

Adult Thoracic Intradural Exophytic Mature Teratoma: Case Report and Literature Review

Abstract

Mature thoracic intraspinal teratomas are rare tumors in adults. In this case study, we present a case of intradural, extramedullary teratoma, which was surgically resected. A 50 year old man presented with progressive bilateral leg pain, severe myelopathy and weakness. Magnetic Resonance Imaging (MRI) revealed a cystic mass lesion in the T11-12 region. Microsurgical resection of the tumor using CO² laser with neuromonitoring was performed. Postoperatively, the patient had a remarkable clinical improvement. Mature spinal teratomas are rare, slow growing spinal tumors. Surgical resection provides excellent recovery, and recurrence rates are low.

Keywords: Spinal teratoma, teratoma, thoracic tumor

Introduction

Teratomas are rare in the spinal column representing <0.5% of spinal tumors.^[1] Most teratomas are identified in children and are even more rare in the adult population. These tumors are histologically made of one to all three germ layers.^[2]

Teratomas are classified as mature, immature, or malignant depending on the amount of cell differentiation.^[3] Most reported cases occurring in the pediatric population are the immature or malignant subtypes and are usually in the sacrococcygeal region. Teratomas in adults are typically the mature subtype and occur most commonly at the level of the conus.^[1,4] These tumors are slow-growing in adults and patients usually present with progressive pain and declining neurological function.^[5]

We present an unusual case of an intradural intramedullary mature teratoma in a 50-year-old male who presented with worsening back pain, urinary incontinence, and progressive lower extremity weakness. Clinical characteristics, pathological features, and surgical nuances for resection of these lesions will be discussed.

Methods

We conducted a retrospective review of initial clinical presentation, pathology,

surgical treatment details, and clinical status of the patient at follow-up. An institutional review board approval was obtained, and the patient consented for the records to be reviewed and the findings to be published in this case report.

Case Report

A 50-year-old male presented with progressive bilateral leg pain, weakness, urinary retention, bowel and bladder dysfunction, and erectile dysfunction for 7 years. He developed progressive lower extremity weakness over a 2-month period, leading to him becoming wheelchair-bound. The motor function in the right hip flexion, knee extension, knee flexion, dorsiflexion, and plantar flexion was 2/5 and 1/5 strength in great toe extension. His left lower extremity strength was 4/5 in hip flexion, 4-/5 in knee extension, knee flexion, dorsiflexion, and 4/5 in plantar flexion and great toe extension. While his sensation was decreased on the right below the L2 dermatomal distribution, all other dermatomes of the bilateral upper and lower extremities were intact to pinprick. Bilateral upper extremity deep tendon reflexes were 2+, whereas lower extremity reflexes were absent. The Babinski reflex was absent bilaterally, whereas the patient had positive bilateral Hoffman's, Tromner and pectoralis reflexes.

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Magnetic resonance imaging (MRI) imaging revealed the presence of a heterogeneous, intradural mass with cystic components, and expansion of the cord/theal sac T11–T12. The findings on the MRI also suggested possible lumbarization of the sacral vertebra, making the case for cord to conus transition at the level of the mass. The lesion exhibited heterogeneous T1 enhancement [Figure 1].

He was subsequently taken to the operating room for tumor debulking. Thoracic laminectomies at T11–T12 were performed, and a midline durotomy was made, exposing a yellow, fatty, exophytic intramedullary tumor with large cystic components. Neural motor units were found widespread over the lesion, which triggered electromyographic muscle activity (EMG) activity. A flexible CO₂ laser was used to debulk the fatty components exophytic to the cord, from medial to lateral, carefully without causing any compression on the cord/conus, and traction on the surrounding nerve roots. The intramedullary cysts were subsequently exposed, revealing mucous products and hair [Figure 2]. A small amount of residual tumor was present that triggered EMG activity. This was left in place for concern of possible viable nervous tissue.

Pathology confirmed the presence of a mature teratoma. The lesion consisted of disorganized, heterogeneous collections of tissue derived from all three germ layers. All elements were noted to be mature, which included skin and adnexal structures, adipose tissue and bone, disorganized fragments of peripheral nerve and ganglion, neuroglial tissue, squamous and columnar epithelium, mucous glands, hair shafts, and keratin debris. There was no evidence of immature elements or malignancy [Figure 3].

Postoperatively, the patient reported drastic improvement in pain. He continued to have lower extremity paresis, however, was able ambulate short distances with a walker. Both 1 and 6-month postoperative MRIs demonstrated small areas of residual tumor that remained stable compared to immediate postoperative imaging and no new recurrence was noted. At 1 year, his strength had improved and was able to walk with a walker up to five blocks. His right leg motor function was 4/5 throughout, except for 2–3/5 dorsiflexion and 0/5 Extensor hallucis longus (EHL). His left lower extremity was 4- to 4+/5 strength overall. The bladder motor function did not improve, and he continued to require intermittent catheterization at 1-year follow-up.

