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Clinical Case Studies

Complex surgical reconstruction for spinopelvic instability caused by a giant Tarlov cyst eroding the sacrum: A case report

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ABSTRACT

Background: Tarlov cysts (TC), also known as perineural cysts are meningeal dilations of the posterior nerve root sheath that typically affect sacral nerve roots. TC are usually asymptomatic and found incidentally. We present the case of a patient with an enlarging sacral TC causing pain from spinopelvic instability secondary to extensive bone erosion. Such illustrative case is intended to increase awareness of the potential need for complex spinopelvic reconstruction in atypical instances of large TC.

Case Description: A 29-year-old female presented to clinic reporting progressive bilateral sacroiliac joint pain that was essentially mechanical in nature. The patient had a normal neurological exam except for a known left drop foot with numbness in the left sural nerve distribution, both attributed to a previously resected peripheral nerve sheath tumor. Magnetic resonance imaging revealed a large multilobulated lesion with imaging characteristics consistent with TC adjacent to the left side of the sacrum, extending outward from the left S1 and S2 neural foramina and measuring 6.7 × 3.7 cm in the axial plane and and 5.6 cm in the sagittal plane. Six weeks of conservative management consisting of physical therapy and pain management was unsuccessful, and the patient reported worsening pain. Surgical reconstruction consisting of L5–S1 transforaminal lumbar interbody fusion, L4 to pelvis navigation-guided instrumentation and posterolateral fusion, and bilateral sacroiliac joint fusion was successfully performed.

Outcomes: At 12 weeks follow-up appointment after surgery, the patient reported resolution of sacroiliac mechanical pain.

Conclusions: Sacral TC are asymptomatic in their vast majority of cases but may occasionally cause neurological deficits secondary to mass effect. Rarely, however, giant TC can also lead to significant bone erosion or the sacrum with secondary spinopelvic instability. In this brief report, we describe a giant TC generating significant spinopelvic instability, which was successfully treated with complex spinopelvic reconstruction, leading to complete resolution of the reported axial mechanical pain.

Background

Tarlov cysts (TC), also known as perineural cysts, are meningeal dilations of the posterior nerve root sheath that typically affect the sacral nerve roots [1,2]. The cyst communicates with the cerebrospinal fluid (CSF) and usually has a small size [2]. In the vast majority of cases, TC are asymptomatic and incidentally discovered on imaging of the lumbossacral region [3]. Nevertheless, TC can enlarge due to net inflow of CSF resulting in stretching or compression of adjacent nerve roots, which can lead to progressively painful radiculopathy, sacral nerve root dysfunction or localized pain secondary to mass effect [4]. When symptomatic, invasive management can be considered. However, best surgical practice remains controversial [2,5–7]. Previously proposed therapeutic strategies include percutaneous image-guided cyst aspiration and fibrin glue injection, CSF shunting, and decompressive laminectomy with various forms of interventions to the cyst such as fenestration, imbrication, excision, and wrapping [5].

We present the case of a patient with an enlarging sacral TC causing symptoms of refractory mechanical SI joint pain. The was treated with complex surgical reconstruction which lead to complete resolution of the symptoms.

FDA device/drug status: Not applicable.

