

COMPLETE, SUSTAINED RESOLUTION OF PAIN WITH PSEUDOARTHRECTOMY FOR BERTOLOTTI SYNDROME: A CASE REPORT

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- Background:** Bertolotti syndrome, characterized by lumbar pain due to a lumbosacral (LS) transitional vertebra, is a known but still controversial and overlooked pain generator despite an explosion of information on the condition in the last decade. Current management includes plain radiography, advanced imaging, minimally invasive interventions, and LS fusion.
- Case Report:** We present a unique case of a middle-aged man with Castellvi Ila Bertolotti syndrome consisting of a unilateral enlarged, dysplastic L5 transverse process forming a pseudoarticulation (PA) with the ilium and normal anatomy contralaterally. He was diagnosed on a plain radiograph and following a diagnostic injection of the PA, he received complete, sustained pain relief from a pseudoarthrectomy.
- Conclusions:** This case illustrates an economical and thoughtful approach to diagnosis and management that may offer pain relief rather than invasive management with surgical fusion that comes with its own morbidity and mortality and consequences, such as adjacent-level disease.
- Key words:** Bertolotti syndrome, pseudoarticulation, low back pain, transverse process, case report, lumbosacral transitional vertebra

BACKGROUND

Bertolotti syndrome is a disorder characterized by low back pain (LBP) due to a lumbosacral transitional vertebra (LSTV) (1). There has been an explosion of literature on Bertolotti syndrome in the last 10 years (2). It remains a controversial diagnosis since a direct correlation between the presence of a transitional segment and pain has not been definitively demonstrated (3). It has also been implicated as a cause of sciatica, but hip pain can be a prominent symptom (2,4,5).

An LSTV is a congenital anomaly in which there is either lumbarization of the first sacral vertebra or sacralization of the last lumbar vertebra (1,3). The variations are described by the Castellvi classification where type Ia is a unilateral enlarged, dysplastic transverse process (TP) (with the other side being normal) while

Ib is bilateral; IIa is a unilateral pseudoarticulation (PA) between the dysplastic TP and the sacral ala (SA), ilium, or both (with the other side being normal) while IIb is bilateral; IIIa describes a unilateral fusion between the dysplastic TP and the SA, ilium, or both (with the other side being normal) while IIIb is bilateral; and IV describes a hybrid between IIa and IIIa (6). More recently, a new classification called the Jenkins classification has been introduced to determine treatment, which allows for the categorization of more complex variants (5,7). It is divided into 4 categories: type 1, 2, 3, and 4 with subclassifications of L (left) or R (right) prominence, A (unilateral) or B (bilateral), or C (type 2 with type 1 and contralateral type 2 anatomy or type 4 with a gap > 10 mm and contralateral type 3 anatomy), and L (left) or R

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(right) iliac contact (5,7). In type 1, there is a dysplastic TP that shows a gap of < 10 mm, but > 2 mm across the LS junction. In type 2, there is a PA between the adjacent TP with a gap of < 2 mm. In type 3, there is complete fusion lateral to the facet at the SA, without spontaneous fusion of the disc or facet joints, of the LS junction. In type 4, there is presence of a unilaterally fused side with varying PA on the contralateral side (type 4A, 4B, or 4C) (5,7).

Studies (3,8-12) have shown that this is a frequently occurring anatomic variant, affecting anywhere from 4.6% to 36% of the population with an average prevalence of 12% depending on the study. In a large cohort study (13) of patients with symptomatic femoroacetabular impingement requiring hip arthroscopy, the prevalence of LSTVs was 13.9%. Another population-based study (14) involving 5,860 individuals reported one of 15.8%, with a significant association between certain LSTV and LBP. Lastly, a study (15) on patients with LBP and/or sciatica showed an LSTV prevalence of 15%. These latter 3 studies indicate that the incidence of symptomatic LSTVs ranges from 13.9% to 15.8%.

