International Journal of Medical and Pharmaceutical Case Reports

10(4): 1-7, 2017; Article no.IJMPCR.39033
ISSN: 2394-109X, NLM ID: 101648033

Tarlov Cyst Causing Sacral One Nerve Compression, a Rare Presentation: Case Report and Literature Review

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Authors' contributions

This work was carried out in collaboration between all authors. Author EM designed the study, performed the statistical analysis, wrote the protocol, and wrote the first draft of the manuscript. Authors AMB and LKT managed the analyses of the study. Authors EB and ER managed the literature searches. All authors read and approved the final manuscript.

Article Information

DOI: 10.9734/IJMPCR/2017/39033
Editor(s):
(1) Priya Singh Rangey, Assistant Professor, Department of Physiotherapy, L. J. Institute of Physiotherapy, Ahmedabad, India.
Reviewers:
(1) Janani Kumar, USA.
(2) Shuli Zhao, Nanjing First Hospital, Nanjing Medical University, China.
(3) Imtiaz Wani, Sher-I-Kashmir Instiute of Medical Sciences, India.
(4) Jianjun Sun, Peking University Third Hospital, China.
Complete Peer review History: http://www.sciencedomain.org/review-history/23129

Case Study

Received 17th November 2017
Accepted 31st January 2018
Published 10th February 2018

ABSTRACT

Aims: We describe the clinical, radiological and pathological characteristics of a symptomatic Tarlov cyst in a female adult patient who presented with severe low back pain of prolonged duration from Kigali, Rwanda.

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Presentation of Case: We report a 48-year-old Rwandan female patient who presented with a history of progressive low back pain and numbness of S_1 and S_2 distributions for the last 15 years. A magnetic resonance imaging (MRI) was carried out to evaluate for disc degenerative disease, however, we found an incidental symptomatic Tarlov cyst in front of the first sacral vertebra. The symptomatic cyst was surgically excised and the excision site after repair covered with durafoam. The patient reported a remarkable improvement in her symptoms in the post-operative period and follow up of up to 8 months duration.
Discussion: Tarlov cyst (TC) or sacral perineurial cyst is a cystic lesion of the nerve root that is common in the sacrum. TCs are typically located at the junction of the dorsal ganglion and the posterior nerve root and usually develop between the endoneurium and perineurium of the nerve root. Perineural cysts are commonly detected as incidental findings during magnetic resonance imaging of the lumbosacral spine in the imaging of suspected disc degenerative disease.
Conclusion: Tarlov cysts very rarely occur and when they do they are commonly asymptomatic. They cause sacral radiculopathy and sacral pain syndrome, particularly in women. To our knowledge, this is the first case of asymptomatic Tarlov cyst causing unbearable pain in Rwanda.

Keywords: Tarlov cyst; perineural cysts; sacral nerve root 1.

1. INTRODUCTION

Tarlov cyst (TC) or sacral perineural cyst is a cystic lesion of the nerve root that is common in the sacrum [1]. The first report of a Tarlov cyst was made by Isadore Tarlov in 1938 as an incidental finding at autopsy [2], and the lesion was subsequently classified as a Type II meningeal cyst by Nabors et al. [3]. Tarlov cysts are typically located at the junction of the dorsal ganglion and the posterior nerve root and usually develop between the endoneurium and perineurium of the nerve root [4].

The incidence of sacral perineural cysts has been estimated to be 1.5% to 4.6% however, symptomatic cases are rare and account for <1% of the total. These cysts cause a variety of symptoms, including urinary or bowel dysfunction, radicular pain, and paresthesias [5].

