Acta Orthopaedica et Traumatologica Turcica 53 (2019) 77–80

Contents lists available at ScienceDirect

Acta Orthopaedica et Traumatologica Turcica

journal homepage: https://www.elsevier.com/locate/aott

A rare cause of type 1 complex regional pain syndrome: Osteoblastoma of the talus

Ozan Beytemür [a, *], Ümit Seza Tetikkurt [b], Serdar Yüksel [a], Mehmet Öncü [c]

[a] Bağcılar Training and Research Hospital, Department of Orthopedics and Traumatology, İstanbul, Turkey
[b] Bağcılar Training and Research Hospital, Department of Pathology, İstanbul, Turkey
[c] Bağcılar Training and Research Hospital, Department of Radiology, İstanbul, Turkey

ARTICLE INFO

Article history:
Received 31 October 2017
Received in revised form
15 June 2018
Accepted 27 June 2018
Available online 9 August 2018

Keywords:
Talus
Osteoblastoma
Complex regional pain syndrome
Foot
Pain

ABSTRACT

We report a 14-year-old boy who presented with pain in the left foot and ankle for about 9 months. The clinical symptoms of the patient suggested complex regional pain syndrome (CRPS). The radiographs and magnetic resonance imaging studies ravealed a bone tumor in the talus, consistent with an osteoblastoma. The patient underwent operative treatment with curettage and grafting. The complaints of the patient completely resolved by 6 months after surgery, and there was no recurrence at the postoperative 23th month follow-up.

Introduction

Complex regional pain syndrome (CRPS) is one of the most common conditions seen during orthopedic practice. The condition can actually be divided into two types. Type 1 CRPS is usually observed following a trauma or surgical procedure without any nerve injury,[1,2] while type 2 CRPS is observed after nerve injury.[1,2] Although type 2 CRPS may be detected in all age groups, it is more common among adults.[1] However, cases in the pediatric age group have been reported in the literature.[3]

Osteoblastomas account for approximately 1% of all primary bone tumors observed in the body.[4] They are usually observed on the spine and long bones.[5] Hence, osteoblastoma of the talus is considered a very rare condition.[5–8] Prior cases reported concerning the talus involve the neck of the talus.[5–9] To the best of our knowledge, osteoblastoma-dependent complex regional pain syndrome has previously only been reported in the literature by Schmidt et al in 1994.[10] Thus, the present case represents just the second report in the literature. This case involving osteoblastoma and CPRS is presented with the aim of increasing awareness of the cause–effect relationship between a bone tumor and CRPS.

Case report

A 14-year old male patient was referred to our clinic due to swelling and pain in the left foot and ankle. The patient's history included pain that had started without any trauma and gradually increased for about nine months. The patient reported that he had been unable to bear weight on that foot during the nine-month period. While the patient reported increasing pain with activity, the pain pattern had also become continuous in recent times. The patient had received non-steroid anti-inflammatory drug therapy and been told to rest; however, the complaints increased.

Upon physical examination, swelling and peripheral edema of the left foot and ankle, hyperhidrosis, and increased brightness and atrophic changes to the nails were found to be present, along with a speckled pattern on the skin. The excessive sensitivity of the skin and muscles, especially cold intolerance, hyperalgesia, allodynia, and related extremity coldness, was detected during the palpation examination. A limitation to the range of motion of the left ankle (dorsiflexion 10°, plantar flexion 25°), weakness of the muscles, and

* Corresponding author. Mimar Sinan Cad.6.sok.34200, Bağcılar, İstanbul, Turkey. Tel.: +905308200992.
E-mail addresses: beytemur@yahoo.com (O. Beytemür), umitseza@gmail.com (Ü.S. Tetikkurt), Serdar84yuksel@gmail.com (S. Yüksel), drmehmetoncu@yahoo.com.tr (M. Öncü).

