

# Multilocular Disseminated Tarlov Cysts: Importance of Imaging

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## Abstract

With technological advancements and wider availability of multimodality imaging, incidental lesions are frequently identified in patients undergoing various imaging studies. We report here a case of multiloculated disseminated perineural or Tarlov cysts (TCs). The primary aim of our study was to (1) provide a comprehensive review of the clinical, imaging and histopathological features of TCs (2) to draw attention to the fact that multiple lumbo-sacral and dorsal TCs can produce nerve injuries and serious movement disturbances (3) to document the usefulness of the magnetic resonance imaging (MRI) and bone scan in noninvasive diagnosis and guiding management in such cases. These cysts are clearly identified by MR and computerized tomography imaging of the lumbosacral spine. However, there are no reports on the scintigraphic findings of TCs in literature. TCs are typically benign, asymptomatic lesions that can simply be monitored. Until date, no consensus exists about the best surgical strategy to be followed for their management.

**Keywords:** Magnetic resonance pelvis, perineural sacral cysts, Tarlov cyst, Tarlov cyst methylene diphosphanate bone scan

## Introduction

Tarlov cysts (TCs) also known as perineural cysts, are pathological dilatations,<sup>[1-3]</sup> located in between the peri and endoneural spaces of the spinal posterior nerve root sheath.<sup>[4,5]</sup> They affect the sacral roots and cause a progressive painful radiculopathy. TCs usually are found in the spine and do not require surgical intervention unless symptomatic. The typical clinical presentation includes back pain, coccyx pain, low radicular pain, bowel or bladder dysfunction, lower limb weakness, sexual dysfunction and infertility. These cysts are usually diagnosed by magnetic resonance imaging (MRI) and can often be demonstrated by computerized tomography (CT) to communicate with the spinal subarachnoid space.

## Case Report

Our patient is a 62-year-old Indian male patient who presented to Orthopedics Department with a vague history of low back ache, gradually increasing in intensity with no other associated symptoms. Bowel and bladder habits were normal. There is no history of trauma. Patient was otherwise healthy and well-nourished. On local examination, no pelvic bone tenderness was elicited. No motor deficits were noted on neurological examination. A whole body bone scan was advised to rule out any skeletal pathology. 99m Technetium methylene diphosphanate (MDP) three-phase whole body bone scan was performed with 555 MBq given intravenously. Initial dynamic and soft-tissue phase images of pelvis were acquired using a dual head variable angle E Cam Gamma camera. Three hours later, high resolution anterior and posterior whole body images were also obtained. An abnormal focal site of MDP uptake was noted in the sacrum S2 level indicating sacral pathology from uncertain etiology [Figure 1]. On further evaluation with contrast enhanced MRI of the lumbosacral spine, cystic lesion was reported in right neural foramen of S2 of 25 mm × 15 mm × 25 mm size whose intensity was the same

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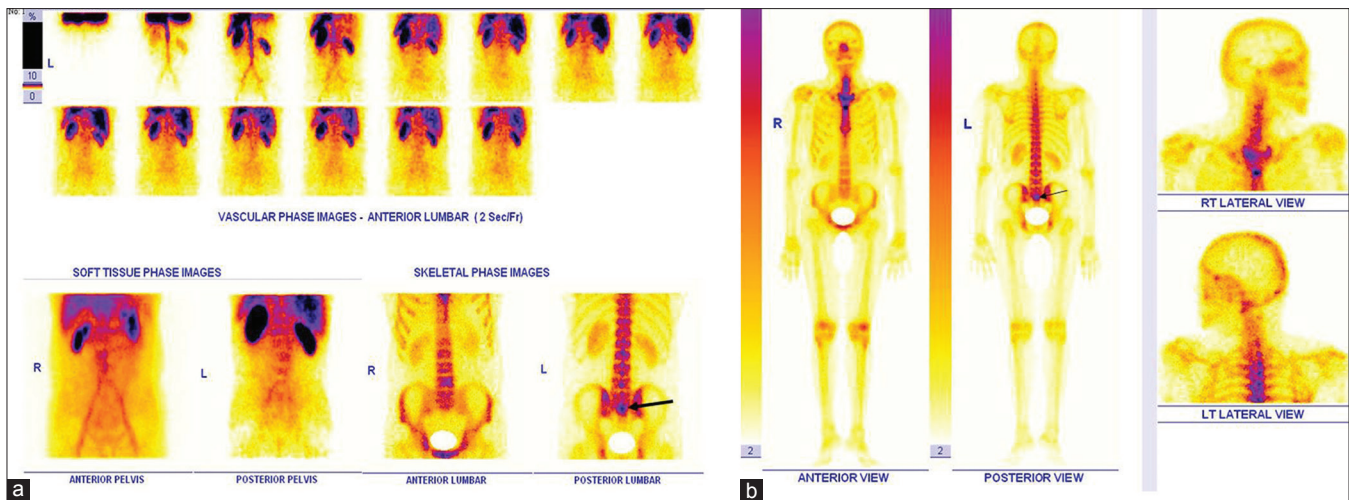


