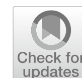
Spine Deformity (2022) 10:727–731
https://doi.org/10.1007/s43390-021-00449-6

CASE REPORT



Severe contracture in the lower extremity resulting from an osteoid osteoma of the lumbar spine in close proximity to neural elements in an adolescent: a case report

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Received: 30 March 2021 / Accepted: 20 November 2021 / Published online: 6 January 2022
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Abstract

Case We report a rare case of a right-sided L4-5 facet osteoid osteoma in a 11-year-old female who presented with lower extremity pain, contractures, and gait disturbance in the setting of failed radiofrequency ablation. After open excision and single-level posterolateral fusion with instrumentation, her symptoms resolved and her spinopelvic parameters normalized.

Conclusions This case highlights the contracture that may occur as a result of neurogenic pain from an osteoid osteoma in close proximity to neural structures.

Keywords Osteoid osteoma · Adult spinal deformity · Scoliosis · Spine · Adolescent

Introduction

Osteoid osteoma (OO) of the spine is rare, accounting for 10% of OO cases [1, 2]. Radiofrequency ablation (RFA) is safe and effective in treating spinal OO when medical management fails, and thermal protection techniques during RFA may be used when a lesion is within 1 cm of a neural structure [1–10]. Open surgery is reserved for situations in which OO is not accessible via RFA or is too close to neural structures such that thermal protection cannot be performed safely [11]. We present a patient who underwent open excision with single-level posterolateral instrumented fusion for a right-sided L4-5 facet OO in close proximity to neural structures, and discuss her outcome through 2-year follow-up.

The patient and her parents were informed that the data concerning the case would be submitted for publication, and they provided consent.

Case report

Clinical presentation

Our patient was an otherwise-healthy 11-year 11-month-old female when she was referred to our clinic. Preoperatively, she reported leg cramping and right lower back pain which was present at night and relieved by NSAIDs. She had knee contractures with spasticity and numbness of bilateral lower extremities. Her symptoms had been present for 9 months. She failed nonoperative treatments and underwent RFA with 6 weeks of relief, but her symptoms returned. On exam, she ambulated with a crouched gait. She had bilateral knee flexion contractures and asymmetry on Adams forward bend. She had 5/5 strength throughout bilateral upper and lower extremities and sensation was intact in all dermatomes. She had 1 + left patellar and bilateral Achilles reflexes. Her right patellar reflex was absent and she had 4–5 beats of clonus bilaterally. Straight leg raise reproduced pain in her legs and back.

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Diagnostic imaging and assessment

Preoperative radiographs showed that the patient was leaning forward in an effort to relieve her pain; her spinopelvic parameters normalized postoperatively (Table 1, Fig. 1). MRI was negative for neural impingement or intradural pathology. Single photon emission computed tomography (SPECT) and conventional CT showed a 0.8 × 1.3 × 0.5 cm lesion originating from the right L5 superior facet extending into the right L4-5 neuroforamen consistent with OO (Fig. 2). A second RFA procedure was considered, but in discussion with her interventionalist, it was felt that the lesion was too close to the right L4 nerve root without a sufficient cortical shell to allow for thermal protection and RFA.

Table 1 Summarizing the changes in spinopelvic alignment of the patient from baseline to 1 week and 2 years after surgery

Sagittal alignment	Baseline	1 week post-op	2 years post-op
TK (degrees)	40.3	47.8	46.9
LL (degrees)	32.3	40.7	65.7
PT (degrees)	32.2	31.6	15.9
SVA (mm)	53	13	6
PI-LL (degrees)	24.8	19.0	5.1

All measures are at neutral

TK thoracic kyphosis, *LL* lumbar lordosis, *PT* pelvic tilt, *SVA* sagittal vertical axis, *PI-LL* pelvic incidence minus lumbar lordosis (spinopelvic mismatch)

Operative procedure and postoperative course

As she had failed nonoperative treatment and RFA, surgery was recommended. She underwent open excision of the right L4-5 facet OO and single-level posterolateral fusion with instrumentation. She had an uncomplicated perioperative course and surgical pathology was consistent with OO. She was discharged home on postoperative day 2. By 4 months postoperatively, her pain had resolved and her contractures (Fig. 3) and asymmetry on Adams forward bend had improved. At 2-year follow-up, she was asymptomatic without pain, numbness, or paresthesias. Her SRS scores improved from her preoperative baseline (Table 2). Her neurologic exam is normal. Her imaging shows a solid fusion mass, normal coronal balance, and improvement in her spinopelvic parameters (Table 1, Fig. 1).

