

LARGE ANTERIOR LUMBAR-SACRAL MENINGOCELE :RADIOLOGICAL FEATURES, MANAGEMENT AND OUTCOME.

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Abstract

Background - Anterior lumbar- sacral meningocele is an uncommon type of spinal dysraphism, which can occasionally be linked to syndromes like Currarino and Marfan syndromes. While neurological complications are infrequent with these lesions, secondary conditions such as meningitis, sepsis, obstetric issues, and bowel and bladder difficulties are frequently observed. Surgical intervention is the established treatment approach for symptomatic or enlarging masses since these lesions do not typically resolve spontaneously. Repairing the dural defect can be achieved using either anterior or posterior surgical approaches.

Aims and Objectives - The objective of this investigation was to diagnose individuals exhibiting symptoms related to large anterior lumbar-sacral meningocele and to examine the management difficulties and outcomes associated with these tumors.

Material and Methods - This retrospective study included two patients admitted with radiologically diagnosed anterior lumbar-sacral meningocele under department of neurosurgery , GRMC and associated J.A. group of Hospitals from May 2013 to November 2022 who underwent surgery. No randomisation done.

Results - The primary objective in the management of anterior lumbar-sacral meningoceles is the complete cessation of communication between the spinal subarachnoid space and the meningocele. Several reported procedures encompass the posterior approach involving sacral laminectomy and ligation of the neck of the anterior sacral meningocele, the transabdominal approach with meticulous suturing of the neck of the meningocele, the perineal approach, and the utilisation of lumboperitoneal shunt insertion in instances where surgical closure proves ineffective for large fistulas.

Conclusion- In general, patients diagnosed with anterior lumbar-sacral meningocele exhibit favourable prognosis and surgical outcomes. Surgical closure is considered the optimal therapeutic strategy for addressing anterior lumbar-sacral meningoceles, primarily due to their inherent lack of spontaneous regression and inclination towards progressive enlargement, which increases the risk of complications. However, it is important to acknowledge that surgical intervention for anterior lumbar-sacral meningoceles presents demanding and challenging circumstances. The intricate nature of the procedure, coupled with the rarity of these defects, contributes to the surgical complexities associated with their management.

Key words- Anterior lumbar-sacral meningocele, sacral laminectomy, meningocele, spinal dysraphism

INTRODUCTION:

Anterior lumbar-sacral meningocele is an exceptionally uncommon manifestation of spinal dysraphism characterised by the anterior protrusion of the dural sac through a defect in the anterior aspect of the sacrum ⁽¹⁾. Only a limited number of cases have been documented in early childhood. Anterior lumbar-sacral meningocele primarily arises from the failure of sacral fusion, leading to the subsequent herniation of the sacral meninges into the sacral cavity. The potential mechanisms contributing to the development of anterior lumbar-sacral meningocele, as classified by North et al. ⁽²⁾, include:

1. Congenital:
 - a) Sacral bone defect
 - b) Arachnoid proliferation
 - c) Connective tissue disorders
2. Degenerative: Ischemic lesion
3. Traumatic: Nerve root avulsion or hemorrhage
4. Iatrogenic: Occurring during surgery

Anterior lumbar-sacral meningocele has been observed to potentially coincide with syndromes such as Currarino syndrome, characterised by anorectal malformations, sacral bony defects, and presacral masses. Additionally, it may be associated with Marfan's syndrome, where the underlying cause could be a disruption in collagen biosynthesis and structure at the dural level ⁽³⁾. The familial occurrence of anterior lumbar-sacral meningocele has been attributed to autosomal dominant transmission of the disease ⁽⁴⁾.

Due to their concealed nature, these lesions typically manifest later in life. They may present with symptoms such as constipation, urological issues, or rarely neurological symptoms. Over three-fourths of the cases are observed in women of reproductive age, who are more likely to be diagnosed with asymptomatic anterior lumbar-sacral meningocele during pelvic examinations. Within this patient group, anterior lumbar-sacral meningocele may contribute to infertility and pose challenges during childbirth.

