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Role of screening of whole spine with sagittal MRI with MR myelography in early detection and management of occult intrasacral meningocele

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ABSTRACT

Objective: We evaluated the role of screening of the whole spine by sagittal magnetic resonance imaging (MRI) along with MR myelography in early detection and management of occult intrasacral meningocele.

Materials and Methods: A prospective and retrospective analysis of MRI and MR myelography studies of the whole spine over a period of one year was performed.

Results: Thirty cases with sacral meningeal cysts were seen. On MRI, six patients (three males, three females) fulfilled the criterion of occult intrasacral meningocele. These patients showed a cyst of cerebrospinal fluid (CSF) signal intensity in the sacral canal below the dural sac. This cyst communicated with the thecal sac through a narrow pedicle. Fat signal intensity in the filum terminale and occult sacral dysraphism in the form of an absent or hypoplastic neural arch was observed in all the patients. Low-lying conus medullaris with thick filum terminale was seen in five of these six patients. Excision of the cyst with the release of filum was performed in two patients with a favorable outcome.

Conclusion: Screening MRI with MR myelography of the whole spine may play a role in the early detection and management of occult intrasacral meningocele. The commonly associated thick filum terminale and low-lying conus medullaris may be missed otherwise that may lead to a progression of symptoms.

Key words: MR, myelography, occult meningocele

Introduction

Occult intrasacral meningocele is a rarely described type of sacral meningeal cyst. At times, occult intrasacral meningocele has been used interchangeably in the literature with sacral extradural arachnoid cyst.^[1-5] Patients with occult intrasacral meningocele may continue to have vague symptoms and these are often missed due to a lack of awareness of this entity and inappropriate imaging protocol. Early detection is critical for timely management of this type of occult spinal dysraphism

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Department of Radiology, Shri Guru Ram Rai Institute of Medical and Health Sciences, Patel Nagar, Dehradun, Uttarakhand, India. E-mail: rajivas23@yahoo.com to prevent further progression of symptoms especially in cases with associated magnetic resonance (MR) findings of a tethered cord.^[5-7] Screening of the whole spine with magnetic resonance imaging (MRI) with MR myelography using total imaging matrix (TIM) protocol may improve the visualization, characterization, planning, and optimum intervention of this lesion.^[8]

Materials and Methods

The study was conducted in a referral tertiary care hospital. We prospectively and retrospectively analyzed MRI and MR myelography studies of the spine over a period of one year. The imaging was performed on an 18-channel, 1.5 tesla MRI system (Avanto, Siemens, Erlangen, Germany). As a part of our standardized protocol, screening imaging of the whole spine was done by sagittal T2-weighted sequence (TR: 3500–3800 ms, TE: 80–90 ms) using combined head and spine phased-array coils. The entire spine was imaged using TIM technology by moving the patient table stepwise without repositioning the patient or changing the coil. This was followed by sagittal and axial T1-weighted sequence (TR: 3500–3800 ms, TE: 80–90 ms), sagittal short-tau inversion recovery (STIR) sequence (TR: 3500 ms, TE: 30 ms, inversion time: 160 ms), and axial two-dimensional (2-D) multiple-echo data image combination (MEDIC) sequence (TR: 380 ms, TE: 17 ms, flip angle: 30°) in the region of interest. Slice thickness used was 3-4 mm with a field of view (FOV) of 250-300 mm. Thick slab half-Fourier acquisition single-shot turbo spin-echo (HASTE) MR myelography was performed in coronal and sagittal planes using the following parameters: FOV: 280 mm, slice thickness: 50 mm, TR: 8000 ms, TE: 1200 ms, flip angle: 150°, and averages (NEX): 2. All cystic lesions detected in the sacral region on screening sagittal T2 sequences and MR myelography [Figure 1] were carefully evaluated using the protocol described above to rule out occult intrasacral meningocele, perineural cysts, or facet synovial cysts. The imaging duration of this protocol in each case including sagittal whole-spine screening was 16-18 minutes. The nonsubsidized total cost of MRI for each patient was approximately 80-130 US dollars (4500-7000 Indian rupees). The criterion for the diagnosis of occult intrasacral meningocele was the presence of an extradural cyst of CSF signal intensity below the dural sac having a pedicle communicating with the thecal sac. In addition, we also looked for aplasia/hypoplasia of neural elements, low-lying conus medullaris, or a thick fatty filum terminale. The conus medullaris terminating below the L2-L3 level was labeled as low-lying, whereas a filum terminale of more than 2 mm width on axial images at the L5 level was termed as thickened.^[6,7,9] The perineural cysts had nerve roots in close proximity to the cysts,^[10] whereas the facet synovial cysts were perifacetal in location and generally associated with facet joint degeneration.^[11] Short-term clinical and imaging follow-up was performed.

