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REVIEW ARTICLE

A comprehensive review of adult onset spinal teratomas: analysis of factors related to outcomes and recurrences

G. Lakshmi Prasad[1] · **S. Divya**[2]

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Abstract

Purpose Spinal teratomas are rare tumours noted in adults and are commonly located in the thoracolumbar region. Currently, there appears to be a lack of clear consensus regarding the management and prognosis of these lesions. A comprehensive review along with an illustrative case managed at the author's institute has been presented.

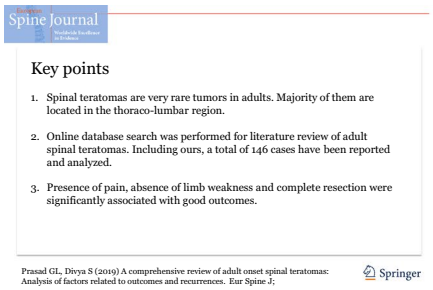
Materials and methods Online database search was performed for literature review.

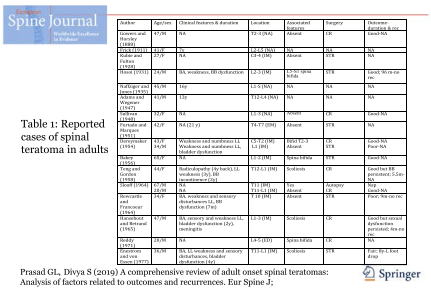
Results A 26-year-old male presented with acute onset neurological deficits and imaging revealed features of conus teratoma. Subtotal resection was performed and patient achieved fair recovery. Including the present one, a total of 146 cases have been reported and were analysed. Mean age was 39.6 years (range 18–85 years) and males predominated. Limb weakness and backache were the commonest symptoms. Majority of the lesions were intramedullary and located in the thoracolumbar region. Complete resection (CR) was achieved in 45% of cases. All, except one, were mature teratomas. Recurrences were noted in nine (6.1%) cases. Outcome was good/excellent in 86 (60%) and fair/poor in 26 (18%) cases. The presence of pain, absence of limb weakness and CR were significantly associated with good outcomes. Furthermore, subtotal resection (STR) had significantly higher recurrence rates than CR. Four deaths (2.7%) were noted.

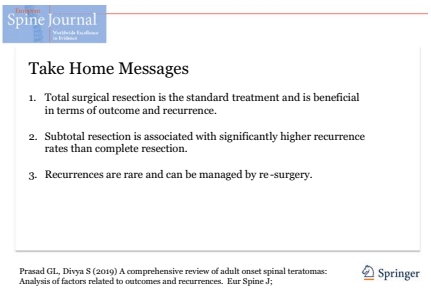
Conclusions Total surgical resection is the standard treatment and appears to be beneficial both in terms of outcomes and recurrences. Overall, recurrences are rare and may be managed by re-surgery. In addition, the present report is the eighth case of adult spinal teratoma with an acute onset presentation.

Graphic abstract

These slides can be retrieved under Electronic Supplementary Material.







Keywords Teratoma · Mature teratoma · Adult · Spinal · Spinal teratomas

Introduction

Spinal teratomas (ST) are rare tumours, constituting less than 1% of central nervous system tumours [1–3]. They are mostly noted in the paediatric age group and most commonly in sacrococcygeal spine [1, 2]. However, the thoracolumbar

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✉ G. Lakshmi Prasad
lakshmi.prasad@manipal.edu

Extended author information available on the last page of the article

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(TL) region is the most frequent location in adults and spinal teratomas presenting in adults are a rare entity. Till date, less than 150 cases of adult spinal teratomas have been reported in the literature [1–100]. The most common symptoms include pain, progressive limb weakness and bowel/bladder disturbances. An acute onset presentation has been rarely described in earlier reports [22, 28, 32, 33, 40, 82, 98]. In the present report, a rare case of a mature cystic teratoma of the conus–cauda region in a 26-year-old male is described. The patient presented with an acute onset paraplegia and bowel/bladder dysfunction and was managed by surgical resection. In addition, a thorough literature review has been presented on this topic of adult spinal teratomas reported in the world literature till date.

Materials and methods

All previous published reports (case reports, letters to editor, review articles and original articles) of adult onset spinal teratomas (with no language restriction) were retrieved and analysed in the current paper. Adult onset was defined as age ≥ 18 years. The first case that was reported in the year 1888 and all the cases that have reported till the present date were included in the analysis. Cases in which no information could be gathered (because of language barriers or missing data) were excluded. Sacrococcygeal teratomas and paediatric onset teratomas were excluded from the analyses. The following keywords were searched using online databases (Pubmed, Index Scopus, EMBASE, Google Scholar): 'teratoma', 'spinal', 'adult', 'spinal teratoma', 'intramedullary', 'intradural', 'extradural', 'mature teratoma', 'cystic teratoma' and 'review'. Hospital records were retrieved for our case report. The following characteristics were analysed: demographics (age, gender), presenting features, duration of symptoms, spinal level of the lesion (cervical/thoracic/thoracolumbar/lumbar/lumbosacral/sacral), axial level of the lesion [extradural (ED)/extramedullary (EM)/intramedullary (IM)], the presence of associated spinal anomalies, radiological and histopathological features, type of surgical procedures, completeness of resection, recurrences, mean time to recurrence (TTR) and outcomes at last follow-up (FU). Acute/rapid progression of deficits was taken as those occurring within 2 weeks of presentation. Outcome was considered good/excellent if functional or complete motor recovery was achieved, while outcome was considered fair/poor when the motor recovery was non-functional or there was non-improvement of symptoms or in cases of neurological deterioration after surgery. Chi-square test was applied to predict the correlation of the above-mentioned characteristics with outcomes and recurrences. A p value of < 0.05 was taken as significant.