Peer review under responsibility of Turkish Association of Orthopaedics and Traumatology.

https://doi.org/10.1016/j.aott.2018.06.011

a 2 cm atrophy of the left calf muscles were present. Radiological examination revealed a diffuse osteopenia on the bones of the left foot and ankle (Fig. 1). These symptoms were used to diagnose CRPS using the Budapest Criteria.[11] There were no other findings revealed on the direct X-ray due to the diffuse osteopenia, although a 28 × 26 mm lesion and an intense hypointense contrast uptake in the T1 scans, as well as an intense hyperintense contrast uptake in the T2 scans, were detected on the lateral process of the talus of the left ankle by means of magnetic resonance imaging (Fig. 2). An increase in effusion in the tibiotalar joint, diffuse bone marrow edema on all the bones of the foot and ankle, an increase in the edematous signal, as well as thickening in the cutaneous-subcutaneous tissue were also detected (Fig. 2). Computed tomography imaging was performed in order to clearly reveal the anatomic margins of the lesion. A 26 × 25 mm lesion, including calcifications in patches in the center surrounded by a thin reactive sclerotic bone, was detected in the lateral side of the tarsal sinus of the left talus (Fig. 3). The bone scintigraphy revealed a lesion with a significant increase in activity during all three phases corresponding to the talus area of the left foot (Fig. 4). As a definite diagnosis could not be established based on these findings, an open bone biopsy was also performed.

After osteoblastoma was diagnosed histopathologically, a permanent surgical treatment was planned for the patient. A pneumatic tourniquet was applied at elevation and the lateral process of the talus of the left foot was accessed using the anterolateral approach during the surgical procedure. The lesion was curetted away using a curette, while the lesion walls were enlarged with a high-speed burr. The procedure continued with the cauterization of the lesion walls using an argon cautery, and grafting was performed using an autologous bone graft removed from the iliac crest. The histopathological examination of the pathological material removed during the surgery reported a lesion consisting of anastomizing woven bone trabeculae, which were lined with a single but prominent layer of osteoblasts, and intervening hypervascular intertrabecular spaces (H.E. × 100). Thus, the diagnosis of osteoblastoma was confirmed (Fig. 4).

During the early postoperative period, a non-weight-bearing range of motion exercises were begun. The patient was allowed to load onto the operated foot after six weeks, and he was referred to a physiotherapy clinic. After six weeks, the patient was allowed to bear weight on the operated foot, and treatment with a physical therapist was planned. The patient underwent elevation, retrograde massage, TENS (transcutaneous electrical nerve stimulation), and contrast bathing three times per day, and an active and passive range of motion exercises were applied. The physical therapy modalities were applied for a total of six weeks. Following the surgery and physical therapy protocol, the swelling and color changes, as well as the associated hyperalgesia and allodynia, reported by the patient during the early months were regressed. Within six months, all the symptoms were resolved. Recurrence was assessed by means of direct X-ray and clinical examination every three months for the first year, and then once per year thereafter. During the final physical examination, which was performed in the 23rd

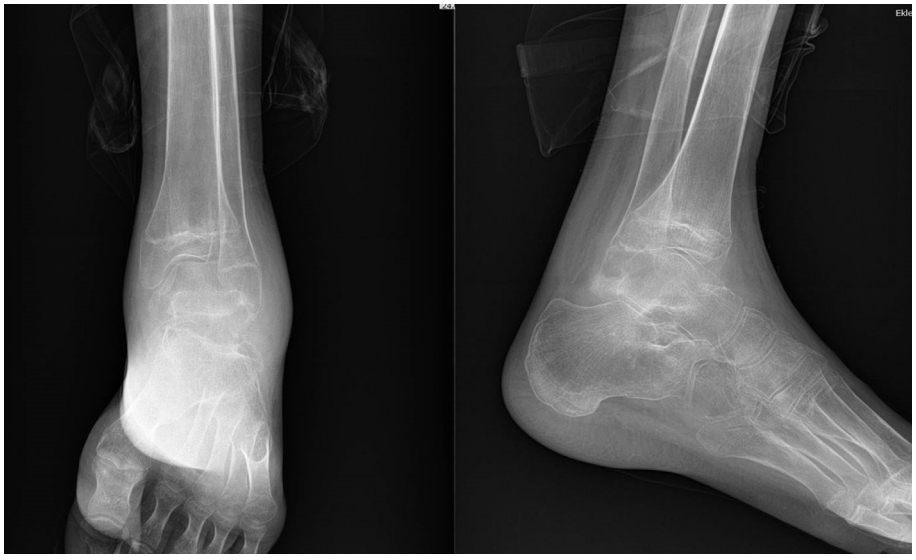

Fig. 1. Preoperative X-ray images.

