

Spina Bifida Occulta in an Adult Presenting as Perilesional Hemorrhage

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Abstract

Occult spinal dysraphism with spinal lipoma, is a rare congenital spinal dysraphic anomaly which usually involves lumbosacral region and manifests in childhood. In the present article, we discuss a case of a 26-year-old man who developed sudden low back pain following intercourse. Magnetic resonance imaging of the lumbosacral spine was suggestive of spina bifida occulta with hemorrhage. The patient underwent a L4–5 laminectomy in the emergency. There was the presence of altered blood in the vicinity of the lesion suggestive of recent hemorrhage. Histopathological examination of the lesion was suggestive of the lipoma. In the present case, the patient had occult spina bifida due to lumbosacral lipoma leading to the tethering of the dura. Strenuous activity resulted in overstretching of the tethered structures and subsequent hemorrhage and intense low back pain which responded well to the surgical treatment.

Keywords

- ▶ spina bifida occulta
- ▶ spinal dysraphism
- ▶ lipoma
- ▶ intraspinal lipoma

Introduction

Occult spinal dysraphism with spinal lipoma, is a rare congenital spinal dysraphic anomaly which usually involves lumbosacral region and manifests in childhood.^{1–3} These lesions can be present clinically due to the mass effect leading to the compression of the neural structures or due to the tethering of the cord.^{3–5} In the present article, we discuss a case of a young adult patient who had had spina bifida occulta and developed sudden low back pain following intercourse.

Case Report

A 26-year-old man presented with sudden onset of low back radiating to both the lower limbs of 1-day duration. He was recently married and the pain precipitated during intercourse. There was no history of fever, trauma, or similar episodes of pain

in the past. There was no history of focal weakness or bowel and bladder disturbances. His general and systemic examination was normal. Higher mental functions, cranial nerves, motor and sensory examinations were normal. All the deep tendon reflexes were normal. Planters were bilateral flexor. There was no neck rigidity; however Kerning sign was positive. Spine examination revealed local tenderness over lumbosacral (L4–5) spinal region. There were no deformities or gibbous. Magnetic resonance imaging (MRI) of the lumbosacral spine axial T2-weighted images showed spina bifida of L4 and paraspinal muscle soft tissue hyperintensities; sagittal images showed displacement of the dura and cauda equina anteriorly by T2 hyperintense collection. The L4/L5 interspinous ligament is showing hyperintensities (▶ Fig. 1). The findings were suggestive of spina bifida occulta, lipoma associated with hemorrhage. The patient underwent a L4–5 laminectomy in the emergency. There was a partly absent spinous process of L4 vertebral body and it

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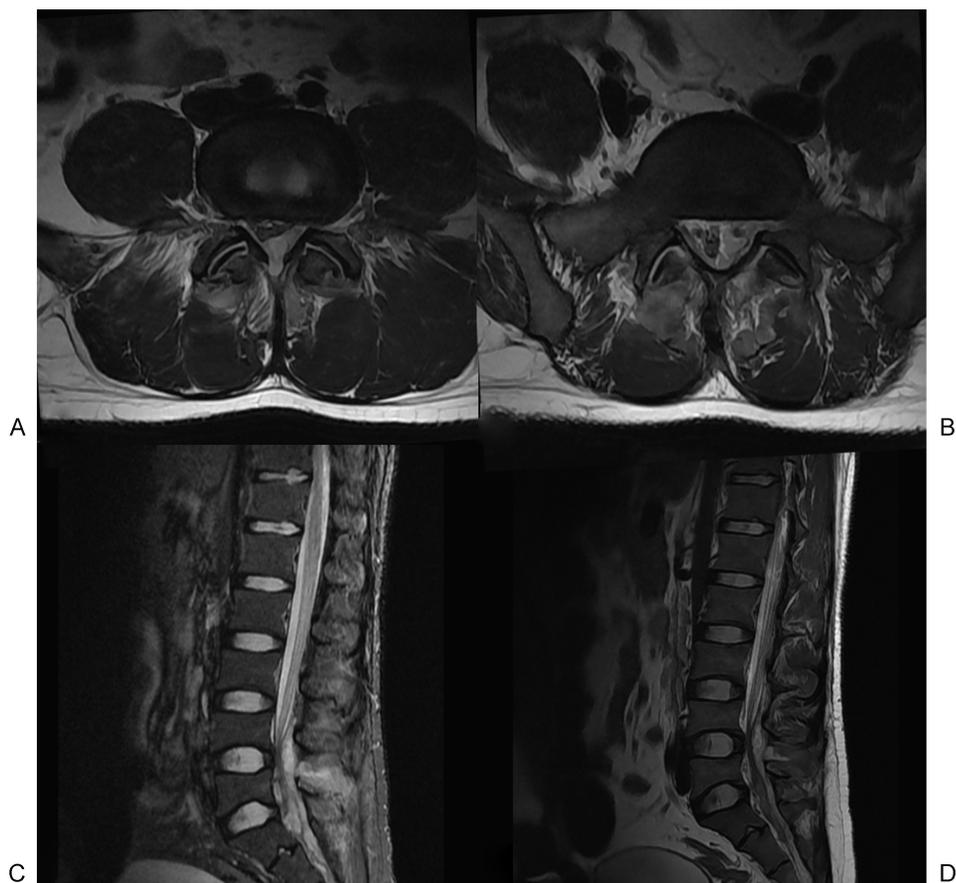