Results

Case illustration

A 26-year-old male presented with acute onset weakness (over 48 h) of both lower limbs 1½-months back, associated with bladder and bowel dysfunction. Neurological examination revealed flaccidity in both lower limbs and complete paraplegia (MRC grade 0/5). Lower limb deep reflexes were non-elicitable. There were absent and reduced tactile sensations, respectively, on the left and right lower limbs below the L1 dermatomes. No cutaneous stigmata of spinal dysraphism were evident on examination. Magnetic resonance imaging (MRI) of the spine showed a heterogeneous lesion consisting of 2 components (cranial and caudal) extending from L1 to L2 vertebrae at the level of conus–cauda region. There was no evidence of associated spinal anomalies. The cranial part was intramedullary and appeared hyperintense on T1 and profoundly hypointense on T2 sequences. The caudal part was extramedullary and was isointense and hyperintense on T1 and T2 sequences, respectively. On fat-saturated sequences, the cranial component showed suppression, suggestive of a fat containing lesion, while both the components showed mild enhancement on contrast administration (Fig. 1a–f). Radiological diagnosis was a mature cystic teratoma. A L1-L2 laminectomy and tumour resection were performed. Intraoperatively, the spinous elements were normal. Caudally, an EM cystic tumour was noted attached to the conus containing calcium deposits and straw-coloured fluid. The cyst could be easily separated from the cauda equina roots and was totally removed. There was no breach in the pia over the nerve roots. The cranial part was intramedullary and contained predominantly fat and blood vessels. An ultrasonic aspirator was used for debulking the lesion. Since it was relatively difficult to differentiate from the conus, subtotal resection was performed. Intraoperative electrophysiological studies were not available and hence were not utilized in this case. Later, a watertight dural closure was performed. Post-operatively, patient had improvement in his lower limb tone and could appreciate tactile sensations from around the second week of surgery and also had mild improvement in his weakness (wiggle his toes and hold his limbs on the bed when kept in a flexed posture) when discharged from hospital. Histopathological examination showed pseudostratified ciliated columnar epithelium overlying fibrocollagenous stroma containing sebaceous glands, smooth muscle, nerve fibres, lobules of adipocytes and congested vessels—overall features suggestive of a mature teratoma. At 15-month follow-up, he had modest improvement in his power and was able to

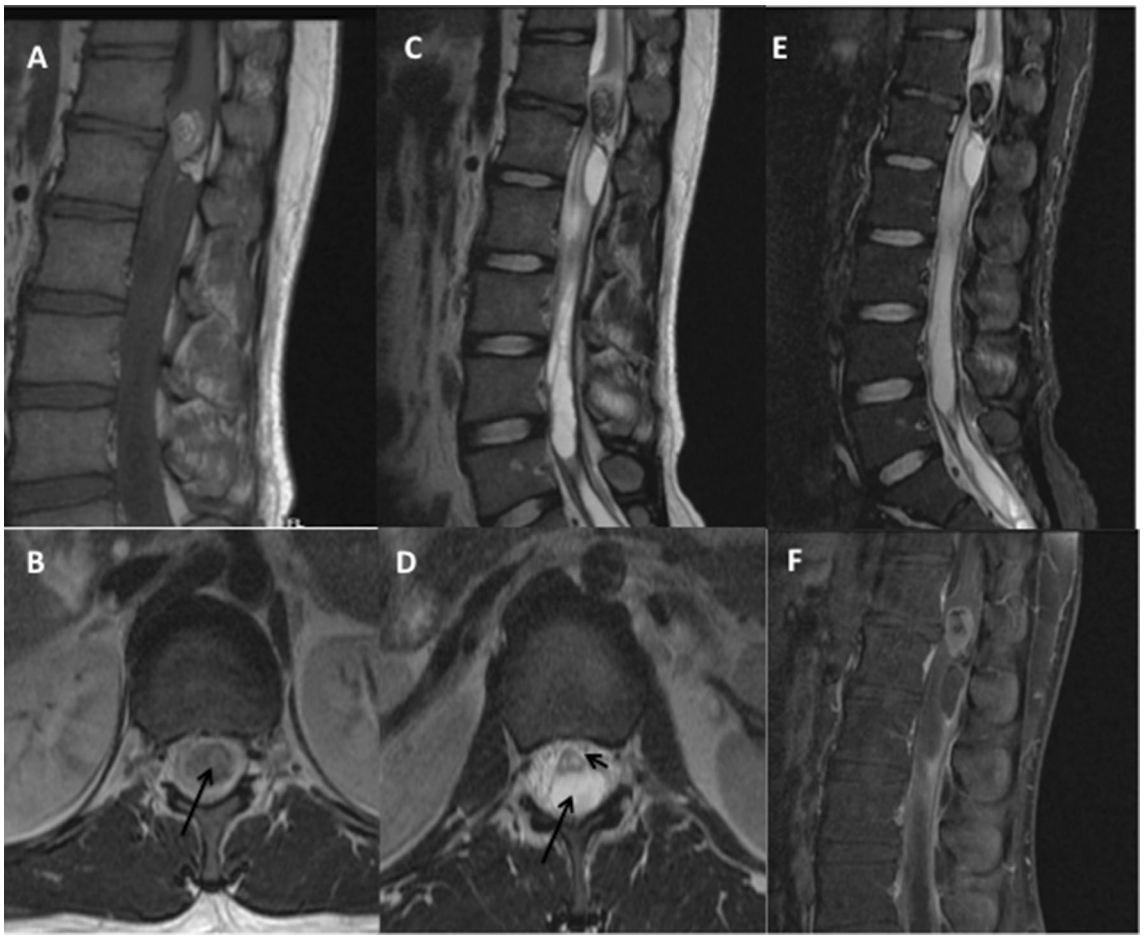

Fig. 1 Pre-operative MRI of lumbosarcal spine showing the heterogeneously appearing conus–cauda lesion with cranial and caudal components. Sagittal and axial T1 WI (**a**, **b**), sagittal and axial T2 WI (**c**, **d**) and STIR images (**e**) showing the cranial component as hyperintense on T1, hypointense on T2 WI and suppressing on STIR images, while the caudal cystic part is hypointense and hyperintense on T1 and T2 sequences and does not show fat suppression. The entire lesion is showing heterogeneous enhancement (**f**). The cranial part is intramedullary (arrow in **b**), and caudal one is extramedullary (long arrow in **d**). The cord is seen to be compressed and displaced anteriorly (short arrow in **d**)

stand with support, but was mostly dependent on others for his daily activities. However, his urinary problem did not recover and was on a catheter for voiding that was changed to clean intermittent catheterization (CIC) few months later. A follow-up MRI could have been helpful in determining the extent of resection, but could not be done because of non-willingness of the patient.

Including ours, a total of 146 cases of adult spinal teratomas have been reported till the present date and have been analysed in this present report. The mean age of the cohort was 39.6 years (range 18–85 years), and the median age was 36 years. The male/female ratio was 1.33:1. The mean duration of symptoms was 46.7 months (3 days to 20 years). Limb weakness and pain were seen in 76 subjects, bladder/bowel disturbances in 61 subjects and meningitic symptoms in three cases. Of those presenting with pain, axial pain (neck pain/backache) was seen in 47 cases and radiculopathy in 24 cases and site of pain was not mentioned for remaining 18 cases. History of minor trauma was reported in two cases. Acute/rapidly progressive symptoms were noted in eight cases (including ours) [22, 28, 32, 33, 40, 82, 98]. Details of clinical features were not available (NA) for a total of 28 cases.

