

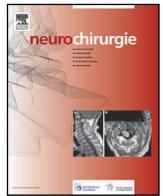


Disponible en ligne sur

ScienceDirect  
www.sciencedirect.com

Elsevier Masson France

EM|consulte  
www.em-consulte.com



Short clinical case

# Bertolotti's syndrome in children: From low-back pain to surgery. A case report

C. Cuenca<sup>a</sup>, J. Bataille<sup>a</sup>, M. Ghouilem<sup>a</sup>, Q. Ballouhey<sup>a</sup>, L. Fourcade<sup>a</sup>, P.-S. Marcheix<sup>b,\*</sup>

<sup>a</sup> Department of Pediatric Surgery, Dupuytren University Hospital, 2, Avenue Martin Luther King, 87042 Limoges cedex, France

<sup>b</sup> Department of Orthopedic Surgery, Dupuytren University Hospital, 2, Avenue Martin Luther King, 87042 Limoges cedex, France

## ARTICLE INFO

### Article history:

Available online 10 July 2019

### Keywords:

Bertolotti's syndrome  
L5 transverse mega-apophysis  
Children  
Low-back pain  
Surgery

## ABSTRACT

Bertolotti's syndrome is a little-known and little-discussed pathology. We report the case of a 13-year-old child diagnosed with Bertolotti's syndrome after several years of functional complaints. Conventional radiography was used to diagnose the transverse mega-apophysis of L5, while sectional and functional imaging confirmed a lumbosacral-iliac impingement. In view of the transient efficacy of medical management, surgical resection of the transverse mega-apophysis was performed. The medium-term decline in symptoms was excellent and the patient resumed physical activities without limitation or pain.

© 2019 Elsevier Masson SAS. All rights reserved.

## 1. Introduction

Bertolotti's syndrome, first described in 1917 [1,2], comprises lumbosacral or gluteal pain and transitional lumbosacral morphological anomaly. This anomaly, which is always congenital, is characterized by a transverse mega-apophysis of L5 merging or articulating with the iliac wing and/or sacrum, and can be uni- or bi-lateral [1,3–6].

The radiological diagnosis of lumbosacral transitional vertebrae (LSTV) is quite straightforward. However, the difficulty in managing this pathology lies in the problem of finding and defining the exact origin of the pain. Complementary investigations, such as magnetic resonance imaging (MRI) and bone scintigraphy can be conducted, and sometimes an infiltration test may be required for both diagnostic and therapeutic purposes [7].

The treatment of Bertolotti's syndrome in children remains controversial and ranges from physiotherapy to surgical treatment by resection or fusion, including medical treatment by infiltration [4,6–9]. No decision-tree has been described for children or adolescents in the current literature. Furthermore, while the adult management is well-described from a diagnostic and therapeutic point of view, there are no published case-reports of surgical management in children. Here, we report the first case of a child surgically treated for proven Bertolotti's syndrome.

## 2. Observations

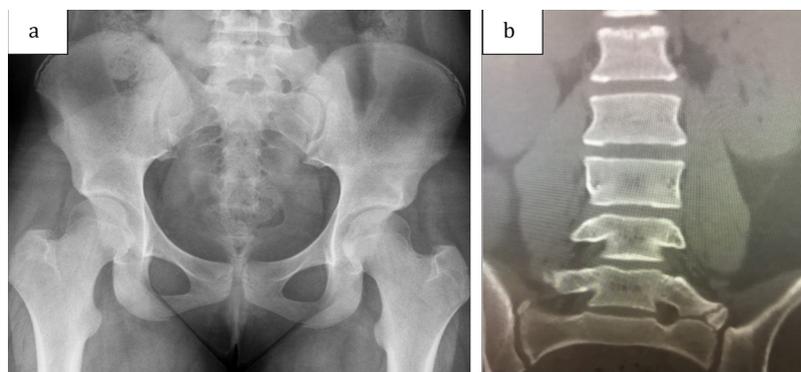
We report the case of a 13-year-old child (weight, 60 kg; height, 156 cm; body mass index, 24) who presented to the pediatric emergency department in October 2015 for undiagnosed mechanical pain in the left hip that had progressed over the course of 4 years. Anamnesis revealed a worsening of the painful symptomatology over the past 2 weeks, leading to inability to participate in regular sports activities. The child also described periods of painful seizures of the left hip associated with reduced-intensity low-back pain. On clinical examination, pain was noted on palpation of the paravertebral lower lumbar muscle masses, without spinal stiffness. The remainder of the orthopedic and neurological examination was normal.

