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Case Report

Thoracic Radiculopathy Unveiling Giant Bilateral Thoracic Tarlov Cysts

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Background

Tarlov cysts, also known as perineural cysts, are fluid-filled sacs that typically arise between the covering layers of the perineurium and endoneurium at the junction of the dorsal root ganglion. They are usually asymptomatic and typically located at the lumbosacral spine. Thoracic localization is rarely reported and is a challenging condition both clinically due to its misleading wide range of symptoms and therapeutically, given the lack of management guidelines.

Case

We report a case of a man in his seventies complaining of chronic back pain with worsening bilateral thoracic radiculopathy with tingling and burning sensations following T11-T12 dermatomes. The neurological examination was without abnormalities. Magnetic resonance imaging revealed bilateral, extradural, and large cystic formations. The diagnosis of thoracic bilateral giant cysts was established. The patient denied surgical treatment. Topical non-steroidal anti-inflammatory drugs and acetaminophen led to partial improvement of symptoms.

Conclusion

This case highlights clinical and imaging features of thoracic Tarlov cysts. The MRI is considered the gold standard for diagnosing Tarlov cysts and effectively ruling out differential diagnoses. The initial therapeutic approach for Tarlov cysts typically involves conservative management.

Keywords: *Tarlov cyst, perineural cysts, thoracic radiculopathy*

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1. Introduction

Perineural cysts, also referred to as Tarlov cysts (TCs), are fluid-filled lesions [1]. According to Nabros' classification, they are categorized as type II meningeal nerve root cysts [2]. These cysts typically develop between the covering layers of the perineurium and endoneurium at the junction of the dorsal root ganglion and posterior nerve root [3]. Although usually asymptomatic and incidentally discovered, TC can be responsible for different symptoms with varying degrees of severity. Symptomatic TCs are rare, and symptomatic thoracic TCs are even rarer. Their diagnosis is sometimes challenging given their rarity and wide range of symptoms, leading to diagnosis delay and potentially worsening outcomes. On the other hand, their therapeutic management is not codified.

Herein, we present a case of symptomatic thoracic TCs. We emphasize clinical and imaging features, diagnostic challenges, and management of TCs.

2. Case Report

A man in his mid-70s presented with a one-year history of back pain in the thoracic spine with bilateral intercostal radiation, described as a burning sensation in dermatomes T10-T11. The patient denied any history of trauma or malignancy and did not report other additional complaints. He was a smoker and had a history of hypertension and dyslipidemia. He had no fever, night sweats, or weight loss.

Physical examination revealed dorsal scoliosis and paravertebral muscle stiffness. No pain was reported upon the percussion of the spinous processes. Pressure over the intercostal region of T10, T11, and T12 reproduced a shooting pain associated with a tingling sensation on both sides. Cardiovascular and pulmonary examinations were without abnormalities, and no skin lesions were found. Neurological examination showed a normal gait, no muscle weakness, no tendon or plantar or abdominal reflexes abnormalities, and no sensory discrimination. Inflammatory biomarkers, calcium, total alkaline phosphatase, and albumin levels were within the normal range. Spine Radiographs showed osteophytes. No osteolytic or osteosclerotic lesions were found.

Magnetic resonance imaging (MRI) demonstrated bilateral extradural, oval-shaped, multiseptated cystic masses. The cysts measured 38 x 20 mm on the right side and 20 x 20 mm on the left side. These lesions were well-defined and exhibited homogeneous hypo-intensity on the T1-weighted image. On fat-suppressed T2-weighted images, they appeared hyperintense, indicating the presence of cerebrospinal fluid (CSF) within the cysts. Additionally, the imaging showed enlargement of the neural foramina of the T10, T11, and T12 vertebrae. No intraspinal extension nor the involvement of nerve fascicles was observed (Figure **1**).

MRI findings were consistent with TC's characteristic features. The absence of solid components within the cysts, invasion into adjacent structures, and abnormalities of soft tissues helped rule out differential diagnoses, especially nerve sheath tumors such as schwannoma.