With regard to the spinal level, the locations of the tumour were as follows: cervical: $n=15$, thoracic: $n=15$, thoracolumbar: $n=72$, lumbar: $n=38$, lumbosacral: $n=6$ and NA: $n=6$ cases. With regard to the axial location, the teratoma was located as follows: IM: $n=69$, EM: $n=60$, combined IM + EM: $n=8$, ED: $n=4$ and NA: $n=5$ cases.

Associated cutaneous abnormalities and spinal anomalies such as spina bifida, split cord malformations and scoliosis were present in 55 cases, absent in 84 cases and details NA for 6 cases. Of those, SCM-1 and SCM-2 were seen in 7 cases each and were noted in all levels, except the cervical

spine. Complete resection (CR) was achieved in 65 (45%) cases, while 54 (37%) cases underwent subtotal resection (STR). Details of resection were NA for 26 (18%) cases. In intramedullary teratomas, CR was achieved in 37.6% ($n=26$), STR in 46.3% ($n=32$) and details NA in 16% ($n=11$) cases. All, except one, were mature teratomas on histopathological examinations.

The mean FU duration was 37 months (2 weeks–208 months). Nine recurrences were noted, and the overall recurrence rate was 6.1%. One patient had six recurrences in 6 years [39]. Overall, the mean time to recurrence (TTR) was 88.2 months (4–180 months). With the exclusion of the single case of immature teratoma that recurred at 4 months, the mean TTR for mature teratomas was 98.8 months (12–180 months). With regard to the extent of surgical resection, there were one and five recurrences, noted for CR and STR, respectively. On statistical analysis, this difference was significant ($p=0.029$). Details of surgical resection were not reported (NR) for the remaining three recurred cases. No malignant changes were evident in the recurred cases.

Outcomes at last FU were as follows: good/excellent in 86 (60%) cases, fair/poor outcome in 26 (18%) cases and NA for 33 (22%) cases. Four deaths (2.7%) were reported, and one case was diagnosed at autopsy. Of the four deaths, two cases each were in the cervical and conus regions. Three cases were IM in location, and the reported case of teratoma with CSF leak/meningitis was EM. The causes of death were intracranial fat dissemination in two cases, CSF leak/meningitis and respiratory failure in one case each. On applying Chi-square test, the presence of pain ($p=0.019$), absence of limb weakness ($p=0.001$) and CR ($p=0.015$) were significantly associated with good outcomes. Also, EM tumours had higher rates of CR as compared to IM tumours ($p=0.025$). The remaining variables such as age (<40 years and >40 years, mean age being 39.6 years), duration of symptoms (<46 months, >46 months, mean duration being 46.7 months), gender, bowel/bladder involvement, location of tumour and associated anomalies did not have a significant correlation with outcomes or recurrence rates on statistical analyses. Table 1 summarizes all the reported cases of adult onset spinal teratomas.

Discussion

Epidemiology and clinical features

In general terms, teratomas are classically defined as neoplasms consisting of multipotential cells of all the three germ cell layers (according to Russell and Rubinstein) or as neoplasms with the power of autonomous growth (according to Wills) [1, 38, 78, 82, 101–103]. Spinal teratoma is an entirely separate entity from that of the intracranial and sacrococcygeal teratomas [39]. Excluding sacrococcygeal teratomas that are predominantly found in neonates and young children, STs are very rare tumours [1, 2]. They account for only 0.2–0.5% of all spinal cord tumours, and only 2% of all teratomas are found to occur in the neuraxis as reported in various studies [2, 3, 48, 72, 73, 104]. Sloof and Kernohan [15] noted only two cases of teratomas in their cohort of 1322 intraspinal tumours. In one previous report of 256 spinal tumours by Tapper and Lack [105], only four cases were teratomas. Sawamura [106] analysed 34 cases of CNS teratomas, and none were intraspinal.

Gowers and Horsley [4] described the first case of a ST in 1888. Majority of the STs are located in the TL region involving the conus, while thoracic and cervical ones are relatively uncommon [1, 48]. Adult onset presentation is rare, and only 145 cases have been reported till date [1–100]. Among adults, STs are most frequently seen in the 4–5th decades. They are more common in males; however, there appears to be a female preponderance in children [1, 40]. Spinal teratomas may remain asymptomatic or may present with non-specific features similar to that of any spinal mass lesion [1, 40, 54]. The most common presenting features include back pain, limb weakness and bowel/bladder disturbances. Except eight cases, all cases have reported a gradual progression of symptoms with the mean symptom duration being 46.7 months. There may be periods of remissions and exacerbations in the symptomatology, as noted by few authors [40]. However, in this present review, none of the cases have reported this type of presentation. With regard to the axial location in the spine, they can either be IM, EM or ED [1, 3, 40, 82, 104]. Intramedullary location appears to be the rarest, and conus medullaris appears to be the most frequent site for IM teratomas [49, 58]. Park et al. [104] compared ED versus ID teratomas and concluded that there were no differences between them in terms of male preponderance, lumbar location, young age, associated anomalies and pathogenic mechanisms.

Imaging

Plain radiographs have no significant role in the diagnosis of these lesions and are limited to detecting vertebral changes such as erosion or widening of the interpedicular space and other vertebral anomalies [49, 60, 107]. There are no characteristic features that can differentiate ST from other spinal neoplasms. Computed tomography (CT) is a sensitive method for differentiation based on the heterogeneity of the teratoma contents such as fat, calcification and cystic areas [40, 60]. MRI is the most sensitive imaging modality; however, in many cases, the diagnosis can be made only after histopathological examination. Heterogeneous solid-cystic masses consisting of fat and calcium with or

