TRANSITIONAL LUMBOSACRAL VERTEBRAE AND LOW BACK PAIN

Diagnostic pitfalls and management of Bertolotti's syndrome

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Abstract – Objective: Bertolotti's syndrome is a spine disorder characterized by the occurrence of a congenital lumbar transverse mega-apophysis in a transitional vertebral body that usually articulates with the sacrum or the iliac bone. It has been considered a possible cause of low back pain. Method: We analyzed the cases of Bertolotti's syndrome that failed clinical treatment and reviewed the literature concerning this subject. Results: Five patients in our series had severe low back pain due to the neo-articulation and two of them were successfully submitted to surgical resection of the transverse mega-apophysis. Taking into account the clinical and surgical experience acquired with these cases, we propose a diagnostic-therapeutic algorithm. Conclusion: There is still no consensus about the most appropriate therapy for Bertolotti's syndrome. In patients in whom the mega-apophysis itself may be the source of back pain, surgical resection may be a safe and effective procedure.

KEY WORDS: low back pain, lumbosacral region, spine.

Dor lombar associada à vértebra de transição lombossacra: dificuldades no diagnóstico e manejo da síndrome de Bertolotti

Resumo – Objetivo: A síndrome de Bertolotti é uma desordem congênita da coluna vertebral caracterizada pela ocorrência de uma mega-apófise transversa lombar em uma vértebra de aspecto transicional, que geralmente se articula com o sacro ou com o osso ilíaco. Tal síndrome tem sido considerada possível causa de dor lombar. Método: Análise dos casos de síndrome de Bertolotti que apresentavam dor lombar sem melhora com tratamento conservador e revisão dos artigos publicados. Resultados: Foram revisados cinco pacientes que não apresentaram melhora com o tratamento clínico, sendo que dois foram submetidos à ressecção cirúrgica da mega-apófise transversa. Considerando a experiência adquirida com estes casos, os autores propõem um algoritmo para diagnóstico e tratamento da Síndrome de Bertolotti. Conclusão: Ainda não há consenso sobre qual é a terapia mais apropriada para a Síndrome de Bertolotti. Em pacientes em que a mega-apófise parece ser a origem da lombalgia, a ressecção cirúrgica parece ser um procedimento seguro e efetivo.

PALAVRAS-CHAVE: dor lombar, região lombossacral, coluna vertebral.

Bertolotti's syndrome is characterized by the presence of a large transverse apophysis in one or both sides of a lumbar transitional vertebra that articulates with the sacrum or with the iliac bone. "Transitional vertebra" is anatomically defined as a vertebra that shares similar features with both upper and lower vertebral segments. In Bertolotti's syndrome, the posterior arch or transverse apoph-

ysis of the vertebra usually has both lumbar and sacral characteristics, occurring most commonly at L5, but can also occur at L6.

The majority of patients are asymptomatic^{1,2} and despite its first description dates from 1917³, little is yet known about the biomechanical effects of such abnormal vertebra and its relation to low back pain. It has been

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Received 25 August 2008, received in final form 5 December 2008. Accepted 18 February 2009.

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assumed that progressive modifications in the biomechanics of the spine can occur, generating abnormal weight overload in the articular facets and adjacent intervertebral discs resulting in degeneration⁴. Although Bertolotti's syndrome is a congenital abnormality, it is often clinically recognized only after the second decade of life⁵. Nevertheless, there is evidence that a typical transverse megaapophysis may be the cause of low back pain in the region of the neo-articulation⁶.

Due to the multifactorial etiology of low back pain in patients with Bertolotti's syndrome, there are several therapeutic options, whereas the most appropriate treatment for each case, as well as the specific role of surgical resection of the transverse mega-apophysis in the therapeutic armamentarium, still remain a matter of controversy.

METHOD

A literature review of the Bertolotti's syndrome was carried out through the Medline $^{\rm TM}$ database. The following keywords were used: Bertolotti's syndrome, lumbosacral transitional vertebra and lumbar back pain.

