrowheads* in **A**).
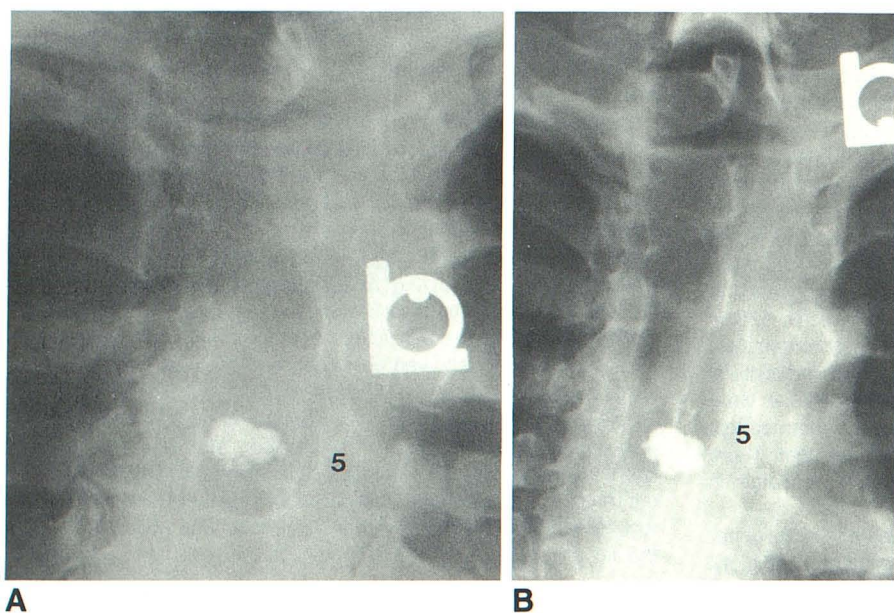

Fig. 2.—**A**, Anteroposterior radiograph obtained after injecting 2 ml of Pantopaque and 5 cm^3 of air. Patient is in a prone, 90° head-down position. A "complete block" to the cephalad flow of contrast is present at T5 level, precisely at upper margin of abnormal crescent-shaped lucency identified on plain radiographs (see Fig. 1A). Configuration of obstruction is unusual in that it is not characteristic for either an intra- or extradural block. **B**, Anteroposterior radiograph obtained after injecting 5 ml of metrizamide from a lateral C1–C2 puncture. Patient is in a prone, erect position. There is complete obstruction to caudal flow of contrast material at C7 level, with a smooth intradural configuration to the block. Pantopaque remains at T5 level even though patient is in a 90° feet-down position; however, it now appears to have moved caudally and lies precisely at lower margin of abnormal lucency.

layers tend to overgrow the others and that the total number of germ layers may be difficult to ascertain. We agree with these authors that tumors with recognizable tissue from only two germ layers should be termed "teratoids," and that only those tumors with identifiable tissue from all three germ layers should be labeled "teratomas" [17]. Willis [18] defines "teratoma" as "a true tumor or neoplasm composed of multiple tissues of kinds foreign to the part in which it arises." The most common components are skin, teeth, CNS tissue, respiratory and alimentary mucosa, and glands [18]. Although

many authors have referenced this definition in their own discussions, this definition does not require the presence of all three germinal layers to make the diagnosis of teratoma. Hence, a number of bigerminal tumors ("teratoids") have been included in the literature as teratomas [19–24]. This same observation was made many years ago by both Hosoi in 1931 [9] and Masten in 1940 [6]. Furtado and Marques, in 1951 [22], claimed that classification of germinal tumors as mono-, bi-, or trigerminal merely represented "a confession of inadequate examination." They believed that, if multiple serial
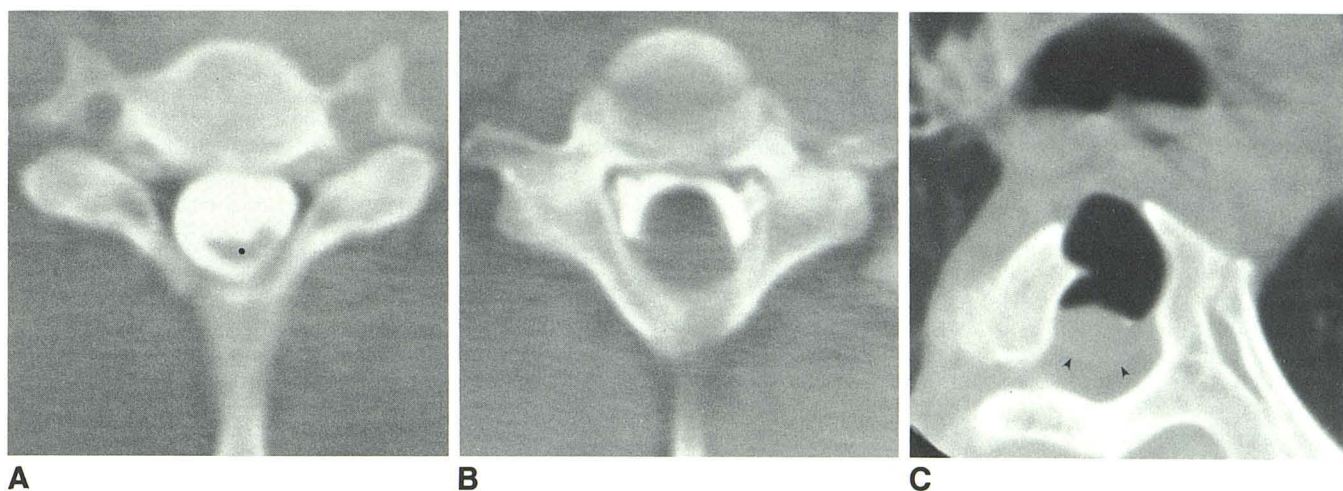
A   B   C