Results

MR studies of 602 patients were analyzed. Of these, 305 patients underwent the above protocol. The age of the patients ranged from 1 to 83 years. There were 175 males and 130 females. Conus medullaris of the spinal cord terminated at the twelfth dorsal (D12) and first lumbar (L1) vertebral level in 278 patients (91.1%). Mean thickness of the filum terminale was 1.12 mm [standard Deviation (SD): 0.11 mm, range: 0.7–1.6 mm]. Fat signal intensity in the filum terminale was detected in 12/200 patients (6%) in whom T1 imaging was done. Transitional vertebra was seen in 58 patients (19.01 %).

Thirty cases of sacral meningeal cysts were seen. The age range of this subset of patients was 13–83 years (mean: 37.85 years, SD: 19.01). Twenty patients had perineural cysts and four patients had facet synovial cysts. The perineural cysts ranged from 0.5 to 3.5 cm in size (mean: 1.30 cm, SD: 0.80 cm). These cysts were seen at the first and second sacral (S1, S2) vertebral levels in 18/20 patients (90%). Five patients had multiple perineural cysts. The facet synovial cysts in our study ranged from 0.2 to 0.5 cm in size (mean: 0.4 cm, SD; 0.14 cm) and were located posterolaterally at the L4-L5 and L5-S1 levels.



Figure 1: Screening sagittal whole-spine magnetic resonance myelography shows the sacral meningeal cyst

Six patients fulfilled the criterion of occult intrasacral meningocele described above. Three patients were males and three were females with age ranging from 13 to 45 years (mean: 27 years, SD: 12.31 years). The accuracy of the above criterion in the detection of occult intrasacral meningocele was 100%. The sensitivity and specificity in a limited subset of data was also 100%. Amongst all the patients, only three patients had lower back symptoms and others had symptoms which were not corresponding to the site. Two of the patients had previously MR-diagnosed tuberculous spondylitis and the occult intrasacral meningocele could only be detected on whole-spine imaging protocol. Low-lying conus medullaris with thick filum terminale was seen in five of the six patients. The mean thickness of the filum terminale in these patients was 2.83 ± 1.1 mm (range: 1.1-4.5 mm). Fat signal intensity in the filum terminale and occult sacral dysraphism in the form of absent or hypoplastic neural arch was also observed in all the patients. A sacral dimple was found in one patient retrospectively. Whole-spine imaging showed lumbarization of S1 vertebra in two of the six patients. Two patients underwent excision of the cyst with the release of thickened filum terminale. Clinical and imaging follow-up of the four patients who did not undergo surgical excision was done. On imaging, stable lesions were seen in three of these four patients. There was a persistence of symptoms in two of the patients, whereas in one patient, improvement was seen. The fourth patient had tuberculous spondylitis and reported improvement following resolution of the disease process. The summary of clinical, imaging, operative, and follow-up data is presented in Table 1.

Case 1

A thirty-year-old male presented with weakness in upper limbs, cervical pain, and mild fever. Whole-spine sagittal T2-weighted MRI showed mild spondylosis in the cervical region. However, an intrasacral cyst measuring approximately $3.8 \times 3.7 \times 1.8$ cm was observed. On further detailed imaging, lumbarization of the S1 vertebra was seen with a low-lying

