Case Report

Pediatric Chronic Fibular Pandiaphyseal Osteomyelitis: Case Report With Literature Review

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Received on: 04-Oct-2020 Accepted for Publication: 22-Feb-2021

ABSTRACT

Background: Chronic Osteomyelitis is a devastating infection causing severe disability in the pediatric population due to its often late presentation. Fibular osteomyelitis is relatively uncommon, with only a few documented reports worldwide.

Case Presentation: We present a case of a 9-year old male presenting with left leg pain subsequently diagnosed to have chronic pandiaphyseal osteomyelitis of the fibula, who was then treated with En bloc resection and culture guided antibiotic therapy for Methicillin-resistant Staphylococcus aureus. Full functional recovery of the left leg was noted at two weeks follow-up with normal infection markers after four weeks.

Conclusion: Diffuse osteomyelitis of long bones are a challenge to treat, as the gold standard of sequestrectomy and debridement may result to poor limb function. Chronic fibular osteomyelitis may be treated just like a tumor of the fibula, wherein en bloc resection can result to good functional outcome.

INTRODUCTION

Osteomyelitis is an inflammatory condition affecting the bone and bone marrow, brought about by an infection [1-3]. Left untreated, this condition will result to the formation of dead, cortical bone associated with abscess formation, and surrounding bone formation; the sequestrum and involucrum [1]. Delay in presentation in children usually results in chronic osteomyelitis, requiring surgery and chemotherapy. Staphylococcus aureus is the most commonly isolated organism in these cases [4]. However, the Methicillin-resistant strain (MRSA) now accounts for more than one-third of positive cultures [4-6]. Fibular osteomyelitis is relatively uncommon, with only a handful of cases reported in literature with only segmental involvement treated successfully with sequestrectomy and antibiotic therapy [2, 7-14]. Here we present a case of MRSA positive chronic osteomyelitis involving the entire diaphysis of the fibula and the subsequent treatment involving en bloc resection.

CASE REPORT

1. History and Physical Examination.

A 9-year old previously well, immunocompetent male presented at our institution with a seven month history of left leg pain and swelling after a fall from standing height. He was not brought for consult until persistence of pain and difficulty in ambulation. Consult was done at a local hospital, where he was diagnosed with cellulitis after normal radiographs, and was given Amoxicillin/Clavulanic acid for ten days with noted improvement of pain. Five months later, there was sudden recurrence of the swelling, progressively increasing in severity. Repeat laboratories revealed an elevated erythrocyte sedimentation rate (ESR), and he was again given Amoxicillin/Clavulanic acid for ten days, with complete resolution of swelling. Two weeks prior to consult, the symptoms returned, and on physical examination, the left leg (Fig. 1) was warm, non-erythematous, with generalized tenderness, but without pain on knee and ankle range of motion. Neurovascular status was normal,

and infection and tumor workup returned with negative results, except for elevated alkaline phosphatase and ESR.

Fig. 1 (A, B and C)
Clinical picture of the patient, showing swelling of left lateral leg without any surrounding erythema or skin lesion. No sinus tracts.

2. Imaging

Radiographs of the left leg showed expansion and cortical thickening mixed with areas of lysis in the left fibular shaft, suggestive of chronic osteomyelitis (Fig. 2). Tubular, obliquely oriented lucencies in the mid and distal diaphyses were thought to represent sinus tracts and an oblique lucency noted in the proximal fibular shaft was suggestive of a pathologic fracture malunion, with irregular callus formation. MRI of the left leg showed non-union fracture at the proximal shaft of the left fibula, with marked periosteal thickening throughout the fibula, as well as intraosseous abscess, consistent with hematogenous osteomyelitis (Fig. 3).