Fig. 3.—**A**, CT metrizamide myelography (C6). Posterior displacement of spinal cord (*dot*) and large ventral subarachnoid space. **B**, CT metrizamide myelography (C7–T1 disk space). Posterior aspect of subarachnoid space is obliterated by an intradural lesion. **C**, CT metrizamide myelography (mid-body of T5). Air instilled at myelography is identified in anterior subarachnoid space filling cleft in T5 vertebral body. There is a suggestion of the cord outline (*arrowheads*).
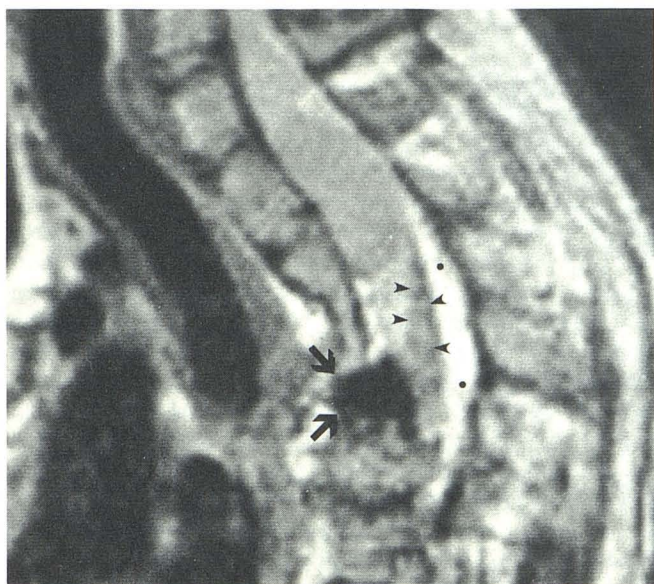

Fig. 4.—Sagittal T1 MR image more clearly shows craniocaudad extent of tumor extending from C7–T1 level to T5–T6 level. While majority of lesion is cystic, a smaller solid component is located at T4 level (*arrowheads*). Spinal cord is displaced posteriorly (*dots*). Air fills cleft of T5 vertebral body (*arrows*).

sections covering the entire tumor were obtained, elements originating from all three germ layers would be identified [22].

We reviewed all reported cases of intradural tumors termed teratomas [3, 5–8, 10, 13, 14, 19, 20, 22, 23, 25–31], teratomatous cysts [4, 32–35], cystic teratoid tumors [36], teratoid cysts [37], cystic teratomas [1, 2, 11, 12, 21], and teratoid tumors [1, 2, 9, 24, 25, 38, 39]. From the histologic descriptions, we could identify only 20 lesions that were trigerminal and could properly be termed intradural "teratomas" [1–16]. (We also acknowledge the "intramedullary teratoma" reported by Garrido and Stein [40], the "intraspinal teratoma" reported by Frazier [41], and the "conus medullaris teratoma" reported by DeSousa et al. [42]. Unfortunately, no histologic details were included for these cases, and their inclusion or exclusion as trigerminal lesions is not possible. Although the intradural cervical lesion described by Adams [43] has been included by some authors under discussion of intraspinal teratomas [11, 35, 37], the lesion was monogerminal in origin and clearly an enterogenous cyst. Similarly, the intradural ependymal cyst described by Hyman et al. [44] has inappropriately been included as a teratomatous cyst by some authors [32, 37].) In addition to the present case, only one reported intradural teratoma has been primarily located in the upper thoracic region [13] and, with the exception of the case reported by Teng and Gordon [8], all the intradural-extramedullary lesions have been dorsal in location. The majority of reported intradural teratomas are cervical or thoraco-lumbar in location.

