Case Report

Metastatic epidural spinal cord compression from testicular yolk sac tumor: case report and literature review

Christine Anne T. Galang[1]^, Nicholas Hernandez[2], Courtney S. Lewis[3], Martin H. Pham[2]

[1]University of Hawaii John A Burns School of Medicine, Honolulu, HI, USA; [2]Department of Neurosurgery, University of California San Diego School of Medicine, San Diego, CA, USA; [3]Touro College of Osteopathic Medicine, Harlem, NY, USA

Contributions: (I) Conception and design: All authors; (II) Administrative support: MH Pham; (III) Provision of study materials or patients: MH Pham, N Hernandez; (IV) Collection and assembly of data: CAT Galang, N Hernandez, CS Lewis; (V) Data analysis and interpretation: CAT Galang, N Hernandez, CS Lewis; (VI) Manuscript writing: All authors; (VII) Final approval of manuscript: All authors.

Correspondence to: Martin H. Pham, MD. Department of Neurosurgery, UC San Diego Health, 9300 Campus Point Drive, MC7893, La Jolla, CA 92037, USA. Email: mhpham@health.ucsd.edu.

Background: Yolk sac tumor (YST), or endodermal sinus tumor, is classically associated with pediatric populations. Metastasis to the spine rarely occurs, usually involving the lower thoracic or lumbar vertebrae. The objective of this report is to present a rare case of YST metastasis to the lower cervical and upper thoracic vertebrae in an adult male. A case-based review of the literature on metastatic YSTs was also performed as an update to the relevant literature.

Case Description: A 28-year-old male with a history of YST presented to our institution with urinary retention, increasing weakness in the upper extremities, and acute onset lower extremity weakness. Computed tomography (CT) and magnetic resonance imaging (MRI) scans confirmed evidence of metastasis from a known YST with symptomatic cord compression. The patient was treated with surgical excision via decompressive laminectomies with instrumentation as described, and histopathologic analysis of the specimen confirmed YST metastasis. His disease recurred one year after index surgery. He succumbed to his disease despite repeated debulking.

Conclusions: Metastasis of YST is rare, but metastasis to lower cervical and upper thoracic vertebrae is possible. YSTs are usually treated via primary surgical resection. Systemic chemotherapy and radiation may prevent recurrence. However, individualized treatment is imperative for improved patient outcomes.

Keywords: Cord compression; germ cell tumor; yolk sac tumor (YST); spinal metastasis; case report

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Introduction

Yolk sac tumor (YST), or endodermal sinus tumor, is a malignant nonseminomatous germ cell neoplasm that primarily originates from primordial gonocytes in the gonads that undergo extraembryonic differentiation (1,2). YST are classically associated with pediatric patients. YST presents in males with a bimodal distribution, under three years old and post-puberty, while the median age for females is 19 years old (3). The first case of metastatic YST in the literature is reported in a 35-year-old female in 1957 (4). While testicular malignancies make up less than 1% of male pediatric cancers, YST is the most common of pediatric testicular malignancies (5). Conversely, testicular germ cell tumors (T-GCTs) make up 14% of all neoplasms

^ ORCID: 0000-0001-8004-3921.

in males 15–29 years old (6).

YST in adults is rare, comprising <20 cases reported in the literature (7-16). Extragonadal metastasis of YST is even more uncommon. Furthermore, metastasis to the spine is seen in less than 1% of pediatric cases (17). In adults, spinal metastasis accounts for 1–5% of YST invasive disease (1,18,19). Spinal metastasis, when present, is generally encountered in the lower thoracic to lumbar spine (8,9,12-14).

Here we present a case of an adult male with YST metastasis to the upper thoracic spinal epidural space, with spinal cord compression and myelopathy, in accordance with the CARE reporting checklist (available at https://jss.amegroups.com/article/view/10.21037/jss-24-28/rc).