Plain radiographs can easily visualize a potential LSTV and the Ferguson view (an AP with a 30° cranial tilt) can best visualize a potential transitional segment (Fig. 1). This view is used in performing sacral transforaminal epidural steroid injections (TFESI). Single-photon emission computed tomography (CT) can help distinguish a symptomatic PA between an enlarged TP and the SA or ilium (Castellvi type II) from other pain generators in the lumbar spine and pelvis (16-18). Three-dimensional (3D) zero-echo time and 3D T1-weighted gradient-echo magnetic resonance imaging (MRI) sequences are effective alternatives to CT for evaluating lumbar facet joints and LSTVs (19). Moreover, F-sodium fluoride (NaF) positron emission tomography/CT can demonstrate a strong correlation between NaF uptake and symptomatic LSTVs (20). Furthermore, 3D MRI LS radiculography can identify symptomatic extraforaminal stenosis in LSTVs by detecting morphological changes in the L5 nerve roots (21). However, in patients with Castellvi type IIa and contralateral LBP, CT appears to be the most sensitive for the evaluation of contralateral facetogenic pain (17). In such patients, the unilateral PA can restrict movement at the ipsilateral L5-S1 facet joint, but not at the contralateral one. If the history and physical exam identify pain contralateral to the PA, this would suggest a symptomatic variant. EOS imaging system (Montreal,

Quebec; Canada) is a new radiographical method that creates 3D reconstructions, but is not widely available (22).

We present a case of apparent Bertolotti syndrome identified by history, physical examination, radiographic findings, and a diagnostic injection with resolution of pain following PA resection. We will discuss the incidence, pathophysiology, diagnosis, and treatment options for Bertolotti syndrome none of which have consensus albeit more literature than in the past (22). This case is unique because the diagnostic approach is efficient and economical, and the results are superlative and sustained despite the surgical approach increasingly falling out of favor (5,7).

CASE PRESENTATION

A 56-year-old man presented with progressively worsening right LBP and hip pain with no known inciting event. The patient gave written consent for this publication. Per Baylor College of Medicine policy, this study is exempt from Institutional Review Board approval due to it being a case report. The pain was described as constant and achy in quality, and 6 out of 10 in intensity on a Visual Analog Scale (VAS). It was worse in the back than the hip with occasional radiation to the right groin. The pain was exacerbated with sitting, bending, and walking. Identification of the right PA (Castellvi IIa) was made with a plain radiograph. There was no significant relief with nonsteroid anti-inflammatory drugs or physical therapy (PT). Fluoroscopically guided injections, including L5 lumbar TFESI and S1 sacral TFESI, as well as lumbar facet joint injections, provided no relief. On physical exam, the patient had right lateral hip pain elicited with internal rotation and Patrick test. Manual muscle testing revealed strength was fully intact in the lower extremities with a negative Gaenslen test.

Initial radiographs demonstrated a PA of the right TP to the ilium (Fig. 1). The orthopedic surgeon requested a diagnostic injection to confirm that this PA was the principal pain generator. We localized the PA under fluoroscopic guidance, injected 0.25% bupivacaine (Fig. 2), and asked the patient to keep a pain diary. On the follow-up visit, the patient noted resolution of the pain, with return of the pain concordant with the half-life of the anesthetic.

The PA was resected. Radiographs postoperatively showed successful resection of the PA (Fig. 3). Currently, the patient remains asymptomatic, including being pain free.

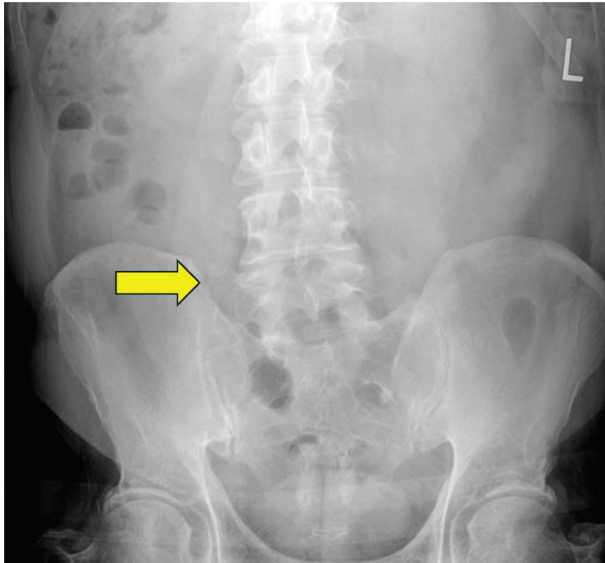


Fig. 1. AP view of the lumbar spine showing a fusion of the TP to the ilium (yellow arrow). AP, anteroposterior; TP, transverse process.

