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*Case Report*

# Surgical management of Bertolotti's syndrome in two adolescents and literature review

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## ABSTRACT

**Background:** Bertolotti's syndrome is defined by back pain and/or radicular symptoms attributed to a congenital lumbosacral transitional vertebra (LSTV). There are few studies that discuss the surgical management of Bertolotti's syndrome. Here, we report long-term outcomes after resecting a pseudoarthrosis between the sacrum and L5 in two teenage patients, along with a review of literature.

**Case Descriptions:** Surgical resection of a lumbosacral bridging articulation (LSTV type IIa) was performed in two patients, 15 and 16 years of age who presented with intractable back pain. The adequacy of surgery was confirmed with postoperative studies. In both patients, pain and functional status improved within 6 weeks and have remained improved at last follow-up.

**Conclusion:** Surgical removal of a pathologic L5 transverse process fused to the sacral ala in two young patients with Bertolotti's syndrome improved postoperative pain and increased overall function. Given the progressive nature of Bertolotti's syndrome, surgical intervention in young patients should be considered to mitigate years of chronic pain and attendant morbidity.

**Keywords:** Bertolotti's syndrome, Lumbosacral transitional vertebrae, Sacralized pseudoarthrosis

## INTRODUCTION

Bertolotti's syndrome is defined as a congenital lumbosacral transitional vertebra (LSTV) that is responsible for disabling low back pain. It most commonly occurs at the L5 level, followed by the L6 level, and is characterized by various morphologic presentations [Table 1]. Several medical and surgical therapies are available to treat this syndrome: for example, physical therapy, corticosteroid injections (many risks/complications without documented long-term efficacy), laminectomy, spinal fusion, and removal of the pathologic bone segment.[2,10,14] Here, we demonstrate the outcomes after surgical resection of LTSV in two pediatric patients and have reviewed the relevant literature.

## CLINICAL PRESENTATION

### Patient 1

A 14-year-old female with midline low back pain and the right hip/leg pain was treated for 9 months with physical therapy and nonsteroidal anti-inflammatory drugs (NSAIDs) without

**Table 1:** Castellvi classification schema of Bertolotti's syndrome by laterality (unilateral vs. bilateral) and morphological characteristics (size and anatomy involved) of the abnormal vertebral articulation.

| Castellvi classification system for lumbosacral transitional vertebra | | |
|---|---|---|
| **Type** | **Characteristics** | **Specification** |
| I | Enlarged, dysplastic transverse process (>19 mm craniocaudal width) | Ia: Unilateral<br>Ib: Bilateral |
| II | Pseudoarticulation of the transverse process and sacrum with incomplete lumbarization/sacralization; enlargement of the transverse process w/pseudoarthrosis with a diarthrodial joint between itself and the sacrum | IIa: Unilateral<br>IIb: Bilateral |
| III | Transverse process completely fused with the sacrum with complete lumbarization/sacralization | IIIa: Unilateral<br>IIIb: Bilateral |
| IV | Type IIa pseudoarticulation on one side of vertebrae and Type IIIa fusion on the contralateral side | |

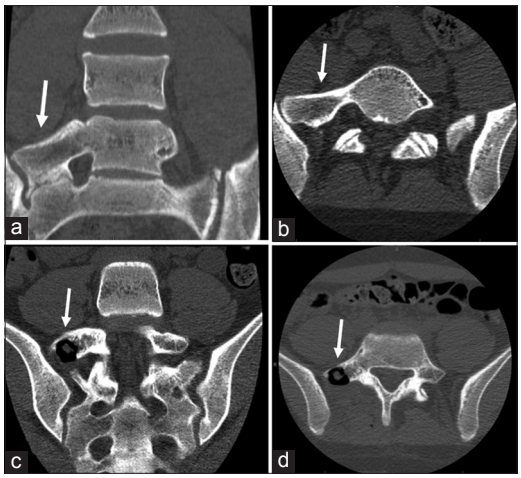


**Figure 1:** Preoperative coronal computed tomography (CT) (a) and sagittal CT (b), both demonstrating a sacralized L5 with pseudoarthrosis from L5 to S1 and the ilium (white arrows). Postoperative coronal CT (c) and axial CT (d), indicating removal of the abnormal articulation (white arrows).

