An Unusual Case Report of Bertolotti's Syndrome: Extraforaminal Stenosis and L5 Unilateral Root Compression (Castellvi Type III an LSTV)

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What to Learn from this Article?

Although being a congenital lesion, lumbosacral transitional vertebrae and Bertolotti's syndrome may be presented in elderly population with sciatica and low back pain.

Abstract

Introduction: Castellvi Type III lumbosacral transitional vertebrae (LSTV) is an unusual case of Bertolotti's syndrome (BS) due to extraforaminal stenosis, especially manifesting in elderly patients.

Case Report: We report a case of BS in a 62 years old Greek female. The signs of the clinical examination are low back pain, sciatica, hypoesthesia, and pain to the contribution of L5 nerve. Imaging techniques revealed an LSTV Type III a (complete sacralization between LSTV and sacrum).

Conclusion: Despite the fact that LSTV is a congenital lesion, the clinical manifestation of BS may present in the elderly population. The accumulative effect of the gradual degeneration of intervertebral foramen (stenosis) may lead to the compression of extraforaminal portion of the nerve root.

Keywords: Bertolotti's syndrome, lumbosacral transitional vertebrae, low back pain, extraforaminal stenosis, classification of lumbosacral transitional vertebrae.

Introduction

In 1917, Mario Bertolotti described the presence of an anatomical variation of the most caudal lumbar vertebrae, with an abnormally enlarged transverse process which articulated or fused unilaterally or bilaterally with the sacrum basis or iliac crest. This "mega" apophysis or the so called lumbosacral transitional vertebrae (LSTV), when associated with chronic, persistent low back pain (LBP) or sciatica, composes the "Bertolotti's Syndrome" (BS) [1, 2, 3, 4, 5]. The prevalence of LSTV varies from 3% to 35% in the general population [1, 2, 6, 7, 8, 9]. Although a 13% of the patients with LSTV are asymptomatic, the incidence of BS is diagnosed in

Access this article online

Website: www.jocr.co.in

DOI: 2250-0685.782

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Journal of Orthopaedic Case Reports | pISSN 2250-0685 | eISSN 2321-3817 | Available on www.jocr.co.in | doi: 10.13107/jocr.2250-0685.782

4-8% of patients with LBP. BS is more frequently encountered in patients <30 years old, in a percentage of 18.5% [2, 3, 4]. The etiology of LBP in BS is multifactorial. LBP is associated with changes at the intervertebral disk, such as early degeneration or herniation, at the spinal canal above the level of the LSTV, at the contralateral facet joints in cases of unilateral fusion, or extraforaminal changes at the site of articulation between LSTV and sacrum ala/iliac crest [3, 4, 5, 6]. We present an unusual case of Castellvi Type III an LSTV, which causes extraforaminal stenosis and impingement of the L5 nerve root, leading to BS.

Case Report

A 62-years-old woman presented to our Department in November 2016, with a history of LBP for 3 months and sciatica for the previous month. She mentioned the systematic use of analgesics (paracetamol), with no subsequent pain relief. Her past medical history was unremarkable for comorbidities, except of the presence of a cardiac pacemaker, due to sinus bradycardia. The use of magnetic resonance imaging (MRI) was, thus, contraindicated. The patient was subjected to a complete clinical examination and laboratory testing. During the physical examination, hypoesthesia and pain found at the distribution of L5 nerve, without signs of muscular weakness in the lower extremity (all reflexes were normal). Afterward, plain radiograph of lumbar spine region depicted a unilateral LSTV on the right side, which was in contact with the base of sacrum (Fig. 1). To elucidate the morphology and the possible presence of pseudoarthrosis or fusion between the LSTV and the sacrum, a three-dimensional reconstruction of computed tomography scans (three-dimensional [3D] CT) was conducted (Fig. 2, 3). 3D CT scan images revealed a complete unilateral sacralization (Type III a, according to Castellvi *et al.* classification). Nevertheless, the findings were consistent with extraforaminal stenosis due to the accumulative effect of the gradual degeneration of intervertebral foramen and subsequent nerve root compression at the corresponding level. Inflammatory markers were normal in the blood routine examination. Our findings from physical examination and CT scan imaging were suggestive of BS. The patient was initially prescribed with nonsteroidal anti-inflammatory drugs (NSAIDs) for 7 days, followed by a 2-week therapy with methylprednisolone. She was subjected to follow-up to detect any deterioration of the neurological symptomatology or the LBP. In follow-up, she did not report any symptoms, and in the clinical examination, we found a light mubness at the distribution of L5 nerve.