In 1978, Rosenbaum et al. [32] reported a case of a bigerminal cystic intraspinal tumor, which they termed "teratomatous cyst." In their discussion they stated, "Although the intraspinal teratomatous cysts may be associated with other congenital anomalies of the spinal axis, the association is not as frequent and the anomalies are not as severe with the intraspinal teratoma as they are with the trigerminal teratoma" [32]. In addition to being confusing and redundant, we believe this is an erroneous statement. In our review of the reported intradural (trigerminal) teratomas, we found no spinal axis congenital anomalies in half of these patients, and, among the remaining patients, spina bifida was the most common congenital spinal axis anomaly associated with these tumors.
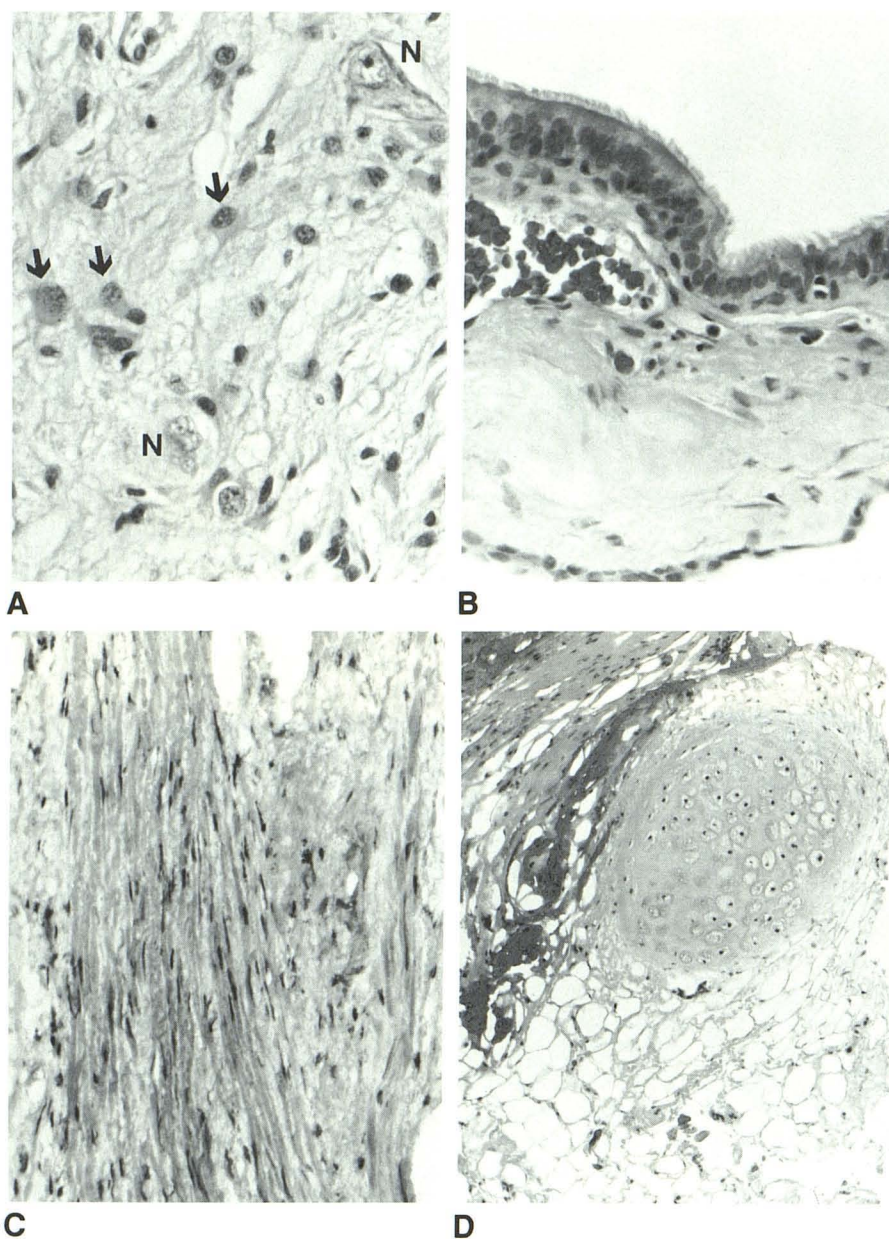
Fig. 5.—**A**, The ectodermal portion of this spinal teratoma is represented by disorganized neuroglial tissue found in cyst wall, distinctly extramedullary in location. Seen here are variably shaped neurons (N) and reactive astrocytes (*arrows*) in a background of neuropil. **B**, The endodermal derivative is respiratory-type epithelium, which lined cystic portion of tumor. Pseudostratified, columnar, ciliated epithelium lines cyst lumen (*top*). A fibrovascular stroma separates ciliated epithelium from arachnoid cap cells (*bottom*). **C** and **D**, Mesodermal derivatives include bundles of smooth muscle, cartilage, and fat. These tissues were located exclusively in solid portion of tumor.

