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Case Report

Intramedullary mature teratoma with an exophytic component in an adult: Report of a case and literature review

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ABSTRACT

Background: Intramedullary mature teratomas (IMMTs) are rare. This is particularly true in the adult population.

Case Description: A 49-year-old female developed progressive paraparesis due to a T6 intramedullary mature teratoma with an exophytic component. She was successfully managed, utilizing a laminectomy with microsurgical tumor removal. The literature review documented 57 similar cases.

Conclusion: Here, we presented a 49-year-old female with a T6 intramedullary mature teratoma accompanied by an exophytic component who underwent total tumor resection with an incomplete recovery.

Keywords: Adult, Intramedullary epidermoid, Intramedullary teratoma spinal cord, Thoracic spine

INTRODUCTION

Spinal teratomas account for approximately 0.1–0.5% of all spinal tumors.[1-6,9,10] They display a mixture of tissues derived from three primitive germ layers and are classified as mature, immature, or malignant.[1-6,9,10] Intramedullary mature teratomas (IMMTs) are the least common subtype found in adults.[1-6,9,10] Here, we present a 49-year-old female with a T6 IMMT who underwent surgical extirpation of the lesion with a resolution of the right lower extremity weakness, but residual left leg paresis. The literature review revealed 57 similar cases.[1-6,9,10]

CASE DESCRIPTION

A 49-year-old female presented with progressive weakness of both lower extremities of 2 years duration (i.e., bed-ridden). She was originally misdiagnosed with multiple sclerosis (MS). On examination, she exhibited a severe spastic paraparesis with hyperactive reflexes/bilateral Babinski signs. The thoracic MRI demonstrated a focal intramedullary lesion with an exophytic component at the T6 level with a mixed T2 signal intensity [Figure 1]. Utilizing an operating microscope, the patient underwent a laminectomy and midline myelotomy resulting in gross-total tumor excision [Figure 2]. The lesion proved to be a mature teratoma [Figure 3]. On the 10-day postoperative, MRI confirmed complete tumor resection [Figure 4]. Ten months later, her

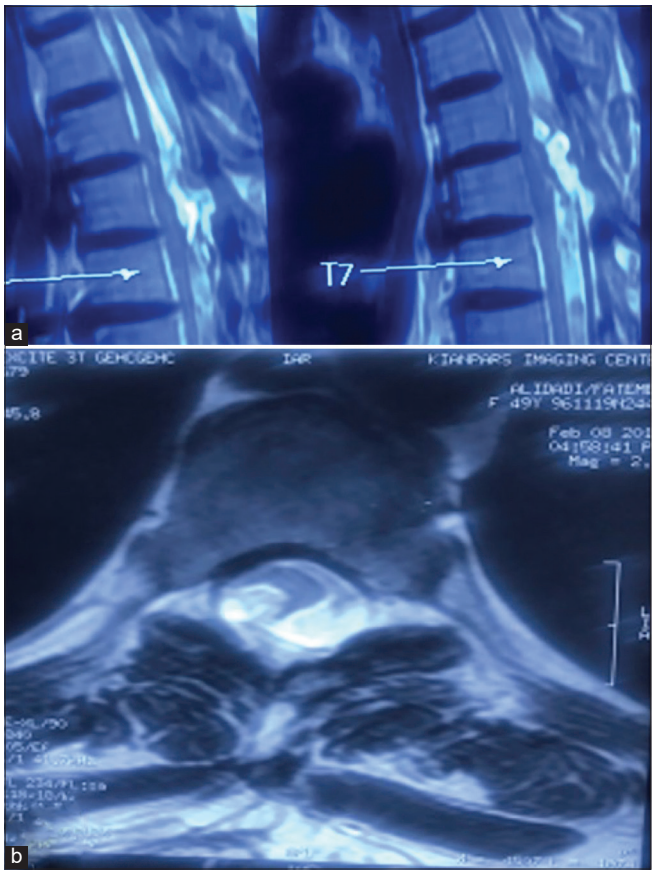

Figure 1: T2-weighted magnetic resonance imaging of the thoracic spine: (a) sagittal image showing a heterogeneous mass with an exophytic component at T6 level and (b) axial image.

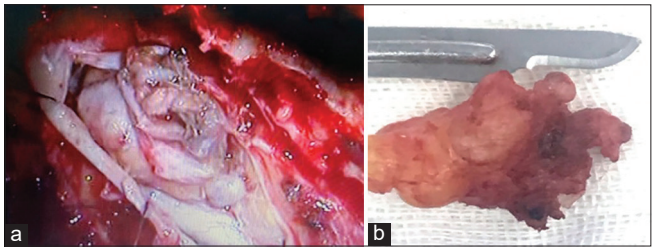

Figure 2: Intraoperative photographs (a) during the excision of a creamy yellow tumor which is fungating from the cord; (b) an irregularly shaped solid-soft tumor mass measuring 3 × 1.5 × 1 cm tumor after total removal.

