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Fluoroscopic OnabotulinumtoxinA injection for Bertolotti Syndrome in refractory back pain: A letter to the editor

Dear editor,

Lumbosacral transitional vertebra (LSTV) also known as Bertolotti segments are congenital anatomical variants of the lumbosacral junction with reported incidence of 4–30 % in the general population [2]. LSTVs are classified utilizing the Castellvi system ranging from I-IV where type I indicates enlargement or dysplastic L5 transverse process (TP), type II indicates incomplete lumbarization or sacralization, type III indicates fusion of the L5 TP and sacral ala, and type IV indicates type IIa on one side and IIIa on the contralateral side [3]. They are further divided into type "a" if unilateral or "b" if bilateral aside from type IV LSVTs [3]. Pain from any LSTV is known as Bertolotti Syndrome and with a reported incidence of 4–8% [2]. Symptomatic LSTVs are most likely to present with lower back pain making diagnosis challenging as nearly 80 % of adults will present with low-back pain at some point in their lives [3]. Furthermore, unilateral LSTVs result in notable biomechanical changes of the spine causing significant adjacent segment findings without having significant changings at the level of the LSTV itself [3]. Treatment options may range from conservative treatment to surgery with case reports of interventional procedures utilizing fluoroscopic guided epidural steroid injections and anesthetic agents (ESIs) to the pseudo-articulations [3]. Corticosteroids are a common medication used in interventional injections, however, not always well tolerated. Systemic side effects from local corticosteroid injections are well documented including elevated blood pressure, altered glucose metabolism, and Cushing syndrome [4–6].

We present a case of a patient who suffered from long-standing back pain resistant to conservative measures found to have Bertolloti syndrome. Interventional procedures utilizing corticosteroids were trialed resulting in significant side effects, thus, BoT was trialed and demonstrated relief in her pain with return of function.

This case involves a 54-year-old female with a history of bilateral total hip arthroplasty (THA) who presented with right lower back, buttock, groin, hip, and posterior thigh pain present for several years, recently worsening and causing severe activity limitation. Her pain was rated as severe and caused her to difficulty with transfers, restricted ambulation, and aggravated with nearly every activity. There were no associated neurologic symptoms or red flags. She previously trialed conservative measures including physical therapy and medications including multiple neuropathic agents and muscle relaxers from which she experienced unfortunate side effects including diaphoresis, general malaise, and insomnia. On exam, the patient demonstrated tenderness with palpation over her right posterior superior iliac spine (PSIS) and sacrum, reduced lumbar extension and hip range of motion, and pain to the right groin, hip, and back with flexion-abduction-external-rotation (FABER) and flexion-adduction-internal-rotation (FADIR), and pelvic compression tests. Logroll test was negative. Spring test was positive over her lumbar spine. Neurovascular exam of the bilateral lower extremities was negative. Chart review of prior CT imaging demonstrated right sided lumbosacral transitional anatomy (Fig. 1). A newly ordered MRI demonstrated unilateral right-sided transitional lumbosacral anatomy, degenerative disk findings at L5-S1 with moderate right foraminal narrowing at that level, and moderate lumbar facet arthropathy at L4-5 and L5-S1; Bertolloti segment noted on right only and classified as a type IIa LSTV due to her pseudo articulation with the sacral ala as seen in Figs. 1 and 2. Her differential diagnosis included myofascial pain, lumbar facet arthropathy, lumbar spondylosis, right SIJ pain, pain related to her right total hip replacement, and Bertolotti segment pain.

Over the course of several months, the patient underwent several procedures including psoas bursa injections, a fluoroscopic guided injection to the right sacroiliac joint (SIJ) with a solution of 1 mL (ml) of 6 mg (mg) betamethasone and 2ml of 2 % lidocaine; right L3, L4, L5 medial branch block (MBB); a radiofrequency ablation (RFA) to right L3, L4, and L5 nerve roots with partial relief in hip and groin pain but not with back pain; and a right L4/5 transforaminal epidural steroid injection (TFESI) containing 1ml of 10mg/ml dexamethasone and 1ml 2 % lidocaine. After each steroid injection, the patient experienced side effects including hypertension requiring medication, headaches, nausea, and general malaise for up to two weeks, making steroid exposure undesirable. She did receive 50 % relief of her back and buttock pain from the TFESI indicating a possible axial source as her pain generator yet with incomplete relief. Alternative axial structures were thus considered for the patient's pain generator.

