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Original Article

# Six cases of occult sacral meningocele

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#### **ABSTRACT**

Background: Intrasacral occult sacral meningoceles (OSM) are uncommon congenital lesions that rarely become symptomatic, even over a patient's life time.

Methods: We operated on six patients with symptomatic OSM diagnosed on MR studies (all six) and/or CT examinations (four cases).

Results: All six patients had uneventful postoperative recoveries. Nevertheless, despite the resolution of low back pain and radiculopathy, preoperative bladder dysfunction improved postoperatively in only one patient.

Conclusion: Few cases of patients undergoing surgery for symptomatic OSM are reported in the literature. Here, we recount our experience with six patients with symptomatic OSM who demonstrated significant postoperative neurological recoveries except for rare improvement in bladder function.

Keywords: Extradural arachnoid cyst, Low back pain, Meningocele, Occult sacral, Sacrum

#### INTRODUCTION

Occult sacral meningoceles (OSM) are cerebrospinal fluid (CSF) containing cystic masses that are typically devoid of neural elements.[1,2,5-9] The radiographic documentation of these lesions is still best-defined utilizing lumbar CT-myelography. [3] Here, we present six patients who presented with symptomatic OSM whose clinical symptoms (i.e., low back pain/radiculopathy) improved with surgery except for bladder dysfunction.

#### MATERIALS AND METHODS

Six patients, including four females and two males ranging in age from 17 to 58 (mean of 41.5), presented with OSM [Table 1]. Symptoms at presentation included low back pain (six patients), unilateral sciatica/radiculopathy (four patients), urinary dysfunction (two patients), and perineal dysesthesias in (one patient). MR documented OSM in all six patients [Figures 1-6]. Another four patients had noncontrast CT scans, while one additional patient had a CT myelogram [Figures 1-4]. Here, we reviewed the clinical presentation, diagnostic evaluations, and surgical management of these six patients with symptomatic OSM (i.e., en block sacral laminotomy, opening/marsupialization of the cyst, water tight closure of the ostium, and a reconstructed bone flap) [Figures 2-6].

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#### **RESULTS**

All six patients had an uneventful postoperative course. Low back pain and radiculopathy resolved in all patients, but bladder dysfunction remained in five of the six.

Table 1: Information of all six cases, including; gender, age, clinical picture, and outcome.

S. No. Sex	Age	Clinical picture	Outcome
0.1.0.			Cateome
1. F	45	LBP and Gluteal pain	Good
2. F	42	LBP sciatica [Figure 2]	Good
3. F	49	LBP and gluteal [Figure 3]	Good
4. M	58	Incontinence and perineal dysesthesia [Figure 4]	Fair
5. F	17	LBP and urinary dysfunction [Figure 5]	Good
6. M	38	LBP and sciatica [Figure 6]	Good

#### Illustrative case

A 45-year-old female with the lower back and right radiculopathy of 4 month's duration was diagnosed with an OSM based on X-ray, MR, and CT-myelographic studies. X-rays displayed scalloping of the sacrum. The MRI revealed a well-defined intrasacral cystic lesion (hypointense in T1 and hyperintense on T2-weighted images), extending from L5 to S3 with scalloping and thinning of the sacrum [Figure 1a and b]. Metrizamide CT myelography showed a block of dye at upper border of L5, while delayed studies showed dye at the bottom of the sacral cyst [Figure 1c and d]. Surgery was performed utilizing a reverse Y incision to first perform a sacral en block laminotomy. This exposed a large cyst with a thin blue capsule filling an enlarged sacral canal. Opening the uppermost portion of the cyst; revealed the ostium that was closed with interupted silk sutures and a muscle graft. The final step was to replace the bone flap that was secured in place with