Thereafter, we analyzed five patients diagnosed with Bertolotti's syndrome and taking into account the clinical and surgical experience acquired with these cases, we propose a diagnostictherapeutic algorithm in order to guide the surgeon through the decision making process involved in the evaluation of back pain and its possible relationship with a transitional vertebra.

RESULTS

A total of 31 articles were retrieved from the MedlineTM database. Of them, we selected only studies written in English regarding diagnosis and treatment of Bertolotti's syndrome, which resulted in 18 articles revised^{1,2,6-21}.

Among the patients with low back pain and transverse mega-apophysis followed in our service from 2000 to 2006, five of them did not improve with clinical treatment, including non-steroidal anti-inflammatory drugs (NSAIDs), physiotherapy and physical exercises. These patients were thus submitted to a therapeutic test with anesthetic block at the contact of the mega-apophysis with the sacrum, which allowed temporary pain relief in all of them. Thereafter, they underwent a radiofrequency neurolisys, whereby three had only a partial control of the pain and two had significant improvement. The latter two patients had the mega-apophysis surgically resected and became asymptomatic after the procedure and during the one year-follow up. Their cases are reported in order to illustrate the possible pathways proposed in the diagnostictherapeutic algorithm of Bertolotti's syndrome as follows.

Case 1

A 39 year-old woman was admitted to the hospital complaining of a 5-year severe low back pain. The pain

was described as being constant, dull, and localized on the paramedian right side. It worsened when the patient performed extension or lateralization of her back to the same side of the mega-apophysis, stood for more than 30 minutes or after physical exertion. Despite the use of NSAIDs and opioids, the pain remained unchanged. Imaging exams showed a transverse mega-apophysis on the right side at L5 that corresponded exactly to the point of pain and tenderness. Magnetic resonance imaging (MRI) depicted no major disc degeneration or other vertebral abnormalities. An anesthetic lumbar block test was performed at the neo-articulation point, which ensured total analgesia for 2 hours. Only a small volume (2 ml) of lidocaine was injected into the site, avoiding any spreading of the solution. After 2 hours of relief, the pain returned and remained exclusively in the region related to the right transverse mega-apophysis. It was then decided to perform a surgical resection of the transverse megaapophysis. There were no complications, and the patient was discharged from the hospital two days after the procedure with 90% improvement in low back pain; at the six month-follow up, she had no low back pain at all.

Case 2

Female, 32 years of age, presented a history of unilateral right-sided low back pain lasting for the previous 4 years. The pain increased when climbing stairs and sitting, and was relieved when lying down. There were no complains of paresthesias or hypoesthesias and no major radiation. Neurological examination was normal. The imaging exams included lumbosacral radiographs, a computed tomography (CT)-scan and a MRI, which revealed bilateral transverse mega-apophysis of L5, articulating with the sacrum. Despite the bilateral image findings the patient showed only major symptoms on her right side (Figs 1 and



Fig 1. Anteroposterior X-ray film showing a transverse mega-apophysis of L5 in the right side (circle).

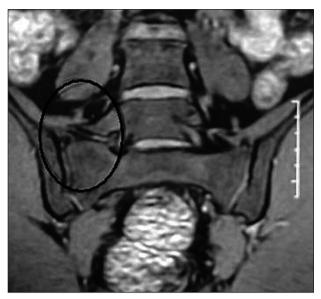


Fig 2. Coronal section magnetic resonance imaging of the lumbosacral region demonstrating contact of the transverse mega-apophysis in the right side of L5 with the sacrum (circle).

2). She was clinically treated without success for three years at which point she underwent an anesthetic block of the lumbosacral articulation on the symptomatic side, resulting in complete remission of pain for two hours. Following this outcome, a radioscopy-guided radiofrequency denervation of the abnormal articulation was performed, resulting in 70% decrease in pain for four months. It was then decided to resect the symptomatic transverse megaapophysis, which led to total resolution of the pain. After this procedure, the patient remained asymptomatic.

