ABSTRACT

Tarlov cysts are pathological cerebrospinal fluid-filled sacs located in the space between the perineum and endoneurium of the nerve roots. Symptomatic Tarlov cysts are extremely rare. There is no consensus regarding the optimal surgical treatment for it up to date. We encountered a recurrent symptomatic sacral Tarlov cyst of a patient whose symptoms resolved after undergoing partial cyst resection, fabrication, imbrication, and duraplasty of sacral Tarlov cysts. A 53-year-old man was initially presented with worsening lower back and buttock region pain, sensory changes involving S1 – S3 distribution of the left lower limb in 2014. The initial magnetic resonance imaging (MRI) lumbosacral had been carried out and revealed a perineural cyst at the level of S1 – S3. The patient did S1 – S3 laminectomy, fabrication, and imbrication after failed conservative treatment and his symptoms resolved for three years. However, similar symptoms recurred in 2017 and the repeated MRI revealed a recurrent well-defined multiloculated cystic structure was seen arising from the spinal canal of S1 – S3 level. The second time, the patient underwent laminectomy S1 – S3, partial cyst resection, fabrication, imbrication, and duraplasty of the sacral region. Many proposed surgical options are available for treating the symptomatic Tarlov cysts. There is no literature reviewed on the best surgical option for the recurrent symptomatic Tarlov cyst. We proposed sacral laminectomy, partial cyst resection, imbrication, fat graft packing, fabrication, and duraplasty in recurrent symptomatic sacral Tarlov cyst.
INTRODUCTION

Tarlov cysts are also known as perineural cysts (Tarlov, 1970), are pathological cerebrospinal fluid-filled sacs situated in the space between the perineurium and endoneurium of the nerve roots (Goyal et al., 1987). It has been estimated that around 1.5% of people have more than one Tarlov cyst, with about 13% of them being symptomatic (Langdown et al., 2005). They are commonly found near the junction of posterior and dorsal root ganglion, and they are bordered by the nerve of reticular fibres. Tarlov cysts are commonly found in the sacral region. Individuals may be affected by multiple cysts of various sizes. The cause of Tarlov cysts is still unknown, it may be either congenital or acquired. Tarlov proposed that these cysts could form due to an increase in cerebrospinal fluid hydrostatic pressure, where the pressure is exerted by the fluid due to the force of gravity (Tarlov, 1970).

The larger the cyst is, the more likely it is to cause symptoms. Although they are asymptomatic typically, 1% may grow and contribute to symptoms include chronic pain at the lower back, especially below the waist, spreading to the buttocks and legs, paresthesias, bowel, and urinary incontinence, impotence, and rarely, weakness in the legs (Tani et al., 2013).

Computed tomography (CT) myelography and lumbosacral MRI are the most common investigations of diagnosing Tarlov cysts. However, dedicated sacral MRI has proven to be more sensitive (Murphy et al., 2016).

Many treatment options are available for symptomatic Tarlov cyst treatment that was being described in the literature ranging from non-surgical, minimally invasive procedures (lumbar cerebrospinal fluid drainage and CT guided aspiration of the cyst) and surgical procedures (such as fenestration of cyst, shrinkage of the cyst by using cautery technique, marsupialization, partial excision and oversewing of the cyst wall, and total excision of the cyst together with nerve root) (Medani et al., 2019).

Surgical techniques such as lumbo-peritoneal shunting, shunting of cyst to subarachnoid, and decompressive laminectomy were uncommonly being practiced, herein, we report a case of recurrent sacral Tarlov cyst compressing the S1 – S3 perineural sheath and eventually requiring S1 – S3 laminectomy, partial resection of Tarlov cyst, nerve root imbrication, fat grafting packing, followed by fabrication and duraplasty.

CASE PRESENTATION

A 53-year-old man with no significant medical illness and family history came to IIUM@SASMEC neurosurgical department in 2014, initially presented with worsening lower back and left buttock region pain with left-sided S1 mapping radicular pain which worsened upon prolonged standing and sitting position, paresthesia in S1 to S3 distribution of the bilateral lower limb without motor weakness over the left lower limb. The patient also had no urinary or bowel incontinence. The initial MRI lumbosacral had been carried out and revealed a perineural cyst at the level of S1 – S3. The patient did S1 – S3 laminectomy, fabrication, and imbrication after failing a conservative treatment trial for 6 months (oral pregabalin and physiotherapy) and his symptoms resolved for three years.

He started to have similar symptoms as described in the initial presentation and required occasional usage of analgesics in 2017. However, his symptoms became worse and warranted for another MRI to reassess the lesion. His MRI revealed a recurrent well-defined multiloculated cystic structure that follows cerebrospinal fluid signal intensity in all sequences (hypointense on T1, hyperintense on T2) (Figures 1 and 2) was seen arising from the spinal canal of S1 – S3 level, measuring about 1.9 × 3.0 × 4.3cm. Thin internal septations were seen within. The sacral exiting nerve roots were displaced laterally bilaterally.
Figure 1 T1-weighted lumbosacral MRI images. Sagittal (a), axial (b, c) views show the well-defined cystic structure (yellow arrow) at the S1 vertebra (b) and the S2 vertebra (c). Preoperative magnetic images demonstrating a hypointense lesion in T1-weighted MRI.