Case presentation

History

A 28-year-old male was originally diagnosed in 2013, initially presenting with debilitating low back pain. Computed tomography (CT) imaging revealed a 12 cm retroperitoneal pelvic mass and left supraclavicular adenopathy, concerning for a nonseminomatous germ cell tumor. At the time of diagnosis, measured beta-human chorionic gonadotropin (b-HCG) was 445 mIU/mL, alpha fetoprotein (AFP) was 513 ng/mL, and lactate dehydrogenase (LDH) was 5,126 U/L. Left radical orchiectomy with partial retroperitoneal lymph node dissection was completed five months after completion of four cycles of neoadjuvant bleomycin, etoposide, and platinum (BEP) chemotherapy, and five months after initial diagnosis. Despite treatment, the patient's subsequent CT imaging showed evidence of progressive metastasis to the left low neck in 2019 (B-HCG <1 mIU/mL, AFP 305.2 ng/mL, LDH 177 U/L) and the patient therefore underwent three cycles of paclitaxel plus ifosfamide followed by high-dose carboplatin plus etoposide (TI-CE) chemotherapy. The patient achieved stable remission for several years, though in 2021, patient developed urinary retention, increasing weakness in the upper extremities, and acute onset lower extremity weakness at which time serum markers were noted B-HCG <1 mIU/mL, AFP 42,889 ng/mL, and LDH 159 U/L. The patient was referred to our institution's neurosurgery clinic for further evaluation and consultation.

Highlight box

Key findings

- We present the case of a yolk sac tumor metastasis to the lower cervical or upper thoracic region in an adult after left radical orchiectomy and chemotherapy with autologous stem cell transplants.

What is known and what is new?

- Yolk sac tumors have metastasized to the spine in previous literature, but metastasis commonly occurs in the lower lumbar region.
- This case illustrates an example of yolk sac tumor metastasis to the lower cervical or upper thoracic region of the spinal cord, resulting in cervical myelopathy.

What is the implication, and what should change now?

- Though yolk sac tumor metastasis is rare, surgeons should be aware of the possibility if a patient returns with urinary retention and upper extremity weakness.

Examination

On examination, cranial nerve function was intact. Motor strength in the left upper extremity was grade 4/5 for deltoid, 4-/5 for biceps, 3/5 for triceps, and 1/5 for wrist extension, wrist flexion, and the interossei muscles. Motor strength in the right upper extremity was grade 4+/5 for deltoid, 5/5 for the biceps, triceps, wrist extension, wrist flexion, and interossei muscles. Motor strength in the left and right low extremities was identical bilaterally, with 4-/5 in quadriceps, hamstring, extensor hallucis longus, and plantar flexion, and 2/5 in iliopsoas muscles. Sensation was diminished bilaterally in fourth and fifth fingers, right medial thigh, and lateral plantar surfaces. The patient had intact passive rectal tone but diminished active rectal tone. Cervical myelopathy was suspected from spinal cord compression.

Imaging

A T2-weighted and T1-weighted magnetic resonance imaging (MRI) of the cervical spine revealed cervical stenosis extending from C3–C7 (*Figure 1*). There was T2 hyperintensity within the C6 and C7 vertebral bodies and signal change within the cord spanning from C6 and C7. The C7 vertebra was significantly affected with 25–50% loss of height, with both anterior and posterior protrusion of bony and soft tissue material causing displacement of the anterior longitudinal ligament anteriorly, and cord compression posteriorly. There was also widening of the interspinous spaces from C6–T1 indicative of mass effect from the rapidly growing tumor mass (*Figure 1*).