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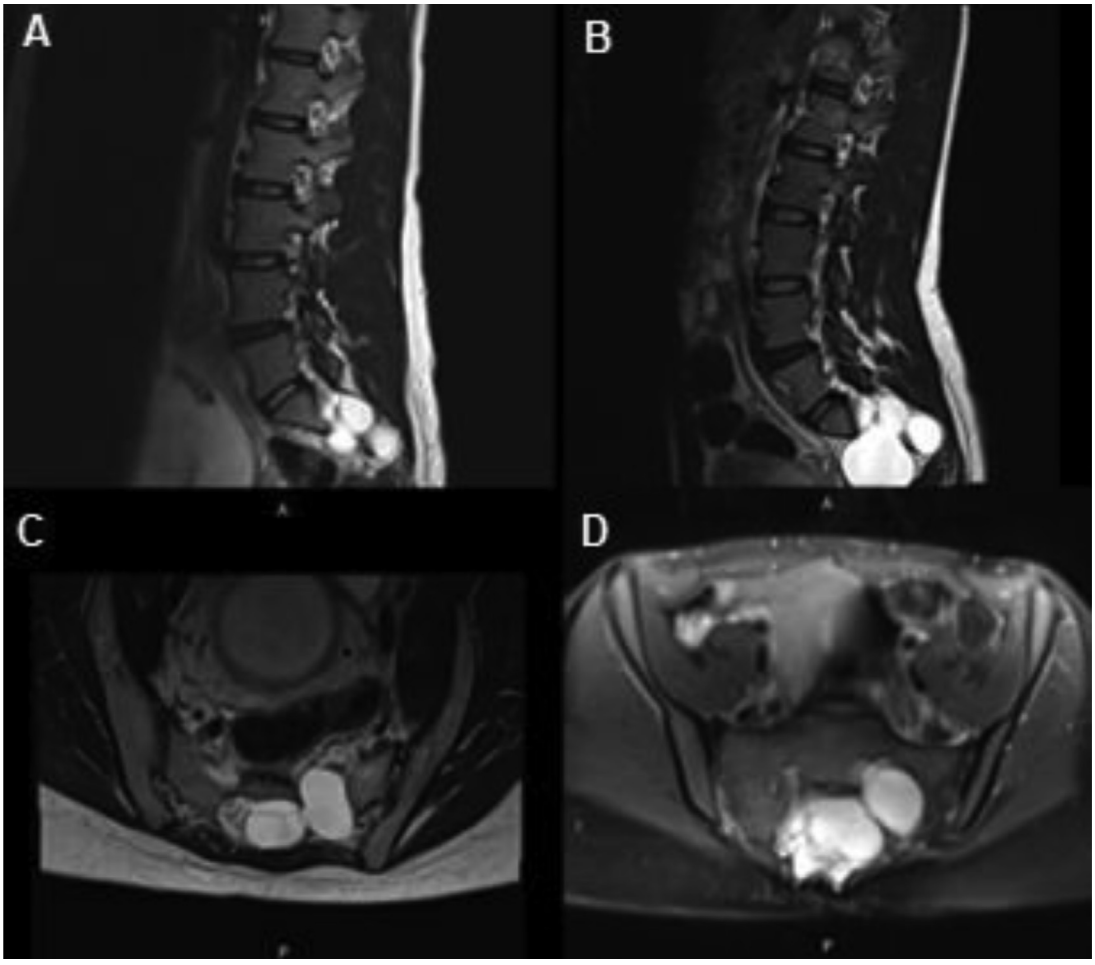

Fig. 1. A large multilobulated T2-hyperintense lesion consistent with perineural/Tarlov cyst is seen in the center to left side of the sacrum, extending along from the left S1 and S2 neural foramina. (A) Sagittal T1 of the first MRI, (B) sagittal T2 of the most recent MRI , (C) axial T1 of the first MRI, and (D) axial T2 of the most recent MRI showing growth of the cyst.

Clinical presentation

A 29-year-old female, with past medical history of mosaic Turner syndrome and status post resection of a low-grade peripheral nerve sheath tumor of the left sciatic nerve, presented to clinic reporting progressive,mechanical SI joint pain worse on the left side. The patient endorsed no recent spinal or pelvic trauma. The patient had a normal neurological exam except for a stable left drop foot with numbness in the left sural nerve distribution, which were both secondary to a previously resected peripheral nerve sheath tumor. Ganslen's, FABER, and distraction tests were used to positively identify SI joint pain.

The patient had a known sacral TC, first discovered incidentally on an MRI obtained 5 years before. Repeat MRI a few years later demonstrated growth of the cyst (Fig. 1). Latest imaging revealed a large, multilobulated lesion consistent with TC adjacent to the left side of the sacrum, extending outward from the left S1 and S2 neural foramina. The lesion was characterized by fluid signal intensity, no central enhancement, and a thin peripheral and septal enhancement. It measured 6.7 × 3.7 cm in the axial plane and 5.6 cm in the sagittal plane, an increase from its previous size of 6.1 × 3.4 × 4.2 cm in the MRI from 5 years before. Given her new symptoms of SI joint pain and the enlarging TC, a CT of the pelvis and lumbar spine were obtained. The CT revealed extensive erosion of thesacrum (Fig. 2).