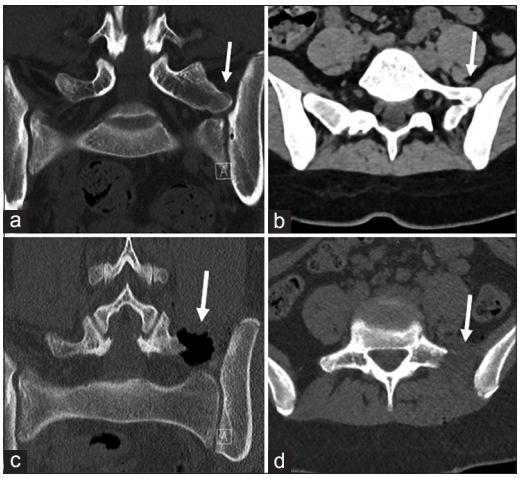


**Figure 2:** Preoperative coronal computed tomography (CT) (a) and sagittal CT (b), both demonstrating an extended left L5 transverse process fused with the ilium and sacrum (white arrows). Postoperative coronal CT (c) and axial CT (d), indicating removal of the abnormal articulation (white arrows).

relief. Her neurological examination was normal. Lumbar X-ray, computed tomography (CT), and magnetic resonance imaging (MRI) studies showed Bertolotti's syndrome, characterized by a right-sided partially sacralized L5 vertebra with pseudoarthrosis between L5 to S1 and ileum, with areas of irregularity/sclerosis [Figure 1a and b]. She underwent resection through a posterior midline approach confirmed on the postoperative CT [Figure 1c and d]. She was asymptomatic within 6 postoperative weeks and remains symptom-free 2 years later.

### Patient 2

A 16-year-old female presented with 2 years of low back pain and 3 months of pain radiating into her left hip refractory to NSAIDs and physical therapy. Comorbidities included recurrent migraines, major depressive disorder, obstructive sleep apnea, and restless leg syndrome. Her neurological examination was normal. The lumbar CT and MRI studies showed Bertolotti's syndrome on the left at the L5-S1 level characterized by an enlarged left L5 transverse process fused with the ilium and sacrum, with mild degeneration/sclerosis of the left L5 pars interarticularis [Figure 2a and b]. After surgical resection, confirmed on postoperative CT, she was intact and remained so 1 year postoperatively [Figure 2c and d].

## DISCUSSION AND REVIEW OF RELEVANT LITERATURE

### Diagnosis and pathophysiology of Bertolotti's syndrome

Bertolotti's syndrome is found in 10% of patients presenting with back and leg pain under 30 years of age.[16] The biomechanics of LSTV is attributed to an alteration or reduction of movement between the transitional vertebra and the sacrum that can ultimately lead to pain from stress in the facet joint and/or is exacerbated by disc degeneration.[9]