Table 1 Reported cases of spinal teratoma in adults

Author	Age/sex	Clinical features and duration	Location	Associated features	Surgery	Outcome; duration and rec
Gowers and Horsley [4]	47/M	NA	T2-3 (NA)	Absent	CR	Good; NA
Frick [5]	41/F	7 years	L2-L5 (NA)	NA	NA	NA
Kubie and Fulton [6]	27/F	NA	C3-4 (IM)	Absent	STR	NA
Hosoi [7]	24/M	BA, weakness, BB dysfunction	L2-3 (IM)	L5-S1 spina bifida	STR	Good; 96 months-no rec
Naffziger and Jones [8]	45/M	16 years	L1-S (NA)	NA	NA	NA
Adams and Wegener [9]	41/M	13 years	T12-L4 (NA)	NA	NA	NA
Sullivan [10]	32/F	NA	L1-3 (NA)	Absent	CR	Good; NA
Furtado and Marques [11]	42/F	NA (21 years)	T4-T7 (EM)	Absent	STR	NA
Dereymaker [12]	43/F 34/M	Weakness and numbness LL Weakness and numbness LL, bladder dysfunction	C5-T2 (IM) L1 (IM)	Bifid T2-3 Absent	CR STR	Good; NA Poor-NA
Bakey [13]	65/F	NA	L1-2 (IM)	Spina bifida	STR	Good; NA
Teng and Gordon [14]	44/F	Radiculopathy (4 years back), LL weakness (3 years), BB incontinence (2 years)	T12-L1 (IM)	Scoliosis	CR	Good but BB persistent; 5.5 months-NA
Slooff et al. [15]	67/M 20/M	NA NA	T11 (IM) T11-L1 (IM)	Yes Absent	Autopsy CR	Nap Good; NA
Rewcastle and Francoeur [16]	34/F	BA, weakness and sensory disturbances LL, BB dysfunction (7 months)	T 10 (IM)	Absent	STR	Poor; 9 months-no rec
Hansebout and Betrand [17]	47/M	BA, sensory and weakness LL, bladder dysfunction (2 years), meningitis	L1-3 (IM)	Scoliosis	CR	Good but sexual dysfunction persisted; 4 months-no rec
Reddy et al. [18]	20/M	NA	L4-5 (ED)	Spina bifida	CR	NA
Enestrom and von Essen [19]	36/M	BA, LL weakness and sensory disturbances, bladder dysfunction (4 years)	T11-L1 (IM)	Scoliosis	STR	Fair; 8 years-L foot drop
Rosenbaum et al. [20]	49/M	BA, weakness and sensory disturbance, bladder and sexual dysfunction (4 year)	T8-10 (IM)	SCM-2	CR	Good; 48 months-NA
Garrison and Kasdon [21]	23/M	BA, bladder dysfunction (5 months)	Conus (IM)	Absent	CR	Good; 4 months-no rec
Padovani et al. [22]	21/F	Pain (5 months), LL weakness (10 days)	C6-T1 (IM)	Absent	STR	Poor; 4 years-no rec

Table 1 (continued)

Author	Age/sex	Clinical features and duration	Location	Associated features	Surgery	Outcome; duration and rec
Padovani et al. [23]	33/F	BA, weakness LL, bladder dysfunction (2 years)	T12-L1 (IM)	Absent	CR	Good but bladder dysfunction persistent; 24 months-no rec
Garza-Mercado [24]	26/M	LBA (1 year) spastic weakness R LL and numbness (2 weeks),	L4 (ED)	Tuft of hair, SCM-1, spina bifida, T12 IM epidermoid	CR	Good; NA
Nakayama et al. [25]	51/F	R LL paresis, numbness (14 years)	T12-L3 (IM)	NA	STR	Good; NA
Conti et al. [26]	24/F	BA, R LL weakness, paraesthesia (1 year)	L1-2 (EM)	SCM-2	CR	Good; NA
Ironside et al. [27]	34/F	NA	L2-4 (NA)	Absent	STR	Poor; 8 years-rec with carcinoid tumour
Smoker et al. [28]	26/M	LL weakness (5 days), urinary hesitancy (1 days), sensory disturbance	C7-T4 (IM)	Vertebral non segmentation, spina bifida occulta, scoliosis	STR	Good; 2 months-NA
Giacomini et al. [29]	31/M	BA, sensory disturbances	T12-L1 (IM)	Yes	CR	Good; 1 year-no rec
Monajati et al. [30]	52/M	NA	L3-5 (EM)	Yes	CR	Good-NA
Pelissou-Guyotat et al. [31]	33/M	Sensory and urinary disturbances	L4 (IM)	Spina bifida	STR	Good; NA
Hamabuchi et al. [32]	54/M	Paraparesis, numbness LL, acute BB incontinence and meningitic symptoms	T12-L1 (EM)	Absent	STR	Meningitic symptoms after 6 days, excellent; 12 weeks-NA
Üstün et al. [33]	23/M	Neck pain, spastic quadriparesis (1 week), urinary hesitancy	C4-5 (EM)	Spina bifida, dermal sinus	CR	Excellent; NA
Kaji [34]	79/F	NA	T11-12 (IM)	NA	NA	NA
Nicoletti et al. [35]	47/M	Weakness, numbness LL, neuropathic ulcers, bladder dysfunction (2 years)	T12-L4 (IM)	Yes	STR	Good; 6 months-NA
Eide and Skullerud [36]	45/M	Peripheral sensory, motor neuropathy (20 years)	Conus (IM)	NA	Resection	NA
Caruso et al. [37]	41/M	Pain, numbness LL, bladder disturbance (15 years)	Conus (IM)	Absent	CR	Good; 4 years-NA
Koen et al. [38]	31/F	Worsening of paraparesis; BA, T10-L1 radiculopathy (1 year)	L3-S1 (EM)	Butterfly vertebra, SCM-1	CR	Poor; 1 year-no rec

Table 1 (continued)

