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Case report

Osteosarcoma of the distal fibula and reconstruction of the ankle using inverted fibula, a case report

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ABSTRACT

Introduction and importance: Osteosarcomas are malignant primary bone tumors of mesenchymal origin producing osteoid material and has peak incidence in adolescents. Distal lower limb tumors are rare and can negatively affect ankle joint stability.

Case presentation: A 24-year-old female who has newly graduated from college presented with distal fibular mass measuring around 5 × 15 cm located on the lateral aspect of the right ankle over a period of 2 months. The mass located on the lateral aspect of the right ankle that was hard, oval and measuring around 5 × 15 cm and originating from the fibula. The overlying skin was normal with no discharging sinuses. Distal neurovascular examination was normal with no lymphadenopathy. Imaging using X-rays and MRI as well as pathological examinations thereafter has proven the diagnosis. She was planned for wide surgical resection at distal fibula and ankle reconstruction after neoadjuvant chemotherapy, then for adjuvant chemotherapy. Ankle reconstruction using fibular autograft was used after its reversal and was then stabilization by syndesmotic screws. She has clinically good outcome.

Clinical discussion: Surgery with extensive and meticulous dissection remains the cornerstone for treating osteosarcomas affecting distal fibula. Neoadjuvant and adjuvant chemotherapy are important for managing micrometastasis. Ankle reconstruction and be performed using different methods with good outcomes.

Conclusion: Lesson learnt is that ankle reconstruction using fibular autograft can be used after reversal and stabilization by screws with good outcome for managing distal fibular osteosarcomas. However, this finding needs to be strengthened with future reports.

1. Introduction

Osteosarcomas are malignant primary bone tumors of mesenchymal origin that produce an osteoid material and have a peak incidence in adolescents [1]. Tumors affecting the distal parts of the lower extremities are uncommon tumors with most tumors being synovial cell sarcoma, osteosarcoma, and Ewing's tumor [1] with a significantly lower mortality rates than tumors in other sites [2].

The standard diagnostic approach for suspected bone lesions includes a preoperative imaging assessment. It includes radiographic imaging in form of minimum of two views of X-rays for the whole affected bone and the underlying or nearby joint which can reveal a lesion with osteoblastic or osteolytic features, a periosteal reaction (Codman's triangle) and possible soft tissue mass. MRI is more preferred in detailing the extension of the mass into the adjacent structures of joints, soft tissues or neurovascular structures. CT scan and bone scintigraphy are helpful in detailing bony features of any cortical abnormalities, fractures and bone mineralization. CT scan for other common areas of metastasis like the chest can also be done to rule out distant metastasis. While biopsy can be performed pre-operatively; intra-operative biopsy is of no doubt performed confirming the suspicion [3].

2. Patient information

The patient is a 24-year-old female who has newly graduated from college with distal fibula osteosarcoma. She has no known allergies and

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not known to have diabetes or hypertension or any other chronic illnesses. She does not take any chromic medications. She does not smoke nor drink alcohol. She has no family history of similar presentation nor taking chronic medications.

3. Clinical findings

Physical examinations revealed an unwell, vitally-stable patient who was pale but not jaundiced or cyanosed. She has no lymphadenopathy and all other systems were normal. The mass was located on the lateral aspect of the right ankle that was oval in shape measuring around 5 × 15 cm, hard in consistency originating from the fibula. The skin overlying the lesion was normal with no discharging sinuses. Distal neurovascular bundle examination was normal with no lymphadenopathy. Decreased ankle ROM.

4. Timeline

Her condition started 2 months prior to presentation with left distal leg and ankle pain. The pain characterized by with a deeply seated nature and severe. It started gradually with progressive increase in swelling. The pain has not decreased with rest and disturbed her sleep at night and her work performance. It affected her walking distance.

The pain was associated with swelling on the lateral side of the ankle, measuring around 5 × 15 cm. It was rapidly increasing in size and associated with decreasing ankle's ROM. There was no associated sinus discharge and the skin over and around it showed no remarkable features. She also complained of fever, weight loss and appetite loss and fatiguability. She has no significant past or family history and her other systems were clear.