Management

Given her abrnomal lumbosacropelvic anatomy and progressive symptomatology, the patient's case was presented and discussed in our institutional combined neurosurgery and orthopedic spine conference. It was determined that the extensive bony erosion from the TC and subsequent weakening of the SI joint leading to mechanical back pain was the most likely etiology of the patient's symptoms. After discussing the options of conservative versus surgical management, the patient and care team opted to first attempt conservative management. After 6 weeks of physical therapy and pain management, the patient was seen again in clinic where she reported progressive, unremitting SI pain while sitting and standing which had been significantly affecting her quality of life. Due to failed conservative management, surgical reconstruction consisting of L5–S1 transforaminal lumbar interbody fusion (TLIF), L4 to pelvis navigation-guided instrumentation and posterolateral fusion, and bilateral SI joint fusion was discussed in our institutional combined neurosurgery and orthopedic spine conference. Even in the absence of an overt fracture, the patient's clear symptoms of mechanical SI joint pain with a left-side predominance was persuasive enough that the spine team decided that a bone-scan was not necessary [8]. The patient was counseled on the operative plans as well as any potential adverse effects and she ultimately decided to proceed with the proposed surgical management.

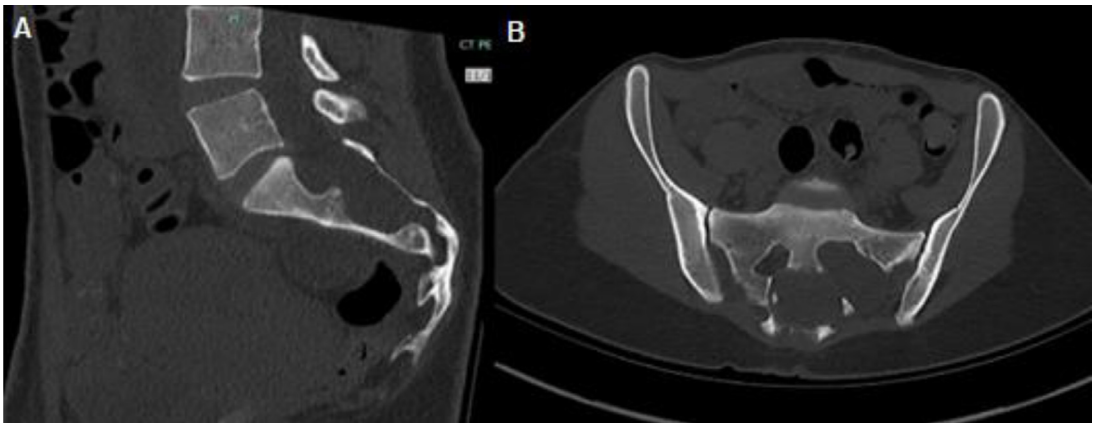

Fig. 2. Multilobulated Tarlov cyst affecting the left side of the sacrum and extending through the left S1 and S2 neural foramina. There is extensive osseous remodeling of the sacrum with enlargement of the sacral spinal canal and neural foramina. (A) Sagittal view, (B) axial view.

Fig. 3. (A) Lateral and (B) anteroposterior views of postoperative upright X-rays.

Surgery was performed starting with navigated insertion of pedicle screws bilaterally at L4 and L5, and on the right pedicle of S1. Then, we proceeded with navigation-guided insertion of a right S2AI screw taking care to tap the size of the screw in order to avoid fracturing what remained of her sacrum. On the left, we placed an S1AI screw and an S2AI screw using navigation guidance, which allowed us a better bony purchase. Next, we performed an L5–S1 TLIF, inserting 2 interbody implants with morselized autograft from the bilateral facetectomy at that level. We believe that by expanding the surface area of vertebral bone exposed to the implants was preferrable in order to reduce the chances of subsidence and providing the best chances for a successful arthrodesis at L5-S1, a level known to have significant rates of pseudarthrosis. Finally, a cobalt chrome rod was used to connect the screws bilaterally. There were no surgical complications. The patient had an uneventful post-operative course and was discharged home 5 days after surgery when incision pain was under control. Upright x-rays of the lumbar spine were obtained prior to discharge (Fig. 3). At the 12-week post-operative follow-up appointment, the patient reported completed resolution of mechanical SI joint pain. Upright x-rays of the lumbar spine at that time demonstrated intact hardware (Fig. 4).