Fig. 1 Axial T2-weighted images (A, B) showing spina bifida of L4 and paraspinal muscle soft tissue hyperintensities, sagittal images (C, D) showing displacement of the dura and cauda equina anteriorly by T2 hyperintense collection. The L4/L5 interspinous ligament is showing hyperintensities.

was replaced by the fatty tissue. The dura was attached to the spinous process and fatty tissue suggestive of tethering. There was the presence of altered blood in the vicinity of lesion suggestive of recent hemorrhage. The dura was detethered and it became lax and pulsatile. Histopathological examination of the lesion showed lobules of mature adipocytes with eccentrically placed nuclei separated by fibrovascular septa which were containing congested blood vessels suggestive of the lipoma

(**Fig. 2**). The patient recovered well after surgery and is doing well at follow-up.

Discussion

The spina bifida occulta can be associated with local cutaneous markers (i.e., midline hypertrichosis, cutaneous hemangiomas, dermal sinus, and midline subcutaneous lipoma).^{1,6} If the lesion

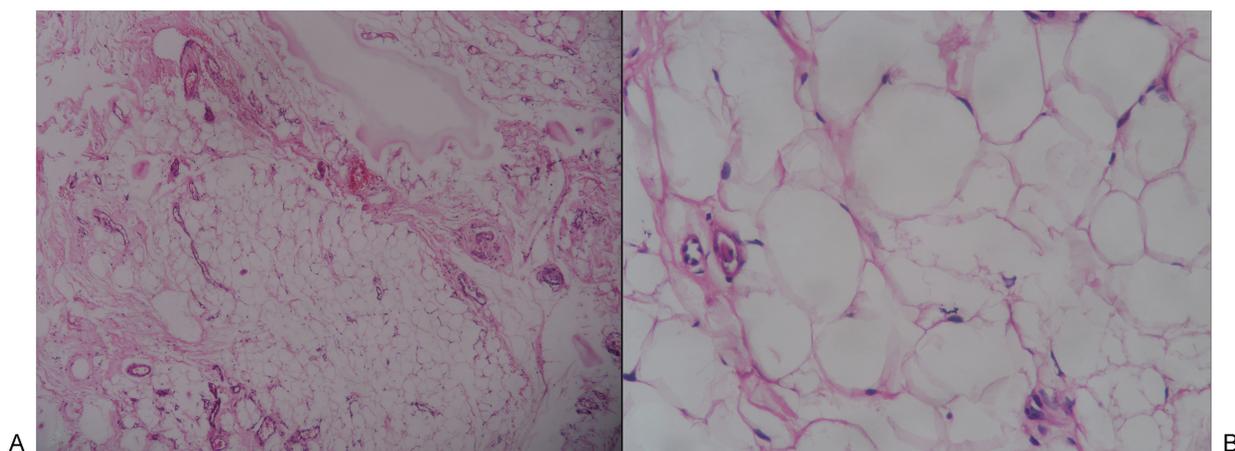


Fig. 2 (A) Lobules of mature adipocytes separated by fibrovascular septa containing congested blood vessels (H&E, $\times 100$) and (B) mature adipocytes with eccentrically placed nuclei (H&E, $\times 400$). H&E, hematoxylin and eosin stain.