In evaluation of her pain generator, the patient consented to, and receive a diagnostic fluoroscopic injection into her L5/S1 Bertolloti segment containing 2ml of 0.25 % bupivacaine and 2ml of 2 % lidocaine after confirmation of adequate intra-pseudo articular needle placement with Omnipaque 300 contrast. On follow up, she reported about 75 % relief of her symptoms lasting as long as expected for the anesthetics. Given her previous adverse reactions to corticosteroids, onabotulinumtoxinA (ONA) was suggested as an alternative. She subsequently trialed an injection containing 100 units ONA in 1ml preservative free normal saline (PFNS) and reported 75 % relief for about 2 months including the return of functional movements and range of motion. 100 units ONA was chosen as a starting point due to findings of previous literature published by Dykstra et al. [7]. 1ml of PFNS was chosen to maintain an adequate target ONA dilution of 1:1 with the aim of 100 units of toxin per 1ml of solution. As she did not receive a full 3 months of relief, a solution of 1:1 dilution 150 units ONA in 1.5ml PFNS was trialed at a subsequent visit. On follow up the patient reported 75–80 % relief for 3 months with minimal residual dull pain. The patient now continues to undergo fluoroscopic injections every 3 months containing a solution of 150 units ONA in 1.5ml PFNS as seen in Fig. 3 below. She

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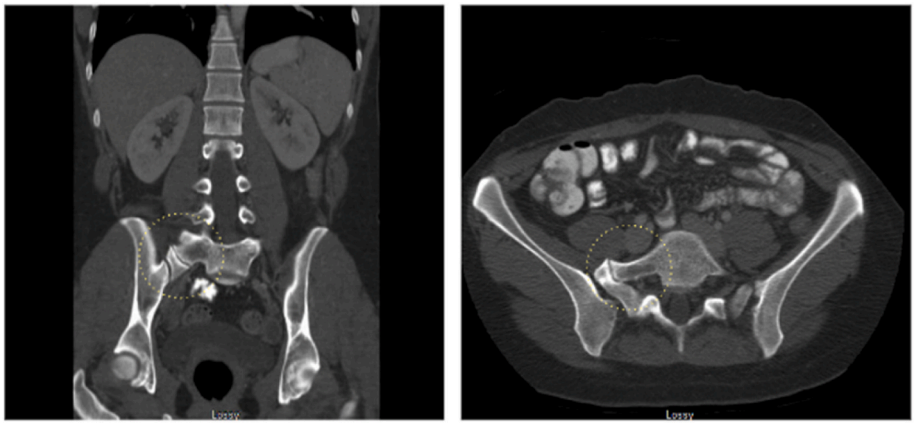

Fig. 1. Right sided Bertolloti segment on Coronal and Axial CT Abdomen and Pelvis with Contrast.

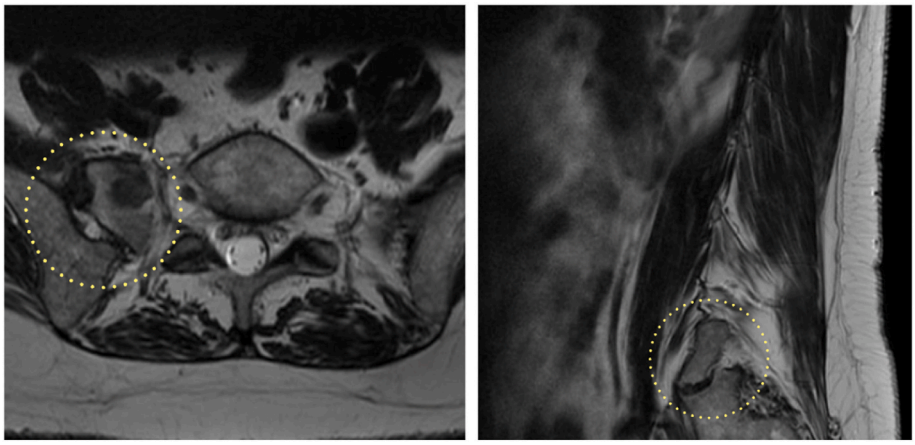

Fig. 2. Axial and Sagittal T2 MRI sequence of right lumbar Bertolloti segment including pseudo-articulation.

continues to receive substantial pain relief, return of functional movements, and resumption of home exercise program (HEP). She has not had any apparent side effects of ONA which may include antibody formation and immune-related complications if it were to enter the circulatory system [1].

