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Case Report

Subtotal resection of an intradural mature teratoma in an adult presenting with difficulty initiating micturition

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Abstract

Background: Teratomas are tumors comprised of tissues from all three germ layers. Teratomas within the spine are exceedingly rare especially in the absence of either spinal dysraphism, congenital abnormalities of the spine, spinal surgery, or history of lumbar punctures. Virchow was the first to describe this occurrence in the spine in 1863 and since then, only a handful of cases have been reported. **Case description:** A 41-year-old male presented with a longstanding history of difficulty initiating micturition and lower back pain with recent onset of saddle paraesthesia and bilateral leg pain. He did not have a history suggestive of spinal trauma nor congenital abnormalities. Neurological examination was unremarkable. Magnetic resonance imaging (MRI) confirmed the presence of an intradural extra axial lesion in the region of the cauda equine. At surgery, the lesion was found to be densely adherent to the conus and a subtotal resection was performed. Histological examination confirmed the lesion to be a mature teratoma. Postoperatively, he made a good recovery and there is no evidence of recurrence at one year follow-up. **Conclusion:** This case demonstrates that a teratoma without immature elements can be subtotally excised to reduce the risk of neurological morbidity.

Key Words: Extramedullary, intradural, spinal tumor, teratoma

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INTRODUCTION

Teratomas consist of tissues that are derived from all three germ layers and can be classified according to their histological characteristics.[1-5] Immature teratomas consist of relatively undifferentiated elements, which can resemble fetal tissue, can be aggressive tumors and have a tendency to recur. Mature teratomas consist of fairly well-differentiated tissues and tend to behave in a comparatively benign fashion.[5,6,10] Malignant teratomas are derived from the yolk sac or endodermal sinus and carry a poor prognosis.[2]

The occurrence of teratomas within the spine is exceedingly rare and only a handful of cases have been reported since their initial description by Virchow in 1863.[10] They can occur anywhere midline from the cephalic to the caudal extremity, however, they have most frequently been reported in the region of the cauda equine and are usually associated with either spinal dysraphism, congenital abnormalities of the spine, spinal surgery or history of lumbar punctures. Indeed it is rare for any one of these features not to be present.[8]

We describe a case of an intradural extramedullary mature teratoma in an adult male who presented with difficulty initiating micturition.

CASE REPORT

A 41-year-old male presented with a longstanding history of difficulty initiating micturition and lower back pain with recent onset of saddle paraesthesia and bilateral leg pain, which had been associated with decreased sensation on ejaculation and occasional fecal incontinence. There was no history of spinal dysraphism, congenital spinal abnormalities, previous spinal surgery, or lumbar puncture.

Clinical examination revealed no evidence of cutaneous abnormalities. Neurological examination was unremarkable.

Computed tomography (CT) lumbar spine revealed no bony abnormalities. Magnetic resonance imaging (MRI) confirmed the presence of an intradural, extramedullary lesion at T12/L1, which was causing marked compression of the distal conus. The tumor measured 35 × 14 mm and displayed hyperintense signaling on pregadolinium T1 and low signal on T2 with a small crescentic element of fat on the dorsal superior border Figures 1a and 2a.

The surgical management consisted of a laminectomy from T11 to L1 and subtotal excision. At surgery, once the dura was opened the capsule was initially decompressed and a considerable amount of proteinaceous fluid was drained. Thereafter, the majority of the capsule was excised, however, the base of the tumor was found to be densely adherent to the distal conus. It was decided that this could not be safely dissected from the conus and therefore a thin rim of capsule was retained.

Postoperatively, the patient developed transient urinary retention, however, he went on to make a good recovery with resolution of his back and leg pain. His bladder function improved although he does report occasional fecal incontinence.

Histopathologic examination of the lesion demonstrated elements of endoderm, mesoderm, and ectoderm with fragments of degenerate keratin, cystic spaces lined with stratified squamous epithelium, adipose tissue, disorganized neural tissue as well as mucus secreting epithelium Figure 3a and b. There were no immature elements or malignant cells seen.

Postoperative imaging at one year confirmed successful decompression of the conus and no evidence of tumor recurrence Figures 1b and 2b.