DISCUSSION

In 1917, Mario Bertolotti, an Italian surgeon, was the first author to put forward a possible relationship between low back pain and congenital anatomical abnormalities in the last lumbar vertebra, described as "sacral assimilation of the lumbar vertebrae"³. Since then, the term Bertolotti's syndrome has been used to define the presence of a transverse mega-apophysis in a lumbar vertebra with transitional characteristics associated with low back pain²².

Four to seven percent of patients with low back pain present transitional vertebrae^{1,21,23,24}. Castellvi et al.²³ analyzed 200 patients with myelographic findings of lumbar disc herniation and discovered vertebrae with transitional characteristics in 60 cases. The authors proposed a classification of four groups based on the morphological characteristics of such abnormal vertebra.

In a recent study including patients with degenerative disc disease (DDD), a higher incidence of transitional vertebra was found in the youngest group (present in up to 10% of the patients under 30 years of age with DDD) in comparison to the general population⁵.

There is a paucity of studies on biomechanical effects associated with the occurrence of transitional vertebrae. It has been hypothesized that motion between the transitional vertebra and the sacrum is reduced and asymmetrical. This asymmetry can result in early degenerative changes within the "neoarticulation" or in the normal contralateral facet joint², giving rise to facet pain⁴.

In addition to the reduction in motion, it is assumed that biomechanical stress transferred to the upper mobile vertebral segment can accelerate the disc degeneration^{22,23,25}. Elster et al.² reported higher incidences of disc herniations linked to degenerative disc disease above the transitional segment. Conversely, the disc between the transitional vertebra and the sacrum is usually fibrotic and contains little nuclear material. Tini et al.²¹ have demonstrated that very few disc herniations occur at this level. In fact, recent series have confirmed that lumbar discs immediately above the transitional vertebra have a higher incidence of degeneration and those between the transitional vertebrae and the sacrum are significantly less likely to degenerate²⁵.

Diagnostic imaging of low back pain usually includes X-rays, CT-scan and MRI. For the evaluation of transitional vertebrae, X-ray films in anteroposterior, lateral and oblique incidences are of special importance. Computed tomography may be helpful in the identification of associated stenosis, osteophytes and areas of sclerosis surrounding the contact point of the mega-apophysis with the lateral iliac or sacral bone. Magnetic resonance imaging is the standard method for studying DDD and possible associated disc herniations that have been demonstrated to occur more frequently in Bertolotti's syndrome.

Bone scintigraphy may reveal an inflammatory process within the articular facets, specifically at the level of the mega-apophysis⁸, although some authors believe that arthritis is not the direct cause of pain in all patients, as many subjects with Bertolotti's syndrome are young and have "cold" bone scans²². Single-photon emission computed tomography may be useful in the identification of possible candidates for local anesthetic infiltration and future radiofrequency blocks²⁶.

The initial treatment of Bertolotti's syndrome, as with other causes of low back pain, is clinical, including a combination of NSAIDs and rehabilitative physical therapy. The performance of anesthetic blocks at the articulation level between the mega-apophysis and the sacrum or iliac bone may be effective in temporarily relieving pain in some cases. This was useful in the identification of the origin of the pain, although no previous study confirmed a prognostic value of this procedure. These blocks should be performed with a minimal amount of anesthetic delivered precisely to the point of interest.