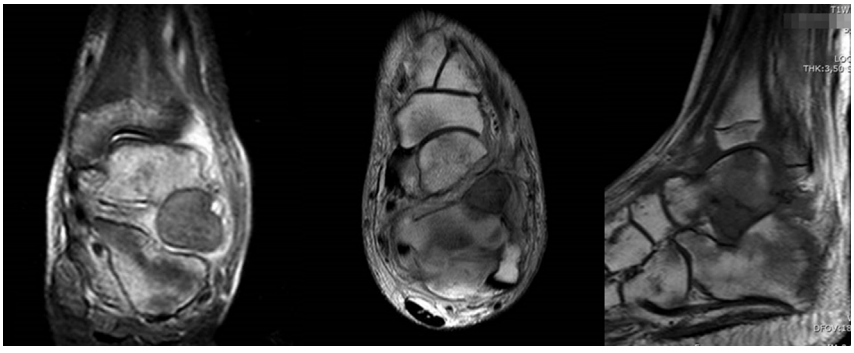

Fig. 2. Preoperative CT images.

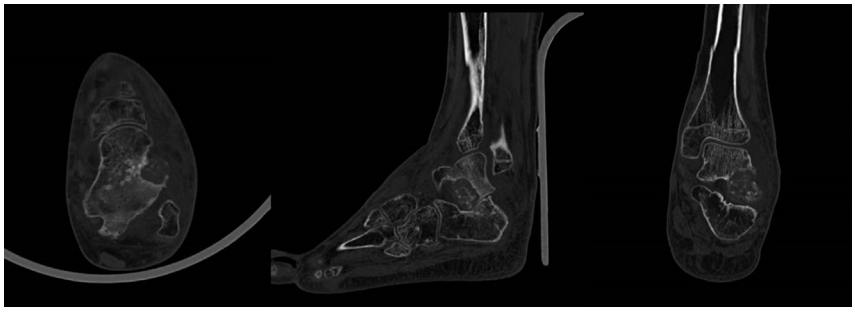

Fig. 3. Preoperative MR images.

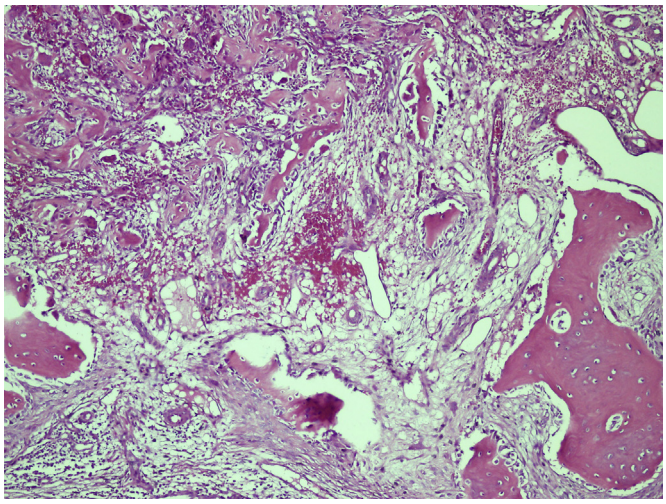

Fig. 4. Consisting of anatomosing woven bone trabeculae, lined by a single but prominent layer of osteoblasts, and intervening hypervascular intertrabecular spaces (H.E × 100).

postoperative month, the extensive osteopenia was also found to be completely resolved upon radiological examination, and no evidence of recurrence was seen (Fig. 5). The patient signed an informed consent form so that his data could be used and published in the present case report.