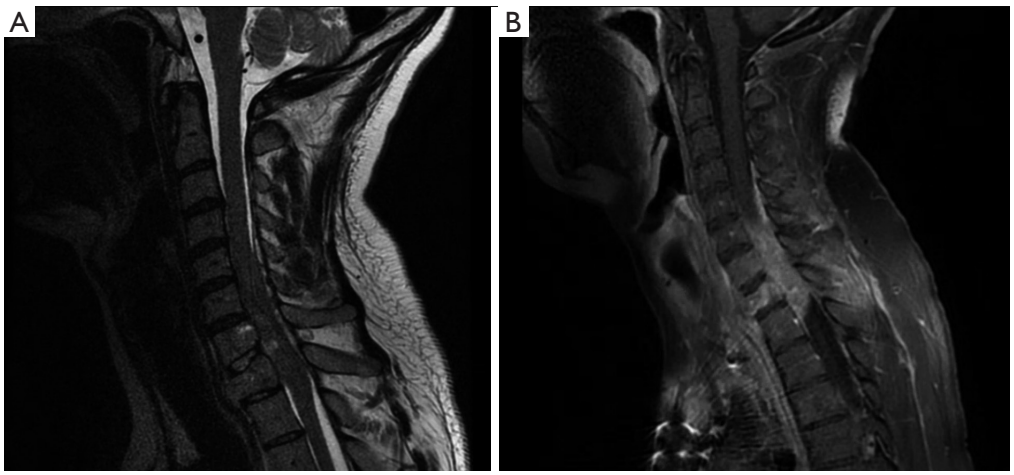

Figure 1 Pre-operative T1- and T2-weighted MRI. (A) T2-weighted MRI demonstrates a mass extending from C3–T1 exerting severe cord compression; (B) T1-weighted MRI demonstrates a mass extending from C3–T1 with hyperintensity. MRI, magnetic resonance imaging.

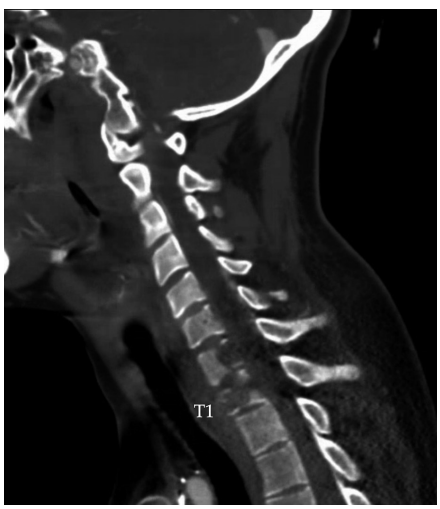

Figure 2 CT of the cervical spine. Cervical spine CT demonstrates lytic lesions affecting the C7 and T1 vertebral bodies. CT, computed tomography.

A pathologic fracture of the T1 vertebra was noted, with ~25% loss of high, and was without significant bony retropulsion.

CT of the cervical spine showed lytic lesions of the C6–T1 vertebrae, predominantly affecting C7 and T1. These lesions affecting the cervico-thoracic junction induced mild kyphosis at C6–C7 and reduced overall cervical lordosis (*Figure 2*).

Chest CT demonstrated a large right chest wall heterogeneous mass measuring 1.5 cm × 1.45 cm, occupying the majority of right hemithorax and exerting considerable mass effect and compression on the trachea, atelectatic right lung, right mainstem bronchi, right lobar bronchi, right pulmonary artery, and the left atrium.

Operation

The patient underwent an open reduction and internal fixation of T1 pathologic fracture, decompressive laminectomies from C5–T2, and extradural resection of the metastatic tumor from C5–T2 (*Figures 3*,*4*). Left sided T1 transpedicular partial corpectomy for ventral decompression of the spinal cord was performed to address the extensive compression of the left side. The patient subsequently underwent a C5–T4 fusion using 5.5 cobalt chrome rods, allograft Morselized bone, and demineralized bone matrix. Spinal nerve root monitoring was conducted throughout the procedure, using somatosensory evoked potentials and motor evoked potentials. There was no evidence of nerve root damage at all spinal levels bilaterally through surgical resection and primary fusion.

Pathologic findings

Histopathologic sections showed fibrovascular and adipose connective tissue invaded by solid sheets and lobules of malignant polygonal epithelioid cells with ample amphiphilic cytoplasm. Large rounded vesicular nuclei containing conspicuous, often multiple, eosinophilic nucleoli were present, with abundant mitotic spindles.

