Symptomatic Tarlov cysts as a cause of low back pain

A case report and literature review

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Tarlov cysts are perineural cysts that were first described in 1938. Most of the cases were asymptomatic, and discovered coincidentally during investigation for other reasons. Different cases often presented different symptoms: lower back pain, buttock or leg pain, and neurological claudication. In those cases with severe neural element compression, neurological deficit occurred. It is often difficult to evaluate the causes of these symptoms, and thus make decisions concerning investigations or treatment. No guidelines for definite diagnoses exist, and no definitive treatments have been agreed upon.

Keywords: *Tranexamic acid, blood loss, laminectomy, spinal fusion, spinal stenosis*

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Case report

A 44-year-old Thai female presented with lower back pain, and pain radiating from the buttock to the right thigh and calf area. The symptoms worsened after prolonged sitting, or long distance walking. Sometimes the pain was referred to the perianal area. In addition, there was non-specific, slight weakness of ankle dorsiflexion and great toe extension in both feet. There was no significant numbness or bladder dysfunction. A pain chart drawn showed its location to be along the S1-2-3 dermatome bilaterally. The initial diagnosis was spinal stenosis. After conservative treatments that included bed rest, medication (NSAIDs, muscle relaxants, tricyclic antidepressants, and gabapentin, or pregabalin), and physical therapy for 6 months, her symptoms remained unresolved. Magnetic resonance imaging (MRI) of the lumbosacral spine showed mild degeneration of the intervertebral discs at L2-3, 3-4, and 4-5, with mild central canal stenosis, a mildly thickened ligamentum flavum, and facet hypertrophy, but no significant nerve root compression. However, a large midline cystic lesion measuring 1.9 x 1.2 x 1.6 cm (Fig. 1) was detected in the sacral area within the posterior sacral cortex (S2-3). Electromyography (EMG) studies showed only right S1 radiculopathy. An evaluation from a neurologist was requested, but no other neurological conditions were found. At our request, an intervention radiologist performed a computed tomography myelogram of the lumbar spine, following which a needle was guided into the sacral space to aspirate the cystic contents. Prior to the aspiration, the patient's the pain score was 9/10 (VAS); after release of the cystic contents (2 ml of central spinal fluid), there was immediate relief of the pain (VAS = 2-3/10). Three ml of normal saline with contrast media was then pushed into the cystic lesion causing pain symptoms (VAS = 8-9/10), but no contrast media leaked into the dural space. The radiologist then injected contrast media into the lumbar spine (L4-5). Contrast media leakage into the cyst didn't show immediately (Fig.3), but after 4 hours an x-ray showed minimal filling of contrast media into the cyst. All of this evidence demonstrated that the cystic lesion was the cause of the symptoms, and it acted as a one-way valve. In other words, the CSF could leak into the cyst but could not exit because of an increase in intra-cystic pressure. Computed tomography showed a thin wall of sacral bone at the posterior cortex - this caused by pressure effect (Fig. 2). Two questions remained unanswered: (i) was the cystic lesion the major cause of all symptoms? and, (ii) was the cyst connected to the subdural space or not? The lumbar myelogram was useful in proving the connection between the dural space and the cystic contents. If the cyst was connected to the dural space and filled with CSF, post-operative CSF leakage could develop from excision of the cyst alone. To prevent leakage following cyst excision, our pre-operative planning included sacral laminectomy and decompression, with imbrications of the cyst wall.

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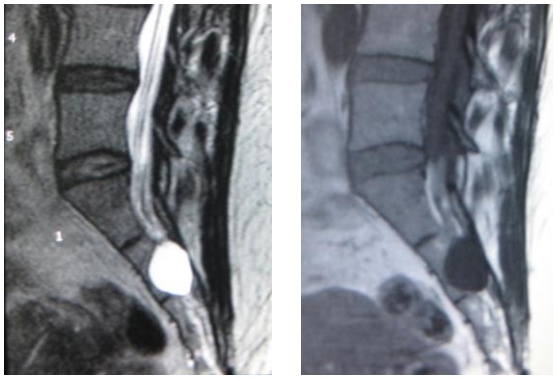


Fig. 1 MRI of the lumbosacral spine with mild intervertebral disc degeneration without protrusion at L4-5 and L5-S1. A cystic lesion can be seen in the area of S2-3 (white arrow).