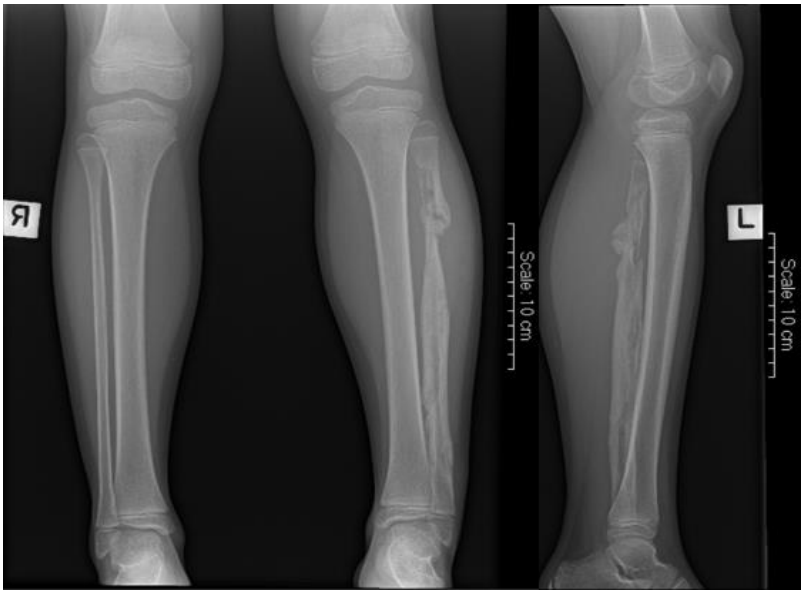


Fig. 2a Anterior-posterior left leg radiograph of the patient
Fig. 2b Lateral left leg radiograph of the patient

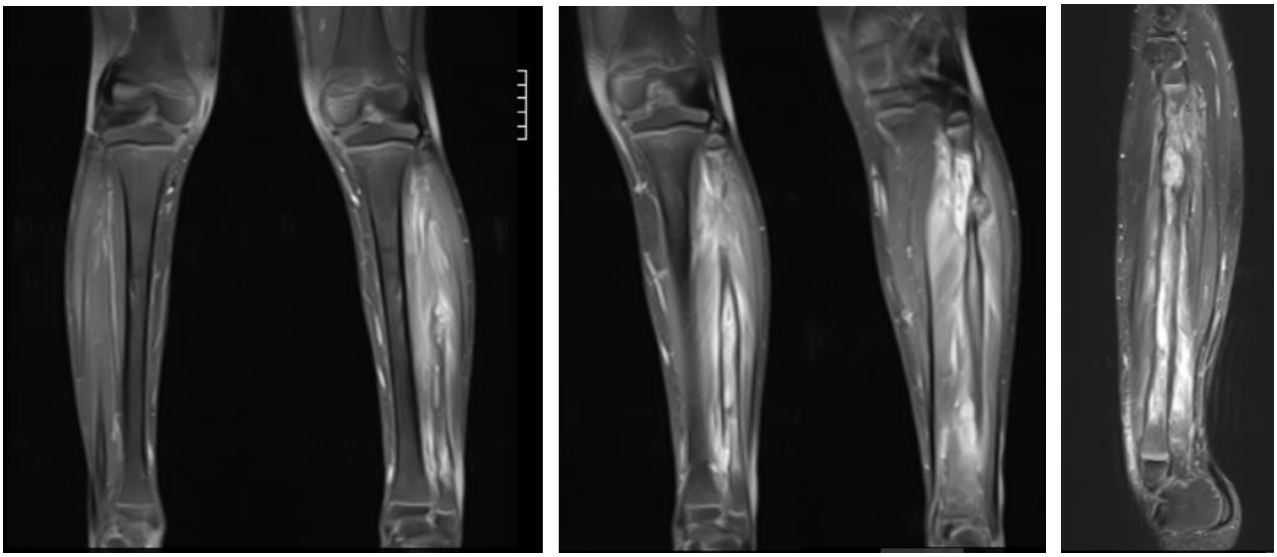


Fig. 3 Left leg MRI showing hyperintensity representing bone marrow edema, as well as intraosseous abscess, characteristic of hematogeous osteomyelitis. There is a non-union fracture at the proximal shaft of the left fibula as well as marked periosteal thickening throughout the fibula