Discussion

Complex regional pain syndrome is a painful condition that particularly affects the limbs and thereby restricts the patient's daily life.[1] Although its etiology remains unclear, CRPS can be classified into two types.[1] Type 1 CRPS usually starts with a traumatic event or surgical procedure,[1] while type 2 CRPS is observed with nerve injury.[1,11] The clinical presentation includes temperature differences and color changes in the skin, subcutaneous thickening, peripheral edema, pain, hyperalgesia, restricted movement of the joint, and osteopenia detected radiologically.[1,2,11] The presented case involved gradually increasing pain in the left foot and ankle, sensitivity to environmental stimuli, restricted movement, edema, and color changes to the skin for approximately one year. Since diffuse osteopenia was detected in the direct X-ray imaging, the patient was diagnosed with type 1 CRPS along with the clinical findings. The advanced radiological tests performed on the patient due to the unknown etiology detected a lesion in the talus; therefore, such a tumoral formation was considered to be the cause of the patient's CRPS. Complex regional pain syndrome is most commonly detected in adults,[1,2] although reports concerning the pediatric age group can also be found in the literature.[3] The etiology usually includes trauma or a surgical procedure.[1,2,11] Nevertheless, the exact etiology cannot be determined in some cases.[1] As our case involved a teenager who does not have any trauma in his history, the case represents is a rare example of CRPD.

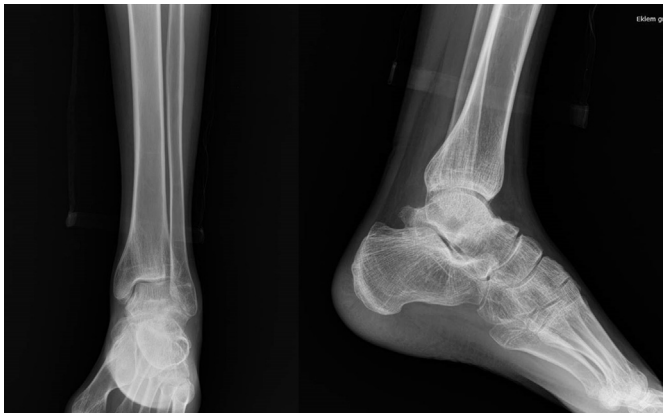

Fig. 5. Postoperative 23th months X-ray images.

Osteoblastomas are a very rare form of tumor.[5–7,9] They account for approximately 1% of all bone tumors and 3.5% of all benign bone tumors.[12] They are most commonly detected on the spine and long tubular bones, while foot lesions are rare.[5,9] However, the most common area of invasion of the foot has been reported to be the talus.[5] The involvement of the talus neck is more common, whereas the involvement of the talus body is quite rare.[5,9] Capanna et al reported that they detected only one case with osteoblastoma of the talus body among seven cases with osteoblastoma.[9] Osteoblastoma was detected on the lateral process of the talus body in the presented case. Such an area very rarely experiences osteoblastoma invasion. Osteoblastomas are more common during the second and third decades of life, as well as in men.[5,9,13] The presented case involved a 14-year old male. Pain during night that is relieved by salicylates in osteoid osteoma is not usually observed in osteoblastoma.[5,6] Our case experienced increasing pain with activities, and he did not report any pain relieved by salicylates during the night.

Such a tumor is radiologically observed as a lytic lesion over 2 cm in size, which may include calcification in patches and be surrounded by a thin sclerotic rim.[9] The most common criterion used for the differential diagnosis of osteoid osteoma is the size.[5] In the past, lesions larger than 2 cm were defined as osteoblastoma, whereas lesions smaller than 2 cm were called osteoid osteoma.[9] However, following the detection of osteoblastoma lesions smaller than 2 cm, the limit was reduced to 1.5 cm.[5,14] Computed tomography is superior to other imaging methods in terms of showing the anatomic formation and calcifications of the lesion.[7] Although magnetic resonance imaging is better at showing the association of a lesion with the soft tissue, it may be considered a malign formation due to a clamorous image.[5] (Figs. 2 and 3)

