Incidental Finding of a Simple Dysraphic State of a Closed Spinal Dysraphism and Tethered Cord in an Adult Patient with Low Back Pain: A casereport

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Case Report


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The recognition of a closed spinal dysraphism without a subcutaneous mass and cutaneous stigmata depends on the development of relevant neurological and urological symptoms and signs. It can also be an incidental finding when symptoms that do not originate from it are investigated. We describe a 39-year-old man who presented with spondylolysis-related low back pain. Plain film showed posterior spina bifida of S1 and magnetic resonance imaging of the L-spine revealed one intradural lipoma at the L5 level and cord tethering. Conservative treatment was suggested because no relevant symptoms or signs of a mass effect of the lipoma or tethered cord syndrome were observed. Surgery should be considered only in patients with neurological dysfunction of the lower limbs or urological deficits. We emphasize the role of patient education when monitoring the symptoms and signs of patients with tethered cord in order to prevent permanent neurological deficits. (Tw J Phys Med Rehabil 2013; 41(3): 195-199)

Key Words: intradural lipoma, low back pain, spinal dysraphism, tethered cord

INTRODUCTION

Low back pain, which is a health problem with a point prevalence that has been estimated to be as high as 12% in the general population, causes disabilities and imposes substantial economic burdens on the community. Patients with serious spine pathologies, such as malignancies or infections, initially present with low back pain. Uncommonly, low back pain helps clinicians find silent spine pathology, such as closed spinal dysraphism (CSD), which might not cause pain directly. CSD refers to fully epithelialized lesions of the spine or spinal cord, without exposure of neural tissues. Some CSD patients remain unrecognized until clinically relevant neurological, urological, or orthopedic symptoms occur later in life. We describe a case with spondylolysis who presented with low back pain and an incidental finding of CSD. This case illustrates the necessity of careful evaluations of patients with CSD and coexisting low back pain in determining their prognoses and optimal treatments.

CASE REPORT

A 39-year-old man who denied a family history of spinal dysraphism complained of acute low back pain for...
the previous 2 weeks that was not relieved by rest or over-the-counter analgesics. He recalled several episodes of acute low back pain in recent years, and the pain was eliminated in 1 week without medication. He had no history of previous spinal trauma or spine surgery. No skin pigmentation change, nevus, or cystic lesion was noted on his back. The low back pain was aggravated by trunk extension, and local tender points were identified over the lower lumbar paraspinal muscles. Straight-leg-raise and crossed straight-leg-raise tests were negative, and the lower limb deep tendon reflexes were normal. No evidence of lower limb motor or sensory deficits was disclosed by a physical examination. He denied bladder or bowel incontinence, urinary retention, or saddle anesthesia. A L-spine lateral-view X-ray showed a fracture of the pars interarticularis of L5 (Figure 1A), and a kidney, ureter, and bladder (KUB) X-ray showed incomplete fusion of the S1 vertebral body (Figure 1B). L-spine magnetic resonance imaging (MRI; Figure 2) was performed because CSD was suspected from the X-ray findings (posterior spina bifida of S1), and it revealed one fusiform intradural lipoma (1.2 × 1.2 × 4.0 cm) at the L5 level with a tethered cord (conus medullaris at the level of the lower half of the L4 vertebrae). Because no relevant neurological or urological dysfunction was observed, conservative treatment was prescribed, and activity modification and regular monitoring were suggested. The patient received a 1-week course of a nonsteroidal anti-inflammatory drug and skeletal muscle relaxant, and the pain was eliminated completely.

**DISCUSSION**

For patients with low back pain, a detailed history and physical examination cannot be overemphasized, and the clinical characteristics are imperative for confirming the diagnosis and determining management. Because the physical findings that were compatible with L5 spondylolysis showed on plain film and were not indicative of serious spine pathology, advanced diagnostic imaging might have been unnecessary in our patient.[4] However, we performed a L-spine MRI due to a suspected simple or complex dysraphic state of CSD based on the posterior spina bifida found with KUB. The patient’s low back pain was not associated with radicular pain or other neurological or urological symptoms, and the pain responded well to rest and drugs. Because of the above reasons, we attributed the patient’s pain to the L5 spondylolysis that was seen on X-ray, and not to tethered cord syndrome (TCS). Accordingly, we diagnosed our patient with adult CSD with tethered cord, which was diagnosed coincidentally.