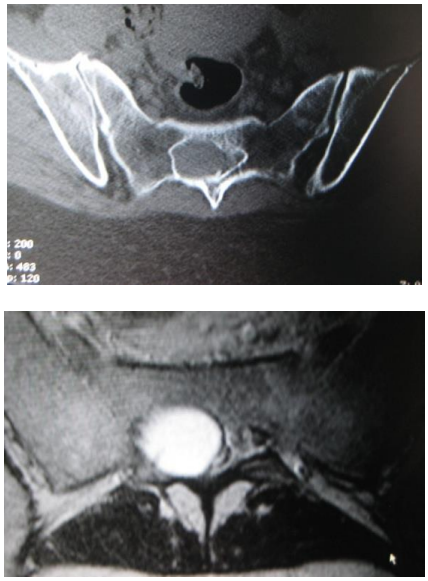
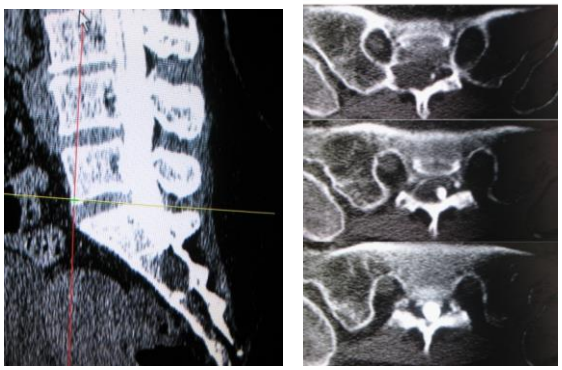


Fig. 2 a large cystic lesion can be seen in the sacral area with a pressure effect, and compression of the cauda equina, causing a thin cortex in the posterior part (white arrow).



A **B**

Fig. 3 CT myelogram after injection of contrast media. (A) No immediate contrast leakage into the cyst (black arrow). (B) 4 hours post injection, contrast media leakage can be seen in the cyst (white arrow)

After a mid-line sacral incision, a laminectomy was performed after burring of the cortex until it was thinned. A cystic lesion filled with CSF was seen, with a nerve fiber attached to the cyst surface. We identified the upper and lower borders of the cyst, but it was difficult to determine normal dura from the cyst wall. The cyst wall was then partially resected and imbricated. The CSF was released prior to cyst imbrication. A valsalva test was performed by partially awakening the patient and using suction in the endotracheal tube. The patient immediately increased intra-abdominal pressure by coughing. We observed the site of the imbrications and found no CSF leakage. Gel foam was inserted and a drain placed above the dural sac, followed by skin closure, layer by layer.

After the operation, all symptoms of the patient were fully resolved: no buttock pain or radiculopathy, and no neurological deficit. There was improvement of sensation around the perianal area, and minimal pain at the surgical wound. There was no CSF leakage and none from the Redivac drain. The patient was ambulatory in a few days. At two, four, and six months follow-up, the patient was able to resume almost full activity with only minimal pain at the surgical wound site.