Based on clinical and radiological features, a bilateral giant thoracic TC diagnosis was established. The patient denied surgical treatment. Topical non-steroidal anti-inflammatory drugs, acetaminophen, and physiotherapy were prescribed. After three years of follow-up, the patient reported partial alleviation of symptoms. Neurological examinations remained without abnormalities.

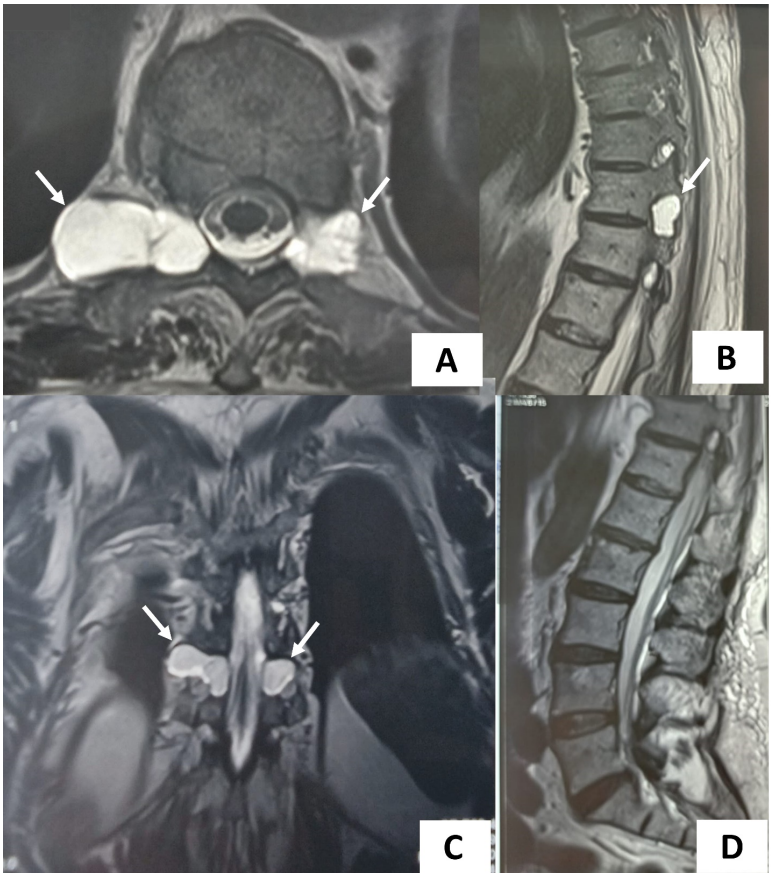

Figure 1. (A): Dorsal spine MRI demonstrated bilateral extradural oval-shaped cystic lesions (arrow) with hyperintensity signal on Fat-suppressed T2-weighted images, corresponding to a CSF-containing cyst. These cysts measured 38x20mm on the right and 20x20mm on the left of T10 on the axial section. **(B):** MRI sagittal section showing enlargement of neural foramina of T10 (arrow). **(C):** MRI coronal section did not reveal cyst extension within the spinal canal, nerve fascicle involvement, and solid components within the cysts. **(D):** Sagittal lumbar spine MRI reveals the absence of the Tarlov Cyst in the lumbosacral region on T2-weighted image.

3. Discussion

We presented a case of bilateral giant thoracic TCs [1] revealed by back pain and worsening bilateral thoracic radiculopathy. These cysts, also known as perineural cysts, are fluid-filled cystic dilatations arising between the perineurium and endoneurium layers of the spinal roots, particularly from the dorsal root [3]. According to Nabros' classification of spinal cysts, our patient had type II meningeal cysts, which consist of extradural meningeal cysts containing neural tissue [2]. These cysts were considered giant because they exceeded 3 cm in diameter [4].

The prevalence of TCs is not well established due to their predominantly asymptomatic nature, with estimates ranging from 1.5% to 13.2% [5]. Symptomatic cases, on the other hand, are scarce and account for less than 1% [6]. They are more frequent in women, occurring in 84% of cases [7]. The average age of diagnosis is estimated to be in the mid-fifties [5]. Nevertheless, it is worth noting that TCs have also been described in children [8]. Although commonly located in the lumbosacral region [9], their thoracic occurrence is rare, and only a few cases of thoracic TCs have been reported [3, 8-11]. Furthermore, giant TCs are infrequently encountered [4].