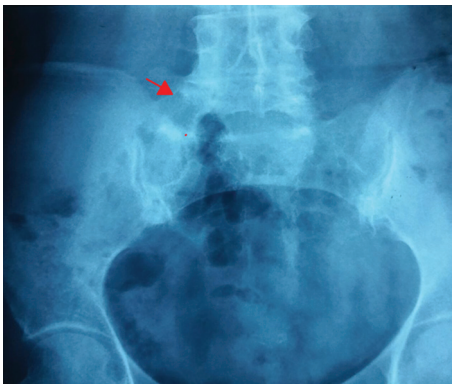

Figure 1: Antero posterior pelvic radiograph. Lumbosacral transitional vertebrae (red arrow).

Discussion

According to Castellvi *et al.*, LSTV is classified with a Ferguson radiograph, which allows a 30° angled anteroposterior (AP) view of the lumbosacral joint, into four types (Fig. 4) based on its morphological characteristics (Table 1). Type I exhibits unilateral (I a) or bilateral (I b) dysplastic transverse processes, measuring at least 19 mm at the craniocaudal axis. Type II includes incomplete unilateral (II a) or bilateral (II b) lumbarization/sacralization with an enlarged transverse process that has a diarthrosis between itself and the sacrum. Type III involves unilateral (III a) or bilateral (III b) lumbarization/sacralization with complete osseous fusion of the transverse process (es) to the sacrum. Type IV describes a unilateral Type II transition with the coexistence of a Type III at the contralateral side [1, 4, 5, 6]. As it was remarked in the literature, Types II-IV appear to be related with BS, while Type I remains asymptomatic and in the majority of cases incidentally found. The prevalence of Type III corresponds to a percentage of 8.3-11.5% [1, 8]. The anatomical variations of LSTV are present in varying frequencies, and their prevalence seems to alter among males and females [1, 8]. Especially, Nardo *et al.* commented on the higher prevalence of LSTV among males rather than females.

According to various authors, the appearance of LSTV is associated with adjacent level disc degeneration, potentially due to the distribution of abnormal biomechanical stresses above the fusion [1, 2, 10, 11]. Otani *et al.* noticed that the prevalence of disc herniation was higher in patients positive to LSTV (17%), compared to the control group without LSTV (11%) [10]. Castellvi *et al.* claimed that disc degeneration seems to be associated with LSTV. In addition to this, the incidence of disc degeneration was higher in the supradjacent level to LSTV (63.5%) and lower in the infradjecent level to LSTV (34%) [1]. Elster found that the incidence of disc degeneration and spinal stenosis was significantly higher in the disc superior the LSTV, compared to other discs [2]. In addition, Aihara *et al.* investigated the role of the iliolumbar ligament in the disc degeneration. According to the study, the iliolumbar ligament

Table 1: Classification of LSTV as proposed from Castellvi

Types	Morphology
I	Dysplastic transverse process (unilateral or bilateral)
II	Incomplete lumbarization/sacralization (unilateral or bilateral)
III	Complete lumbarizarion/sacralization (unilateral or bilateral)
IV	Combination (Types II and III)

LSTV: Lumbosacral transitional vertebrae

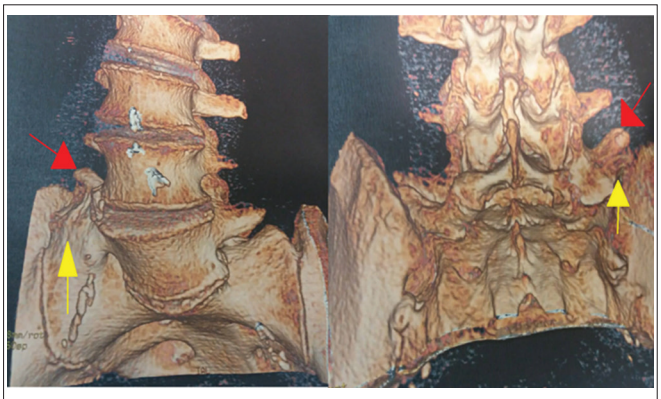

Figure 2: Three-dimensional reconstruction of computed tomography (anterior-posterior surface). Fusion between lumbosacral transitional vertebrae (LSTV) and sacrum (yellow arrows), LSTV (red arrow).

Figure 3: Three-dimensional reconstruction of computed tomography (anterior surface). Sacralization (red arrow), lumbosacral transitional vertebrae (yellow arrow).