Discussion

Mature spinal teratomas are usually adherent to nervous tissues and at times are composed of cystic structures that compress the neural elements.^[6,7] These tumors have very low rates of recurrence in the setting of both gross (9%) and subtotal (11%) resection.^[8,9] The leading theory of teratoma development is cell migrational error. Misplaced germ lines migrate to the caudal mass that usually forms at the conus and lower thoracic regions of the developing embryo.^[5]

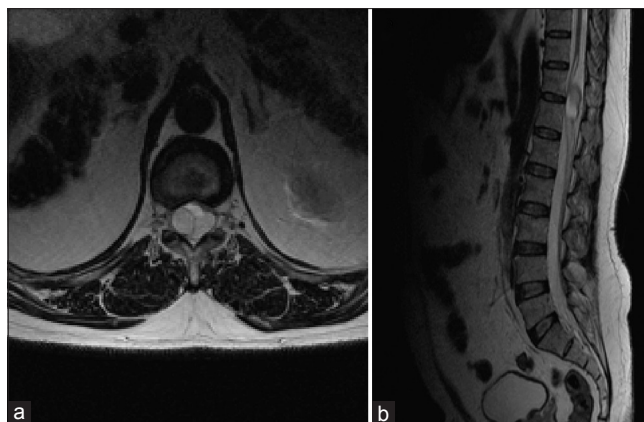


Figure 1: Magnetic resonance imaging. (a) Axial and (b) Sagittal T2 images show a heterogeneous cystic lesion at the level of the conus. The lesion appears intradural extramedullary

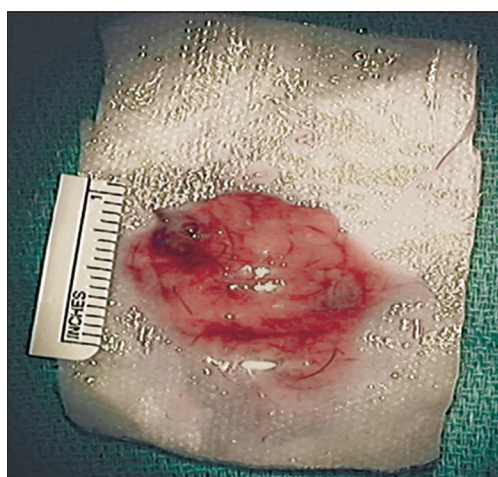


Figure 2: Firm fatty tumor with cystic components containing mucous products and hair

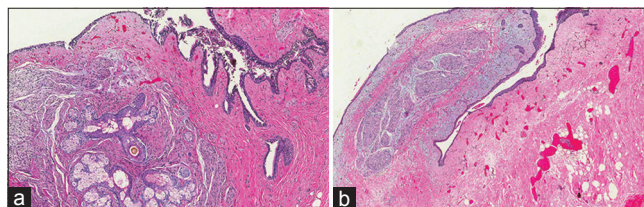


Figure 3: (a) Adipose tissue, peripheral nerve, sebaceous glands, hair shaft, and columnar epithelium lining the cyst wall. (b) Disorganized fragments of peripheral nerve and ganglion, fragments of bone, and squamous epithelium

This portends the common location of mature teratomas at or near the conus, the inclusion of several, but not all, well-differentiated germ cell layers and their relatively benign growth.^[10] However, intradural mature teratomas are rare, and the thoracic location has been described in the literature even less frequently.^[7,9,11-16] Furthermore, these lesions usually present in isolation and unlike teratoma of childhood, association with spinal dysraphism is rare.^[15]

In contrast, immature teratomas usually originate in the sacrococcygeal region and are less well

differentiated.^[17] These tumors exhibit rapid growth and secrete several biomarkers of proliferation. Immature teratomas are more commonly observed in the pediatric population and many case reports argue for a dysembryologic origin composed of chaotically differentiating pluripotent cells.^[10,18] These tumors require closer surveillance and more radical resection may be warranted.

Microsurgical techniques for tumor resection are used to dissect the tumor from the normal neural tissue. Intraoperative microscopy is used to develop a plane around the tumor. At times, as was the case in our patient, the tumor may invade the spinal cord parenchyma. The use of a CO₂ laser has many advantages in this setting. The CO₂ laser uses focused energy to coagulate and cut with pen-like precision without having to manipulate the tissues. Using the laser allows removal of infiltrating tumors in small areas next to critical structures. In addition, multiple adaptations have been made to the structure of the laser holder, including the neck, which resembles a pen and has the flexibility to allow alterations in the angle of use. Since no cleavage plane is present, the laser can be used to vaporize the tumor from the core to its periphery until white matter of the spinal cord is seen. A thin rim of tumor may be adherent to the parenchyma, which can be gently vaporized at a low voltage with the goal of disrupting viable tumor cells. The primary objective of surgical resection of intradural intramedullary spinal cord tumors is the decompression of neural elements without further damaging the neurological structures. Given the relatively benign nature of mature teratomas, we believe CO₂ laser for intramedullary tumor cell vaporization with preservation of the surrounding nervous tissue is paramount.

There is a small risk of recurrence as has been shown by Stevens *et al.*^[19] and annual evaluation of neurological function and spinal imaging is recommended. Adjunctive radiation or chemotherapy is usually not recommended except in cases of malignant cell components.^[8] This underscores the importance of complete pathological analysis of the whole tumor species as these tumors are heterogeneous and malignant cell populations may be interspersed among benign tissue.

Conclusion

Thoracic spinal intradural mature teratomas are exceedingly rare, and thus, the prognosis for this disease following surgery has not been well defined. While these teratomas are typically slow-growing, neurological decline warrants surgical intervention. Here, we propose the use of the CO₂ laser for microsurgical resection. As there is a risk of recurrence after surgery, routine clinical and MRI follow-up are necessary.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have

given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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