Author	Age/sex	Clinical features and duration	Location	Associated features	Surgery	Outcome; duration and rec
Al-Sarraj et al. [29]	43/F	NA	Conus (EM)	Absent	NA	5 years-no rec
	47/F	NA	T10-L1 (EM)	Spina bifida	NA (adenoCa in teratoma)	5 years-no rec
	41/M	NA	Conus (EM)	Absent	NA	2 years-no rec
	32/F	NA	C2-7 (EM)	Spina bifida	NA	6 years-six rec
	23/M	NA	Conus (EM)	Spina bifida	NA	NA
	32/M	NA	Conus (EM)	Absent	NA	9 months-no rec
	35/M	NA	Conus (IM)	Absent	NA	10 years-rec
Poeze et al. [40]	23/M	Acute urinary retention; voiding problems (1 year); foot drop	T12-L1 (IM)	Absent	STR	Good (foot drop persistent); 6 years-no rec
Natarajan [41]	60/M	R thigh myokymia, quadriceps weakness and LBA (10 years),	Conus-cauda (IM + EM)	Absent	Resection	Excellent-NA
Bloch et al. [42]	56/M	NA	T12-L1 (IM)	Absent	CR	Poor; 8 months-NA
Allsopp et al. [43]	64/M	LL weakness and sensory loss, urinary disturbance (10 months)	T11-L1 (EM)	Spina bifida	CR	Fair; 3 years-rec; 2nd Sx-fair (60 months)
	56/F	LBA and radiculopathy, R distal weakness and sensory loss (2 years)	L1-2 (EM)	Spina bifida, tuft of hair	CR	Poor; 12 months-NA
Okuyama et al. [44]	50/F	Pain, focal sensory loss, weakness LL (6 years)	L3-4 (ED, dumb-bell shaped)	Absent	CR + fusion	Good; 3 years-no rec
Arai et al. [45]	54/F	LBA (6 years), LL weakness and numbness (1 year), neurogenic bladder	L2-5 (EM)	Spina bifida, tuft of hair	CR	Good; 6 months-NA
Fan et al. [46]	43/F	LBA with radiation to R LL (several m), patchy sensory loss R LL	L1-2 (IM + EM)	Tuft of hair, small distal syrinx	CR (carcinoid in teratoma)	Excellent; 13 months-no rec
Elmaci et al. [47]	42/M	Pain, L foot weakness, neurogenic claudication (1 month)	L4-5 (EM)	Spina bifida, SCM-1 (L5)	Excision	Fair; (foot weakness persistent); 1 year-NA
Nonomura et al. [48]	33/F	Feet numbness (5 years), LL weakness (1 year),	T12-L1 (IM)	Spina bifida	STR	Fair; 8 years-no rec
	56/M	R LL paresis (2 years) worsened since 1 month, numbness all 4 limbs (2 years)	Multiple (medulla to L2) T12-L2 (IM)	Absent	STR	Poor; NA

Table 1 (continued)

Author	Age/sex	Clinical features and duration	Location	Associated features	Surgery	Outcome; duration and rec
Hejazi and Witzmann [49]	45/F 20/M	LBA (2 years) with LL rad (1 year), paraparesis (8 months), urinary incontinence (4 months) LL radiculopathy, weakness and urinary retention (20 days)	L1-2 (IM) L2-4 (IM)	Absent Absent	CR CR	Excellent; 1 year-no rec Excellent; 3 months-NA
Maiuri et al. [50]	NA	Radicular pain, U/L leg weakness	Cauda equina (EM)	NA	STR	Good; 42 months-NA
Fernández-Cornejo et al. [51]	46/M	CES (4 years)	L1-2 (IM)	Absent	CR	Good, saddle hypesthesia persistent; 9 months-NA
Ates et al. [52]	30/M	LBA, constipation, urinary retention and sexual problems (4 years)	L3-4 (EM)	Thick filum	CR	Excellent; 6 months-no rec
Kao et al. [53]	24/F	LBA with LL radiation	L2-4 (ED with retroperitoneal spread)	NA	CR	Good; 9 years-no rec
Kumar et al. [54]	19/F (pregnant)	BA, myelopathy × 3 months	T5-6 (EM)	Spina bifida	STR (ER, PR + ve)	Excellent; NA
Stevens et al. [55]	85/M	LBA, paraparesis (9 months-prior minor trauma)	L1-2 (EM)	Absent	CR	Excellent; 2 months-no rec
Ak et al. [2]	43/F	Neck pain, numbness, paraparesis (1 year)	C2-3 (IM)	C3 spina bifida, C5 level midline cutaneous mass till dura	CR	Good; NA
Paterakis et al. [56]	62/M	Radiculopathy (3 years) quadriparesis (2 months)	C2-C5 (IM)	Kyphoscoliosis, C3-5 body fusion	STR	Tracheostomy and died after 3 months due to respiratory failure
Kahilogullari et al. [57]	42/F	LBA (1 year), urinary incontinence (1 month)	L1-conus (IM)	Absent	CR	Excellent; no rec
Tsitsopoulos et al. [58]	44/F	L trunk and LL paraesthesiae, left LL weakness and numbness (5 years),	T8-10 (IM + EM)	SCM-1 (L2-3), spina bifida	STR (SCM-not removed)	Good; 15 months-no rec
Caruso and Colonesse [59]	41/F 40/M	Pain, weakness, sensory and urinary disturbances Weakness, numbness LL	Conus (IM) Conus (IM)	Absent Absent	STR STR	Good; 7 years-no rec Good; 6 years-no rec
Makary et al. [60]	46/F	Neck pain (several years), progressive radiculomyelopathy	C1-C2 (IM)	C2 spina bifida, incomplete segmentation, DS, scoliosis	CR	Excellent; 6 months-no rec
Mut et al. [61]	34/F	LBA with LL rad, R LL paresis (6 weeks), urinary retention (recent)	L1-2 (IM)	SCM-2	STR	Excellent; 3 months-no rec

Table 1 (continued)

Author	Age/sex	Clinical features and duration	Location	Associated features	Surgery	Outcome; duration and rec
Sung et al. [62]	38/M	BA (8 years), urinary incontinence (2 months)	T11-L1 (EM)	Absent	STR	Excellent; 2 weeks-NA
Mohindra et al. [63]	35/M	BB problems (2 years)	L4-5 (IM + EM)	Absent	CR	Excellent; 1 year-NA
Han et al. [64]	35/F	LBA, leg pain, weakness, sensory changes, BB dysfunction (22 weeks)	T12-L2 (IM)	Absent	STR	Good, 21 months-no rec
Benes et al. [65]	52/F	LBA, weakness (4 months)	L2-L5 (IM)	Tuft of hair	STR	Good; 12 months-NA
Ijiri et al. [66]	68/F	BA, LL numbness (4 months); urinary dysfunction	L1-2 (EM)	Absent	CR (ossified)	Transient deterioration, excellent-NA
Ghostine et al. [67]	65/F	Dizziness, ataxia (1 year), B/L hand tremors	C1-C2 (IM)	Absent	STR	Good; 3 months-NA
Gu et al. [68]	23/M	LBA (2 years), LL rad (2 weeks)	L2-3 (EM)	Absent	STR	Excellent; 4 months-NA
Arvin et al. [69]	34/M	Transient Lt finger numbness (8 years back), rec of numbness and radiculopathy	C4-6 (IM)	Spina bifida	CR	Excellent; 2 years-no rec
Jeong et al. [70]	56/M	R leg weakness, numbness, and voiding problems (14 years)	L2-3 (EM)	Spina bifida	CR	Good; no rec
Oh et al. [71]	44/M	Urinary problems, LL weakness	L3-5 (IM)	Absent	STR	Intracranial fat dissemination, hydrocephalus-Died
Sharma et al. [1]	51/F 31/M 28/M 30/M 32/M 45/F 32/M 30/M	BA, numbness and weakness LL, bladder dysfunction (20 years) Numbness and gait unsteadiness (5 months) Pain both LL (1.5 years) LBA, bladder dysfunction (3 years) Pain, weakness LL, urinary incontinence (2 years) Weakness, sensory changes, bladder dysfunction (6 years) Radiating BA, bladder dysfunction (2.5 years) Left hemiparesis and sensory loss (3 years)	D10-L2 (IM) D11 (IM) L4-5 (EM) L1-3 (IM) D10 (IM) D1-3 (EM) D11 (IM) C2 (EM)	Spinal bifida, lipo-MMC, tethered cord SCM-1 Tuft of hair SCM-1 SCM-2 Tethered cord Absent Spina bifida	NA (CR to STR)	Exact details NA, 1 rec after 9 years