Discussion

Sacral TC are generally benign, and incidentally found [2]. Previous studies have suggested that TCs can be found in up to 13% of the general population, the vast majority in asymptomatic patients [9–11]. Approximately 1% of the population with TC become symptomatic [11,12]. There is a lack of consensus on the best therapeutic approach for patients with symptomatic TC. Percutaneous treatment can beperformed through CSF drainage or fluoroscopy-guided percutaneous fibrin gel injection, and conservative treatment with physical therapy, anti-inflammatory and drugs for neuropathic pain [6,13]. Previously proposed operative

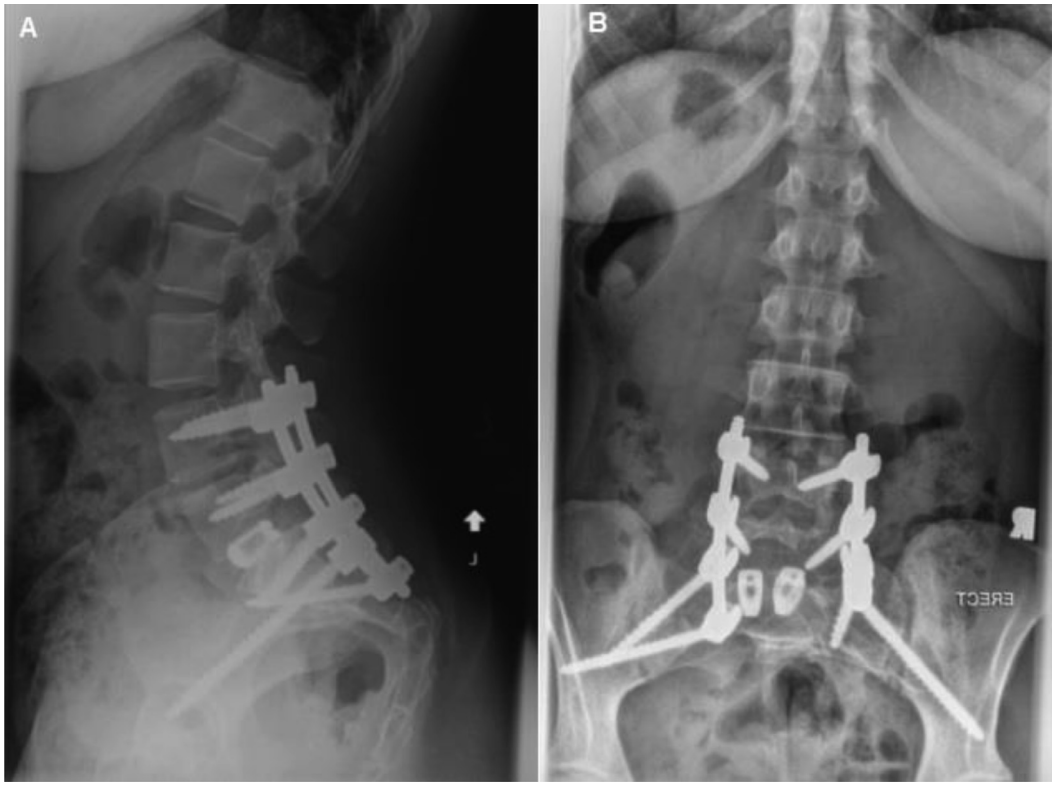

Fig. 4. (A) Lateral and (B) anteroposterior views of upright X-rays at 12-week follow-up.