Young paediatric patients predominantly present with symptoms of chronic constipation or urinary retention ⁽⁵⁾. In some instances, laxatives may be administered over extended periods or a misdiagnosis of Hirschsprung's disease may occur. To avoid such erroneous conclusions, the inclusion of digital rectal examinations, which are frequently overlooked, can prove invaluable. Complications may ensue if the cysts become secondarily infected, potentially leading to the development of meningitis ⁽⁶⁾ or the formation of a pyocele. Concurrent tethered cord syndrome with anterior lumbar-sacral meningocele may manifest as neurological manifestations ⁽⁷⁾. Epidermoid cysts may uncommonly occupy these cystic structures or rupture into the rectal cavity. Importantly, anterior lumbar-sacral meningocele has been reported to be misdiagnosed as an ovarian cyst on numerous occasions ⁽⁸⁾.

Thorough clinical examination and radiological investigations play a pivotal role in distinguishing anterior lumbar-sacral meningocele from other aetiologies of cystic presacral masses in paediatric and adult patients, such as sacrococcygeal teratoma, tumors like dermoid, lipomas, neuroblastoma, neuroectodermal cyst, rectal duplication cyst, and ovarian cyst.

Surgical intervention serves as the primary approach for managing anterior lumbar-sacral meningocele. The objectives of surgery encompass the closure of communication between the meningocele and the spinal subarachnoid space, decompression of pelvic structures through meningocele excision, and potential untethering of the spinal cord, if indicated ⁽⁹⁾. The posterior sacral laminectomy technique represents the standard approach for addressing anterior lumbar-sacral meningocele. This surgical route allows for the ligation of the meningocele's base, thereby disrupting its connection with the thecal sac. Additionally, if a tethered cord is present, it can be managed concurrently. Closure of the defect can be achieved using a dural fibrin patch ⁽¹⁰⁾. Care must be exercised to preserve nearby nerve roots to mitigate the risk of postoperative neurological complications. The utilisation of laparoscopic techniques for the surgical management of anterior lumbar-sacral meningocele is progressively gaining prominence. This approach offers several advantages and is increasingly being adopted. In cases where anterior lumbar-sacral meningocele is concomitant with anorectal malformations, such as in Currarino syndrome, a posterior sagittal approach may be advantageous and useful for effective management ⁽¹¹⁾.

MATERIAL AND METHODS:

This was a retrospective study on two patients who were admitted under department of neurosurgery, GRMC and associated J.A. group of Hospitals, Gwalior from May 2013 to November 2022 who underwent surgery and after analysing the patient's outcome the results were obtained.

A retrospective analysis was conducted to evaluate patients who underwent surgical intervention for the diagnosis of anterior lumbar-sacral meningocele. The study collected relevant data encompassing the patients' clinical manifestations, including symptoms and signs before and after surgery, as well as imaging results (Figure 1 and 2) obtained prior to and following the surgical procedure. Additionally, surgical findings were documented. The patients' status and progress were assessed at the time of discharge, with further evaluations performed at 3 months and 6 months post-discharge during follow-up.

The clinical outcomes were primarily assessed by comparing postoperative MRI results with the corresponding preoperative MRI scans at the 3-month follow-up. This comparison allowed for the analysis of any changes observed. Furthermore, the study evaluated the improvement or deterioration of patients' signs and symptoms following surgery.

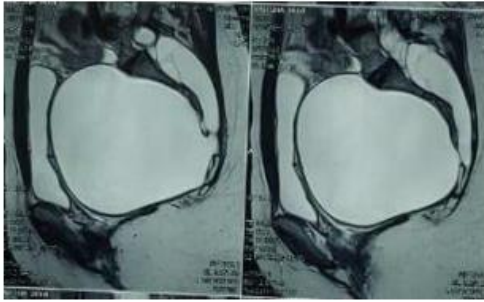


Fig.1

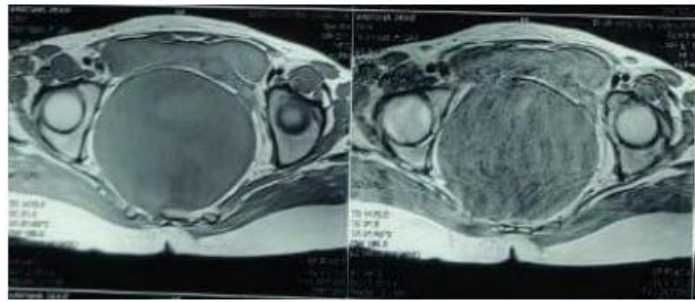


Fig. 2

Fig 1. And Fig 2. - Pre-op Lumbosacral MRI midsagittal and axial cuts.

