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Thoracic epidural lipomatosis as a cause of progressive myelopathy: A case report

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Abstract
Case: A 57-year-old patient diagnosed with panhypopituitarism, chronically managed with steroids, developed a dorsal lipomatosis combined with multiple vertebral fractures, leading to progressive dorsal myelopathy. Surgical management was necessary, and the follow-up showed significant improvements in symptoms and quality of life.
Conclusion: Spinal epidural lipomatosis is a condition resulting from excessive fat deposit in epidural space. Patients can be asymptomatic or complain from myelopathic or radicular symptoms. Many etiologies are possible and exogenous steroids excess is one of the most frequent, having a predilection for male sex and thoracic spine. Surgical management is frequently necessary and may provide good results.

Keywords: Epidural lipomatosis, myelopathy, exogenous steroids, spine decompression

Introduction
Spinal epidural lipomatosis (SEL) is a rare condition resulting from excessive fat deposit in epidural space [1, 2]. Clinical presentation may range from asymptomatic patients to complaints resembling spinal stenosis and even cauda equine [3]. Since it was first described by Lee *et al*. in 1975 many etiologies have been described and disagreements still exist about idiopathic cases [1]. Keonhee Kim *et al*. divided SEL in 5 categories according to etiology [1]: Exogenous steroid use, hormonal disease producing excessive endogenous steroid, obesity, surgery induced, and idiopathic. Males are more commonly affected and the thoracic SEL is encountered in around 75% of the cases, especially if caused by exogenous steroids excess [2]. Conservative treatment is an option in the cases where the causative factor may be stopped. However, most of the cases end up developing neurological symptoms and surgical management is required [1, 4]. In this paper, the authors report a case of dorsal lipomatosis in a patient with pan hypopituitarism chronically submitted to a high dose of exogenous steroids that developed progressive myelopathic symptoms and required surgical treatment.

Case Report
A 57-year-old male diagnosed with panhypopituitarism, body mass index 19.6 kg/m^2 (166 cm, 54 kg) followed by an endocrinology team at our hospital and treated with prednisolone 15mg/ day and testosterone 250 mg/ month, came to us with a complaint of back pain and progressive lower extremity weakness for the last six months with no history of trauma.
Physical examination revealed an important walking limitation requiring wheelchair for longer dislocations, the Oswestry Disability Index (ODI) was 40 (80%). There was a decreased pin-prick sensation and sensory level was at T_9/ T_{10}. Deep tendon reflexes were pathologically brisk. He denied any bowel or bladder dysfunction.
On spine X-Ray several, dorsal and lumbar vertebral body fractures were present, but it was not possible to identify if any was recent. (Figure 1)
Magnetic Resonance Imaging (MRI) demonstrated a posterior epidural lipomatosis extending from T_3 to T_9 vertebral levels with a maximum thickness of 8mm at T_5. The collection caused severe compression of the anteriorly displaced spinal cord. Hyper intense signal, consistent with myelopathy, was also noted within the cord at T_4/T_5 and T_8/T_9. (Figure 2)
A Terminal Fillum lipoma was also found and fat deposit at L5 level conditioned a left deviation of sacral roots. (Figure 3) The fractures were not recent based on MRI images.

On investigation, routine laboratory blood tests including complete blood count, liver function, and renal function analysis were within normal ranges. Because of his clinical course and imaging findings, surgical decompression was recommended.

De compressive laminectomies along with ligamentum flavum resection were performed from T_3 to T_9. The adipose tissue was dissected free of the dura, thoroughly coagulated, and removed in a piecemeal fashion. Hemostasis was achieved using thrombin-based, flow able, hemostatic agents. A subfascial wound drain was left in place and removed 2 days later. Histopathological analysis demonstrated mature, adipose tissue.