Figure 2 T2-weighted lumbosacral MRI images. Sagittal (a), axial (b, c) views show the well-defined multiloculated cystic structure which follows cerebrospinal fluid signal intensity (yellow arrow) at the S1 vertebra (b) and the cyst (yellow arrow) at S2 vertebra (c). Preoperative magnetic images demonstrating a hyperintense lesion in T2-weighted MRI.
A decompressive laminectomy over S1-S3, partial cystectomy, imbrication, fat graft packing, fabrication of Tarlov cyst, and duraplasty was performed. At surgery, laminectomy was done over 3 levels at the sacrum region, involving S1 – S3 using a Kerrison Rongeur, partial resection of the cyst wall was performed. An inlet was identified from the subarachnoid space (Figure 3), the adipose tissue was harvested from the abdomen earlier on was used to seal the subarachnoid connection and further reinforced with BioGlue (A composition of purified bovine serum albumin and glutaraldehyde) as a sealant to prevent cerebrospinal fluid fistula formation (Figure 4). The residual of the cyst wall was imbricated, and the obliteration was confirmed by a Valsalva manoeuvre. Subsequently, duraplasty was done using a dura patch to seal the defective area (Figure 5). No cerebrospinal fluid leakage postoperatively, and hence spinal lumbar drainage was not inserted. The patient’s lower back and buttock region pain, and S1 – S3 sensory disturbance distribution resolved postoperatively and remained asymptomatic 6 months later.

**DISCUSSION**

Tarlov cysts are extrathecal cerebrospinal fluid-filled cavities in the perineurial recesses that are frequently found at the S2 and S3 levels. They are meningeal dilatation in between the endoneurium and perineurium in the spinal nerve root sheaths and are communicating with the subarachnoid space.

Tarlov discovered this perineural cyst in 1938, he postulated that the process of inflammation within the sheath of the nerve root leads to the formation of the cyst with the inoculation of fluid (Tarlov, 1938). Tarlov cysts are usually asymptomatic, only 1% may contribute to symptomatic Tarlov cysts (e.g., lower back/perineal pain, urinary and bowel incontinence, radiculopathic pain, and rarely infertility) (Tani et al., 2013). A ball-valve effect of the cerebrospinal fluid from the subarachnoid space has caused the growth of the Tarlov cysts which leads to symptomatic Tarlov cyst.

Varieties modalities of imaging tools are available to detect this type of lesion. CT, MRI and myelography are the common tools used to detect Tarlov cysts. MRI has shown to be more efficient in investigating the relationship of the cyst with the surrounding soft tissues (Langdown et al., 2005).

Many neurosurgical treatment options proposed for symptomatic Tarlov cyst (Fibrin glue obliteration, partial cyst resection + imbrication, cyst fenestration only, partial cyst resection + imbrication, cyst wall clipping, cyst...
fenestration + paraspinal muscle pedicle flap, imbrication + fat graft packing, partial cyst resection) (Sunday et al., 2018). The surgery aims to obliterate the connection with the subarachnoid space and prevent further communication with cerebrospinal fluid pathways, which will lead to a reduction of the size of the cysts and subsequently relieve the symptoms. In the literature available, the recurrent rate of surgical technique with cyst fenestration + paraspinal muscle pedicle flap had reported the highest recurrence percentage (74%) (Potts et al., 2016), followed by imbrication + fat graft packing (15%) (Weigel et al., 2016), partial cyst resection + imbrication (8%) (Xu et al., 2012), cyst fenestration alone (6%) (Smith et al., 2011), fibrin glue obliteration (Cantore et al., 2013) and cyst wall clipping (Patel et al., 1997) had reported the lowest recurrent rate of 0% respectively. Potts et al. (2016) reported after cyst fenestration technique in symptomatic Tarlov cysts had shown promising outcomes; however, it suffers from a high recurrent rate of 74%. The rest of the studies had reported comparatively low recurrent rates.

In this report, we decided to perform combined surgical techniques (partial cyst resection, imbrication, fat graft packing, and duraplasty) in this patient with recurrent Tarlov cysts. This combined approach has shown promising outcomes. We would suggest further study by using combined surgical techniques in handling the cases of Tarlov cysts and to further analyze the efficacy and the rate of recurrence.

CONCLUSION

Many proposed surgical options are available for treating the symptomatic Tarlov cysts. Sacral laminectomy, partial cyst resection, imbrication, and adipose tissue packing to seal the subarachnoid space inlet reinforced with Biogluue with duraplasty in this case report appeared to be a viable and optimal surgical option.

CONFLICT OF INTEREST

The authors declare that they have no competing interests in publishing this case.

CONSENTS

Written consent was obtained from the patient to publish this case report. A copy of the written consent is available for review by the Chief Editor.

REFERENCES