Since a final diagnosis could not initially be established due to the rare location of the lesion, an open bone biopsy was performed, as well as a permanent surgical procedure. The treatment method usually preferred for osteoblastoma cases is open curettage and grafting.[9] A wide resection may also be preferred if the location is not anatomically problematic for functional purposes.[15] We preferred curettage and grafting as the surgical approach in the present case. Moreover, lower recurrence rates have been reported with adjuvant therapy methods (high-speed burr, phenol, liquid nitrogen, argon cautery, etc.) performed in addition to the curettage.[16] We routinely additionally perform argon light cauterization after the dilatation of the lesion walls using a high-speed burr following the curettage of benign tumors. Hence, we also did so in the present case. We did not detect any findings indicting relapse during the follow-up visit of the patient at the 23rd postoperative month.

Consequently, musculoskeletal tumors should also be considered in cases with an unknown etiology for treatment resistance in instances of type 1 complex regional pain syndrome along with a history of trauma or surgery.

Conflict of interest

There is no conflict of interest.

Funding

There is no funding.

References

1. Shah A, Kirchner JS. Complex regional pain syndrome. *Foot Ankle Clin*. 2011;16:351–366.
2. de Mos M, Sturkenboom MC, Huygen FJ. Current understandings on complex regional pain syndrome. *Pain Pract*. 2009;9:86–99.
3. Rogers JN, Valley MA. Reflex sympathetic dystrophy. *Clin Podiatr Med Surg*. 1994;11:73–83.
4. Robert K, Heck, Canal ST, Beaty JH. *Campbell's Operative Orthopaedics*. Benign/Aggressive Tumors of Bone. 11th ed. Mosby Elsevier; 2008:892. ISBN: 978-0-8089-2361-9.
5. Kroon HM, Schurmans J. Osteoblastoma: clinical and radiologic findings in 98 new cases. *Radiology*. 1990;175:783–790.
6. Patil KS, Somayaji HM, Nandi SS. A rare case Report of A Young male patient presenting with osteoblastoma of the talus. *J Orthop Case Rep*. 2014;4:22–24.
7. Mir NA, Baba AN, Maajid S, Badoo AR, Mir GR. Osteoblastoma of body of the talus–Report of a rare case with atypical radiological features. *Foot Ankle Surg*. 2010;16:e24–e26.
8. Duan XJ, Yang L. Removal of osteoblastoma of the talar neck using standard anterior ankle arthroscopy. A case report. *Int J Surg Case Rep*. 2016;23:52–55.
9. Capanna R, Van Horn JR, Ayala A, Picci P, Bettelli G. Osteoid osteoma and osteoblastoma of the talus. A report of 40 cases. *Skelet Radiol*. 1986;15:360–364.
10. Schmidt A, von Gontard A. Sudeck's disease in osteoblastoma of the right talus and depression in a 12-year-old boy. Case report. *Z Kinder Jugendpsychiatr*. 1994;22:123–129.
11. Soylev GO, Boya H. A rare complication of total knee arthroplasty: type l complex regional pain syndrome of the foot and ankle. *Acta Orthop Traumatol Turc*. 2016;50:592–595.
12. Unni KK. *Bening Osteoblastoma in Dahlin's bone tumours. Generel aspect and data on 11087 cases*. th ed. Philadelphia: Lippincott-Raven; 1996.
13. Marsh BW, Bonfiglio M, Brady LP, Enneking WF. Benign osteoblastoma: range of manifestations. *J Bone Jt Surg Am*. 1975;57:1–9.
14. McLeod RA, Dahlin DC, Beabout JW. The spectrum of osteoblastoma. *AJR Am J Roentgenol*. 1976;126:321–325.
15. Atesok KI, Alman BA, Schemitsch EH, Peyser A, Mankin H. Osteoid osteoma and osteoblastoma. *J Am Acad Orthop Surg*. 2011;19:678–689.
16. Saglik Y, Atalar H, Yildiz Y, Basarir K, Gunay C. Surgical treatment of osteoblastoma: a report of 20 cases. *Acta Orthop Belg*. 2007;73:747–753.