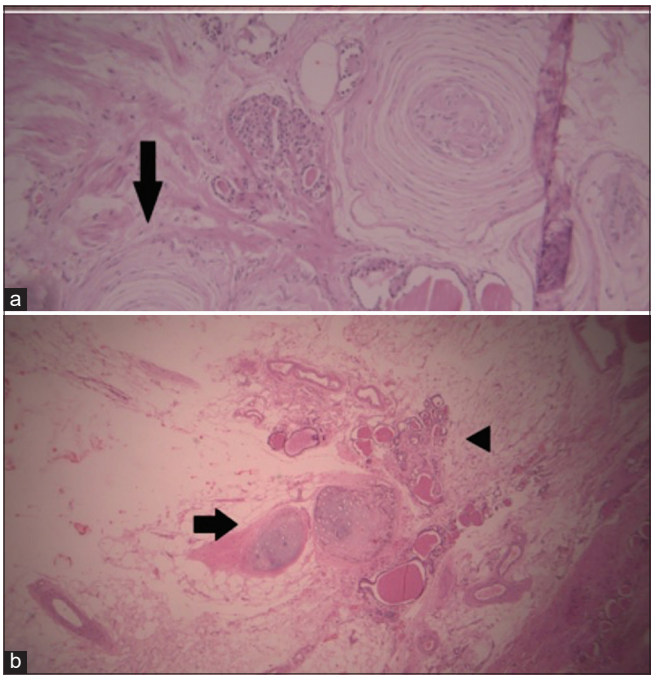

Figure 3: Pathology of the tumor, (a) presence of different components including thyroid like glands along with organoid corpuscle like nerve bundles (arrow) and (b) abundant adipose tissue along with focal mature cartilage (arrow) and some glandular structures (arrowhead).

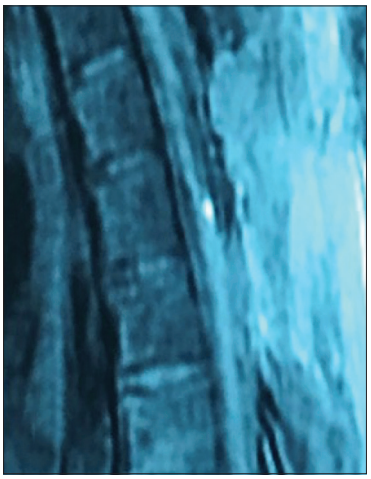

Figure 4: Postoperative T2-weighted sagittal magnetic resonance imaging of the thoracic spine shows total removal of the teratoma.

right lower extremity strength had dramatically improved, but left lower paresis remained unchanged.

DISCUSSION

Etiology

Teratomas originate from pluripotent stem cells which are derived from all three germ cell layers. In IMMTs, all three embryonic layers are misplaced within the dorsal aspect of the spinal cord (e.g., in the midline during their normal migration from the primitive yolk sac during neural tube closure).[1-6,9,10] Associated congenital anomalies include spina bifida, split cord malformation (SCM), meningomyelocele, and Klippel-Feil syndromes.

Incidence

IMMs are very rare in adults; there are 57 similar cases reported in the literature [Table 1]. In those studies, patients

Table 1: Approximately 57 reported cases of IMM in adults within medical literature; IMMs in adults are very rare.