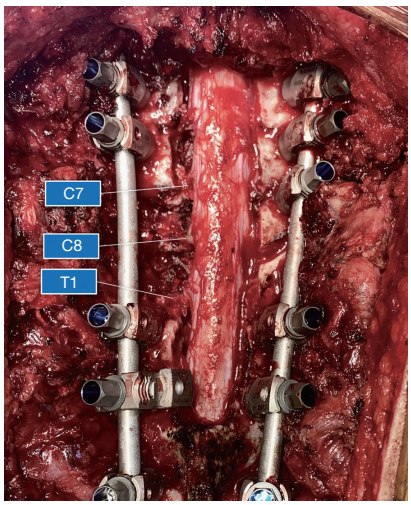

Figure 3 Intraoperative imaging demonstrating the described decompression and fusion. Intraoperative imaging indicating C5–T2 decompression laminectomies, and primary fusion construct extending from C5–T4.

Eosinophilic hyaline globules were spread through the sheets of tumor cells. The neoplasm stained positive for glypican-3 (Glyp-3) and spalt-like transcription factor 4 (SALL4), and negative for CD30 and CD117. Positive detection of Glyp-3 and SALL4 with negative test for CD30 served as diagnostic confirmation of a YST (20,21).

Outcome

His immediate postoperative course was uncomplicated, and he was discharged on postoperative day 5. There were no wound complications, and he was treated with a post-operative regimen of paclitaxel, ifosfamide, and cisplatin (TIP) at which time AFP levels were measured >60,500 ng/mL. Unfortunately, there was recurrence at the operative site 1 year after this index surgery and despite repeated debulking, the patient expired shortly thereafter.

Ethical statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent for publication of this case report and accompanying images was not obtained from the patient or the relatives after all possible attempts were made.

Discussion

YST, also called endodermal sinus tumors, are malignant germ cell neoplasms that typically originate in the gonads, testis, or ovaries. YSTs are the seventh most common pediatric neoplasm occurring in the gonads of young children and are extremely rare in adults (22,23). A suggested reliable marker to diagnose YSTs is AFP, though some studies have shown YSTs can occur with normal AFP levels (24,25). Schiller-Duval bodies and tumor tissue arranged in a sinusoidal pattern are common histopathological findings. Possible pathogenesis mechanisms include activation of germ cells at the midline during early embryological days and distribution of germ cells to other organs after this embryonic period (18,26). While YSTs are usually confined to the gonads, hematogenous metastasis can commonly occur in the lungs, retroperitoneal lymph nodes, liver, and bone (23,27).

Spinal metastasis of YSTs is rare. Previous studies have shown that testicular tumors may metastasize to the spine from the retroperitoneal lymph nodes (8,28). Another possible mode of metastasis of pelvic or testicular tumors to vertebrae is via Batson's venous plexus, which is a valveless vertebral venous plexus system that communicates between the pelvis and vertebral bodies (28,29). Batson's venous plexus was previously shown to play a role in various metastatic diseases like colorectal carcinoma (28). Once YSTs metastasize to the spine via lymphatic or hematogenous routes, spinal cord compression may occur (11-13,17,30). Cases describing metastatic YSTs with spinal involvement, or spinal cord compression are summarized (*Table 1*).

The most common location of spinal cord compression in the literature is the lower thoracolumbar region (T9–T12) likely due to the location of YSTs and spread via Batson's plexus. Interestingly, cervical or upper thoracic involvement is less common (9,12,16). One study reported a metastatic YST in an infant, leading to a cystic mass and swelling of the neck without cervical or upper thoracic involvement (16). While YSTs are a malignancy most common in pediatric populations, a few cases have discussed metastatic YSTs with spinal cord compression in adults (10,13,14). There have been few reports of sacral involvement (9,12). We have been unable to find any previously published literature on testicular YSTs in an adult, with metastasis to or compression of the cervical or high thoracic spine.