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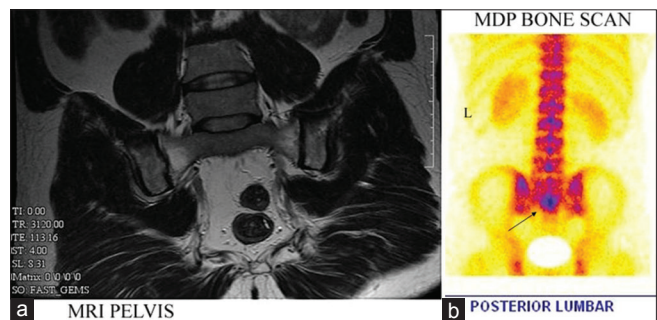
**Figure 1:** (a and b) A three-phase whole body 99mtechnetium labeled methylene diphosphanate bone scan showing focal hot spot in sacrum (arrow) with no increased vascularity. There were no other abnormal hot spots in rest of skeletal survey

as cerebrospinal fluid (CSF) suggesting TC [Figure 2]. No contrast-enhanced findings were observed. Similar lesions were also noted in S1, right T3-T4, right T7-T8 and left T6-T7 neural foramina, but with no corresponding hot spots on MDP bone scan. Initial and a 6-h delayed CT myelography showed no free communication between cyst and subarachnoid space [Figure 3]. However, there was surrounding sacral bony erosion around this cyst which explains the abnormal focal increased MDP uptake.

### Discussion

Incidence of TCs in adults is between 4.6% and 9% respectively,<sup>[6]</sup> with no sex predilection, but is more prevalent in younger age groups.<sup>[7]</sup> TCs 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. Although the terminology of TC is synonymous for any cystic spinal lesions, the most common incidental lumbosacral lesions identified by MR are vertebral hemangiomas, followed by perineural cysts, fibrolipomas, synovial cysts and sacral meningoceles. TCs are distinctly different from various other benign etiologies and their pathological confirmation rests on the fact that their cyst walls are composed of perineurium and neural tissue. The cysts show membranous tissue walls, with peripheral nerve fibers and ganglionic cells embedded into connective tissue.<sup>[6,8]</sup> Voyadzis *et al.* found nerve fibers in the walls of the cysts in 75% of their patients.<sup>[9]</sup>

The cysts in TCs are usually formed by the dilated sheaths between the peri and endoneural spaces of the spinal posterior nerve root sheath having microconnections to the subarachnoid space. Thus when pulsatile, the hydrodynamic forces of CSF act through a ball-valve mechanism, causing these perineural cysts to fill and



**Figure 2:** (a and b) Lumbo-sacral contrast enhanced magnetic resonance imaging and Tarlov cyst methylene diphosphanate bone scan images showed a cystic lesion in right neural foramen lesion at S2 level, 25 mm × 15 mm × 25 mm in size with hot spot in bone scan, whose intensity was the same as cerebrospinal fluid. Similar lesions were also noted in S1, right T3-T4, right T7-T8 and left T6-T7 neural foramina but with no hot spots in methylene diphosphanate bone scan

expand in size, thus compressing the neighboring nerve fibers, resulting in neurological symptoms,<sup>[10]</sup> The ball-valve theory has been previously postulated as the reason why some large TCs cause symptoms that progress, whereas others cause only mild symptoms. The cysts are often multiple and can erode surrounding sacral bone structures, causing irritation of the periosteal pain fibers and insufficiency fractures,<sup>[11]</sup> The other type of TCs, so-called un-valved cysts (with no CSF cross connections) are usually asymptomatic.

Spinal meningeal cysts have been recently classified by Nabors *et al.*,<sup>[5]</sup> into three different types:

- Type I (extradural meningeal cysts without spinal nerve root fibers);
- Type II (extradural meningeal cysts with spinal nerve root fibers [that is, TCs]); and
- Type III (spinal intradural meningeal cysts).