5. Diagnostic assessment and interpretation

Lab investigation showed anemia with elevated ESR and normal CRP levels. RFT, LFT and Bone profile was normal. X-ray of the left ankle showed a lytic/sclerotic distal fibular mass with periosteal reaction and sunburst appearance along with Codman triangle; findings consistent with conventional intermedullary osteosarcoma. An MRI of the whole leg and foot showed a heterogenous distal fibular mass (Low T1 and High T2) with soft tissue involvement and no skin or neurovascular involvement. CT scan of the chest revealed no features of metastasis. Core needle biopsy showed satellite cells that had highly malignant

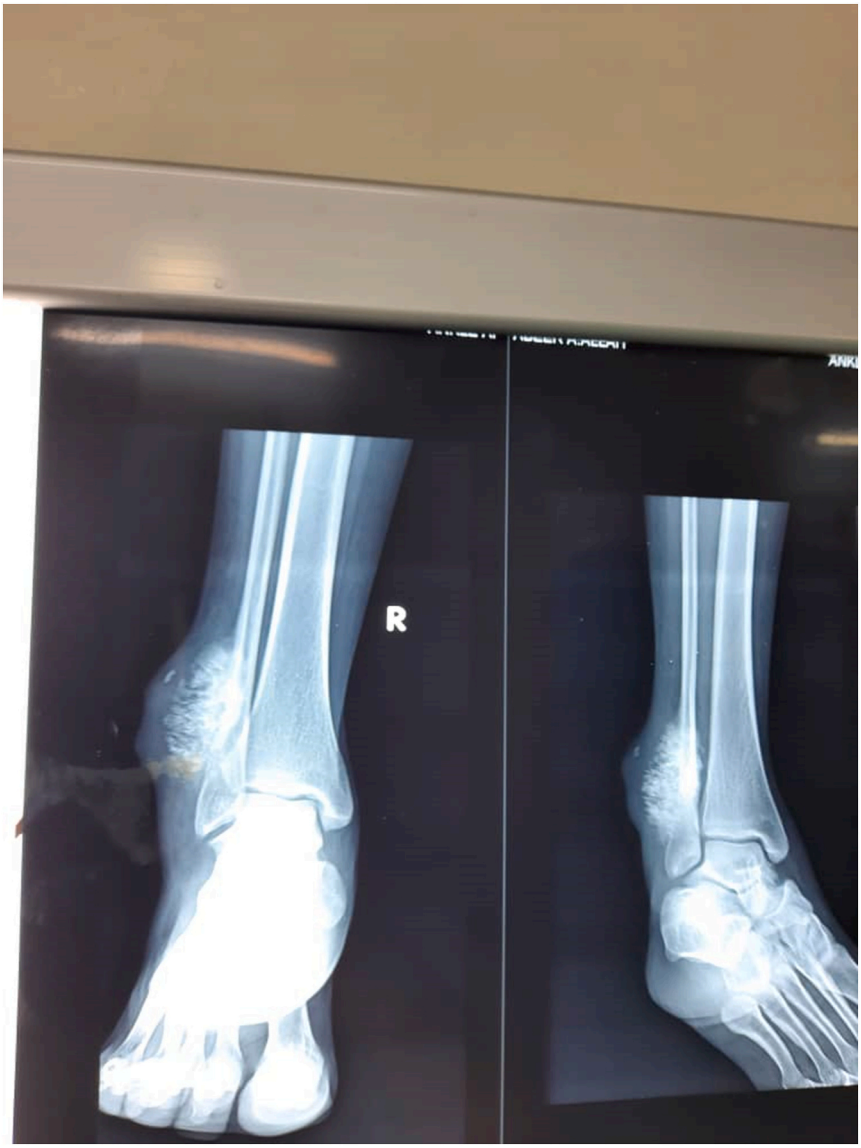

Fig. 1. X-ray of the left ankle showing a lytic/sclerotic distal fibular mass with periosteal reaction and sunburst appearance along with Codman triangle.

features evident by high mitotic figures, and increased nuclear: cytoplasmic ration and high cellular atypia in addition to malignant cells that produce osteoid materials; features consistent with conventional osteosarcoma (Fig. 1).

6. Intervention

The patient was planned for wide surgical resection at distal fibula and ankle reconstruction after neoadjuvant chemotherapy, then for adjuvant chemotherapy.

Surgery was done after proper counselling and informed consent; after completion of the neoadjuvant chemotherapy. It was done under the effect of spinal anesthesia using a tourniquet without exsanguination. The fibula has been approached through the lateral approach. A wide marginal excision of the distal fibula was done along with the overlying skin with meticulous dissection and homeostasis.

The proximal part (15 cm) of the fibula was resected and inverted upside down with the head becoming the most distal part of the lateral malleolus to reconstruct the ankle. It was then fixed to the remaining part of the fibula by 1/3 fibular plate 10 holes with 2 syndesmotic screws 4 mm 4 cortices under imaging intensifier. This resulted in a stable ankle with stable full range of motion. The skin was then closed in layers after insertion of the drain that was removed after 24 h. A posterior slap was put to allow syndesmotic healing (6 weeks). The patient was mobilized immediately post-operatively using Zimmer frame of low touch weight bearing.