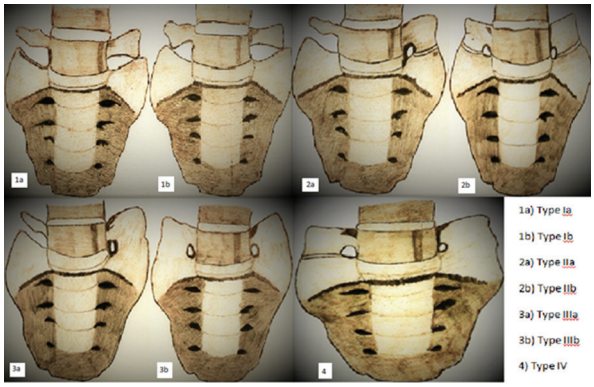

Figure 4: Lumbosacral transitional vertebrae Types I-IV.

was thinner and weaker in the level above LSTV, in patients who suffered from disc degeneration in this exact level [11]. The knowledge of the pathophysiology can explain the mechanism of the disc degeneration.

The presence of LSTV interrupts the anatomy of the spine. Altered anatomy imposes difficulties in the surgical approach of the lumbosacral region [5]. Chithriki *et al.* conducted a prospective study of 441 patients, estimating the prevalence of LSTV at 8, 4%. In patients with sacralization of the L5 vertebra, the level of aortic bifurcation was found at L3 in 59%, at L4 in 23% and at the mid-vertebral level L3/L4 in 14%, while in those with lumbarization of the S1 vertebra, at L4 in 47%, at the mid-vertebral L4/L5 level in 33% and at the L5 in 13% [7]. Furthermore, Kim *et al.* concluded that the Tuffier's line (line connecting the highest points of iliac crests) was not found in its ordinary position, which is at the L4 or L5 body. The finding is of great clinical significance during needle insertion in the lumbar region [12]. Weiner *et al.* mentioned that the junction between common iliac veins and inferior vena cava was located lower than its ordinary location at the L5 intervertebral level, anteriorly to the vertebral body and right to the midline, leading to potential complication during the anterior approach of the L4-L5 intervertebral disc [13].

The diagnosis of BS is based on clinical examination and imaging of the lumbosacral spine (AP radiographs, CT, MRI, and skeletal scintigraphy). AP radiographs are useful for identifying the presence of LSTV but are nonspecific to determine whether the source of pain is related to LSTV [5, 7, 9, 11, 14]. On the other hand, MRI scans are highly reliable [3, 6, 9, 12]. CT-scan provides information on the degenerative lesions of the osseous structures (hypertrophy or bone sclerosis) [4]. In addition to CT-scan, there is single-photon emission computed tomography/CT for the detection of bone abnormalities due to stress [4, 5, 14, 15]. MRI reveals the disc herniation or potential edema of the articulation of the LSTV with the sacrum, which may coexist in patients suffering from BS [4. 10, 11, 14]. 3D MRI and CT are highly reliable for diagnosing L5 nerve compression due to extraforaminal stenosis by LSTV [4]. Skeletal scintigraphy depicts an increase on bone metabolic activity in certain foci [4, 5, 14].

The therapeutic approach of BS is conservative treatment and surgery [5, 6, 9, 14, 15]. The first-line therapeutic option is the conservative treatment, which consists of physical therapy, NSAIDs, corticosteroids and muscular relaxants [4, 5, 6, 14, 15]. Especially, corticosteroids and local anesthetic drugs can be injected into the articulation for both diagnosis (determination whether or not the pain originates from the pseudarthrosis) and treatment [6, 14, 15]. If so, and if conservative therapy is unsuccessful, surgical resection or fusion of LSTV is required [6, 14, 15].

Conclusion

Despite the fact that LSTV is a congenital lesion, the clinical manifestation of BS may present in the elderly population. The accumulative effect of the gradual degeneration of intervertebral foramen (stenosis) may lead to the compression of extraforaminal portion of the nerve root. Spine physicians should bear in mind that the symptomatology from LSTV may not only be presented in young population. BS should be a part of differential diagnosis in patients with LBP and spinal stenosis in the most caudal level of spine.

Clinical Message

LSTV is a congenital lesion that leads to subsequent nerve root impingement and BS. Although it is more commonly encountered in young people, it should be included in the differential diagnosis in elderly population who present with LBP and sciatica.

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Conflict of Interest: Nil
Source of Support: None

How to Cite this Article

Kapetanakis S, Chaniotakis C, Paraskevopoulos C, Pavlidis P. An Unusual Case Report of Bertolotti's Syndrome: Extraforaminal Stenosis and L5 Unilateral Root Compression (Castellvi Type III an LSTV). Journal of Orthopaedic Case Reports 2017 May-Jun;7(3):9-12.