Discussion

In 1938[1], Tarlov first described meningeal cysts at the sacral nerve root. All were located posterior to this nerve root or dorsal root ganglion and sympathetic neurons[2]. Most cases were asymptomatic and found accidentally. Meningeal cysts are found in 4.6 to 9.0 % [3] of the normal adult population. Although the pathogenesis of these cysts is not known, Tarlov[2] discovered inflammatory cells on the cyst wall leading him to postulate that these cysts develop from an inflammation around the nerve root, and are the cause of pain. Tarlov[4] also suggested that the cysts may develop following trauma because intra cystic blood cells were found in those patients with a history of trauma. In some cases, cystic lesions were found after an intra-operative dural tear during spinal surgery. An alternative pathogenesis of the Tarlov cyst might be congenital malformation or abnormality[5]. Tarlov cysts can produce two issues: first, neurological deficit from neural element compression, e.g. nerve root dysfunction or cauda equina syndrome; and second, pain -- both localized and radicular. Localized pain is caused by an increase in intracystic hydrostatic pressure, with erosion of the sacral cortex. Radiculopathy is caused by nerve root compression or irritation. Symptoms include lower back pain with or without leg pain, claudication, coccydynia, dysuria, or buttock pain. Severity of the pain varies from mild to severe. Not known is the natural history, but some studies indicate that cysts may have a long life, and that cyst enlargement may be caused by hydrostatic pressure (valsava force), and

hence an increase in symptoms. MRI studies are the investigative tool of choice, but myelograms are useful in detecting communication between the cyst and the subarachnoid space. A positive filling defect is a good indicator of success following surgical treatment[6].

Three classifications of Tarlov cysts exist. The first was described by Tarlov in 1938[1,2,4].

1) Tarlov's classification

Lesion type	Communicate with subarachnoid space	Filling on myelography	Location about nerve root	Nerve fiber in cyst
Perineural cyst (Tarlov's cyst)	No	Delayed filling or no filling	Just distal to posterior of nerve root or dorsal root ganglion	Yes
Meningeal diverticula	Yes	Rapid filling	Proximal to dorsal root ganglion	No
Long arachnoidal prolongations	Yes	Rapid filling	Continuous prolongations to subarachnoid space	--

Other classifications are Goyal's[7], (1987) and Nabors'[8], (1988)

2) Goyal's classification

Nomenclature	Description
Perineurial cyst (Tarlov's cyst)	Cyst formation within nerve root sheet at DRG
Root sleeve dilation	Enlargement of subarachnoid space around nerve root proximal to dorsal root ganglion
Intradural arachnoid cyst	Arachnoid pockets within thecal sac
Extradural arachnoid cyst	Arachnoid outpouching through dural defect
Traumatic root cyst	Traumatic tear in leptomeninges causing CSF collection

3) Nabors's classification

Type I	Extradural meningeal cyst without nerve fibers
Type II	(Tarlov's cyst) extradural meningeal cyst with nerve fibers
Type III	Intradural spinal meningeal cyst

All Tarlov cysts are a subset of meningeal cysts, and are defined as Type I using the classifications of Tarlov, and Goyal, but Type II in Nabors' classification. Goyal's and Nabors' systems describe the characteristics and locations of the cysts. Tarlov's classification is useful because it defines cystic patterns, communication with the subarachnoid space, and the characteristics of the contrast media filling of the cyst.

Tarlov cysts are rarely the cause of pain. When symptoms do present they depend on the location and and relationship to nerve roots. It is important to rule out other causes of low back or leg pain, e.g. spinal stenosis or a pelvic mass. MRI is the imaging of choice for diagnosis. CT myelogram is useful in giving information as to whether or not cystic communication with the subarachnoid space exists. J.-Y. Lee et al.[9] showed that delayed intra-cystic filling of contrast media after a CT myelogram indicated the presence of a one-way valve: CSF entered the cyst easily but released with difficulty; or there was a positive filling defect[16] that could increase the intra-hydrostatic pressure. This information was helpful in selecting treatment. In those cases with rapid filling of contrast, the surgeon must be aware of post-operative complications from CSF leakage, so that complete imbrications of the cyst must be performed to prevent CSF leakage. Some cases require myofascial flap coverage. In types with any nerve fibers within the cyst, care must be taken to protect those fibers during the surgical procedure.