Discussion

While spinal OO causing nerve root irritation has been described in the medical literature [12–14], our patient's presentation with bilateral lower extremity contractures and spasticity represents a unique constellation of symptoms not routinely seen with OO. OOs have been shown to cause an

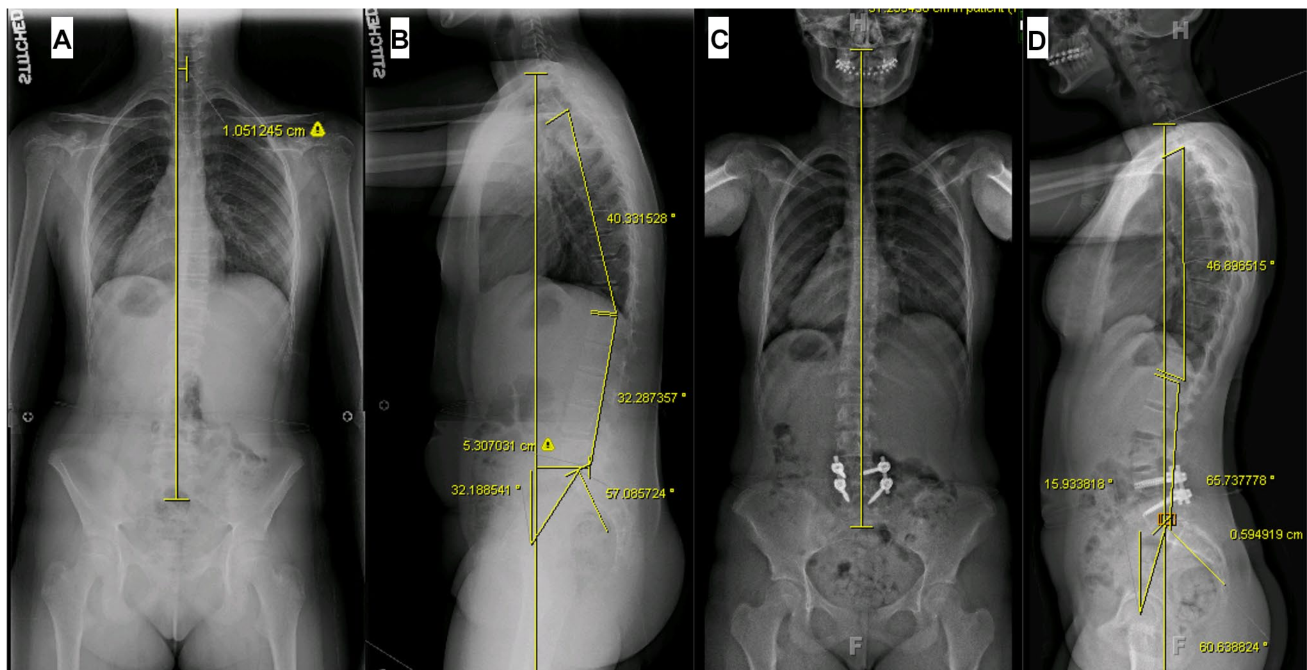

Fig. 1 Standing radiographs from before surgery (**A**, **B**) and 2 years after surgery (**C**, **D**). The patient appears to be leaning forward in her preoperative radiographs in an attempt to relieve pain. This is reflected in her spinopelvic parameters, which normalize postoperatively