causes tethering or mass effect than the patients can present with local and radicular pain, hyporeflexia, spasticity, sensory disturbances, motor weakness, and bowel/bladder dysfunction.^{1,7} Adult patients usually present with low back pain (in contrast to children) and if the lesion is long-standing and progressive in nature than neurological abnormalities including motor weakness, sensory disturbances, abnormal reflexes, and bowel/bladder dysfunction may develop.^{3,8,9} Plain radiographs, computed tomography scan (with bone window), and MRI of appropriate regions have been used in conjunction with each other to make a diagnosis (including details of the lesions and associated anomalies) of spina bifida occulta and to decide the management strategy.^{1,10-12} MRI is the imaging modality of choice and lipomatous tissue appears hyperintense on T1-weighted MR images and hypointense on T2-weighted MR images.^{10,13} Congenital lumbosacral lipomas, which lead to mass effect, neurological symptoms, and/or cosmetic deformity need surgical intervention.¹ The surgical intervention is aimed at to stabilize the neurological functions and untethering of the spinal cord (to prevent delayed neurological deterioration).^{1,3,14-16}

Conclusion

In the present case, the patient had occult spina bifida due to lumbosacral lipoma leading to the tethering of the dura. Strenuous activity resulted in overstretching of the tethered structures and subsequent hemorrhage and intense low back pain which responded well to surgical treatment.

References

- 1 Brkić H, Altumbabić H, Moranjković M. Lipoma and occult spinal dysraphism. *Acta Med Acad* 2006;35:107-112

- 2 James CC, Lassman LP. Diastematomyelia and the tight filum terminale. *J Neurol Sci* 1970;10(2):193-196
- 3 McLone DG. Occult Dysraphism and the Tethered Spinal Cord Lipomas. *Pediatric Neurosurgery*. Philadelphia, PA: Churchill Livingstone; 1999:61-78
- 4 Kanev PM, Bierbrauer KS. Reflections on the natural history of lipomyelomeningocele. *Pediatr Neurosurg* 1995;22(3):137-140
- 5 Soonawala N, Overweg-Plandsoen WC, Brouwer OF. Early clinical signs and symptoms in occult spinal dysraphism: a retrospective case study of 47 patients. *Clin Neurol Neurosurg* 1999;101(1):11-14
- 6 James CC, Lassman LP. Spinal dysraphism. Spinal cord lesions associated with spina bifida occulta. *Physiotherapy* 1962;48:154-157
- 7 Schropp C, Speer CP, Schweitzer T, Krauss J. Congenital skin lesions in occult spinal dysraphism—what is typical? [in German]. *Z Geburtshilfe Neonatol* 2006;210(6):222-227
- 8 Warder DE, Oakes WJ. Tethered cord syndrome: the low-lying and normally positioned conus. *Neurosurgery* 1994;34(4):597-600, discussion 600
- 9 Hirsch JF, Pierre-Kahn A. Lumbosacral lipomas with spina bifida. *Childs Nerv Syst* 1988;4(6):354-360
- 10 Raghavan N, Barkovich AJ, Edwards M, Norman D. MR imaging in the tethered spinal cord syndrome. *AJR Am J Roentgenol* 1989;152(4):843-852
- 11 Smoker WR, Godersky JC, Knutzon RK, Keyes WD, Norman D, Bergman W. The role of MR imaging in evaluating metastatic spinal disease. *AJR Am J Roentgenol* 1987;149(6):1241-1248
- 12 Masaryk TJ, Modic MT, Geisinger MA, et al. Cervical myelopathy: a comparison of magnetic resonance and myelography. *J Comput Assist Tomogr* 1986;10(2):184-194
- 13 Patwardhan V, Patanakar T, Patkar D, Armao D, Mukherji SK. MR imaging findings of intramedullary lipomas. *AJR Am J Roentgenol* 2000;174(6):1792-1793
- 14 Pierre-Kahn A, Zerah M, Renier D, et al. Congenital lumbosacral lipomas. *Childs Nerv Syst* 1997;13(6):298-334, discussion 335
- 15 Kanev PM, Lemire RJ, Loeser JD, Berger MS. Management and long-term follow-up review of children with lipomyelomeningocele, 1952-1987. *J Neurosurg* 1990;73(1):48-52
- 16 Byrne RW, Hayes EA, George TM, McLone DG. Operative resection of 100 spinal lipomas in infants less than 1 year of age. *Pediatr Neurosurg* 1995;23(4):182-186, discussion 186-187