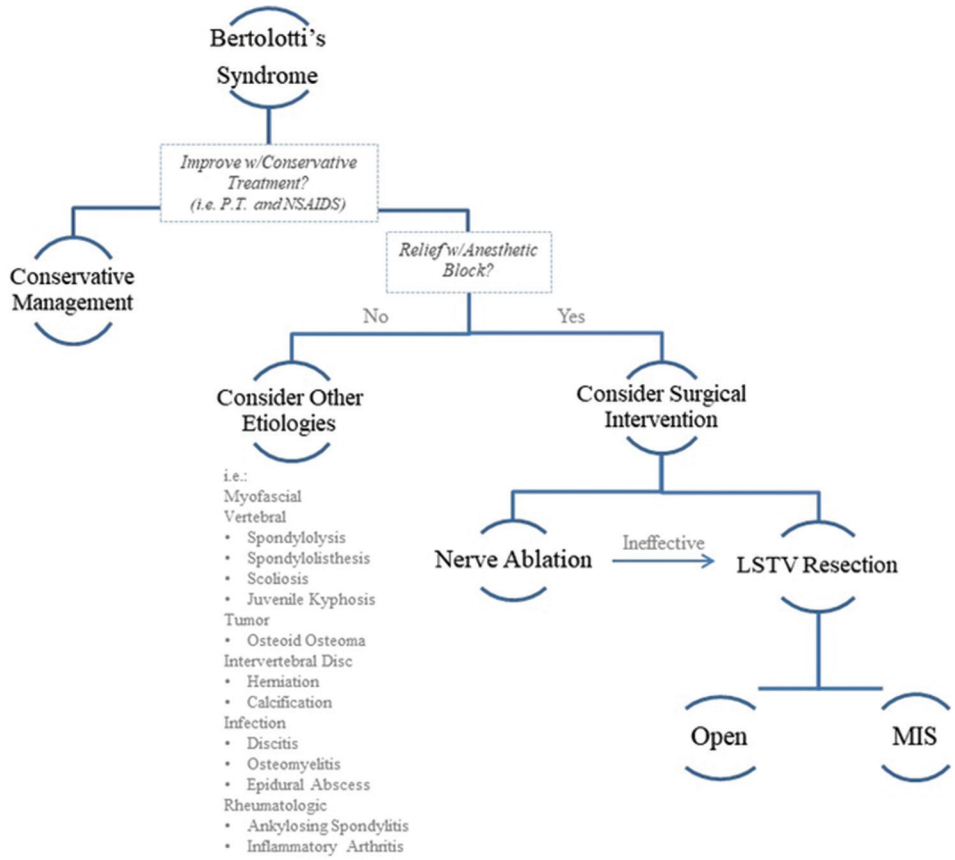


**Figure 3:** Flow diagram of management and treatment considerations for Bertolotti's syndrome.

### Radiographic analysis

Radiographs, including flexion, extension views, and oblique views, confirm the diagnosis of Bertolotti's syndrome. Both CT and MRI also demonstrate Bertolotti's syndrome and also readily identify associated stenosis, osteophytes, and sclerosis adjacent to the articulation between the lumbar segment and ilium and/or sacrum.

### Management of Bertolotti's syndrome

Surgical resection of LSTV should be considered in patients presenting with intractable low back pain despite conservative treatment (e.g., physical therapy, nonsteroidal anti-inflammatory drugs, and localized anesthetic blocks in adults Figure 3).[7]

### Outcomes of surgery with Bertolotti's syndrome

There are few reports for patients under 18 years of age treated for Bertolotti's syndrome [Table 2].[5] In three pediatric studies, two 17 years old experienced relief of their back and leg pain at 6 months and 1 year after surgery; in another, a 13 years old reported no improvement at 6-month follow-up; in a third, an 18 years old (in the same study) reported total alleviation of back pain 2 years after surgery.[3,4,8] Likely, Bertolotti's syndrome is underdiagnosed in the pediatric population. Although surgical outcomes are generally positive, no randomized studies have documented their efficacy versus conservative nonsurgical treatment.

## CONCLUSION

Bertolotti's syndrome is seen in 10% of patients presenting with back and leg pain under 30 years of age.[16] While few surgical cases are reported in pediatric patients, removal of the abnormal transverse apophysis and disconnection from the lumbar spine/sacral ala should be considered as for those who fail conservative treatment.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given her consent for her images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Table 2:** Bertolotti's syndrome surgical intervention and outcome studies.