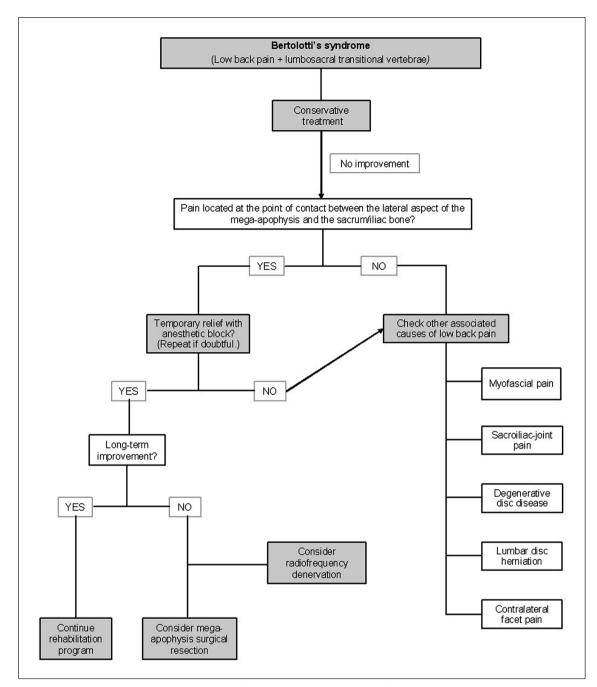


Fig 3. Proposed diagnostic-therapeutic algorithm for evaluation and treatment of Bertolotti's syndrome.

In our experience, conventional radiofrequency neurolysis proved to be a safe, non-invasive procedure for those cases that had improved with anesthetic blocks, providing significant pain relief and aiding future physical rehabilitation programs⁶. Nevertheless, the relief may be temporary, whereby major studies concerning about the efficacy of radiofrequency denervation are required. Surgical resection of the transverse mega-apophysis at the symptomatic site may be indicated in those refractory cases associated with degenerated facets²⁷. Our patients experienced a complete remission of the symptoms fol-

lowing surgery and remained asymptomatic until the end of the follow up period.

Degenerative disc disease is managed according to the indications and particularities of each case. The therapeutic armamentarium for DDD includes surgical microdiscectomy, nucleolysis and arthrodesis⁵. The latter involves pedicular screws, transforaminal lumbar interbody fusion (TLIF), posterior lumbar interbody fusion (PLIF) and, more recently, anterior lumbar interbody fusion (ALIF). (See proposed diagnostic-therapeutic algorithm.)

Despite the existence of a significant number of as-

ymptomatic patients, transitional vertebrae associated with abnormal transverse mega-apophysis may be the cause of low back pain in others. This symptom may be related to progressive modifications in the biomechanics of the spine, generating abnormal weight overload in articular facets and adjacent intervertebral disc degeneration. Abnormal lateral contact of transverse mega-apophysis with sacrum or iliac bone can also be the source of local pain.

Perhaps due to its rare occurrence, the pathophysiology of Bertolotti's syndrome is still obscure and there is no consensus about the most appropriate therapy for each patient. Due to its multifactorial causes and the common findings of low back pain in the general population, it becomes essential to differentiate between low back pain due to transverse mega-apophysis contact with the sacrum from other sources of back pain in patients with Bertolotti's syndrome. Based on our experience we propose an algorithm in order to identify a subgroup of patients with Bertolotti's syndrome in which the transverse mega-apophysis could be assigned as the cause of low back pain; consequently, these patients might benefit from surgical resection.

REFERENCES

- Brault JS, Smith J, Currier BL. Partial lumbosacral transitional vertebra resection for contralateral facetogenic pain. Spine 2001;26:226-229.
- 2. Elster AD. Bertolotti's syndrome revisited. Transitional vertebrae of the lumbar spine. Spine 1989;14:1373-1377.
- Bertolotti M. Contributo alla conoscenza dei vizi di differenzazione regionale del rachide con speciale riguardo all assimilazione sacrale della v. lombare. Radiol Med 1917;4:113-144.
- 4. Avimadje M, Goupille P, Jeannou J, et al. Review article. Can an anomalous lumbo-sacral or lumbo-iliac articulation cause low back pain? A retrospective study of 12 cases. Rev Rhum Engl Ed 1999;66:35-39.
- Quinlan JF, Duke D, Eustace S. Bertolotti's syndrome: a cause of back pain in young people. J Bone Joint Surg 2006;88: 1183-1186.
- Endo K, Ito K, Ichimaru K, et al. A case of severe low back pain associated with Richard disease (lumbosacral transitional vertebra). Minim Invasive Neurosurg 2004;47:253-255.
- Brenner AK. Use of lumbosacral region manipulation and therapeutic exercises for a patient with a lumbosacral transitional vertebra and low back pain. J Orthop Sports Phys Ther 2005; 35:368-376.
- Connolly LP, d'Hemecourt PA, Connolly SA, et al. Skeletal scintigraphy of young patients with low back pain and a lumbosacral transitional vertebra. J Nucl Med 2003;44:909-914.
- 9. Dai L. Lumbosacral transitional vertebrae and low back pain. Bull Hosp Jt Dis 1999;58:191-193.
- 10. Delport EG, Cucuzzella TR, Kim N, et al. Lumbosacral transi-