Standard anteroposterior pelvic radiography found that a left transverse mega-apophysis had developed at the expense of L5 (Fig. 1a). Lumbosacral spinal computed tomography confirmed a lumbosacral transitional anomaly with unilateral sacralization of L5 on the left, associated with a transverse apophysomegaly of L5 with transverso-sacroiliac neo-articulation (Fig. 1b). On the right, it showed the transverse process of L5 to be larger than those of the overlying vertebrae but without direct articulation with the sacrum or the right iliac wing. Lumbosacral spinal MRI showed intramedullary bone edema on either side of the left transverso-sacroiliac neoplastic joint with a hypoplastic L5/S1 intervertebral disc. The diagnosis of LSTV classified as stage 2B of the Castellvi classification was maintained for this patient [7,10].

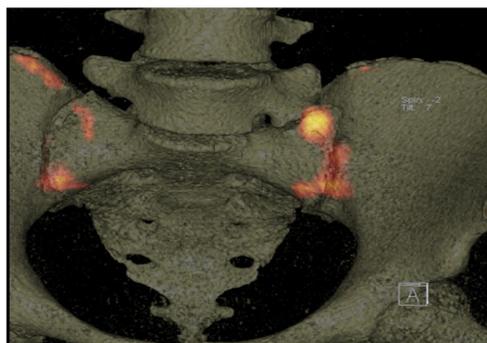
Bone scintigraphy showed a neo-articulated hyperfixation and a transverso-sacroiliac bone conflict, providing additional support for a diagnosis of Bertolotti's syndrome (Fig. 2).

\* Corresponding author.

E-mail address: psmarcheix@orange.fr (P.-S. Marcheix).



**Fig. 1.** a: Anteroposterior pelvic X-ray showing the left transverse mega-apophysis of L5; b: Coronal image of the lumbosacral spine showing the L5 left transverse process and transverse sacroiliac neo-articulation.



**Fig. 2.** Three-dimensional reconstruction of pelvic bone scintigraphy showing hyperfixation in the left transverse sacroiliac neo-articulation related to the bony impingement.



**Fig. 3.** X-ray at 6 weeks after resection of the left transverse mega-apophysis.

To confirm the suspicion of Bertolotti's syndrome, infiltration of the left transverso-sacroiliac neo-articulation was performed in April 2016 under fluoroscopic control by corticosteroids (1 ml of 0.25% cortivazol) and local anesthetic (20 ml 1% lidocaine). A week after the infiltration, the child resumed sports activities. The therapeutic test confirmed Bertolotti's syndrome.

However, a month and a half later the patient experienced recurrence of pain. In light of the positive but short-acting therapeutic and diagnostic test, we amended the diagnosis to Bertolotti's syndrome resistant to medical treatment. Surgical management was proposed and consisted in resection of the left transverse mega-apophysis. The procedure was performed in September 2016 under general anesthesia using the Wiltse approach (left posterolateral paramedian incision), with the patient placed in ventral position on a Jackson table [11]. The results were encouraging and the patient resumed school activities at 10 days postoperatively.

The 6-week follow-up consultation found resolution of the painful symptomatology, and sporting activities were resumed without limitation at 3 months postoperatively. The condition declined over 1 year, with complete disappearance of pain and no radiographic recurrence (Fig. 3).

### 3. Discussion

The adult diagnostic and therapeutic pathway was used for this case because there is no current consensus on the management of this pathology in children. In adults, in a context of chronic low-back pain, the diagnosis of Bertolotti's syndrome is based on lumbar spine radiographs showing a transverse mega-apophysis. Cross-sectional imaging (bone scan) can support diagnostic suspicion by specifying the morphological anomaly and highlighting the neo-articulation impingement (functional imaging: MRI and bone

scintigraphy). A diagnostic test that infiltrates the neo-articulation with anesthetic and anti-inflammatories is used to assess the possible relationship between the painful symptomatology and the morphological anomaly (neo-articulation). If the test is positive, the pathology is almost certainly Bertolotti's syndrome: i.e., pain related to a neo-articulation [3,4]. The test is both diagnostic and therapeutic because it also serves as the medical treatment for this syndrome. There is no established consensus as to the time limit for proposing surgical management. Gepner et al. defined an effective therapeutic test as transient if the effects persisted for less than 8 weeks [12]; surgical management is therefore indicated if the returns within 8 weeks after the infiltration. Diagnosis of Bertolotti's syndrome is based on a range of clinical, radiological, morphological, functional, and therapeutic evidence [4]. This etiology should be noted in the case of atypical or projected pain, such as coxalgia (as in the present case) or abdominal pain [13].