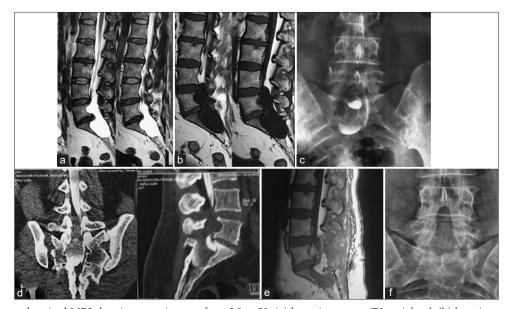


Figure 1: Lumbosacral sagittal MRI showing a cystic mass from L5 to S3; (a) hyperintense on T2-weighted; (b) hypointense in T2-weighted; (c) lumbosacral delayed myelogram shows dye at the bottom of the cyst; (d) CT myelogram demonstrate block of dye at L5 level and dye at the bottom of the cyst; (e) postoperative T2-weighted sagittal MRI, no fluid in the sacrum, hyperintensity is due to free fat graft; and (f) postoperative lumbosacral AP radiograph; note repair of the sacral roof.

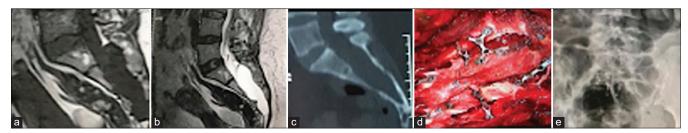


Figure 2: Case 2, (a and b) T1- and T2-weighted sagittal MRI, (c) reconstructed CT scan, (d) intraoperative photograph showing repair of the sacral roof with mini-plate (e) postoperative lumbosacral AP radiograph showing mini-plates.

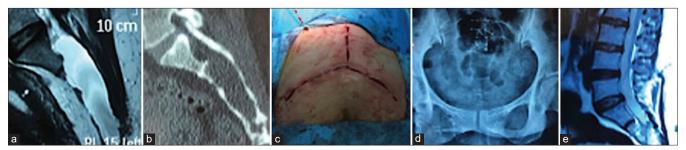


Figure 3: Case 3 - (a) T2-weighted sagittal MRI, (b) reconstructed CT scan, (c) reverse Y incision, (d) lumbosacral plain AP radiograph showing sacral roof repair with mini-plates (e) postoperative T2-weighted sagittal MRI showing disappearance of the sacral cyst.

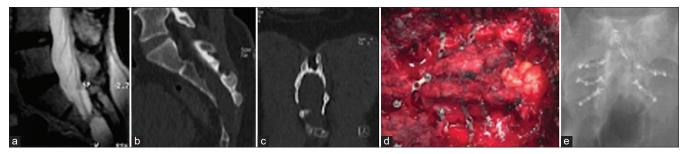


Figure 4: Case 4 - (a) T2-weighted sagittal MRI, (b) reconstructed sagittal, (c) coronal CT scan, (d) intraoperative photograph showing repair of the sacral roof with miniplate, (e)postoperative lumbosacral AP radiograph showing miniplates.

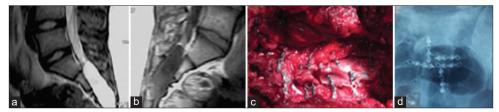


Figure 5: Case 5 -(a and b) T2- and T1-weighted sagittal MRI, (c) intraoperative photograph showing repair of the sacral roof with mini-plate (d) postoperative lumbosacral AP radiograph showing miniplates.

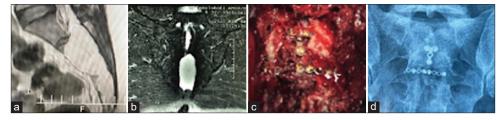


Figure 6: Case 6 - (a and b) T2-weighted sagittal and coronal MRI (c) intraoperative photograph showing repair of the sacral roof with miniplate (d) postoperative lumbosacral AP radiograph showing mini-plates.

a few silk sutures. The postoperative course was uneventful, and the 2-week postoperative MR showed that the cyst had fully resolved [Figure 1e]. One year later, the patient remained asymptomatic, and the radiographic survey documented a now fused/intact sacral roof [Figure 1f].[1-5]