DISCUSSION

The lumbar spine has multiple potential pain generators with several of them playing a role in Bertolotti syndrome due to the hypothesized pathophysiology. It was proposed by Elster (8) that the pathophysiology of Bertolotti syndrome is similar to that following spinal fusion, characterized by the tendency for the disc above or below the fusion to deteriorate over time, which is known as adjacent-level disease. The proposed mechanism suggests when one segment is fused and less mobile, other segments at that level, including the disc and the contralateral facet joint will experience more movement and thus be more prone to degeneration. These hypermobile segments can be potential pain generators. The PA, however, can itself be a source of pain, as can any other articulation. Our case was atypical as the patient experienced LBP ipsilateral to the PA, while it is typically contralateral to the PA in Castellvi type IIa.

The clinical presentation of Bertolotti syndrome is characteristically unilateral or bilateral axial LBP or buttock pain. However, in cases where there is disc degeneration and the potential for disc herniation, there is also the possibility of foraminal stenosis as Vergauwen et al (15) reported, and patients may present with radicular symptoms.

Conservative therapies are the cornerstone of treat-

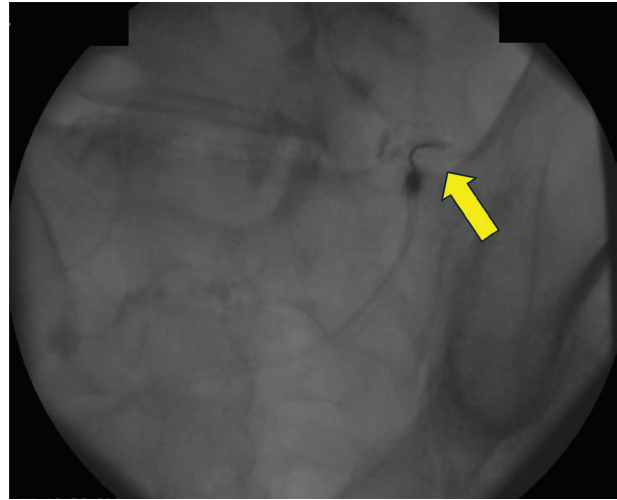


Fig. 2. Diagnostic injection of the PA. PA, pseudoarthrosis.

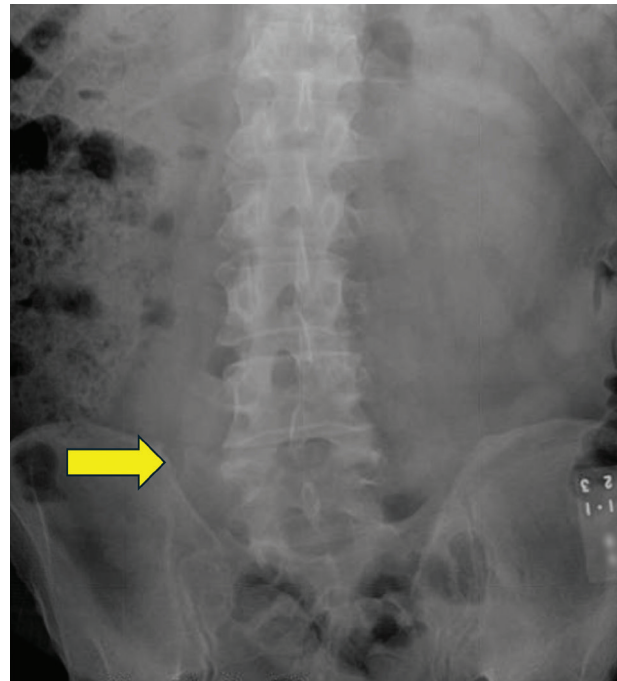


Fig. 3. Postoperative lumbar spine radiograph after successful pseudoarthrectomy (yellow arrow).