Inclusion criteria :

All the patients with clinical and radiological diagnosis of anterior lumbar-sacral meningocele and underwent surgery.

Exclusion criteria :

Patients who did not undergo any surgical procedure, small or unresectable anterior sacral meningocele and those not willing for participation.

RESULTS :

A non-randomised, retrospective, single institute study was conducted between May 2013 to November 2022, in GR Medical College and JA group of hospitals (GRMC), Gwalior in the department of Neurosurgery on two patients diagnosed with anterior lumbar-sacral meningocele. Based on the data collected and data analyses following observations were made.

The study findings revealed that females are more commonly affected and no patients experienced neurological deficits immediately after the surgical intervention. Furthermore, at the 6 month follow-up, there were no abdominal or pelvic pain, constipation, urinary complaints, or hydrocephalus among the participants. The MRI conducted during the 3-month follow-up demonstrated the closure of the dural defect, indicating the absence of communication between the thecal sac and the intra-abdominal portion of the meningocele. Additionally, a significant reduction in the size of the intra-abdominal cavity was observed.

DISCUSSION:

Anterior sacral meningocele is a rare congenital malformation. It was first described in Lancet in 1837 from the Medical Society of London as a part of neural tube defect spectrum. Since then, only 250 cases have been reported in the literature⁽¹²⁾.

In present study, first patient was a 24 years old female who presented with left side abdominal pain and urgency of micturition for past six months. MRI of the abdomen and lumbosacral spine was suggestive of large anterior sacral meningocele with intrasacral cystic component with defect in anterior part of lower sacral vertebra. S1 to S4 laminectomy through posterior approach was done with aspiration of cerebrospinal fluid and closure of the defect by muscular graft.

Second case was a 15 years old female who presented with complaint of progressive fullness of lower abdomen since childhood. There were no associated neurological, gynaecological or urological symptoms. MRI of lumbosacral spine was suggestive of defect in sacrum anteriorly on left side with protrusion of meninges through the defect from L4 to upper sacrum and a large unilocular cyst. L4 to S1 laminectomy was done through posterior approach with aspiration of cerebrospinal fluid and closure of the defect by muscular graft.

Mohta et al⁽¹³⁾ reported a similar case of 19 month old female child presented to the hospital with complaints of constipation since 6 months of age and difficulty in passing urine for last 6 months. During surgery a cystic lesion measuring about 8×4 was seen emerging within the leaves of sigmoid mesocolon from the anterior part of sacrum in the pelvis. The sac was opened at the apex, clear cerebrospinal fluid was aspirated and the sac was excised with watertight closure of the tapering base saving the nerve roots and controlling epidural veins. Closed dural defect was checked for leakage of cerebrospinal fluid and haemostasis and then covered with local tissue.

One such case was seen in a baby girl aged 2 months by Khan Yousuf et al⁽¹⁴⁾, who was admitted with abdominal distension and urinary difficulty for one week. She had a lower abdominal mass which investigated by MRI, turned out to be an anterior sacral meningocele causing her symptoms. The meningocele was excised successfully via an open abdominal approach. Postoperative recovery and follow-ups remained uneventful.

CONCLUSION:

It is crucial to acknowledge that while anterior lumbar-sacral meningocele is an exceedingly rare condition, it can still occur in young patients. Given its nature, surgical closure represents the only viable treatment option, as spontaneous regression is not observed. The condition typically progresses with time, leading to enlargement and an associated increase in complications. The primary objective in the treatment of anterior lumbar-sacral meningocele is to eliminate the communication between the spinal subarachnoid space and the meningocele, effectively achieving closure.