#### **DISCUSSION**

OSMs enlarge with time and often fill the sacral cavity. [1,2,5-9] Within this capsule, the sacral roots are ultimately pressed against the dorsal bony wall of the sacrum. In some cases, these meningoceles will continue to grow resulting in a marked remodeling of the sacral canal (i.e., in coronal/sagittal planes with only a thin shell remaining). Few patients become symptomatic, [1,2,5-9] typically with urinary/fecal dysfunction (nearly uniform), low back pain, radiculopathy, perineal pain, and/or dysesthesias.[1,2,5-9] Notably, those who present with acute/severe local pain may have sustained acute sacral fractures associated with their OSM.

#### Imaging: X-rays, MR, and CT-myelo-CT studies

Plain X-rays of the sacrum typically demonstrate enlargement of the sacral canal with thin and scalloped walls.[2] MRI, the study of choice, typically shows a centrally located and intrasacral meningocele, usually between the S1 and S4 segments.[1,2,5-9] The size, extent of the cyst, bony wall changes, and even the location of the ostium might be depicted on MRI. Other findings may include; a low-lying conus, thick filum terminale, and a lipoma. A blockage of CSF flow from the thecal sac into the cyst, with delayed filling of the cyst itself, is usually observed utilizing CT myelography.

#### Management

We prefer the reverse Y skin incision to achieve maximum blood flow to the skin while facilitating the small en block laminoplasty/laminotomy. The OSM's cyst wall should be incised and aspirated (i.e., demonstrating of CSF), and then followed by, water tight closure of the ostium.<sup>[1,2,5-9]</sup> Reconstruction/fixation of the sacral roof with mini plates may be considered (i.e., the advantage of performing an initial laminoplasty but more difficult to achieve successfully in adults).[4]

#### **CONCLUSION**

Intrasacral occult meningoceles should be considered among the differential diagnoses for patients presenting with the lower back pain, radiculopathy, perineal dysesthesia, and urinary/fecal dysfunction/incontinence in both adolescence and adulthood.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

#### **Conflicts of interest**

There are no conflicts of interest.

#### REFERENCES

- Bozkurt M, Kahilogullari G, Eroglu U, Erdem A. Giant intrasacral meningocele: A case report. Childs Nerv Syst 2013;29:2123-5.
- Doty JR, Thomson J, Simonds G, Rengachary SS, Gunby EN, Nabors MW, et al. Occult intrasacral meningocele: Clinical and radiographic diagnosis. Neurosurgery 1989;24:616-25.
- Enderle C. Occult sacral meningocele (Recealed with Myelography). Riv Neurol 1932;5:418-23.
- Feigenbaum F. Giant sacral meningeal diverticulum containing a thickened filum with lipoma in an adult with spinal cord tethering. Case report and review of the literature. J Neurosurg Spine 2008;9:281-2.
- Hamamcioglu MK, Hicdonmez T, Kilincer C, Cobanoglu S. Intrasacral extradural arachnoid cysts-three case reports. Neurol Med Chir (Tokyo) 2008;48:223-6.
- Huang SL, Jiang HX, Cheng B, Ning N, He XJ. Characteristics and management of occult intrasacral extradural cyst in children. Br J Neurosurg 2013;27:509-12.
- Lohani S, Rodriguez DP, Lidov HG, Scott RM, Proctor MR. Intrasacral meningocele in the pediatric population. J Neurosurg Pediatr 2013;11:615-22.
- Muthukumar N. Sacral extradural arachnoid cyst: A rare cause of low back and perineal pain. Eur Spine J 2002;11:162-6.
- Nishio Y, Hamada H, Kurimoto M, Hayashi N, Hirashima Y, Endo S. A case of accoult intrasacral meningocele presented with atypical bowel symptoms. Childs Nerv Syst 2004;20:65-7.

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