Currently, management of cases resistant to medical treatment, including rehabilitation and infiltration, is surgical, by resection of the transverse mega-apophysis [3,14]. In case of pain related to degeneration of the intervertebral disc or overlying instability, posterior segmental fusion is recommended [7].

We found no publications in the literature regarding the surgical management of Bertolotti's syndrome in children. Indeed, this is a pathology that is rarely mentioned in the pediatric population, although it accounts for 4.6% of low-back pain cases in the general population, and 11–18% in under-30 year-olds [1,15,16].

Bertolotti's syndrome is linked to a congenital anatomical anomaly and is therefore present in childhood. Ossification of the transverse mega-apophysis occurs at the end of bone maturation, at which time the impingement may become symptomatic. Malformations that were previously tolerated and compensated for in children become problematic in young adults, just as tarsal

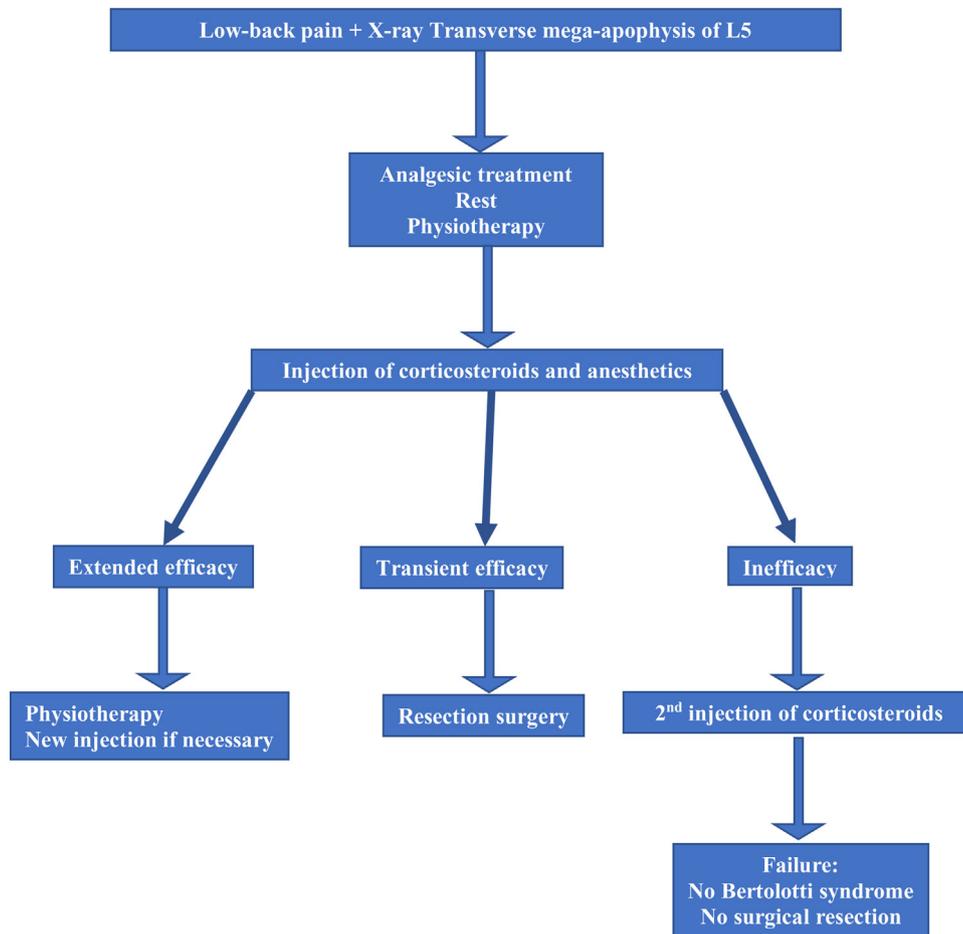


Fig. 4. Decision-tree for the management of proven Bertolotti's syndrome in children.

coalitions are symptomatic of the growth and calcification of previously cartilaginous structures [17].

The chronic low-back pain associated with Bertolotti's syndrome can lead to delayed diagnosis, disabling low back pain, and neurological complications (lower-limb pain) related to impingement between the mega-apophysis and the underlying nerve root. These neurological signs may not completely regress after decompression if they are treated too late. Improved awareness of Bertolotti's syndrome in children on the part of pediatricians and community physicians would undoubtedly prevent diagnostic delay, relieve chronic pain, and reduce the risk of neurological sequelae [18]. We therefore propose a decision-tree for this condition (Fig. 4).