The origin of these lesions is controversial and unclear, with causal evidence supporting inflammation within the subarachnoid space, traumatic hemorrhage or pseudomeningoceles, congenital diverticula from persistent embryonic fissures, or hydrostatic cerebrospinal fluid (CSF) pressures [6,7]

Tarlov cysts are associated with a variety of radiological findings. Initial plain radiographic examination may reveal Tarlov cysts causing erosion of the sacrum, bonescalloping, or a rounded paravertebral shadow [8]. Tarlov cysts exhibited a characteristic delayed filling pattern on oil-based pantopaque contrast–enhanced myelography and do not immediately fill with oil-based contrast but can later be visualized hours, days, or weeks later to contain contrast and it was one of the criteria used by Tarlov to distinguish perineurial cysts from meningeal diverticula [6]. Computerized tomography scanning, both with and without intrathecal contrast material, has improved our ability to diagnose perineurial cysts. These lesions are isodense with CSF on non-contrast CT scans [9,10] and can often be seen to cause various osseous lesions and erosions [11]. Postmyelography CT scanning is effective in demonstrating the presence of a communication of the cyst with the spinal subarachnoid space, and it can also demonstrate surrounding sacral bone scalloping [12]. MRI is considered to be the preferred initial imaging modality when a sacral perineurial cyst is suspected [5]. These cysts have CSF-like characteristics on MR imaging—that is, a low signal on T1-weighted images and a high signal on T2-weighted images [7]. Magnetic resonance imaging can also be used to delineate the exact relationship of the cyst to the the calsac, as well as the total volume of fluid within the cyst [13]. It may also demonstrate bone and pedicle erosion, sacralcanal widening, and neural foramina enlargement [7].

We report a case of symptomatic Tarlov cyst presenting with S_1 nerve root compression, which was successfully treated by cyst excision and the excision site covered with durafoam.

2. CASE PRESENTATION

A 48-year-old female Rwandan, who worked as a secretary in a district administrative office, consulted our outpatient department complaining of lower back pain for last 15 years. Initially, it was on and off mild pain, which was not limiting any daily activities and not radiating. However,

she noticed a progressive increase in pain in the last 8 years, which later started interfering with her daily activities. A few years prior to presentation, she noted that the pain was worsened by acts of sitting and standing for long periods of time and relieved by walking.

However, 6 months prior to presentation to our hospital she noted worsening of pain that caused a lot of difficulties in walking, causing her to limp. The pain radiated to the left thigh and leg and was associated with numbness in dorsal aspect and the sole of her left foot. She denied any history of prior surgery and of any member of her family with the same problem.

She had been treated with analgesics and physiotherapy without much success and she opted to consult our spine clinic for further investigations and management. On arrival at our outpatient department, she was limping and in severe pain with her hand holding her lower back.

On examination, we noted that she had paraesthesia on the dorsal aspect and sole of left foot in the S_1 & S_2 distributions. The muscle power was 4/5 on the left leg and 5/5 on right leg with normal deep tendon reflexes on the upper and lower limbs. Straight leg raise test was positive and Patrick's test negative. She had no urinary or fecal incontinence.