Table 1 (continued)

Author	Age/sex	Clinical features and duration	Location	Associated features	Surgery	Outcome; duration and rec
Moon et al. [72]	35/M	Quadriparesis, numbness, urinary dysfunction (3 weeks)	C6-7 (IM)	Absent	NTE (Immature)	Poor; 4 months-MRI rec, 8 months-clinical rec
Agrawal et al. [73]	45/F 34/F 35/M	C6 paraesthesia C6 radiculopathy and weakness LBA, BB dysfunction	C4-6 (IM + EM) C6 (EM) L2-3 (EM)	Absent Absent Absent	STR CR CR + RT (malignant) transformation)	Good; 1 year-NA Excellent; 2 years-NA Excellent; 8 months-no rec
Jian et al. [74]	18/M 57/M	LBA (1 month), dysuria (3 days) LBA (3 months), weakness (2 months)	L2-4 (IM) L1-2 (IM)	Absent Absent	CR CR	Excellent; 2 months-NA Incomplete; 2 months-NA
Yu et al. [75]	34/M	LBA, LL weakness and numbness (2 months), sexual disturbances (2 weeks)	Conus (IM)	Adjacent AVM-NA	STR	Good recovery but sexual disturbance persistent; 2 months-NA
Maiti et al. [76]	18/M	BA, urinary disturbances (1 year), paraesthesia LL (5 months)	L2-3 (IM)	L1 SCM-1, low conus	STR	Good but bladder problem persisted; 6 months-NA
Conti et al. [77]	38/F	BA with R LL radiation, numbness in R trunk, abdomen (5 years)	L1-2 (IM)	Tuft of hair, kyphoscoliosis, SCM-2	CR	Excellent; 2 months-NA
Bouaziz et al. [78]	38/F	Quadriparesis (2 months-prior trauma)	CMJ (EM)	Absent	CR	CSF leak, meningitis, Died
Musil et al. [79]	60/F	LL weakness, hypesthesia	Conus (IM)	Absent	STR	Fair; 12 months-3 surgeries, rec; *Staph. aureus* growth in 1st surgery specimen
Ben Nsir et al. [80]	70/F	Radiculopathy (2 months)	T12-L3 (IM)	Absent	STR	Excellent; 3 years-no rec
Garg et al. [81]	32/M	HA, neck pain, vomiting, fever (3 days), multiple cranial nerve palsies, neck rigidity (intracranial dissemination with chemical meningitis)	L5-S3 (EM)	Absent	Excision (after 1 month)	NA
Kalani et al. [82]	18/M	Rapid LL weakness	T10 (IM)	Absent	2 stages-STR f/b NTE	Good; 3 months-no rec
Li et al. [3]	22/F	LBA, R LL weakness (3 months), radiation to R LL (2 months)	T12-L2 (IM)	Absent	CR	Excellent; 3 months-NA
Jiang et al. [83]	56/M	BA, radiculopathy (1 year), foot weakness (2 months)	L4-S1 (EM)	Kyphoscoliosis	CR	Excellent; 34 months-NA

Table 1 (continued)

Author	Age/sex	Clinical features and duration	Location	Associated features	Surgery	Outcome; duration and rec
Vanguardia et al. [84]	41/M	CES	T12-L1 (EM)	Absent	STR	Good (occasional faecal incontinence); 1 year-no rec
Babu et al. [85]	66/F	Urinary dysfunction (7–8 years, worsened few months), LL stiffness and weakness (1 year), BA (2 months)	L3-4 (EM)	SCM-2	CR	Excellent (urinary problem too improved); 1 year-no rec
Pandey et al. [86]	30/M	Numbness LL (6 months), paraparesis (2 months)	L4 (EM)	Absent	CR	Excellent; 3 months-NA
Alkherayf et al. [87]	50/M	BA, radiculopathy, leg numbness (2 years), foot weakness (5 weeks)	L1-L2 (IM)	Absent	STR	NA
Gubney et al. [88]	62/M	Paraparesis	L4-5 (EM)	SCM-2	NA	NA
Yilmaz et al. [89]	49/F	Paraparesis, LBA	L1-L5 (IM + EM + ED)	Operated MMC at birth	STR	Good-NA
Oh et al. [90]	67/M	Nocturia, urinary retention, paraparesis (2 months)	Conus (IM)	Absent	Resection	Died-Intracranial fat dissemination, Sepsis, LL power did not improve
Turan et al. [91]	48/M	Lt EHL weakness, LL fasciculations (2 months) f/b (1 year) left foot drop, patchy l/l sensory loss	L1-3 (IM)	Absent	CR	Excellent; 9 months-no rec
Asan et al. [92]	29/F	LBA (4 months)	T12-L1 (IM + EM)	Absent	NTE	Excellent; 8 months-NA
Agay et al. [93]	20/M	BA, numbness, urinary incontinence (1 year)	D11-12 (EM)	Absent	CR	Excellent; NA
Ariñez Barahona et al. [94]	54/M	CES (10 months)	L2-3 (IM)	Absent	CR	NA
Khazender et al. [95]	37/M	LBA (5 years increasing × 3 months) and sexual dysfunction, numbness (3 months)	L1-2 (EM)	Absent	CR	Poor (faecal and urinary incontinence); 1 year-no rec
Wang et al. [96]	30/M 24/M 21/M 27/F 22/F 56/M 24/F	Exact details NA	L3-4 (EM) L2-3 (EM) L4-5 (IM) L2-3 (EM) L3-4 (EM) T12-L1 (EM) T12-L1 (EM)	Cyst Absent LLTC Absent Absent Spina bifida Absent	STR STR STR CR STR STR CR	NA
Scmidt et al. [97]	49/M	Urinary dysfunction (8 months)	L1-L2 (EM)	Absent	STR	Fair; 2 years-no rec

Table 1 (continued)