#### 4. Conclusion

The presentation of this clinical case demonstrated the necessity of identifying Bertolotti's syndrome in young adolescents who are at risk of delayed diagnosis and inadequate management leading to a painful adolescence. Surgical management appears to be safe and effective in the long term, when medical treatment has failed.

#### Disclosure of interest

The authors declare that they have no competing interests.

#### Acknowledgement

The English in this document has been checked by at least three professional editors, native English speakers.

#### References

- [1] Quinlan JF, Duke D, Eustace S. Bertolotti's syndrome. A cause of back pain in young people. *J Bone Joint Surg Br* 2006;88(9):1183–6.
- [2] Bertolotti M. Contribution to the knowledge of the defects of regional differentiation of the vertebral column with special attention to the fusion of the fifth lumbar vertebra to the sacrum. *Radiologique Med* 1917;4:113–44.
- [3] Almeida DB, Mattei TA, Soria MG, Prandini MN, Leal AG, Milano JB, et al. Transitional lumbosacral vertebrae and low back pain: diagnostic pitfalls and management of Bertolotti's syndrome. *Arq Neuropsiquiatr* 2009;67(2A):268–72.
- [4] Jancuska JM, Spivak JM, Bendo JA. A review of symptomatic lumbosacral transitional vertebrae: bertolotti's syndrome. *Int J Spine Surg* 2015;9:42.
- [5] Aihara T, Takahashi K, Ogasawara A, Itadera E, Ono Y, Moriya H. Intervertebral disc degeneration associated with lumbosacral transitional vertebrae: a clinical and anatomical study. *J Bone Joint Surg Br* 2005;87(5):687–91.
- [6] DePalma A, Rothman R. Clinical manifestations of lumbar disc syndrome. *The intervertebral disc* 1970:203–48.
- [7] Konin GP, Walz DM. Lumbosacral transitional vertebrae: classification imaging findings clinical relevance. *AJ.N.R. J Am Neuroradiol* 2010;31(10):1778–86.
- [8] Ju CI, Kim SW, Kim JG, Lee SM, Shin H, Lee HY. Decompressive L5 Transverse Processotomy for Bertolotti's Syndrome: A Preliminary Study. *Pain Physician* 2017;20(6):E923–32.
- [9] Mitra R, Carlisle M. Bertolotti's syndrome: a case report. *Pain Pract* 2009;9(2):152–4.
- [10] Castellvi AE, Goldstein LA, Chan DP. Lumbosacral transitional vertebrae and their relationship with lumbar extradural defects. *Spine (Phila PA 1976)* 1984;9(5):493–5.
- [11] Abelin-Genevois K, Accadbled F, de Bodman C, Bollini G, Bonnard C, Bronfen C, et al. Liste des auteurs. *Orthopédie Pédiatrique Rachis et Thorax*. Elsevier; 2016. p. vii–ii.

- [12] Gepner P, Pierron D, Robine DJR. Lombalgie chronique et vertèbre transitionnelle lombosacrée: un lien pathogénique probable à propos de 5 observations. *Rachis* 1994;6(3):141–6.
- [13] Mercader Rodriguez B, Sanchez RF, Domenech Abellan E, Parra JZ, Canovas CS, Castellon Sanchez MI. Bertolotti syndrome: a little known cause of low-back pain in childhood. *J Pediatr* 2015;166(1):202.
- [14] Jonsson B, Stromqvist B, Egund N. Anomalous lumbosacral articulations and low-back pain. Evaluation and treatment. *Spine (Phila Pa 1976)* 1989;14(8):831–4.
- [15] Dizdarevic I, Bishop M, Sgromolo N, Hammoud S, Atanda Jr A. Approach to the pediatric athlete with back pain: more than just the pars. *Phys Sportsmed* 2015;43(4):421–31.
- [16] Neelakantan S, Anandarajan R, Shyam K, Philip B. Multimodality imaging in Bertolotti's syndrome: an important cause of low back pain in young adults. *BMJ Case Rep* 2016:2016.
- [17] Peyrou P, Moulies D. Les «fausses entorses» de l'adolescent. *Journal de traumatologie du sport* 2006;23(2):96–104.
- [18] Manmohan S, Dzulkarnain A, Nor Azlin ZA, Fazir M. Bertolotti's syndrome: a commonly missed cause of back pain in young patients. *Malays Fam Physician* 2015;10(2):55–8.