Tabl	e 1: Summary of clinic	al, MR imagin	ig, and operative	e findings w	rith follo	p dn-w	ata in	patients with occul	It intrasacral men	iingocele
Age/ sex	Symptoms	MR Region Advised	Location and size	Occult sacral dysraphism	Conus level	FT* (mm)	FFT**	Additional findings	Surgery	Clinical and imaging follow- up from 6 to 14 months
32/M	Progressive weakness in upper limbs, cervical pain, and mild fever	Cervical	S3-54 3.8 × 3.7 × 1.8 cm	~	L3-L4	4.5	≻	Lumbarization of S1	Excision of cyst with release of filum	Good clinical outcome at one year. Mild pain persisting. No imaging follow-up
13/F	Pain, fever, and paraplegia	Dorsal	S2-54 4.9x2.0x1.5 cm	≻	L3	o m	~	Dorsal tuberculous spondylitis with cord compression	Z	Improvement of symptoms due to resolving tuberculous spondylitis at 6 months, no imaging follow-up
Zo/M	Lower limb weakness, urinary incontinence	Lumbosacral	S1-S5 10.1×4.5×2.5 cm	~	L3-L4	3.2	~	Neurogenic bladder	Excision of cyst with release of filum	Recurrent smaller cyst at 14 months on MRI, clinically fair improvement due to release of filum.
M/71	Old dorsal tuberculous spondylitis, low back pain, and mild lower limb weakness, small sacral dimple	Cervicodorsal	S3-54 4.4x2.1x1.7 cm	>	L2-L3	2.7	≻	Resolved dorsal tuberculous spondylitis. Lumbarization of S1	z	Mild pain and persistent weakness at 8 months. On imaging, stable size of lesion in sacrum
45/F	Low back pain, pain in abdomen	Lumbosacral	S3-54 1.7×1.1×0.8 cm	≻	L3-L4	2.5	~	1	Z	Improvement of pain at 6 months. On imaging, stable size of lesion in sacrum
35/F	Neck pain, giddiness, abdominal pain	Cervical	S1-52 3.8x2.7x1.8 cm	≻	L1-L2	1.1	~	1	z	Persistent symptoms at 11 months. On imaging, stable size of lesion in sacrum



conus medullaris terminating at L3-L4 level. In addition, a thickened fatty filum terminale was detected which was adherent to the intrasacral cyst at the S3-S4 level. The cyst had a small communication with the distal end of the thecal sac leading to a diagnosis of occult intrasacral meningocele [Figure 2a-d]. Laminectomy was done along with excision of the cyst and a part of thickened filum terminale. Histopathological examination showed the cyst wall to be formed by nonspecific fibrous tissue lining. The patient showed an uneventful postoperative course and good outcome with only mild pain during follow-up at one year.

Case 2

A twenty-year-old male presented with a history of weakness in the lower limbs and urinary incontinence. Sagittal T2-weighted MRI and MR myelography showed a large biloculated cystic lesion at S1-S5 level measuring approximately $10.1 \times 4.5 \times 2.5$ cm. The upper locule of the lesion was partially communicating with the thecal sac. There was a thin septum between the upper and lower locules. The upper locule displayed CSF signal intensity on all sequences, whereas the lower locule appeared hyperintense on gradient diffusion imaging suggestive of a high protein content. Conus Medullaris terminated at the L3-L4 level with a slightly thickened fatty filum terminale. In addition, significant distension of urinary bladder with diverticulation of its wall was seen. This suggested the possibility of a neurogenic bladder [Figure 3a-e]. Excision of the cyst was performed along with the release of the filum terminale. During follow-up at around fourteen months, the patient had a fair outcome. Some degree of weakness persisted in the lower limbs, whereas urinary incontinence had improved significantly. The follow-up MR imaging showed a recurrent cyst in the sacrum measuring about 7.7×2.2×3.0 cm. However, the urinary bladder exhibited a reduction in the thickening of its wall.

Discussion

The term occult intrasacral meningocele was first suggested by Enderle who diagnosed it on myelography.^[12] The incidence of this type of occult spinal dysraphism is unknown. Occult intrasacral meningocele can be considered as type I extradural arachnoid cyst. The exact pathogenesis of the entity is debatable but the origin may be congenital, related to failure of



Figure 2: (a-d) Sagittal and axial MR (T1-weighted (a, c, d)) and T2-weighted (b) images show a low-lying conus medullaris terminating at the L3-L4 level. Thick fatty filum terminale appears hyperintense (\uparrow) on T1 (a, c) and is adherent to a large intrasacral cyst at the S3-S4 level. Absent neural arches are seen on axial T1 images (c, d)

closure of the neural tube or developmental as a result of increased CSF pulsations. The frequent association of fatty filum terminale and absent neural elements with occult intrasacral meningocele may favor a congenital origin of the lesion.^[1-5]We would also like to suggest a probable congenital nature of the lesion as the above findings were seen in nearly all the patients in this study. In addition, transitional vertebra was seen in two of the six patients.