| Study | Year | #Cases | Mean age (year) | Level/Type | Procedure | Follow-up | Outcomes |
|---|---|---|---|---|---|---|---|
| **Adolescents and adults** | | | | | | | |
| Jönsson *et al.*[8] | 1989 | 11 | 39 (Range: 13–76) | n/a | Resection of anomalous LSTV articulation | 6 months–3 years | Seven patients pain resolved<br>Two patients no change in pain |
| Brault *et al.*[4] | 2001 | 1 | 17 | L6-S1 Type IIa | Resection of the right L6 anomalous transverse process | 1 year | Remission of LBP and leg pain w/no limitations in activity |
| Babu *et al.*[3] | 2017 | 2 | 17<br>38 | L5-S1 Type IIb | Resection of LSTV utilizing O-arm | 6 months<br>9 months | Resolution of pain<br>Resolution of pain |
| **Adults** | | | | | | | |
| Santavirta *et al.*[17] | 1993 | 16 | 34 (Range: 27–58) | 11 L5-S1<br>5 L6-S1<br>13 Type Ia<br>2 Type IIIa<br>1 Type IIIb | 8: posterior-lateral L5-S1 fusion<br>8: resection of LSTV articulation | 2–17 years | 10 patients w/improved LBP<br>Seven patients resolved LBP<br>11 of 13 patients w/continued sciatica<br>Six patients underwent reoperation |
| Abe *et al.*[1] | 1997 | 1 | 37 | L5-S1<br>Type IIb | Extraforaminal decompression of the left L5 transverse process and bony spur through extraperitoneal anterior approach | 2 weeks<br>1 year | Relief of LBP and leg pain; increased hypesthesia, numbness, and mild weakness<br>Resolution of pain, hypesthesia, and numbness; no work limitations |
| Ichihara *et al.*[6] | 2004 | 1 | 34 | L5-S1<br>Type IIb | Resection of LSTV and nerve decompression through posterior approach | 1 year | Improved hip and leg pain |
| Ugokwe *et al.*[20] | 2008 | 1 | 48 | L5-S1<br>Type IIa | Minimally invasive resection of the left L5 transverse process and pseudoarticulation | 6 weeks<br>6 months | 10% improvement in pain<br>90% LBP and leg pain relief |
| Weber *et al.*[21] | 2010 | 1 | 53 | L5-S1<br>Type IIa | Lateral foraminal and extraforaminal nerve decompression through posterior approach | 1 year | No LBP or radicular pain |
| Shibayama *et al.*[18] | 2011 | 1 | 46 | L6-S1<br>Type IIa | Extraforaminal decompression of the right L6 body, transverse process, and upper sacral ala | 30 months | Improved LBP, sciatica, and returned to work |
| Miyoshi *et al.*[15] | 2011 | 1 | 29 | L5-S1<br>Type IIb | Resection of the right LSTV osteophytes/pseudoarticulation | 1 month<br>1 year | Improved R-sided leg and buttock pain<br>R-sided pain resolved |
| Kikuchi *et al.*[11] | 2013 | 2 | 70<br>53 | L5-S1<br>Type IIa | Decompression and LSTV resection through anterior approach | 1 year<br>1 year | LBP and leg pain relieved and functionally improved |
| Malham *et al.*[13] | 2013 | 2 | 27, 49 | L5-S1<br>Type IIa | Resection of LSTV through anterior, retroperitoneal approach | Patient 1 (27 years)<br>4 weeks<br>2 years<br>Patient 2 (49 years)<br>3 months<br>2 years | Improved LBP and return to work<br>No exercise limitations<br>Improved LBP and return to part-time work No limitations with low-impact exercise |

(*Contd...*)

**Table 2:** *Continued.*

| Study | Year | #Cases | Mean age (year) | Level/Type | Procedure | Follow-up | Outcomes |
|---|---|---|---|---|---|---|---|
| Li *et al.*[12] | 2014 | 7 | 40.2 (Range: 26–63) | L5-S1<br>5 Type IIa<br>2 Type IIb | Minimally invasive paramedian tubular-based resection of LSTV pseudoarticulation | 1 year | Three patients complete relief in LBP<br>Two patients improved LBP<br>Three patients complete relief of radicular pain<br>One patient improved radicular pain<br>Two patients had initial relief but had return of LBP at 1–4 years |
| Takata *et al.*[19] | 2014 | 1 | 45 | L5-S1<br>Type IIb | L4-5 discectomy and resection of the left LSTV pseudoarticulation | n/a | Improved LBP and sciatica pain |
| Adams *et al.*[2] | 2018 | 1 | 37 | L5-S1<br>Type IV | Minimally invasive, L5/S1 instrumented fusion, with allograft and bone graft substitute | 2 weeks | Complete resolution in the left-sided SI joint and LBP and the right-sided leg pain |

LBP: Low back pain, LSTV: Lumbosacral transitional vertebra, #The number of cases (patients), as expressed in whole numbers

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**Conflicts of interest**

There are no conflicts of interest.

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