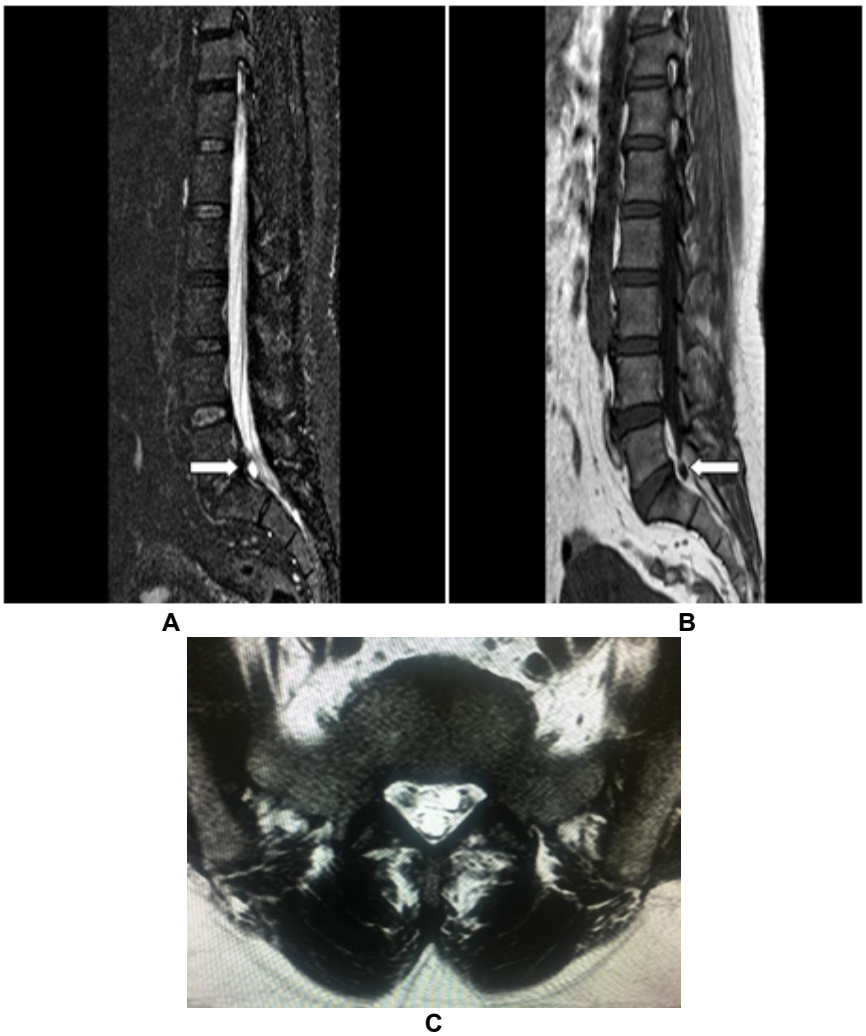


Fig. 1. Lumbosacral MRI. A and B: Sagittal MR T2 & T1-weighted imaging studies respectively of the lumbosacral spine, demonstrating a cystic lesion ventral to the terminal dura in front of S_1. C. Axial MR T1-weighted imaging of the same lesion

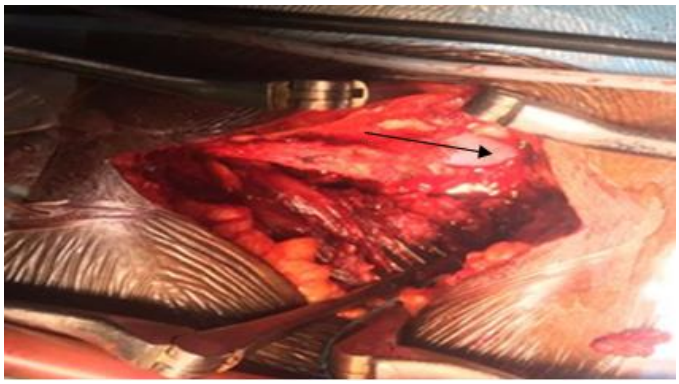
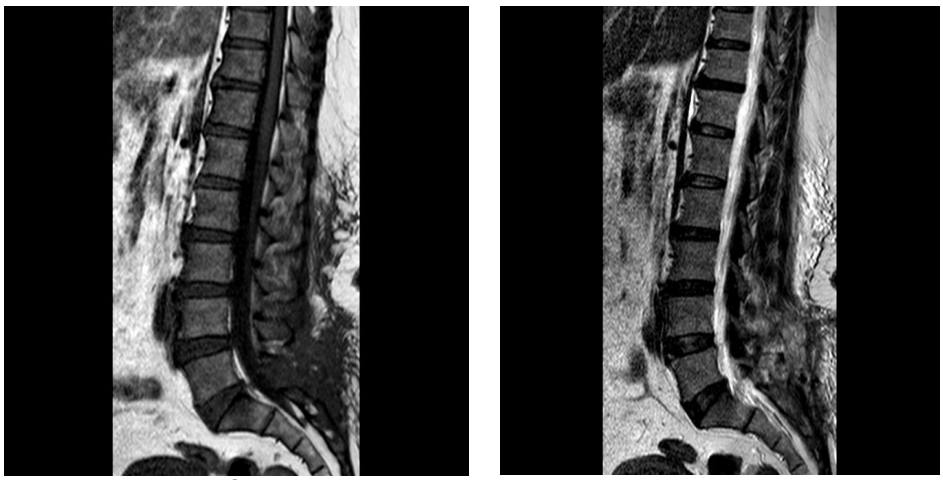


Fig. 2. Intraoperative findings: Tarlov cyst shown by a black arrow at left sacral posterior nerve root of S_1



C **D**

Fig. 3. C & D: Sagittal T1WI&T2WI sequences respectively: A follow-up Lumbosacral MRI 1 month after surgery showed no cyst.