Few of the important reasons put forward in the etiology of TCs are inflammation of nerve root cysts followed

by fluid collection, arachnoidal proliferation along and around the sacral nerve root, post-traumatic disruption of peri and epineural venous drainage secondary to hemosiderin deposition; other developmental or congenital factors apart from trauma have been reported in 40% cases.<sup>[6,9,10]</sup> Patients can have a myriad of symptoms along with low back ache, that is typically accentuated by coughing, standing and change of position explained by the increasing CSF pressure due to the ball valve flow effect. The symptoms can be sudden or gradual. Symptomatic relief can usually be achieved by recumbent position.

MR is the gold-standard investigation in identifying TCs and to study their relationship with surrounding structures. Bone scan is highly sensitive and can easily identify early bony involvement. It may also be used to identify the lesion to be operated first especially in situations where they are multiple. Conventional MRI, shows the cyst to be a fluid-filled lesion with low signal on T1-weighted images and high signal on T2-weighted images (i.e. CSF signal). Single-photon emission computed tomography-CT can help in localizing the lesion better. Bone uptake of MDP is facilitated through a mechanism known as chemisorption. There is a limited role for CT here and is mainly advised for percutaneous aspiration treatment of the cysts. Development of CT myelography, an invasive imaging modality, has led to an improvement in our ability to diagnose "TCs" as a cause of sacral radiculopathy and to show any communication of these cysts with the spinal subarachnoid space. These cysts can enlarge due to inflow of CSF, ultimately producing symptoms due to distorting, compressing, or stretching of adjacent sacral nerve roots.



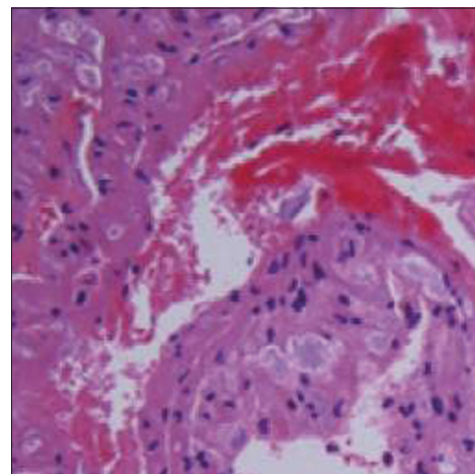
**Figure 3:** Myelo computerized tomography (CT) of pelvis. Initial and a 6-h delayed CT scans, revealed no free communication between the Tarlov cyst (TC) and subarachnoid space. However, surrounding sacral bony erosion around TC in S2 level produced the increased methylene diphosphanate uptake in the bone scan as a result of new bone formation

Optimal treatment for symptomatic TCs is still controversial despite advancements in diagnosis and imaging. Conservative management include analgesics, physiotherapy, lumbar CSF drainage<sup>[12]</sup> and CT guided cyst aspiration, neither of which prevents symptomatic cyst recurrence. Neurosurgical techniques for symptomatic perineural cysts include simple decompressive laminectomy, cyst and/or nerve root excision<sup>[13]</sup> and microsurgical cyst fenestration and imbrication.<sup>[14]</sup> Care must be taken in preserving nervous fibers of the parental nerve roots,

which lie directly on the walls of the cyst. Although no consensus exists on the definitive treatment of symptomatic TCs, surgical methods have yielded the best long-term results to date. Based on patient symptoms, bone scan and MR findings of sacral erosion, our patient underwent S2 cyst fenestration, partial cyst wall resection with myofascial cutaneous flap closure reinforcement surgery. Histopathological sections showed an irregular cystic wall composed of dense collagenous bundles including neural tissue along with vascular structures. Immunohistochemistry stain for S-100 protein showed positivity for neural tissue [Figure 4]. Patient is symptomatically better at 3 months follow-up.

## Conclusion

Sacral TCs are incidentally detected benign pathologies that may or may not require immediate management. However, this case is a reminder that a benign disease entity like perineural TCs can be a cause of nerve roots injury and their lumbo-sacral location can lead to cauda equina syndrome, without disc herniation or other cause



**Figure 4:** Histopathology-showing irregular cystic wall composed of dense collagenous bundles including neural tissue along with vascular structures (H and E,  $\times 200$ ). Immunohistochemistry stain for S-100 protein showed positivity for neural tissue

of the vertebral canal stenosis. Both MRI and bone scan can be used as an effective screening tool and can assist in early management of these cases.

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