Spinal dysraphism or spinal and spinal cord malformations include a variety of congenital neural tube defects, and the skin, subcutaneous tissues, vertebral

**Figure 1.** (A) L-spine lateral view X-ray showing fracture of the pars interarticularis of L5 (arrow). (B) Kidney, ureter, and bladder (KUB) X-ray showing incomplete fusion of the S1 vertebral body (circle).

**Figure 2.** Magnetic resonance imaging (upper row: sagittal views; lower row: axial views) showing one intradural lipoma (arrow) with hyperintensity at the L5 level. The spinal cord is tethered to the anterior surface of the lipoma. (A) T1-weighted images. (B) T2-weighted images. (C) Fat suppression image.
column, and neural elements may be involved separately or in combination. The derangement of embryogenesis between weeks 2 and 6 results in spinal dysraphism. In 2000, Tortori-Donati et al proposed a new classification system based on neuroradiological and clinical findings and categorized spinal dysraphism into open spinal dysraphism (OSD) and CSD according to skin cover. OSD generally has open defects with exposed neural elements, and CSD generally has fully epithelialized lesions without visible neural tissues. CSD is more common and much more heterogeneous than OSD.

By definition, CSD is covered by skin and may not be obvious by inspection, but associated cutaneous stigmata, such as hairy nevus, capillary hemangioma, dimples, and discolored patches, suggest CSD. Most spinal dysraphism patients are diagnosed at birth or in early infancy, although some patients (especially patients with CSD) are recognized later, even in adulthood, after clinically relevant neurological, urological, or orthopedic symptoms develop. Plain films with anteroposterior and lateral views are important for evaluating the associated abnormalities in vertebral bodies. MRI is critical for detecting and evaluating spinal dysraphism and tethered cord and for determining treatment, including surgical planning.

CSD can occur with or without a subcutaneous mass. CSD without a subcutaneous mass can be further divided into simple or complex dysraphic states, and the intradural lipoma in our patient was a simple dysraphic state. Simple dysraphic states of CSDs without a subcutaneous mass are heterogeneous and the most common abnormalities in children who present with TCS without cutaneous stigmata.

Intradural lipomas originate from the mesoderm and are contained within intact dura. They may be huge or multifocal, are commonly located at the lumbosacral level, and are associated with TCS. Congenital lumbosacral lipomas have little or no growth potential. The lipoma is intradural and hyperintense on MRI T1-weighted images and can be separated from the subcutaneous adipose tissue. Posterior spina bifida, which is the simplest variety of CSD and which commonly occurs at L5 or S1, refers to the defective fusion of the posterior neural arch of the vertebrae. It may be an isolated and incidental finding in 15-30% of the general population, with no clinical significance. However, even minor vertebral defects can be associated with urological dysfunction occasionally.