techniques include partial or full cyst resection, imbrication, obliteration, and cyst fenestration. Medani et al. reported similar outcomes in a study comparing cyst fenestration and nerve root imbrication [7]. These microsurgical techniques are associated with high rates of CSF leakage, cyst recurrence and persistent symptoms [7,13]. However, there is evidence suggesting that microsurgical intervention may be more beneficial for patients with serious radicular symptoms [2,13]. Novel microsurgical itechniques have also been developed to limit cyst recurrence and CSF leakage. Huang et al. describe a procedure in which the surgeon drainsthe space-occupying cyst and complete obliterate the communication between the cyst and the subarachoid space by introducing a dumbbell shaped fat graft into the ostium [13]. In addition to microsurgical management, decompressive laminectomies have also been reported to possibly alleviate symptoms of nerve root compression [6,7].

However, there is a paucity in the literature on TC causing SI instability requiring lumbopelvic fixation [14]. Puffer et al. reported a case of a TC causing significant bone erosion leading to an insufficiency fracture of the sacrum [14]. Similar to our case, their patient required lumbosacropelvic fixation due to the extensive sacral bone erosion. In our case, even though there was no evidence of an overt fracture, the lumbosacropelvic stabilization was deemed a suitable option given the progressive and refractory nature of the patient's mechanical SI joint pain secondary to sacral erosiont [15]. A previous study has shown that the combination of multiple SI joint provocative maneuvers has a high predictive value for identifying pain arising from the sacroiliac joint, with a reported sensitivity ranging from 77to 87% [16]. In the present case, we decided to placean S1AI screw on the left because of the degree of bone erosion on the medial aspect of the left S1 pedicle which rendered it suboptimal for the insertion of a pedicle screw. It is well known that the L5–S1 level has unique biomechanical profile that predispose it to pseudarthrosis [17]. In addition to addressing the SI joint pain, the spinopelvic fixation was helped in dispersing the construct load across the pelvis, further reducing the mobility at the L5–S1 level, and increasing the chances of a successful arthrodesis. Creating fixation points anterior to the lumbosacral pivot point, as performed in this case, has been shown to substantially enhance stability of hte construct [18]. Finally, we have been previously demonstrated that the simultaneous use of S1AI and S2AI screwsmay be an interesting salvage technique to achieve a strong distal anchor for a robust pelvic fixation [19].

There are reported cases of patients with giant sacral TC who ultimately presented with sacral insufficiency fractures during conservative treatment [20]. Because our patient had persistent pain and taking into account the risk of such type of secondary insufficient fracutre, we decided to pursue surgical fixation. The operative plan was discussed extensively among neurosurgery and orthopedics teams, which agreed that, despite the young age of the patient and the known complications of a lumbosacropelvic fixation, , maximal distal fixation was necessary to properly stabilize her spine. Though multiple fixation options were available, our team concluded that fusion from L4 to the pelvis with S1 and S2AI screws was the best option. We believe that pelvic fixation was critical to limit motion at the L5–S1 segment and promote arthrodesis at that level. Furthermore, we agreed on implanting dual cages via the bilateral facetectomy at the L5–S1 level. Though evidence on the efficacy of a dual cage placement is mixed [21], we felt it was imperative to maximize the chances of an effective fusion and minimize the risk of subsidence at the L5-S1, especially taking into accoun the patient's pelvic instability.Though short-term follow-up revealed resolution of symptoms, long-term follow-up will be necessary to assess adequate construct stability and persistence of the observed clinical improvement.

Conclusion

Surgical indications for TC are typically secondary to neural compression. This case demonstrates an atypical presentation of a giant sacral TC leading to significant bone erosion of the sacrum and refractory SI pain. We suggest that sacropelvic joint stabilization with complex lumbosacropelvic instrumentation may be a useful tool in cases of large sacral TC with associated sacropelvic instability. This short case report illustrates that spinopelvic reconstruction can be considered as a therapeutic option for treatment of large sacral TC eroding the sacrum in

patients with refractory mechanical SI pain secondary to SI joint instability.

Informed patient consent statement

Complete written informed consent was obtained from the patient for the publication of this study and accompanying images. This manuscript was written consistent with CARE guidelines.

Declaration of Competing Interest

The authors have no conflict of interest to disclose.

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