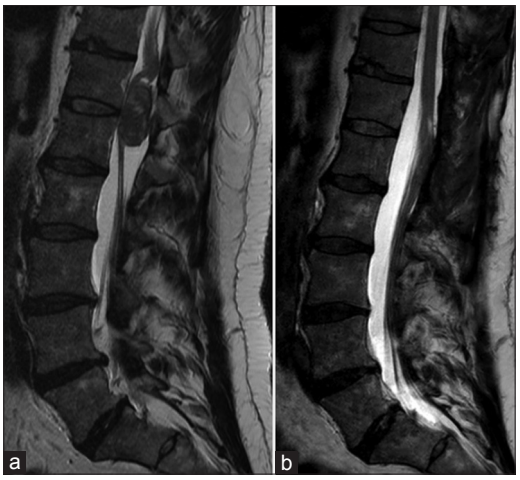

Figure 1: (a) and (b) Sagittal T2-weighted MRI demonstrating intradural extra axial lesion (left). Sagittal T2-weighted MRI at one year demonstrating tumor excision with no evidence of recurrence (right)

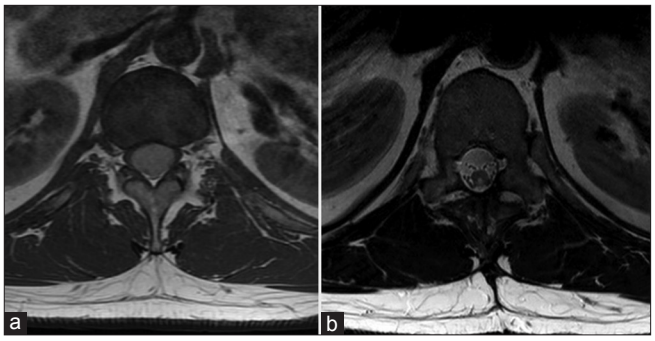

Figure 2: (a) and (b) Axial T1-weighted MRI with gadolinium contrast demonstrating diffuse enhancement with severe canal compromise (left). Axial T2-weighted MRI at one year demonstrating tumor excision (right)

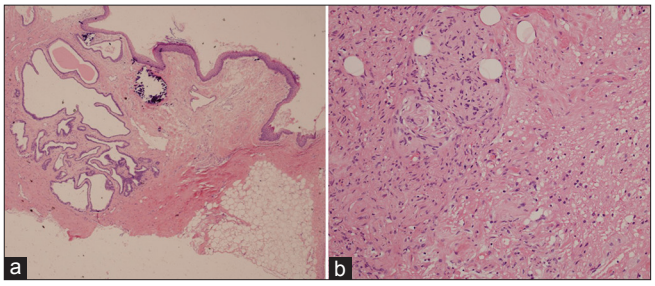

Figure 3: (a) A hematoxilin and eosin stain, ×10 magnification, showing a teratoma with a squamous-lined cyst wall with calcification, fat, and columnar lined epithelial structures. (b) A hematoxilin and eosin stain, ×20 magnification, showing teratoma containing disorganized schwannian cell (left) and glial tissue (right) together with adipocytes

DISCUSSION

The pathogenesis of spinal intradural teratomas remains unclear. The traditional theory is that in the early weeks of embryogenesis, the primordial germ cells are misplaced into the dorsal midline, during migration to the gonadal ridges from the primitive yolk sac.[2,5,9] This is supported by the fact that there is an association between spinal teratomas and midline spinal dysraphic defects.[2,6,10] An alternative explanation is provided by the dysembryogenic

theory, which suggests that pluripotent cells from the caudal cell mass in the developing embryo can result in a teratoma with multiple erroneous gene function and cellular inductive interactions.[6,8]

Given the slow growing nature of these tumors, they generally present with subtle, nonspecific signs and symptoms such as back pain or radiculopathy.[7]

The diagnosis of a teratoma may be suspected if initial plain radiography or CT imaging reveals bony changes such as vertebral bodies erosion, displaced pedicles, thinned laminae, calcifications, or features suggestive of diastematomyelia.[2,10] However, this case has demonstrated that these findings are not always present. MRI is the imaging of choice, however, the imaging characteristics can be variable depending on the degree of cystic and solid components.[1,6]

Although the treatment of choice is complete surgical excision, this case has confirmed that this is not always necessary especially if this can only be achieved at the risk of surgical morbidity. The rate of adherence to the cord is reported to be in the region of 50% and there can be little doubt that dissection of the densely adherent capsule from the conus would have resulted in a neurological deficit.[3] For this reason, previous authors have advocated performing subtotal resections and this would appear to be reasonable given the relatively small difference in the recurrence rates between subtotal and total resections, (11% and 9%, respectively) and the slow-growing nature of these tumors.[10]

There would appear to be little role for adjuvant therapy in the management of patients with mature spinal teratomas and the long-term prognosis for these patients is good.[4,5]

There is insufficient data from which to draw guidelines regarding follow up for patients with mature spinal teratomas, however, given the slow growing nature of these tumors, this patient will require serial clinical and radiological follow up for a number of years.

In conclusion, this case has demonstrated that these rare slow growing tumors can occur even without the more typical skeletal or cutaneous abnormalities and the development of subtle neurological symptoms should lead the clinician to consider the diagnosis. Thereafter if necessary, these tumors can be successfully managed with subtotal excision in order to minimize the risk of neurological morbidity.

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