- tional vertebrae: incidence in a consecutive patient series. Pain Physician 2006;9:53-56.
- 11. Hughes RJ, Saifuddin A. Numbering of lumbosacral transitional vertebrae on MRI: role of the iliolumbar ligaments. AJR Am J Roentgenol 2006;187:59-65.
- 12. Hughes RJ, Saifuddin A. Imaging of lumbosacral transitional vertebrae. Clin Radiol 2004;59:984-991.
- 13. Luoma K, Vehmas T, Raininko R, et al. Lumbosacral transitional vertebra: relation to disc degeneration and low back pain. Spine 2004;29:200-205.
- 14. MacLean JG, Tucker JK, Latham JB. Radiographic appearances in lumbar disc prolapse. J Bone Joint Surg Br 1990;72:917-920.
- 15. Marks RC, Thulbourne T. Infiltration of anomalous lumbosacral articulations. Steroid and anesthetic injections in 10 back-pain patients. Acta Orthop Scand 1991;62:139-141.
- 16. O'Driscoll CM, Irwin A, Saifuddin A. Variations in morphology of the lumbosacral junction on sagittal MRI: correlation with plain radiography. Skeletal Radiol 1996;25:225-230.
- 17. Otani K, Konno S, Kikuchi S. Lumbosacral transitional vertebrae and nerve-root symptoms. J Bone Joint Surg Br 2001;83: 1137-1140.
- Pekindil G, Sarikaya A, Pekindil Y, et al. Lumbosacral transitional vertebral articulation: evaluation by planar and SPECT bone scintigraphy. Nucl Med Commun 2004;25:29-37.
- Peterson CK, Bolton J, Hsu W, et al. A cross-sectional study comparing pain and disability levels in patients with low back pain with and without transitional lumbosacral vertebrae. J Manipulative Physiol Ther 2005;28:570-574.
- Taskaynatan MA, Izci Y, Ozgul A, et al. Clinical significance of congenital lumbosacral malformations in young male population with prolonged low back pain. Spine 2005;30:210-213.
- 21. Tini PG, Wieser C, Zinn WM. The transitional vertebra of the lumbosacral spine: its radiologic classification, incidence, prevalence, and clinical significance. Rheumatol Rehabil 1977; 16:180-185.
- 22. Jönsson B, Strömqvist B, Egund N. Anomalous lumbosacral articulations and low back pain. Evaluation and treatment. Spine 1989;14:831-834.
- Castellvi AE, Goldstein LA, Chan DP. Lumbosacral transitional vertebrae and their relationship with lumbar extradural defects. Spine 1984;9:493-495.
- 24. MacGibbon B, Farfan HF. A radiologic survey of various configurations of the lumbar spine. Spine 1979;4:258-266.
- Aihara T, Takahashi K, Ogasawara A, et al. Intervertebral disc degeneration associated with lumbosacral transitional vertebrae: a clinical and anatomical study. J Bone Joint Surg 2005; 87:687-691.
- 26. De Maeseneer M, Lenchik L, Everaert H, et al. Review article. Evaluation of lower back pain with bone scintigraphy and SPECT. Radiographics 1999;19:912-914.
- 27. Santavirta S, Tallroth K, Ylinen P, et al. Surgical treatment of Bertolotti's syndrome: follow-up of 16 patients. Arch Orthop Trauma Surg 1993;112:82-87.