Currently, there is no consensus on appropriate treatment. Non-operative treatment should be tried for all symptomatic patients. Pharmacological treatment with NSAIDs or other pain killers, and physical therapy are useful. If there is no dramatic improvement, it can be inferred that the cyst caused the symptoms. Minimally invasive surgery has been reported. Paulsen et al.[3] and Voyadzis et al.[10] suggested that CT guided percutaneous release of the cystic contents can provide symptomatic relief of pain for 3 weeks to 6 months. Symptoms. However, surgeons must not immediately turn to surgical intervention, because symptoms can recur in 3 weeks to 6 months. Other methods included a lumboperitoneal shunt with reported success in 1 case [11], and another in which fibrin glue was injected into the cyst with reported improvement of pain for 6 months to 2 years in 4 cases[12]. However, in this study 75% of the patients developed post operative, aseptic meningitis.

Indications for surgical intervention includes the size of the cyst (greater than 1.5 cm), radiating pain, or signs of neural element compression, e.g. cauda equina compression[10]. Tanaka[6] reported that 83% of patients improved after a surgical decompression procedure. The principle surgical treatments are cyst excision, and closure of the dura or connection with the subarachnoid space. Microsurgical techniques [13,14], using surgical fenestration and imbrications have been reported benefits in eight patients out of a series of thirteen (61.5%). To prevent post-operative leakage, a muscle flap may be used to cover the surgical site, or lumbar drainage[13] should be performed.

Conclusion

Most Tarlov cysts can be treated non-operatively. Operative treatment is useful in selected, symptomatic cases. Pre-operative investigation with an MRI and a myelogram help the surgeon to select an appropriate surgical technique for a successful treatment.

Acknowledgement

I would like to thank the patient for permission to report her case.

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รายงาน ถุงน้ำ Tarlov ที่มาด้วยอาการปวดหลังและการทบทวนวรรณกรรม

ชานินทร์ บุญตั้งใจ, พบ.

รายงานถุงน้ำ Tarlov ในผู้ป่วยหญิงอายุ 44 ปี มาพบแพทย์ด้วยอาการปวดเอว ปวดร้าวลงขาและด้านขวา มีอาการชารอบทวารหนักและเริ่มมีอาการอ่อนแรงในการกระดกข้อเท้าและนิ้วหัวแม่เท้าขวา เอ็กซ์เรย์คลื่นแม่เหล็กไฟฟ้าพบถุงน้ำ Tarlov ที่กระดูกเชิงกรานมีการกดทับเส้นประสาท ให้การรักษาแบบอนุรักษ์แล้วอาการไม่ดีขึ้น ได้ทำการเจาะระบายน้ำโดยใช้เข็มเจาะผ่านการใช้เครื่องเอ็กซ์เรย์คอมพิวเตอร์ พบว่าอาการปวดหลังลดลงชัดเจนและได้ทำการฉีดสีที่ถุงน้ำ Tarlov และช่องไขสันหลัง พบว่ามีการเชื่อมในลักษณะทางเดียว (one way valve) และมีปริมาณเล็กน้อย ได้ให้การรักษาโดยการผ่าตัดเปิดช่องและตัดถุงน้ำออกพร้อมทั้งซ่อมแซมปิดทางเชื่อมต่อกับช่องไขสันหลัง ภายหลังการรักษาผู้ป่วยมีอาการปวดลดลงและสามารถกลับมาทำงานได้ตามปกติ ถุงน้ำ Tarlov พบได้โดยบังเอิญและส่วนใหญ่ไม่มีอาการ แต่ในผู้ป่วยบางรายพบว่ามีการกดทับเส้นประสาททำให้เกิดอาการปวดได้ เอ็กซ์เรย์คลื่นแม่เหล็กไฟฟ้าช่วยในการวินิจฉัยและบ่งบอกลักษณะความรุนแรงหรือการกดทับเส้นประสาทได้ดี ส่วนการเอ็กซ์เรย์คอมพิวเตอร์ร่วมกับการฉีดสีจะให้ข้อมูลว่าถุงน้ำ Tarlov มีการเชื่อมต่อกับช่องไขสันหลังอย่างไรและสามารถลดอาการกดทับเส้นประสาทได้หรือไม่เมื่อมีการระบายน้ำในถุงน้ำออก เพื่อช่วยในการวางแผนการรักษาหรือป้องกันภาวะแทรกซ้อนที่อาจเกิดขึ้น