Pathogenetic and pathophysiologic mechanisms of

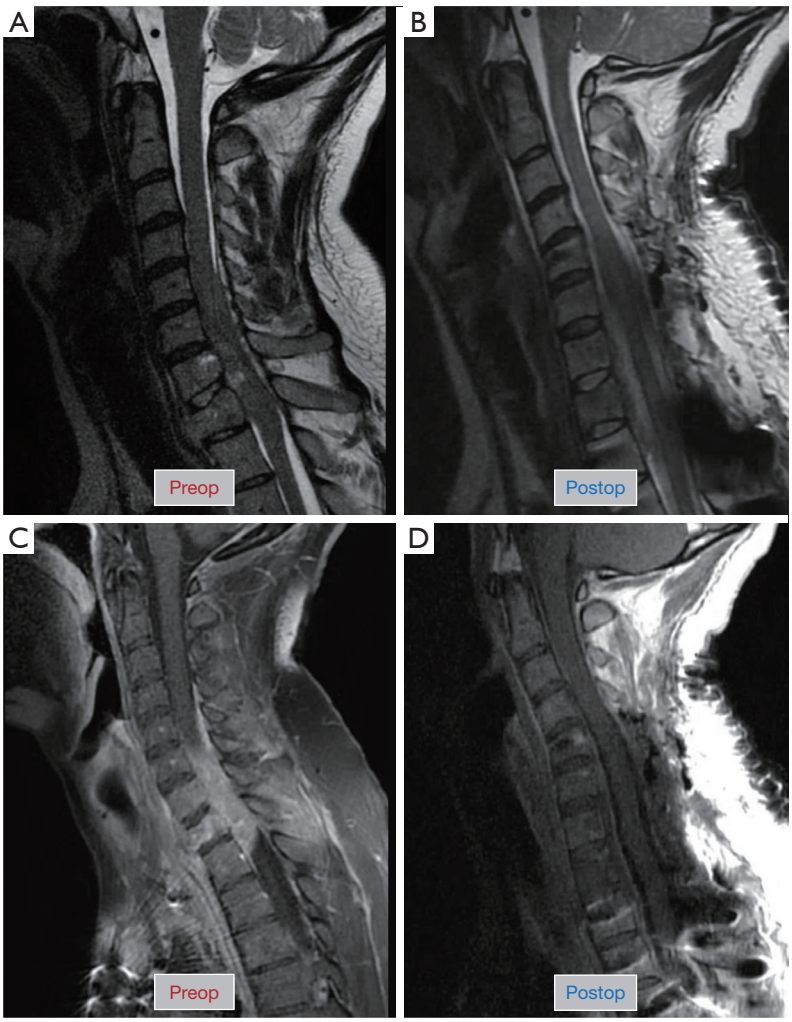

Figure 4 Comparative pre- and post-surgical MRI. (A) Preoperative T2-weighted MRI demonstrating metastatic tumor and cord compression; (B) postoperative T2-weight MRI demonstrating decompression and appropriate widening of the spinal canal. (C) T1-weighted MRI redemonstrating hyperintense mass occupying the canal and exerting significant compression; (D) post-operative T1-weighted MRI again demonstrating tumor resection and decompression. MRI, magnetic resonance imaging.

abdominal or pelvic tumor metastasis to the spine are unclear since YSTs are uncommon. Moreover, metastasis to cervical and upper thoracic spine is rare (31). Previous studies found adenopathy in the left supraclavicular lymph nodes, or Virchow's nodes, are attributed to abdominal or pelvic primary cancers. Vertebral lymphatic vessels may bridge between the left supraclavicular lymph nodes and prevertebral lymph nodes (32,33). YSTs, originating in the pelvis, may metastasize via the lymphatic system and manifest as lymphadenopathy. Patients with known metastasis to these lymph node basins who present with symptoms of spinal cord compression should suspect metastasis via Batson's venous plexus lymphatic drainage mechanisms.

Treatment of YSTs is individualized due to the rarity of the disease. Some case reports highlight radiotherapy as a curative option (34). Other studies show that YSTs are resistant to radiotherapy (35). The nomogram developed by Li *et al.* demonstrated that metastatic YST is highly resistant to postoperative radiotherapy, and that enrollment in postoperative chemotherapy is the main risk factor affecting the prognosis of patients with treatment resistant metastatic YST. In the present case, the patient had disease recurrence despite several surgical debulking procedures