The etiology of TCs remains unclear. Several hypotheses have been proposed to decipher their formation. A traumatic origin has been suggested. Hemosiderin deposits block venous drainage,

resulting in cyst formation. Others speculate an inflammatory mechanism within the subarachnoid space leading to excessive cell proliferation and cystic dilatation between the perineurium and endoneurium layers. Other studies suggest a congenital origin, wherein TCs arise from the persistence of diverticula originating from an embryonic fissure. The most agreed-upon hypothesis is the altered CSF flow dynamics associated with increased pressure. The increased CSF pressure within the subarachnoid space stresses the embedded nerve root, leading to its cystic dilatation. Furthermore, the created pressure gradient contributes to cyst formation and growth.

TCs are typically benign and remain asymptomatic. However, as in our case, some TCs tend to enlarge, leading to a wide range of manifestations with varying degrees of severity. The predominant symptom commonly seen in patients with TCs is back pain. Radiculopathy may also manifest [12]. As a result, patients may experience shooting pain, numbness, and tingling sensations in the affected nerve root. Notably, a prevalent misconception employed to explain nerve root compression involves the assumption that TCs externally compress the nerve, depending on their location and size. Contrary to this notion, Tarlov Cysts do not exert external pressure on the nerve root. They correspond to dilated nerve roots themselves. The elevated pathological pressure of CSF compresses the nerve roots embedded within, independently of their size, consequently leading to radiculopathy [10]. Furthermore, physical examination can reveal scoliosis.

Despite being generally considered benign, TCs are not devoid of potential complications such as cyst rupture with CSF leakage, leading to intra-spinal hemorrhage [1]. They might be associated with a higher risk of infection and potentially life-threatening conditions [14,15]. In addition, instances of intracystic hemorrhage have been documented primarily because of trauma or anticoagulation therapies. In exceedingly rare circumstances, it has been reported during childbirth [16] or even spontaneously [17].

Upon clinical presentation and examination, distinguishing Tarlov cysts from other etiologies is challenging due to their misleading character as they mimic various diseases. Laboratory findings are without abnormality in patients with TC. Tarlov cysts might masquerade as numerous neurological diseases depending on their presentation, such as syringomyelia with sensory disturbances, muscle stiffness, progressive scoliosis, and poor segmental neuropathic pain. It can also mistakenly be attributed to discogenic conditions [12], such as degeneration, protrusions, herniation, or spinal stenosis. Plain radiographs showed osteophytes. In our case, bilateral thoracic radiculopathy in an elderly patient raised clinical suspicion of herpes zoster and postherpetic neuralgia. However, the patient denied any history of prior herpes infection or recent exposure, and no characteristic skin lesions or associated fever were found.

It is noteworthy to mention the association between TCs and bone erosion and remodeling, commonly known as scalloping [4, 12], which predominantly affect the sacral bone. These lesions have been identified on radiographs and may initially be mistaken for malignant osteolytic lesions. MRI enables the exclusion of disc protrusion, herniation, and spinal stenosis. Besides, it provides findings suggestive of TC, including extradural oval-shaped CSF-containing cysts arising from the dorsal nerve root. Complex patterns with irregular walls, lobulation, and internal septation can be observed. An enlargement of neural foramina of the affected root can be seen. Negative signs like the lack of solid components within the cysts, the absence of invasion into adjacent structures, and the absence of abnormalities of soft tissues

help differentiate TCs from other diagnoses. These tumors present a significant challenge clinically and radiologically due to the overlapping symptoms and similar exhibited radiologic characteristics. The main differential diagnoses are nerve sheath tumors such as schwannomas and Neurofibromas [7].

Compared to TC, schwannomas typically appear iso- or hypointense on T1-weighted images because of their solid components. However, hyperintense signals on T2-weighted images secondary to intra-tumor hemorrhage, can pose a diagnostic dilemma when differentiating them from TCs [18]. Additionally, schwannomas exhibit avid post-contrast enhancement, whereas TCs demonstrate only peripheral enhancement.