ment, including activity modification, medication management, and PT that includes core strengthening and stretching. If conservative measures are unsuccessful, more invasive treatment options can be pursued. It is imperative with so many structures that are a potential source of pain to use diagnostic injections into suspected pain generators and have the patient use

a pain diary to record the amount of pain relief they have following a diagnostic injection. Once the pain generator has been identified, therapeutic injections may also be attempted. There is a case report (23) of radiofrequency ablation (RFA) in the literature in which a patient had complete resolution of pain from bipolar strip lesions of the PA. More recent literature (11) also suggests continuous RFA, pulsed RFA, and cooled RFA can be helpful. If conservative measures and interventions fail, surgery can be considered. Studies on the success of surgery done by Santavirta et al (24) and Jonsson et al (25) had mixed results with respect to pain relief, which further highlights the importance of diagnostic injections to identify the pain generator prior to attempting more invasive treatments. A more recent study by Ju et al (26) of 61 patients that all had significant pain relief from a preoperative block to the PA and had a pure L5 transverse processotomy showed benefit for Castellvi type I and Castellvi II short joint type but not for Castellvi II wide joint type. An even more recent study by Afana et al (27) presents a newer surgical approach called the mini-open tubular microsurgical transverse processotomy, which although effective does not offer complete pain relief. More recent data (12) suggests, however, that a pseudoarthrectomy can lead to quality-of-life improvement. Despite all this, Jenkins et al (5) recommend fusion for Castellvi II/Jenkins type 2 patients and for some patients a strategy of shaving down hypertrophic bone. Other groups have made a similar recommendation of fusion for Castellvi IIa patients (28). Part of the reason this recommendation for fusion was made by Jenkins et al (5) and Crane et al (11) is that there is a high rate of recurrent surgery for Castellvi II/Jenkins type 2 patients because the pain relief can wear off. According to Jenkins et al (5), a "good" outcome is to be either a reduction of the original VAS pain level from their preoperative level to 50% of that level, or to a 3 out of 10 or less. Although there is another case report (29) in the literature of a complete, sustained pain relief with a pseudoarthrectomy in a Castellvi IIa

patient, that patient was far younger, 17 years old to be more specific. One of the significant limitations of our case is that it is a case report, but illustrates powerfully the axiom that less is more. In our case, the diagnosis was confirmed with a plain radiograph and a fluoroscopically guided injection of the PA, which is far more economical than advanced imaging, such as MRI. You could argue it could have been far more economical to do the diagnostic injection in the PA before any other injection. Regardless, the patient's symptoms resolved with surgical resection of the PA, even though some authors propose going straight to surgical fusion in such patients.

CONCLUSIONS

Although Bertolotti syndrome is a controversial diagnosis, it must be kept in the differential diagnosis. Regardless, if there exists a correlation between a LSTV and LBP, it is certainly possible for a PA, contralateral zygapophyseal joint, or a degenerative disc in a patient with a LS transitional segment to be a pain generator as any other joint or disc in the spine can be. Furthermore, it is important to use diagnostic injections with a patient pain diary in identifying the source of back pain when pursuing interventional or surgical treatment options since there are many potential pain generators in the spine. Even if the patient responds to the diagnostic injection into the PA, there is no guarantee the pseudoarthrectomy would be successful like it was successful for our patient. Moreover, even if the patient responds to the pseudoarthrectomy, the pain relief may not be complete. If it is complete, it may not be enduring like it has been for our patient. The primary takeaway lesson from this case is that a combination of a plain radiograph and a diagnostic injection can identify a painful PA. The second takeaway lesson is although there are a multitude of surgical treatments for Bertolotti syndrome, a pseudoarthrectomy for Castellvi II should be considered even though it has fallen out of favor for more invasive treatments, such as a surgical fusion in some circles.

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