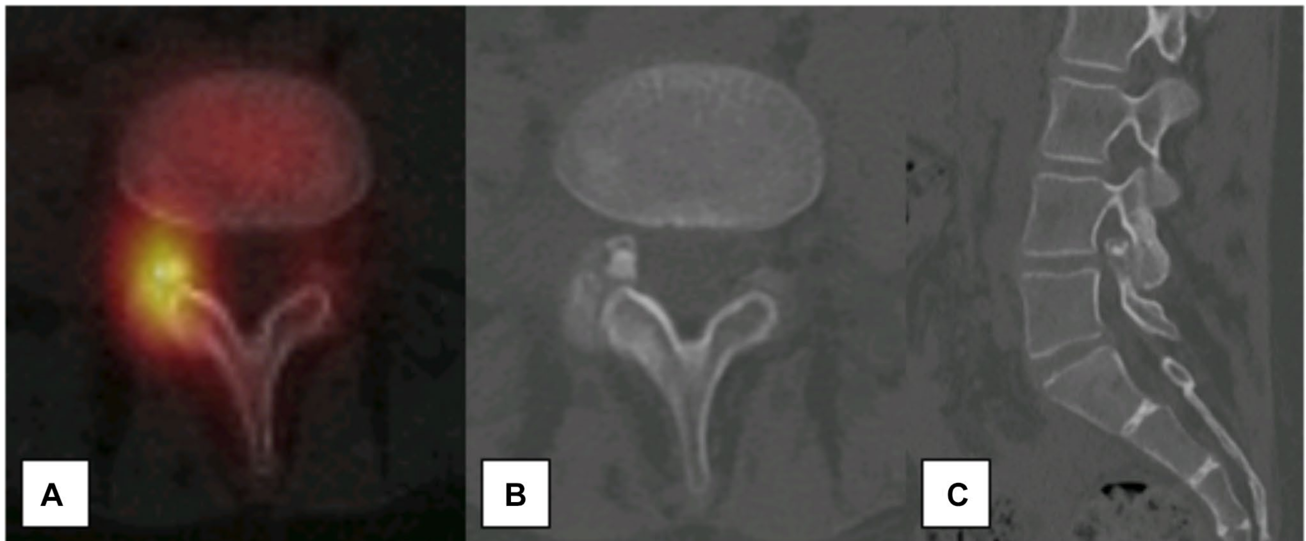

Fig. 2 **A** Axial SPECT image at the level of the L4 inferior endplate. **B** and **C** Axial and right parasagittal CT images post-ablation