Author	Age/sex	Clinical features and duration	Location	Associated features	Surgery	Outcome; duration and rec
Wan et al. [98]	35/M	Numbness, weakness, BB dysfunction (6 months)	T10-12 (EM)	Absent	STR + PF	Good; 208 months-rec
	30/M		L3-4 (EM)	Absent	STR	(180 months)
	60/M	Weakness (36 months)	L1-5 (EM)	Absent	CR + PF	Good; 188 months-rec
	27/F	Pain, numbness, weakness, sexual and BB dysfunction (12 months)	L2-S1 (EM)	Absent	CR + PF	(168 months)
	48/M		L2-3 (EM)	Absent	CR + PF	Fair; 117 months-no rec
	31/F		L3 (EM)	Absent	CR	Good; 108 months-no rec
	26/M	Pain, numbness, bowel dysfunction (108 months)	L1-2 (IM)	TCS	STR + PF	Good; 101 months-no rec
	18/F		T12-L3 (EM)	Absent	CR + PF	Excellent; 92 months-no rec
	43/F	Weakness, dysuria (108 months)	L3-4 (EM)	Absent	CR	Fair; 94 months-no rec
	67/F		L2 (EM)	Absent	CR	Excellent; 84 months-no rec
	24/F	Pain, numbness (0.5 months)	T12-L2 (IM)	Spina bifida	CR + PF	Excellent; 62 months-no rec
	26/F		L1-2 (EM)	Absent	CR	Good; 59 months-no rec
	19/M	Weakness, dysuria, sexual dysfunction (3 months)	L1-2 (EM)	Absent	CR + PF	Excellent; 55 months-no rec
	32/F		L2-3 (IM)	TCS	STR + PF	Excellent; 50 months-no rec
	48/F	Pain (2 months)	L2-3 (EM)	Absent	CR + PF	Excellent; 47 months-no rec
	58/M	Pain, numbness, weakness, BB dysfunction (7 months)	L4-S1 (EM)	Absent	CR + PF	Poor; 40 months-no rec
	34/F		L1-2 (IM)	Absent	CR + PF	Excellent; 39 months-no rec
	50/M		L1-2 (EM)	Absent	CR + PF	Excellent; 33 months-no rec
		Pain, weakness, BB dysfunction (360 months)				Good; 30 months-no rec
		Pain (8 months)				Good; 27 months-no rec
		Pain, numbness, bowel dysfunction (3 months)				
		Numbness, weakness, dysuria (18 months)				
		Pain, numbness, weakness (120 months)				
		Pain, numbness (84 months)				
		Pain (2 months)				
		Numbness, weakness, bowel dysfunction (24 months)				
		Pain, weakness, BB dysfunction (0.5 months)				
Mohammadi et al. [99]	18/M	LBA	L2-3 (IM)	Absent	NTE	Good-NA
Lekgabe et al. [100]	29/F (PP)	Urinary retention	Lumbar (IM)	NA	Resection	Excellent-complete recovery
Present case (2018)	32/M	Paraplegia, numbness, urinary retention (2 days)	L2-3 (IM + EM)	Absent	STR	Fair; 15 months-NA

NA not available, *CE* complete resection, *y* years, *IM* intramedullary, *STR* subtotal resection, *BA* backache, *BB* bowel and bladder, *m* months, *rec* recurrence, *EM* extramedullary, *LL* lower limbs, *Nap* not applicable, *ED* extradural, *L* left, *SCM* split cord malformation, *LBA* low backache, *R* right, *d* days, *Ca* carcinoma, *Sx* surgery, *U/L* unilateral, *CES* cauda equina syndrome, *ER o*estrogen receptor, *PR* progesterone receptor, *+ve* positive, *DS* dermal sinus, *B/L* bilateral, *MMC* myelomeningocele, *NTR* near total resection, *RT* radiotherapy, *Staph* Staphylococcus, *LLTC* low lying tethered cord, *PP* post partum

without haemorrhage are the major radiological clues for a ST on MRI. Contrast enhancement is not a typical feature of mature teratomas; however, they may show marginal rim enhancement or a nodular enhancement [43, 46, 47, 54, 56, 66, 67, 69, 70, 72, 73, 78, 80, 82, 86–88, 90, 92, 100, 102].

In the present review, majority were heterogeneous lesions, but a few homogeneous lesions have also been reported [49, 66, 69, 78, 81–83, 86, 93, 99]. Characteristically, these lesions appear hyperintense on T1 sequences indicating the presence of fat. However, lesions that are predominantly T1 hypointense and isointense (with peripheral fat in a few cases) have also been reported [2, 28, 33, 45, 48, 54, 63, 65–67, 69, 70, 76, 78, 83, 86, 87, 89, 91, 93, 96, 100]. Associated congenital abnormalities may or may not be evident [60, 72, 73, 102, 108]. In our case, the lesion had similar characteristics that pointed towards a possible diagnosis of a teratoma preoperatively. Intradural teratomas are commonly oval or lobulated heterogeneous masses in contrast to the ED teratomas that are more often dumb-bell shaped [3, 104]. Although Sharma and coworkers and Moon and associates pointed out that STs frequently co-exist with dysraphic congenital spinal malformations, most authors have noted that simultaneous presentation of occult spinal dysraphic lesions and a ST appear to be a rare phenomenon in adults [1, 3, 47, 49, 72, 109]. The associated congenital anomalies may include spina bifida (SB), myelomeningocele (MMC), dermal sinus (DS) and split cord malformations (SCM) [102]. These anomalies are mostly associated with IM tumours, and the presence of such congenital abnormalities should raise the suspicion of a teratoma [73, 102]. In the present analysis, these anomalies were present in 55 cases (38%). The occurrence of ST with SCM is extremely rare, especially in adult population, and the split cord might exist at a remote location from the tumour. Hence, in such cases, it has been suggested that the entire spine need to be imaged because of the possibility of dysraphic lesions being present at non-contiguous sites [3, 47, 58]. There were 14 (9.6%) cases of associated SCM (7 cases each of types I and II) noted in the present review.

Embryology and pathogenesis

Several theories have been proposed to account for the embryogenesis of these tumours. Kubie and Fulton [6] believed this tumour to be an ependymal diverticulum. Ugarte and associates proposed that persistence of neurenteric canal resulted in a teratoma [107]. Bucy and Buchanan supported the theory of germinal cell aberration [32, 110]. However, none of the above theories are accepted now. According to the dysembryogenic theory, a teratoma is formed by chaotic differentiation of pluripotent cells in locally disturbed developmental environment in primitive streak or caudal cell mass [1, 3, 73, 82, 104]. The misplaced germ cell theory suggests that certain pluripotent primordial germ cells of the neural tube are misplaced during migration from the yolk sac to the gonad, thus resulting in a teratoma formation [1, 16, 73, 82, 104]. The presence of Barr bodies in teratomas in male patients has been suggested as an evidence of germ cell origin of such tumours [38]. This theory is more likely to be feasible in adult intraspinal teratomas due to the absence of significant dysraphism [3, 104]. However, despite all these theories, the exact pathogenesis of STs remains unclear.