Although largely used for spasticity, botulinum toxin (BoT) has been used as a treatment for muscular and neuropathic pain [1]. BoT works as a treatment for pain through inhibiting the secretion of pain mediators from nerve endings, reducing local inflammation, and deactivating sodium channels [1]. In this patient, BoT likely produced similar analgesic effects through reduction of nociceptive transmission in the pseudoarthrosis as those in facet joints. BoT has demonstrated peak effects in about 3–4 weeks after injection with effects lasting about 3 months [8]. Lee et al. demonstrated its anti-nociceptive effects for axial mediated pain and even efficacy over corticosteroid containing injections for SIJ pain [9]. A case series by Dykstra et al. further investigated the use of BoT injections into several joints including cervical and lumbar facet joints demonstrating superior relief to corticosteroids for up to 2 months [7]. Dykstra also demonstrated the use of 25–100 units ONA with efficacy up to 3.2 months in the SIJ [7]. BoT itself is remarkably safe with potential side effects including muscular fatigue or weakness in the surrounding tissue that are self-limited and reversible; other minor adverse effects may include hypersensitivity reaction [10]. More serious side effects of BoT may include entry into the epidural space causing

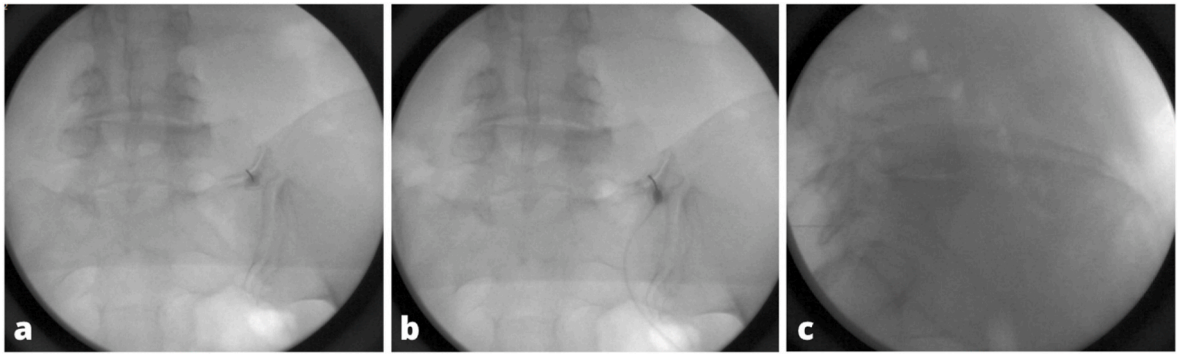

Fig. 3. (a) Fluoroscopic AP view of Bertolloti segment with needle placement; (b) contrast placement into pseudoarthrosis prior to ONA injection into Bertolloti segment; (c) lateral fluoroscopic view Bertolloti segment.

profound multi-limb weakness and central nervous system depression, thus care must be demonstrated when interpreting contrast flow patterns prior to injecting.

This case demonstrates how clinicians must carefully evaluate patients with lower back pain and be vigilant in the constant evaluation of pain generators and treatments. Clinicians should also be aware of the side-effects and harms of their treatments with the ability to find alternative treatments to benefit their patients. In this case, a patient with multiple possible pain generators underwent a thorough evaluation and several procedures before identifying her Bertolloti segment as the pain generator. Given the significant side effects of corticosteroid containing injections during her evaluation, botulinum toxin was used as a therapeutic agent with good relief and no side effects. Further research may help evaluate the role of botulinum toxin into axial joints and spaces for the treatment of lower back pain as an alternative to corticosteroid containing treatments.

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Declaration of competing interest

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