Author	Year	Sex	Age	Location	Surgery
Hosoi	1931	M	24	Conus	Subtotal
Dereymacker	1954	F	43	Cervicothoracic	Total
Dereymacker	1954	M	34	Conus	Subtotal
Bakay *et al.*	1956	F	65	Conus	Subtotal
Sloof *et al.*	1964	M	20	Conus	Total
Sloof *et al.*	1964	M	67	Low Thoracic	Autopsy
Rewcastle and Francoeur	1964	F	34	Low Thoracic	Subtotal
Hansebout and Betrand	1965	M	47	Conus	Total
Caruso *et al.*	1966	M	41	Conus	Total
Eneström and Von Essen	1977	F	36	Conus	Subtotal
Rosenbaum *et al.*	1978	M	49	Low thoracic	Total
Besel,	1979	F	22	Low thoracic	Total
Garrison and Kasdon	1980	M	23	Conus	Total
Padovani	1982	F	21	Cervicothoracic	Partial
Padovani	1983	F	31	L. L. Conus	Partial
Conti,	1984	F	24	Conus	Partial
Giacomini	1986	M	31	Conus	Total
Pelissou-Guyotat	1988	M	33	Conus	Total
Nicoletti *et al.*	1994	M	47	L. L. Conus	Partial
Caruso *et al.*.	1996	M	41	Conus	Total
Al-Sarraj	1998	M	35	Conus	Partial
Koen	1998	F	31	L. L. Conus	Partial
Poeze *et al.*	1999	M	23	Conus	Partial
Fan *et al.*	2001	F	43	Conus	Total
Nonomura *et al.*	2002	F	33	Low Thoracic	Partia
Nonomura *et al.*	2002	M	56	Cervicothoracolumbar	Partial
Hejazi and Witzmann	2003	F	45	Conus	Total
Hejazi and Witzmann	2003	M	20	Conus	Total
Sharma, *et al.*	2003	M	32	T10	Not specified
Sharma *et al.*	2003	M	32	T11	Not specified
Sharma *et al.* 31	2003	F	51	T0-L2	Not specified
Sharma *et al.*	2003	M	30	L. L. Conus	Not specified
Fernández-Comjo	2004	M	46	Conus	Total
Ak	2006	F	43	Cervical	Total
Paterakis	2006	M	63	Cervical	Subtotal
Tsitsopoulos	2006	F	44	Low thoracic	Subtotal
KahiloguL. L.ari, *et al.*	2006	F	42	Conus	Total
Caruso and Colonnese,	2006	F	40	Conus	Subtotal
Caruso and Colonnese,	2006	F	41	Conus	Total
Makary *et al.*	2007	F	46	Upper cervical	Total
Mut *et al.*	2007	F	34	Conus	Total
Mohindra	2008	M	35	Conus.	Total
Oh	2009	M	44	L. L. Conus	Subtotal
Benes	2009	F	52	L. L. Conus	Subtotal
Arvin	2009	M	34	Cervical	Total
Ghostine	2009	F	65	Cervical	Subtotal
Jian	2010	M	57	Conus	Total
Yu	2010	M	34	Conus	Subtotal
Yamomoto *et al.*	2013	F	42	Conus	Subtotal
Alkheray	2015	M	60	Conus	Subtotal
Asan	2016	F	29	Conus	Subtotal
Turan	2016	M	48	L. L. conus	Total Exophyt
Khazendar	2017	M	37	Conus	Total
Barahona	2018	M	54	L. L. conus	Total
Oliveiraa	2019	F	35	L. L. conus	Subtotal
Hrushikesh	2020	M	40	Conus	Total
Rahimizadeh, *et al.*	2020	F	49	Mid thoracic	Total

averaged 39.9 years of age and included 31 males and 26 females [Table 2].

The majority of lesions (39 cases) involved the terminal portion of the spinal cord/conus followed by the lower thoracic region (eight cases), cervical spine (five cases), cervicothoracic junction (three cases), and mid thoracic level (one case) (i.e., decreasing frequency).

Clinical picture

The clinical features of IMMTs reflect their locations. Those with conus lesions typically present with over-flow incontinence and progressive lower extremity paraparesis. More cephalad cervical tumors may result in quadriparesis, while upper thoracic lesions will be associated with higher-level paraparesis.

Imaging with CT/MR

MR scans are the studies of choice, as they readily demonstrate intramedullary heterogeneous cystic and/or solid masses with/without an exophytic component (latter seven patients in the literature). CT may document attendant bony abnormalities (i.e., a variable tumor density or calcification within the tumor).

Histopathology

As they originate from pluripotent stem cells, teratomas can incorporate a wide range of tissues including; skin, muscle, bone, cartilage, intestinal mucosa, fat, teeth, and even hair.[1-6,9,10] The distinction between epidermoid cysts and juxtamedullary lipomas with IMMTs may prove difficult.[7,8]

Table 2: The sex frequency and percentage of 57 adult cases with IMMT.

Sex	Frequency	Percent
Male	31	54.4
Female	26	45.6
Total	57	100.0

Table 3: Management of 57 patients with IMMT with frequency and percentage.

Surgery	Frequency	Percent
Autopsy	1	1.8
Not specified	4	7.0
Partial	9	15.8
Subtotal	16	28.1
Total	27	47.4
Total	57	100.0

Treatment

The best treatment for IMMTs is gross total surgical excision (e.g., found in 27 cases from the literature). Intimate adherence of tumor to the spinal cord parenchyma may preclude total removal (e.g., showing in 25 cases). Notably, 4 out of 57 cases in the literature did not specify what operative approaches were utilized [Table 3]. Postoperative neurological recovery may vary, long-lasting urinary incontinence and/or other long-term permanent deficits may not improve or resolve, and there is always the risk of worsening.[1-6,9,10]

CONCLUSION

The occurrence of an IMMT in an adult is rare. Timely diagnosis and surgical management (e.g., gross total if feasible) of these lesions remain critical in achieving the best postoperative outcomes.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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