Postoperative period went without complications, dorsal pain was greatly reduced and on the second day the patient was able to ambulate on his own. He was admitted to a rehabilitation center and continued to show progressive neurological improvement on the following months. The ODI was 15 (30%) after one year. (Figure 4)

Figure format

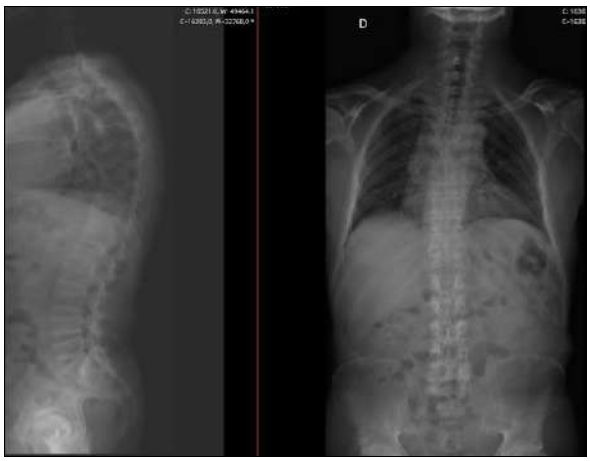

Fig 1: Dorsal and Lombar X-Ray; Multiple vertebral insufficiency fractures, disc space narrowing, osteophytes and body endplates sclerosis contribute to increased thoracic kyphosis

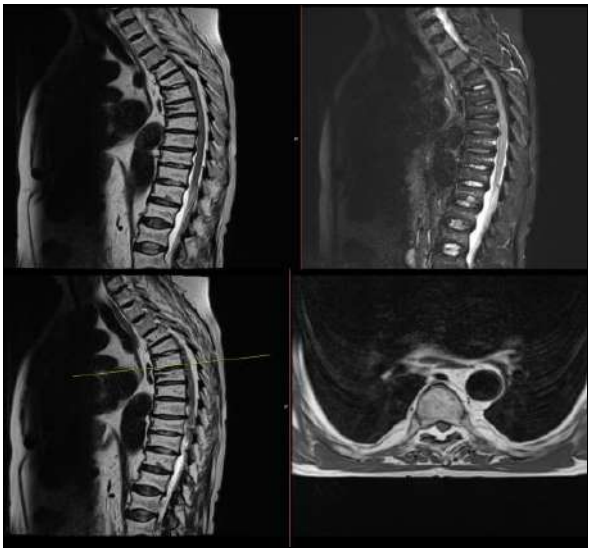

Fig 2: Dorsal MRI; No edema on the fractured vertebral bodies with endplate sclerosis which suggest chronic fractures. Posterior epidural lipomatosis extending from T_3 to T_9 vertebral bodies. At T_4/T_5 and T_8/T_9 an hyper intense signal consistent with myelopathy can be seen

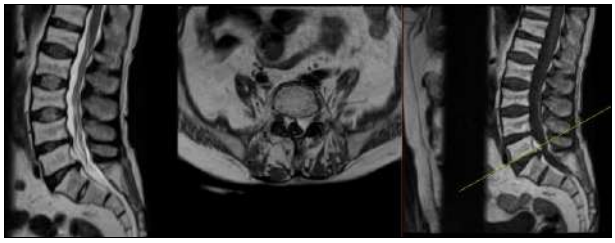

Fig 3: A Terminal Fillum lipoma at L5 level conditioned a left deviation of sacral roots