Table 1 Literature summary for metastatic yolk sac tumors

Author, year	Patient age	Gender	Size (cm) and location of tumor (segment if applicable)	Presenting symptoms	Preoperative alphafeto protein	Treatment	Outcome
Colak, 1991	4 y	Male	T11–L1	Progressive paraparesis, neurogenic bladder	140 ng/mL	Neoadjuvant chemotherapy and radiation; laminectomy; adjuvant chemotherapy and radiation	Inguinal lymphadenopathies, paraparesis and neurogenic bladder resolved
Lee, 2002	14 months	Male	T9–T11, S3	Progressive paraparesis, neurogenic bladder	7,369 ng/mL	Hemilaminectomy; declined adjuvant chemotherapy	Partial resolution of neurogenic bladder; discharged on postop day 12
Guzel, 2007	31 y	Male	22×17×15, L1–L5	Back pain, gait difficulty, spastic paraparesis, hypoesthesia, urinary incontinence	Elevated	Laminectomy; adjuvant chemotherapy	Expired due to sepsis
Kanto, 2007	20 y	Male	L4	Recent aches, vomiting and headaches	79 ng/mL	Right high orchiectomy; right occipital osteoplastic craniotomy; adjuvant chemotherapy	Metastasis to L4 corporal body; spondylectomy; no relapse after 18 months follow up
Unal, 2011	2 y	Male	5.5×3.2, S3	Abdominal swelling	300 IU/mL	Chemotherapy	Regression of caudal lesion after 12 weeks; minimal regression in abdominal lesions; lung lesions unchanged
Ajiboye, 2015	47 y	Male	153×135, T12–L2	Sudden onset BLE weakness, back pain, saddle anesthesia, urinary retention, loss of bowel function	4,770 ng/mL	Laminectomy; adjuvant chemotherapy	Improvement in neurological function; lost to follow-up after 5 months from day of presentation
Almouhissen, 2016	22 y	Male	15×12, T9–T12	Left scrotal mass increasing in size	1,456 ng/mL	Left radical inguinal orchiectomy	Weakness in both lower limbs, decreased oral intake, and nausea after 16 days; metastasis compressing dorsal spine; chemotherapy
Chen, 2019	8 y	Female	12×9.7×7.38, central pelvic cavity	Intermittent lower abdominal pain, fever, constipation, nausea, vomiting	13,220.25 ng/mL	Bilateral salpingo-oophorectomy; pelvic lymph node dissection; omentectomy; appendectomy; radical excision of the implanted tumors	Discharged after 5 days; Referred to oncology for further treatment
Saniasiaya, 2019	Newborn	Male	10.3×16.5×16, cervical	Large left-sided cystic mass, neck swelling, reddish-friable mass protrusion from left ear canal, left-sided facial weakness	143,984 ng/mL	Incisional biopsy; planned chemotherapy	Expired due to sepsis before beginning treatment

BLE, bilateral lower extremity.

of the retroperitoneum, neck, spine, and abdomen, and had failed multiple regimens of salvage chemotherapy. Ultimately, additional radiotherapy was deferred after a multidisciplinary consensus with the surgical, medical oncology, and radiation oncology departments.

To our knowledge, this is the first case of a gonadal YST in an adult male with metastasis to the cervical and upper thoracic spine, resulting in cervical myelopathy. The rarity of this case is a limitation of this case report as there is a lack of literature with similar patient presentations. With history of a resected YST, this patient presented with a retroperitoneal pelvic mass and left supraclavicular adenopathy before undergoing a left radical orchiectomy. The patient unfortunately suffered progression of lymphatic disease, including spinal metastasis which presented as acute onset myelopathy. Laboratory, radiologic, and histologic examination can help to establish a timely diagnosis and to administer effective treatment. YSTs are usually treated via primary resection, but individualized treatment is imperative for better patient outcomes.

Conclusions

YSTs, also called endodermal sinus tumors, rarely metastasize to the spine. We presented here one case of a patient with a previously treated YST that metastasized to the lower cervical and upper thoracic regions of the spine, manifesting as lymphadenopathy and cervical myelopathy. Involvement of Batson's plexus may play a role in the metastasis of YST to the spine. Individualized treatment of YSTs may prevent recurrence or metastasis.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://jss.amegroups.com/article/view/10.21037/jss-24-28/rc

Peer Review File: Available at https://jss.amegroups.com/article/view/10.21037/jss-24-28/prf

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Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent for publication of this case report and accompanying images was not obtained from the patient or the relatives after all possible attempts were made.

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