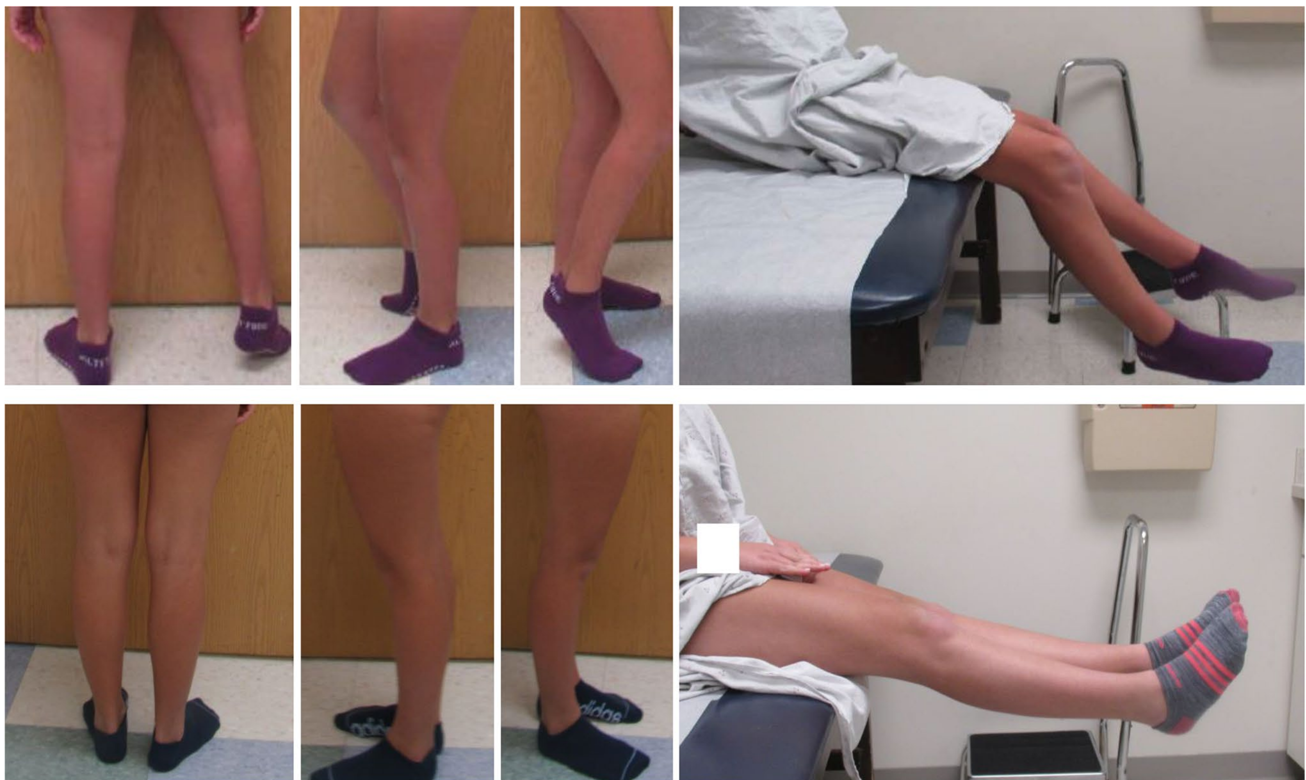

Fig. 3 Clinical photos before surgery (top) and after surgery (bottom)

inflammatory response through increased prostaglandin synthesis [15–17]; it is possible that our patient's symptoms were related to the local inflammatory environment generated by her unilateral facet lesion.

Percutaneous RFA emerged as a treatment option for OO in 1989, and by the late, 1990s had replaced open excision as the dominant approach for lesions in the long bones and pelvis [18]. Numerous clinical series have supported the safety and efficacy of RFA in spinal OO; however, lesions in close proximity to neural elements present a unique treatment challenge [2, 3, 18–21]. Complications associated with RFA of spinal OO are rare but include temporary pain, limited

Table 2 Preoperative and 2-year postoperative scoliosis research society 22-item (SRS-22) scores broken into domains

	Pre-op	2 years post-op
Pain	2.4	5
Function	3	5
Self-image	3.5	5
Mental Health	4	4.8
Satisfaction	2	5
SRS subscore	3.225	4.95

mobility, and extremity paresthesias [2]; thermal protection techniques during RFA may be used to mitigate these risks [19, 22]. OO recurrence rates following RFA have been reported at 0–12.5% [2, 3, 11, 13]. Our patient's symptoms returned 6 weeks after her RFA treatment and after a discussion with her interventionalist, the decision was made to proceed with open excision given the proximity of the lesion to the right L4 nerve root.

Open excision has been shown to be a safe and reliable approach for relieving pain and restoring function in patients with OO who have failed nonoperative measures [13, 14, 23–25]. Risks of open OO excision include incomplete removal and a potentially higher recurrence rate when compared with RFA, iatrogenic spinal instability, and nerve injury [4–6, 8]. We recommend performing an en-bloc resection when possible, instrumenting the area of resection to prevent instability, and using meticulous surgical technique when developing the interval between the lesion and surrounding neural tissues. Technologies including navigation and video-assisted thoracoscopic surgery may also be useful in localizing the lesion and preventing excessive bone removal. [7, 12, 24, 26].

Conclusion

We report a presentation of painful lower extremity symptoms and contractures from a right-sided L4-5 facet OO near neural elements in an adolescent female that was successfully treated using open excision and single-level posterolateral fusion with instrumentation. The authors encourage a multidisciplinary approach to the diagnostic workup and treatment of these lesions to optimize outcomes.

Author contributions All authors contributed to the study conception and design. Material preparation, medical record review, and data collection were performed by JCFC. The first draft of the manuscript was written by JCFC and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

Funding No funding was received for conducting this study.

Declarations

Conflict of interest Neither Dr. Clohisy nor Dr. Rubio have any relevant financial or non-financial interests to disclose. In the past 36 months, Dr. Gupta has received royalties or honoraria, served as a paid consultant or speaker, or had paid travel from DePuy, Medtronic, Innomed, Scoliosis Research Society (SRS), AO Spine, Alphatec, Medicrea, Mizuho, Globus, and OMeGA, and has had stock in Johnson & Johnson and Procter & Gamble.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent Written informed consent was obtained from the parents.

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