A diagnosis of disc degenerative disease at L_5/S_1 was suspected and a Lumbosacral spine MRI was requested.

MRI findings showed degeneration of L_5/S_1 disc but no significant compression at the same level to explain the severity of her symptoms. On analysis of the MRI images, we noted the presence of an incidental cystic lesion in front of the S_1 vertebra highly suggestive of Tarlov cyst.

The patient was informed about the diagnosis and the need for surgery to relieve her symptoms. We obtained her informed consent for the surgery. A posterior decompression with mini laminectomy and cyst excision with the application of durafoam was planned. Preoperative investigations were done and consent signed.

Under general anesthesia in a prone position, posterior sacral approach, a decompressive mini laminectomy was done and Tarlov cyst was found on left sacral posterior nerve root of S_1. We inspected the cyst for origin and for spinal nerve root fibres. The origin was found to be from the nerve root sheath. The armpit of the spinal nerve root and extremity of the terminal pool were examined and not related to the cyst. The cyst was partially resected and found in the cyst cavity spinal nerve root fibers. The defect was oversewn and repaired area covered with duraform (Dural Graft Implant, DePuy Synthes) to prevent cerebral spinal fluid leakage and closed as routinely done. The patient recovered uneventfully.

On the first postoperative day; she reported an improvement in numbness and pain, and also

started physiotherapy. Physiotherapy was continued for 3 weeks postoperatively. On fifth postoperative day, she reported the disappearance of numbness and was discharged to continue with physiotherapy as an outpatient and to be reviewed after two weeks and later after a month. We reviewed her a month later and all her symptoms had completely gone. We have followed up the patient for a period of 8 months and she is well and resumed work as a secretary and is also now able to do her domestic duties at home as well.

3. DISCUSSION AND LITERATURE REVIEW

Tarlov cysts are defined as CSF-filled saccular lesions located in the extradural space of the sacral spinal canal and are formed within the nerve root sheath at the dorsal root ganglion [4].

Tarlov first described cystic lesions of the spinal nerve root in his autopsy studies of the terminal filum. Tarlov cysts are synonymous with Type II meningeal cysts in a classification of meningeal cysts proposed by Nabors and colleagues [3].

Several mechanisms have been proposed to explain the formation and growth of Tarlov cysts. Previous studies have suggested that the cysts are acquired later in life in those spinal segments subjected to trauma and cerebrospinal fluid pressure [14]. Tarlov believed that the cysts resulted from traumatic or spontaneous hemorrhage into the nerve root, a degenerative process involving the posterior nerve root, or the migration of subarachnoid hemorrhage along the root [6]. The major factor contributing to growth and symptomatology of Tarlov cysts is the hydrostatic and pulsatile forces of cerebrospinal fluid [15].

The prevalence of Tarlov cysts has been estimated to be 1 to 4.6% among the general adult population [12]. The majority (70%) of the Tarlov cysts are asymptomatic, therefore symptomatic cases are rare and have been seen to account for only less than 1% in the general population [16]. Tarlov cyst is common in younger patients with a prevalence of 4.0% in people less than 50 years of age compared to1.3% in persons greater than 50 years of age [17,4].

Asymptomatic sacral perineural cysts are quite often a coincidental finding when lumbar magnetic resonance imaging is performed for other reasons. They have been found in approximately 1% of lumbar MRI cases [18].

Park et al. 2011 found 2.1% of Tarlov cyst as incidental findings of the lumbar spine MRI during herniated intervertebral disk disease evaluation [17].