Some situations may require further investigations, such as myelography, in instances where MRI results are inconclusive. Myelography can unveil the presence of a cystic, fluid-filled lesion with contrast leakage located within the nerve root sleeve. Additionally, it may show CSF movement pulsations suggestive of Tarlov Cysts. Nonetheless, in many cases, MRI alone is sufficient, obviating the need for further invasive examinations. The combination of clinical evaluation, MRI findings, and the course of progression typically provides sufficient grounds for establishing a diagnosis, as exemplified in our patient. To date, there is no standardized treatment. However, conservative treatment is recommended in the absence of severe symptoms such as debilitating chronic pain. This approach typically involves physical therapy, analgesic medication, and neurotrophic drugs [19].

Surgical intervention becomes a necessary course of action when such symptoms are present. Various surgical approaches are available, offering a range of treatment possibilities, varying from perineural cyst aspiration to laminectomy with cyst wall fenestration. Microsurgical techniques involving cyst excision combined with duraplasty have recently exhibited promising outcomes. Numerous case series have reported a significant improvement in over 80% of patients following surgical intervention [3].

Another innovative and minimally invasive approach, the transforaminal endoscopic procedure, has emerged. This novel technique offers immediate post-operative symptom relief, a shorter recovery time, and a reduced risk of complications [20].

Additionally, Epiduroscopic neural laser decompression has demonstrated significant improvements while maintaining low complication rates [21]. Similarly, percutaneous cyst drainage has shown substantial symptom resolution in the short term; however, frequent cyst refilling overtime has led to the proposal of using subcutaneous infusion ports [22].

Postoperative complications, although rare, primarily include cerebrospinal fluid (CSF) leaks, neurological deficits, infection, spinal instability, bowel and bladder dysfunction, and nerve root tethering due to scar tissue. Laminectomy is associated with the highest rate of complications. CSF leaks and neurological deficits are most frequently observed in cervical spine surgeries, followed by thoracic and lumbar procedures. Spinal instability is particularly common in the thoracic region, especially after extensive bone removal, which may require a spinal fusion procedure. Autonomic dysfunction is predominantly noted in cases of lumbar Tarlov cysts. In addition, multiple cysts and a history of previous spine surgery have been associated with a poorer outcome [23, 24].

4. Conclusion

Thoracic Tarlov cysts are associated with a broad range of symptoms, spanning from back pain and thoracic radiculopathy to severe life-threatening

complications, such as spinal compression, cyst rupture, hemorrhage, and infection. We emphasize the clinical and radiological features of Tarlov cysts. MRI is considered the gold standard for diagnosing Tarlov cysts and effectively ruling out differential diagnoses. The initial therapeutic approach for Tarlov cysts typically involves conservative management, as in our case. Surgical intervention becomes imperative in case of severe presentation or associated complications. Moreover, there is a growing interest in surgical techniques, with the emergence of innovative minimally invasive procedures.

5. Highlights

1. TC is associated with a broad range of symptoms, from back pain and radiculopathy to severe complications (spinal compression, cyst rupture, and infection).
2. MRI is considered the gold standard for diagnosing TC, excluding differential diagnoses. It demonstrates the cysts' lack of extension within the spinal canal, nerve fascicle involvement, and the absence of solid components within the cysts.
3. The initial treatment approach for TC involves conservative options. Surgery is necessary in case of severe presentation or complications.
4. There is a growing interest in surgical techniques, with the emergence of new minimally invasive approaches such as endoscopic treatment.

Acknowledgement

None.

Statement of Ethics

Ethics committee approval is not applicable because we described a retrospective case. It does not involve a clinical trial. We have ensured informed consent was obtained from the patient before reporting the case.

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Patient Informed Consent Statement

The ethics committee of the Hospital approved this study. The patient has signed the consent to participate form.

Disclosure Statement

The authors declare no competing interests.

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Author Contribution

The following authors were responsible for drafting the text, sourcing and editing clinical images, investigation results, drawing original diagrams and algorithms, and critical revision for important intellectual content: M SLOUMA, S ZARATI,

MD YEDES, I GHARSALLAH. The following authors gave final approval of the manuscript: M SLOUMA.

Data Sharing Statement

The datasets used and analyzed during the current study are available from the corresponding author upon reasonable request.

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