Histopathology

Teratomas are histologically characterized by the presence of tissues derived from all the three germ layers [1, 48, 82]. Although, the presence of derivatives of two germ layers does not exclude the lesion as a teratoma, they are more aptly termed as teratoid tumours [38, 49, 102]. Walter and Kleinert classified teratomas into mature, immature and malignant based on the degrees of differentiation [1, 2, 39, 111]. Mature teratomas are composed exclusively of the fully differentiated 'adult type' tissue elements such as mature cartilage, squamous epithelium, skin appendages, columnar mucosa and nervous tissue and are considered as benign forms. Immature teratomas are aggressive tumours having primitive, undifferentiated components resembling ''foetal'' tissues and tend to recur frequently. Malignant teratomas are derived from the yolk sac or endodermal sinus and are associated with high levels of serum alpha-fetoprotein (AFP) and are associated with very poor prognosis [1, 40, 66, 73, 105, 112]. Histology of a ST shows mature type in most cases, while immature type STs are rarely reported in adults. Nephrogenic and pulmonary differentiations and carcinoid tumour arising in teratomas are very rare [1, 46]. In our present review, all except one were mature teratomas on histopathology. These teratomas may be hormonally responsive as noted in one report, where a sudden worsening of symptoms was noted during pregnancy [54]. Furthermore, because of the heterogeneity of the tissues, few authors have opined that the entire specimen should be thoroughly sampled to look for the components of a teratoma and other immature elements. This is because of the reason that these components may not be evident in the initial sections and are likely to go undetected which might have important prognostic values [1].

Treatment and outcome

Surgical resection is the treatment of choice for STs, and early surgery is recommended before irreversible neurological damage ensues. Complete resection should be aimed on all occasions and whenever possible, they should be excised with the capsule intact in order to reduce the

risk of postoperative complications such as meningitis or myelitis [1, 40, 48, 60, 72]. This is generally possible in EM tumours; however, it might not be feasible in an IM tumours. In such cases, STR with preservation of functioning neural tissue seems to be a feasible option [1, 2, 48, 49, 58, 72]. Intraoperative use of electrophysiological monitoring may help to achieve greater tumour resection with preservation of neural function. In one of the previous reviews, IM tumours could be completely removed in 61.8% of the reported cases and improvement of symptoms occurred in 45.5% of them [40]. However, in this present review, CR was noted in only 37.6% cases of IM teratomas while STR was reported in 46.3% cases. It was noted that 66% of EM tumours underwent CR while 55% of IM tumours had STR, and this difference was statistically significant. Recurrences are uncommon and are mainly seen in the immature forms [40]. Because of the slow growth of these tumours, symptomatic recurrences usually occur after very long periods despite STR [49, 54]. However, few authors have noted STR not to be associated with poor outcomes [1, 49]. Hejazi et al. reported no difference in recurrence rates with CR versus STR (9% and 11%, respectively) at a mean follow-up of 25 months (43, 49). However, as noted in our results section, the overall recurrence rate was 6.1% ($n=9$) at a mean follow-up of 37 months. Subtotal resection and CR were performed for 54 (37%) and 65 (45%) cases. There were one and five recurrences in CR and STR, respectively, and this difference was statistically significant ($p=0.029$). Also, no malignant changes were evident in the recurred cases.

Several serum markers such as β-human chorionic gonadotropin (β-hCG) and AFP may be monitored for detection of recurrences of sacrococcygeal teratomas; however, this is limited in STs, as recurrences may originate from the non-secreting parts of the tumours [1, 3, 43]. Recurrences may be seen after a prolonged latency with one report documenting recurrence as late as 96 months after surgery, thus highlighting the extremely slow growth of these tumours [43]. In this present review, the mean TTR was 98.8 months (12–180 months) for mature teratomas. Hence, long-term clinical and imaging follow-up is recommended, especially after STR. Re-exploration and excision is advisable for symptomatic recurrent cases [1, 43, 49, 72]. Outcome is good to excellent in majority of cases [1, 39]. Immature and malignant forms are uniformly associated with poor outcomes and high recurrence rates. Poeze and associates pointed out that in benign teratomas, 26.3% died during a mean FU period of 38 months, while patients with malignant or immature IM teratomas died within 1 year after surgery [40]. As noted in the results section, there were four deaths reported and one case was diagnosed at autopsy [15, 56, 71, 78, 90]. Adjuvant radiotherapy (RT) is of no utility even in recurrent tumours, and it should be reserved only for teratomas with an immature or malignant component [1, 43, 49].

Limitations

Although this paper is the most extensive review till date on adult onset STs and including even those cases reported in non-English literature, cases reported in other languages are surely to be missed due to indexing problems and technical difficulties in accessing those articles.

Conclusions

This present paper provides a very comprehensive and in-depth analysis of all the adult onset spinal teratomas reported till date and analyses all the parameters possibly related to outcomes and recurrences. Spinal teratomas are uncommon neoplasms and are rarely encountered in the adult population. Majority of them are located in the thoracolumbar region as opposed to sacrococcygeal location in neonates and young children. Total surgical resection is the treatment of choice; however, this might not be possible in intramedullary cases for which subtotal resection might be a good option. Recurrences are uncommon and are mainly seen in immature teratomas and subtotal resections. Outcomes are good/excellent in over two-thirds of cases. Results of this paper show that the presence of pain, absence of limb weakness and complete tumour resection were significantly associated with good outcomes. Furthermore, subtotal resections had significantly higher recurrence rates than complete resections. Adjuvant radiotherapy is of no utility even in recurrent tumours, and hence, it should be reserved only for teratomas with immature or malignant components. This paper also highlights the importance of performing surgery even in those with late clinical presentations and complete neurological deficits, especially if an extramedullary component is present.

Compliance with ethical standards

Conflict of interest The authors report no declarations of interest

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Affiliations

G. Lakshmi Prasad[1] · S. Divya[2]

[1] Department of Neurosurgery, Kasturba Medical College, Manipal, Manipal Academy of Higher Education, Manipal, India

[2] Department of Orthodontics, Manipal College of Dental Sciences, Manipal, Manipal Academy of Higher Education, Manipal, India