Tethered cord, which is mostly asymptomatic, results from fixation of the spinal cord or filum terminale to inelastic structures, which is usually a complication after myelomeningocele repair, or presents due to several CSD forms, like the intradural lipoma in our patient. MRI showed an elongated cord, a low-lying conus medullaris (below L3 vertebrae), and posterior displacement of the conus or filum. During the normal spinal range of motion, the tethered cord cannot move freely in the spinal column, and the spinal cord can be injured by acute trauma or repetitive microtrauma. TCS refers to a constellation of progressive clinical symptoms that arise from excessive spinal cord tension and that result in deficient oxidative neuronal metabolism and ischemic spinal cord damage. TCS can be defined as primary or secondary. Primary TCS is congenital and occurs as an isolated anomaly. Secondary TCS is acquired and occurs as a result of other abnormalities, such as spinal dysraphism or retethering after a previous spinal surgery (e.g., surgery for meningomyelocele or intradural lipomas). History and physical examination are hallmarks for diagnosing TCS, and findings suggestive of TCS vary with age. TCS is usually diagnosed in early ages and uncommonly in adult life. Spinal imaging is needed to confirm the diagnosis. The symptoms and signs in adult patients with TCS include pain, progressive motor and sensory dysfunction of the lower limbs (weakness, sensory changes, gait disturbance, or difficulty running), and colorectal (loss of bowel control) and urological deficits (incontinence or retention). Adult patients with TCS experience unrelenting nondermatomal pain with a shock-like feeling, which localizes to the lumbosacral spinal region, lower limbs, or perineal region. Physical examination shows hyperreflexia, spasticity, clonus, positive Babinski sign, and sensory loss at a stage of advanced dysfunction. Before establishing treatment strategies, we should distinguish patients with TCS who have clinically relevant symptoms from patients with tethered spinal cord who have no relevant symptoms.

Surgical guidelines for intradural lipomas with or without tethered cord have not been well established. Surgery should only be considered in patients with
clinical symptoms and signs from a lipoma mass effect or cord tethering, rather than in asymptomatic patients whose MRIs show intradural lipoma with tethered cord\cite{10,13}. The surgical risks in asymptomatic patients might outweigh the benefits\cite{14,15}. The appearance of upper motor neuron signs and the progression of lower motor neuron signs are indications for surgical untethering\cite{7}. The aim of surgery is to restore distal spinal cord mobility in patients with TCS by total or subtotal debulking of intradural lipomas\cite{7,13}.

**CONCLUSION**

For patients with low back pain and coexisting tethered cord, detailed history taking and physical examination are imperative in order to clarify if the pain comes from TCS, in which early surgery is indicated, or from other etiologies that are responsive to conservative treatments. Clinicians must diagnose TCS in a timely fashion through neurological and urological assessments. Patients with tethered cord should be educated about TCS symptoms and signs. Precautions against repetitive trunk flexion and twisting and heavy lifting are commonly advised. Spinal trauma, such as falls on the buttocks, should be avoided, as they may stretch the cord and induce TCS. For asymptomatic simple dysraphic CSD states, clinicians should advise patients to remain active, seek regular multidisciplinary follow-up examinations and urological and neurological dysfunction monitoring, and provide information about self-care measures.

**REFERENCES**

因急性下背痛意外發現成人之隱性脊柱裂及脊髓牽扯：
病例報告

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診斷無合併皮下腫塊(subcutaneous mass)及皮膚特徵(cutaneous stigmata)的隱性脊柱裂(closed spinal dysraphism)常需依賴已經出現之神經學或泌尿道系統相關症狀或徵象。偶爾，它可能是探究其他病因所得到的意外發現。本病例報告描述一位 39 歲的年輕男性發生急性下後背而被診斷為第五腰椎的椎弓解離症(spondylolysis)，腎臟輸尿管及膀胱 X 光顯示在一層椎有脊柱裂(spina bifida)，腰椎磁振造影則顯示在第五腰椎的椎位有一硬腦膜內脂肪瘤(intradural lipoma)，且合併脊髓牽拉(tethered cord)。由於病患未出現脂肪瘤所造成之腫塊效應(mass effect)及脊髓牽拉症候群(tethered cord syndrome)之相關症狀及徵象，病患接受藥物治療一週後，症狀即有改善。我們建議當病患已經出現下肢或泌尿道系統的神經功能異常時才需考慮接受手術治療，而針對沒有症狀的病患，則需教育病患脊髓牽拉症候群之相關神經學症狀，並建議定期門診追蹤，以避免發生永久性之神經功能損傷。（台灣復健醫誌 2013; 41(3): 195 - 199）

關鍵詞：硬腦膜內脂肪瘤(intradural lipoma)，下背痛(low back pain)，脊柱裂(spinal dysraphism)，脊髓牽拉(tethered cord)

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