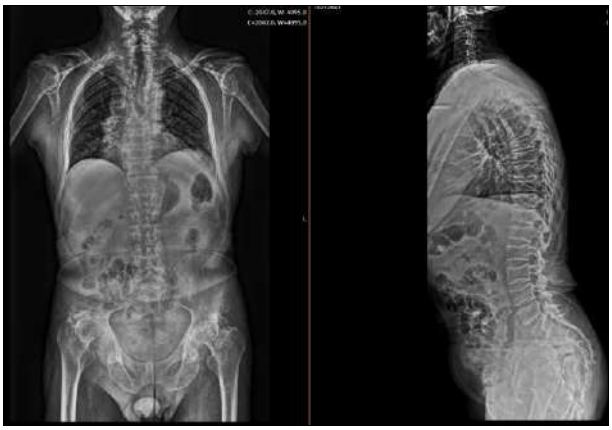

Fig 4: X-Ray one year post-operatively

Discussion

Spinal fatty deposits are rare conditions defined by location and pathologic features that go from lipomyelomeningoceles, intradural spinal lipomas or spinal angiolipomas, to epidural lipomatosis [4]. SEL is the result of an epidural adipose tissue overgrowth within the spinal canal. Despite affecting man more often, lumbar localized deposits tend to appear more commonly in woman [4]. Exogenous steroid use represents more than half of the cases and results from a chronic steroids usage in high doses. Some authors defined these high doses as 30 to 100 mg per day, on average, for a duration of 5 to 11 years (2) but others failed to reach statistical significant values. Moreover, reports of acute development of SEL with doses as low as 15mg/ day of prednisolone for a short period of time may imply that host's sensitivity to steroid as well as medication formulation, and cumulative dose, may also play a role in its development. [1, 5, 6]. Other hormones can also interact with the glucocorticoid receptor, and treatment with testosterone has been associated with cases of SEL [6]. Pan hypopituitarism requires chronic steroid and testosterone administration, and our patient was under a chronic regime of prednisolone plus testosterone that may put him at higher risk of developing SEL.

Exogenous steroid's induced SEL has been found to have a propensity for thoracic spine [3, 4]. The exact mechanism is still unknown, but mechanic compression and ischemia are the most accepted explanations as the hypertrophic adipose tissue, often necrotic, increases epidural pressure and compression of dural sac and nerve roots leading to progressive radicular and/ or myelopathic symptoms [3, 4, 7]. Interestingly, Al-Khawaja *et al*. stated that patients with steroid-related SEL had higher rates of paraplegia on presentation compared with non–steroid-related SEL: 25% versus 5%. [8] Despite not having provided an explanation for that, the propensity for the thoracic spine together with a smaller canal diameter may justify, in part, the higher rates

of paraplegia in patients with excess of steroids. Besides that, these patients are prone to compressive spinal fractures which, in some cases, are the cause of an acute-onset paraplegia and the need for surgical decompression and fixation [1]. Sensory changes with numbness, paresthesias or radicular symptoms are also frequent but bowel and bladder incontinence are rare complaints [4]. Back pain is the most common complain, normally lasting for several months or even years [4, 7]. The subject of our case report presented to us with several insufficiency fractures of the spine that were thought to be the cause of back pain and only with the MRI could we understand that the major concern were the epidural fat deposits.

MRI is the most useful diagnosing method, compression can be seen on different views and adipose tissue is seen as high-signal intensity on T_1-weighted images and a low signal on T_2-weighted images. Epidural adipose tissue that has a thickness greater than 7 mm has been reported to be the diagnostic criterion for SEL [3, 4].

Removal of the causative factor is the basis of the conservative treatment and weaning from steroids has been reported to be successful in numerous cases [1, 4]. However, many patients do not tolerate leaving the steroid medication or the onset of the symptoms is fast and severe, making a surgical decompression the most appropriate treatment [1]. Previous works reported good outcomes with de compressive surgery using laminectomy or laminotomy with half of the patients improving patient-related outcome scores [9]. Is important to notice the high mortality rates, in part, as a result of frail patients submitted to chronic immunosuppressive medication, that make most authors to propose a conservative measure whenever possible [10]. With that in mind, we propose our patient to laminectomies of the spine levels with evident compression. There was a major concern in being the less aggressive possible, we preserved all the articular processes and believed that, despite the extensive ($T_3 – T_9$) decompression, the rib cage would have a major role in stabilizing this segment without progressive deforming kyphosis. The follow-up proved it to be an adequate strategy with reasonable clinical outcomes and good radiological results.

Conclusion

SEL is a debilitating condition that cannot be undermined. The spine surgeon should be aware of this possibility and only by knowing its various etiologies can he ensure the best investigation and treatment strategies. Conservative measures with medication adjustments are crucial to manage an already frail patient. However, onset of symptoms may be sudden, severe, or progressive and prompt surgical intervention with laminectomies may be the most appropriate option. Prognosis after surgical treatment is variable and is impacted by multiple factors, including severity of preoperative neurological deficits though a better functional status is to be expected.

Conflict of Interest

Not available

Financial Support

Not available

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