MRI is the first-choice radiological investigation and allows an accurate evaluation of the relationship between parent roots and cysts. CT scan is useful for showing erosions of the sacral bone caused by cyst expansion [5].Our patient had a symptomatic Tarlov cyst that caused progressive severe low back pain, paraesthesia at S_1 & S_2 distribution but no bowel, bladder or sexual dysfunction. These features are similar to those that are seen in other lumbosacral pathologies where patients may experience pelvic and/or perineal pain; sensory dysesthesias; low-back pain; radicular pain; sciatic pain; and bowel, bladder, or sexual dysfunction [12,17]. The onset of symptoms can be sudden or gradual. Usually, patients report that their symptoms are exacerbated by coughing, standing, and change of position. Symptomatic relief can usually be achieved by recumbent positioning [19,13].

Different authors elaborated different modalities of treatment of symptomatic Tarlov cysts [5,7,14,13-20]. Mitra et al. 2008 suggested that oral and epidurally injected steroids may be a useful adjunct in the conservative treatment of perineural cysts and found that the efficacy of steroids in symptomatic perineural cyst management requires further investigation. The conclusions from their findings were limited because of the small sample size [19].Paulsen et al. [7] 1994 tried percutaneous CT-guided needle aspiration and drainage and he reported a recurrence of symptoms 3 weeks to 6 months in all cases. However ,Voyadzis et al. [21] 2001 and Lee et al. [14] 2004 observed no relief of pain in their series (3 patients in each series.

Zhang et al. [13] 2007 treated 31 patients with a CT-guided percutaneous fibrin glue injection with and without previous aspiration; they described an 80% symptom improvement with cyst aspiration and a 75% symptom improvement without cyst aspiration. There was no recurrence of the treated cysts during a mean follow-up of 23 months. Two cases of transitory aseptic meningitis were reported. Murphy et al. [22] 2008 described a larger series of 122 patients treated by CT guided needle aspirations with fibrin glue

cyst injection. Asymptomatic improvement in 65% of patients was achieved, although 23% experienced a recurrence of symptoms after 7.3 months.

Giampaolo Cantore et al. [5] 2013, treated 19 patients by cyst remodeling around the root using titanium clips. The mean follow-up was 122.6 months, one of the longest in the literature. Sixteen (84.2%) of the 19 patients experienced complete or substantial resolution of preoperative symptoms and neurological deficits after surgery. No patient reported complications.

Our patient's cyst was excised and covered with duraform (Dural Graft Implant, DePuy Synthes). Duraform is a dural graft implant with a collagen-based biocompatible dural substitute that exhibits tensile strength and wet handling capabilities; is intended for use in procedures where the repair or substitution of the patient's dura mater is needed. And this treatment led to complete healing. This is somewhat similar to Tarlovs work where he advocated complete cyst removal and excision of the affected posterior root and ganglion, as a surgical treatment of symptomatic perineural cysts [23].

The complications reported in the literature in the surgical cases include prostatitis, cerebellar bleeding, CSF leakage, dislodgement of the muscle patch, urinary incontinence, dislocation of the catheter, recurrent pain, deep venous thrombosis and superficial wound infections [16,18,24].

Voyadzis et al. [14] 2001. suggest that patients with severe radicular symptoms that are related to Tarlov cysts greater than 1.5 cm in diameter enjoy substantial improvement following cyst resection. Awad TE et al. [1]. 2016 found the complete or substantial resolution of the preoperative local and radicular pain after microsurgical excision of the cyst.

4. CONCLUSION

Tarlov cysts are extremely rare causes of sacral nerve compression however it is now certain that they should be excluded in every patient with sacral radiculopathy and sacral pain syndromes especially women. We have described the successful management of a Tarlov cyst that caused S_1 nerve compression. Surgery should be considered as the primary management for symptomatic Tarlov cysts and great outcomes are expected even in low and middle-income countries.

CONSENT AND ETHICAL APPROVAL

Ethical approval was obtained from the institutional review board of Oshen King Faisal Hospital, Kigali Rwanda and written informed consent was obtained from the patient for publication of this case and accompanying images.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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DOI: 10.4103/2152-7806.85469.

Peer-review history:
The peer review history for this paper can be accessed here:
http://www.sciencedomain.org/review-history/23129