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Author's contribution :

MJ- Definition of intellectual content, Literature survey, Prepared first draft of manuscript, Implementation of study protocol, data collection, data analysis, manuscript preparation and submission of article, **SA**- Correction of tables, Coordination and manuscript revision, **KVK**- Concept, design, clinical protocol, manuscript preparation, editing and revision, statistical analysis, **AK**- Manuscript revision, **PS**- Manuscript revision

Conflicts of interest- nil

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Limitations of the study -

Patients with cranial, cervical, dorsal or isolated lumbar meningocele or meningomyelocele and patients who lost follow up are not included.

REFERENCES:

- Villarejo F, Scavone C, Blazquez MG, Pascual-Castroviejo I, Perez-Higueras A, Fernandez-Sanchez A, et al. Anterior sacral meningocele: Review of the literature. *Surg Neurol*. 1983;19:57–71. [[PubMed](#)] [[Google Scholar](#)]
- Nabors MW, Pait TG, Byrd EB, Karim NO, Davis DO, Kobrine AI, et al. Updated assessment and current classification of spinal meningeal cysts. *J Neurosurg*. 1988;68:366–77. [[PubMed](#)] [[Google Scholar](#)]
- North RB, Kidd DH, Wang H. Occult, bilateral anterior sacral and intrasacral meningeal and perineurial cysts: Case report and review of the literature. *Neurosurgery*. 1990;27:981–6. [[PubMed](#)] [[Google Scholar](#)]
- Marin-Sanabria EA, Nagashi T, Yamamoto K, Nakamura Y, Aihara H, Kohmura E. Presacral meningocele associated with hereditary sacral agenesis and treated surgically: Evaluation in three members of the same family. *Neurosurgery*. 2005;57:E597. [[PubMed](#)] [[Google Scholar](#)]
- Bedi NK, Chadha R, Bagga D, Anand R, Mohta A. Anterior Sacral Meningocele: An uncommon cause of constipation in early childhood. *Indian Pediatr*. 1992;29:1157–60. [[PubMed](#)] [[Google Scholar](#)]
- Blond MH, Borderon JC, Despert F, Laugier J, Maheut J, Robert M, et al. Anterior sacral meningocele associated with meningitis. *Pediatr Infect Dis J*. 1991;10:783–4. [[PubMed](#)] [[Google Scholar](#)]
- Hara Y, Shirane R, Yoshimoto T. Anterior sacral meningocele associated with tethered cord syndrome. *No Shinkei Geka*. 1992;20:1217–21. [[PubMed](#)] [[Google Scholar](#)]
- Erdogmus B, Yazici B, Ozdere BA, Safak AA. Anterior sacral meningocele simulating ovarian cyst. *J Clin Ultrasound*. 2006;34:244–6. [[PubMed](#)] [[Google Scholar](#)]
- Tani S, Okuda Y, Abe T. Surgical strategy for anterior sacral meningocele. *Neurol Med Chir (Tokyo)* 2003;43:204–9. [[PubMed](#)] [[Google Scholar](#)]
- Bayar AM, Yasitli U, Tekiner A, Gokcek C, Edebali N, Erdem Y, et al. Anterior sacral meningocele. A case report. *J Neurosurg Sci*. 2007;51:89–92. [[PubMed](#)] [[Google Scholar](#)]
- Ashley WW Jr, Wright NM. Resection of a giant anterior sacral meningocele via an anterior approach: case report and review of literature. *Surg Neurol* 2006; 66:89-93. [[PubMed](#)] [[Google Scholar](#)]
- Sánchez AA, Iglesias CD, López CD, Cecilia DM, Gómez JA, Barbadillo JG, et al. Rectothecal fistula secondary to an anterior sacral meningocele. *J Neurosurg (Spine)* 2008; 8:487-9.
- Mohta A, Das S, Jindal R. Anterior sacral meningocele presenting as constipation. *J Pediatr Neurosci*. 2011 Jan;6(1):40-3. doi: 10.4103/1817-1745.84406. PMID: 21977087; PMCID: PMC3173914.
- Yousuf A, Tayyaba B, Naima R, Yaqoot J, Quresha H and Nasir S. Anterior Sacral Meningocele. *Journal of the College of Physicians and Surgeons Pakistan* 2010, Vol. 20 (5): 337-338.