The cyst as such may be asymptomatic or may cause symptoms due to mass effect. Patients generally present in middle age with mild or atypical symptoms like back pain, gluteal or perineal pain, radiculopathy, or sphincteric disturbances.^[2-5,12,13] However, in this study we were able to detect half of the patients in the second decade due to the selected imaging protocol. A subset of the patients with this congenital syndrome may develop progressive symptoms and signs which at times may be related to the cervicodorsal region. The progression in clinical features is nearly always due to the associated thickening of the filum which may lead to tethered cord syndrome. The symptoms of a tethered cord are leg or back pain, limb weakness, gait disturbance, bladder-bowel symptoms, and foot deformities. Early diagnosis is imperative in such cases.^[7,13] The symptomatology in our patients was also variable and ranged from neck or low back pain, weakness in the upper or lower limb, or radiculopathy. Two of our patients who had dorsal tuberculous spondylitis revealed additional occult intrasacral meningocele on whole-spine imaging protocol. Abdominal pain was also noted in two patients.



Figure 3: (a-e) (a) Sagittal MR myelography (b) T2-weighted (c) T1-weighted (d) gradient diffusion and (e) axial T1-weighted (d) images show a large biloculated cystic lesion in the sacrum. The upper locule exhibits cerebrospinal fluid signal intensity, whereas the lower locule displays hyperintense signal on gradient diffusion imaging. Neurogenic bladder (1) is also noted

MRI especially whole-spine MRI screening with MR myelography as a part of the standardized protocol may lead to the early diagnosis of these lesions as seen in this study. Early detection is possible only if we stop compartmentalizing the spine into cervical, dorsal, and lumbosacral spine. Even in our study, the clinician advised MR of either cervical or dorsal spine in four of the six patients.

Thick slab MR myelography, being heavily T2 weighted, is especially useful as a screening sequence of the whole spine. It aids in detection of the intrasacral cyst in nearly one to two minutes. Once a cystic lesion is seen, occult intrasacral meningocele needs to be ruled out because a low-lying conus medullaris and thick filum terminale can be commonly associated with this lesion. In addition, patients with this type of cyst in the sacrum have either a small fistula or a narrow communication with the theca through small holes at the end of the dural sac. The demonstration of this communication is also possible on MR myelography as an alternative to conventional myelography.^[5,8,13]

Perineural cysts and facet synovial cysts can be differentiated easily from this entity as they are generally smaller in size compared to occult intrasacral meningocele and are associated with nerve root sheaths and facet osteoarthritis, respectively.^[10,11]

Treatment of the cyst is required in patients with a progression of symptoms. Extensive fenestration or excision of cyst wall is the treatment of choice for symptomatic cysts.^[2,4,5,13] Cyst subarachnoid shunt may be performed if the cyst wall is adherent to the neural sheath or ventral to the cord.^[14] The thick filum terminale needs to be surgically released to relieve symptoms of a tethered cord.^[5,13] Surgery was performed in two of the patients in this study. One of the patients who had cervical symptoms and weakness of upper limbs improved significantly with the excision of the thick filum terminale and the intrasacral cyst. On account of the cervical and upper limb symptoms, it was challenging for the surgeon to offer intervention to this patient in the lumbosacral region of the spine. The second patient also reported a fair outcome despite recurrence of the cyst likely due to the release of the tethered cord. Clinical and imaging follow-up of other patients who did not undergo any surgical intervention is being done as the thick fatty filum terminale and low-lying conus medullaris may lead to symptomatology of a tethered cord in future. The follow-up is limited to six to fourteen months presently. Long-term clinical and imaging follow-up alone will decide the real value of the early diagnosis of these lesions.

The study emphasizes the importance of screening whole-spine imaging using sagittal MRI and MR myelography for timely detection of this abnormality. The screening whole-spine imaging protocol does not add much to the total imaging time. The interesting observation in most of our patients was the presence of the thick filum terminale and low-lying conus medullaris that may require surgical intervention in the future for possible progression of symptoms. The limitation of the protocol is that if a lesion is detected, the patient has to be convinced to undergo a detailed imaging of the lumbosacral region which has fiscal implications. In addition, it was difficult and challenging to offer treatment in all the patients. Further studies with longer clinical and imaging follow-up are needed to develop an optimal management protocol for these rarely described lesions following MRI diagnosis.

Conclusion

MR imaging with MR myelography of the whole spine may play a role in early detection and diagnosis of occult intrasacral meningocele separately amongst a plethora of sacral cysts. The importance is not to diagnose this type of sacral cyst as such but commonly associated thick filum terminale and low-lying conus medullaris. These findings may be missed if one does not scrutinize closely and may lead to a progression of symptoms.

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