The surgery was performed by an orthopedic oncology consultant assisted by an orthopedic oncology trainee.

7. Follow-up and outcome

6 weeks post-operatively, the posterior slap was removed and the patient started gentle R.O.M exercise at the ankle and gradually increasing her mobility. She underwent an uneventful post-operative recovery course with good wound healing and no complications and then she was transferred to receive her adjuvant chemotherapy. The resected part of the fibula was sent to histopathology which confirmed clear margins of the excision and excellent tumor necrosis rate as a response to the neoadjuvant regimen. The last follow-up was performed a week before submitting the manuscript with no complications related to either the wound itself or to the part of the fibula inverted.

8. Discussion

Osteosarcoma is a malignant primary bone tumor. It usually affects patients in their second decade of life [1]. The challenging aim of surgery is to retrieve the functionality of the ankle joint and avoid deformities to prevent future arthrosis. The main reason for recurrence and unfavorite outcomes is thought to be the inadequate margins of resection [4]. Preservation of the lateral malleolus is generally recommended when the resection is done at least 15–20 mm above the distal tibiofibular joint or at a distance of 5 mm above the growth plate; otherwise, reconstruction is better recommended for restore joint functionality. Various reconstruction techniques have been described with good outcomes [5,6].

Regarding ankle joint reconstruction, whether due to distal fibular osteosarcoma or any other cause, literature regarding this is limited with most knowledge here are case reports or series. It has not been performed usually, and only handful of cases has been described. Lublinar et al. managed a case of such procedure in 1985 for an aneurysmal bone cyst with good results 2.5 years after surgery [7], while Sirveaux et al. described a case of a distal fibular chondromyxoid fibroma t in a 15-year-old patient that was managed with cryopreserved allograft with good outcome in 2004 [8]. Jamshidi et al. have used an allograft distal fibula in their reconstruction for their 4 patients with malignant tumors with complete range of motion and ankle stability [9]. Similar to our case, Leibner et al. and Capanna et al. posited the reconstruction to be performed through a fibular graft above the site of the lesion which should be settled in place through a syndesmotic screw and connecting the ligaments to the fibular graft in an attempt to preserve the bone normality as possible and avoid valgus deformity as possible [10,11].

In 2008, Rui Niimi et al. conducted a case series involving 10 patients to assess the prognosis and limb functionality after limb-salvage surgery. They have described an 80 % survival rate, a 90 % limb preservation rate as well as an 88 % mean functional score. They have found skin-related post-operative complications in 3 patients and orthopedic-related problems in 4 patients including fracture, non-union and prosthesis loosening [12].

Adyb Adrian Khal et al. conducted a retrospective case series on 10 patients in 2012, in which they have evaluated the outcome of ankle joint biomechanics after distal fibular resection. In addition to the fact that 6 patients have survived and 4 have died, they have described a stable ankle in all 10 patients with no local recurrence. However, they have described other complications including chronic ankle pain and external peroneal nerve palsy [13].

Not very much different to this case, Saadon et al. [14] reported a case of an 11-year-old boy with osteosarcoma of the distal fibula who underwent a limb salvage surgery with removal of the distal fibula and retaining of his foot instead of radical amputation. In our case; however, we tried to reconstruct the ankle with fibular graft by having the fibular resected and then inverted upside down with its head becoming the most distal part of the lateral malleolus to reconstruct the ankle. The fibula was then stabilized in place using syndesmotic screws.

9. Conclusion

The educational objective for this case is that osteosarcoma of the distal fibula is usually managed through surgery with meticulous dissection and clear margins and possible reconstruction. Adjuvant and neoadjuvant chemotherapy play a significant role in dealing with micrometastasis. In this case we report an ankle reconstruction using a fibular auto-graft that was reversed upside down and then put in place and stabilized using syndesmotic screws; this method yielded a good outcome in our 24-year-old female patient. More future cases are needed to strengthen these outcome findings.

This case has been reported in line with the SCARE criteria [15].

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

Ethical approval was obtained from Ethical committee at Ibrahim Malik Teaching Hospital.

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Author contributions

1. Hassan Mohammed Hassan Elbahri: Involved in study design, data acquisition, drafting the article, revising it critically and finally approved the manuscript.
2. Hozifa Mohammed Ali Abd-Elmaged: Involved in conception of the study design, drafting the article and finally approved the manuscript.

3. Mohamed Abdulkarim: Involved in conception of the study design, drafting the article and finally approved the manuscript.

Registration of research studies

Not applicable.

Guarantor

Mohamed Abdulkarim.

Declaration of competing interest

Authors report no conflict of interest of any sort.

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