Intradural germ cell tumors have been reported in association with a variety of spinal axis congenital anomalies. Spina bifida occulta at the level of the lesion has been reported most frequently. Both diastematomyelia and diplomyelia have been reported in association with these tumors [3, 32]. In 1938, Ingraham [2] reported a case of a 10-week-old infant with bifid T1–T4 vertebral bodies in association with an intradural "cystic teratoma." (The pathologic description of this tumor reveals no clear-cut evidence of mesodermal elements, and we believe this lesion to be a bigerminal "teratoid.") As speculated by Ugarte et al. [45], these defects could, perhaps, be explained on the basis of Gardner's "hydromyelic theory" [46, 47]. The basic defect could be failure of the rhombic roof of the fourth ventricle to become permeable to fluid forming

within the developing neural tube between 6 and 8 weeks of gestation. Resulting overdistention of the neural tube occurs (hydromyelia), which could lead to rents in the roof and floor plates of the neural tube (diastematomyelia). Mechanical displacement of the neighboring sclerotomes could account for their failure to unite anteriorly (cleft vertebrae) or for their joining improperly (spinal canal midline spur). The various manifestations could be attributed to the time of onset of the hypothetical permeability defect, to whether or not the defect becomes compensated, and to the magnitude and timing of such compensation [45].

The origin of intradural germ cell tumors is unknown. Kubie and Fulton [4] suggested that teratomatous cysts represent an ependymal diverticulum of the central canal of the spinal
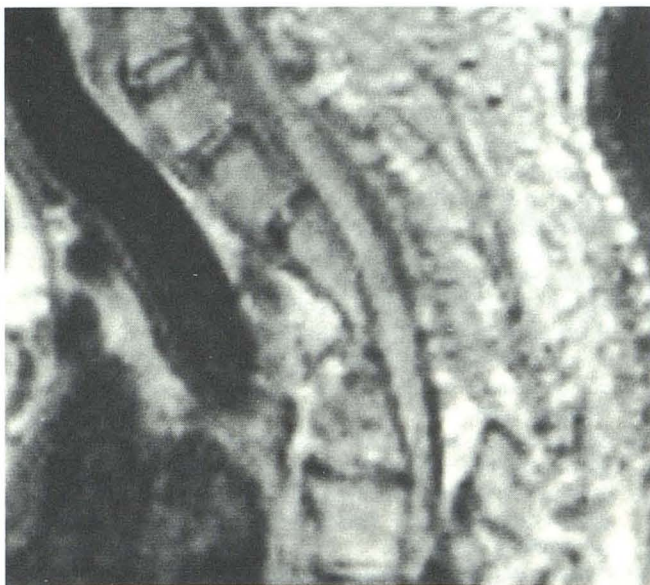
Fig. 6.—Postoperative sagittal T1-weighted MR reveals normal spinal cord size and configuration.

cord. This theory, however, does not explain the presence of bone, cartilage, and adipose tissue present in many of these cysts. Rhaney and Barclay [48] thought that these tumors were congenital malformations. They believed that the primitive streak was capable of forming both endoderm and paraxial mesoderm so that detachment and maturation of cells from Hensen's node, as the ecotoderm migrates caudally, could give rise to cysts lined by ciliated columnar or mucus epithelial cells [48]. In 1964, Rewcastle and Francoeur [33] performed sex chromatin studies on six patients with intraspinal teratomatous cysts. Upon finding sex chromatin in the nuclei of cells lining the cystic cavities in 48% of the male patients with teratomas, they suggested that the cells forming the epithelial lining of these cysts resulted from faulty migration of germ cells from the endoderm to the primitive gonad [33]. The absence of sex chromatin in the fibrous connective tissue would suggest that only the cells lining the cyst were the product of faulty germ cell migration [33]. These sex chromatin studies would support the theory that these cysts represent true teratomas and are not the result of escape of cells from the primitive streak during embryogenesis.

More recent experimental work with the murine teratoma model has shown that primitive embryonic cells, other than germ cells, are multipotent and therefore capable of giving rise to teratomas [49]. Nongerminal (extraembryonic) yolk sac cells in rats can apparently also form teratomas [50]. There is little information available concerning extragonadal human teratomas, but recent data on the cytogenetics of such tumors favor their origin from mitotically dividing diploid cells instead of germ cells [51]. Thus, extragonadal teratomas could arise from cells genetically identical to the somatic cells of the host, being similar to identical twins [51].

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