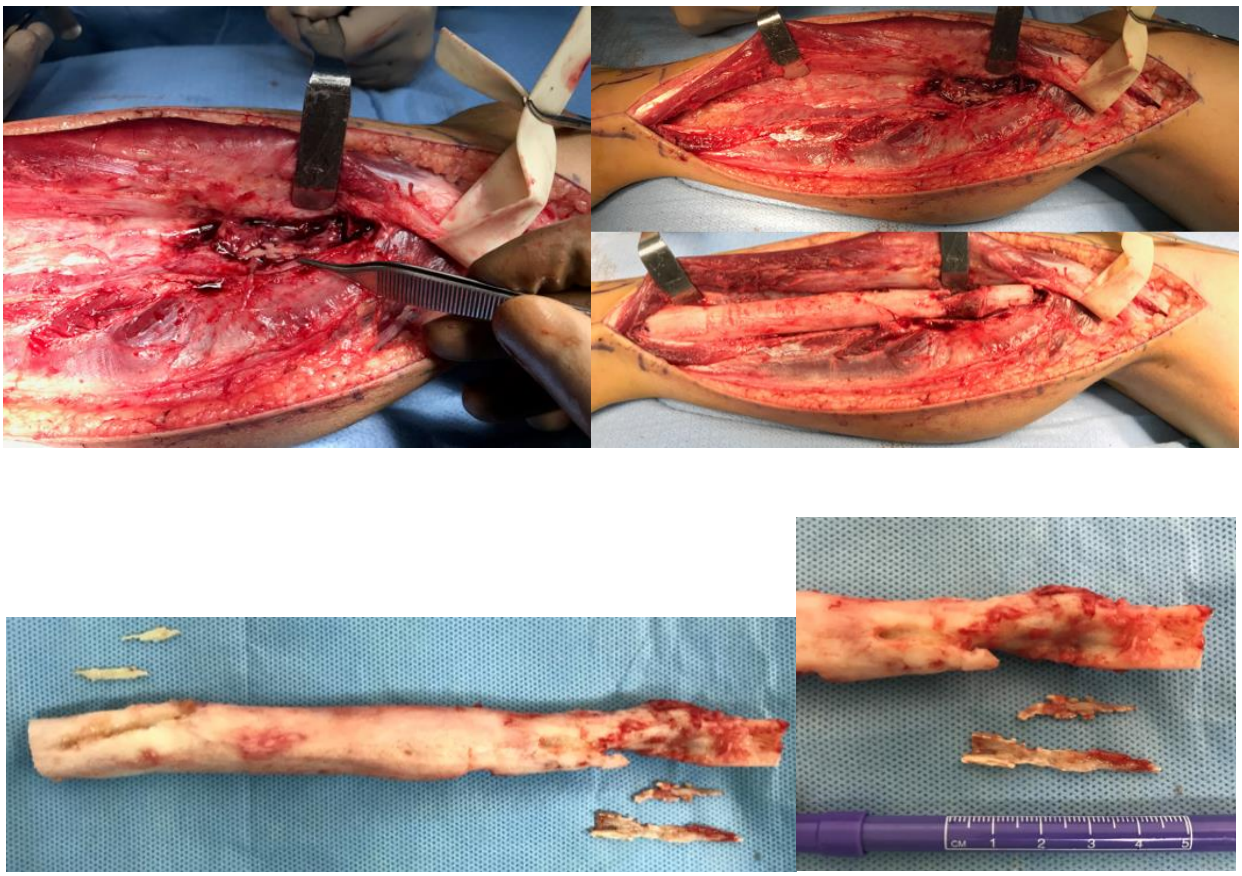


Fig. 4 Intraoperative pictures showing sequestra from the left fibula

3. Operation and Post-Operative Course.

The patient was then planned for sequestrectomy and debridement, with possible En bloc resection of the left fibula, to be done under general anesthesia. Pediatric cardiopulmonary clearance and parental consent was obtained. The patient then underwent the contemplated procedure without any complications (Fig. 4). Intraoperatively, the entire fibular shaft was noted to have multiple cloaca draining purulent material, with

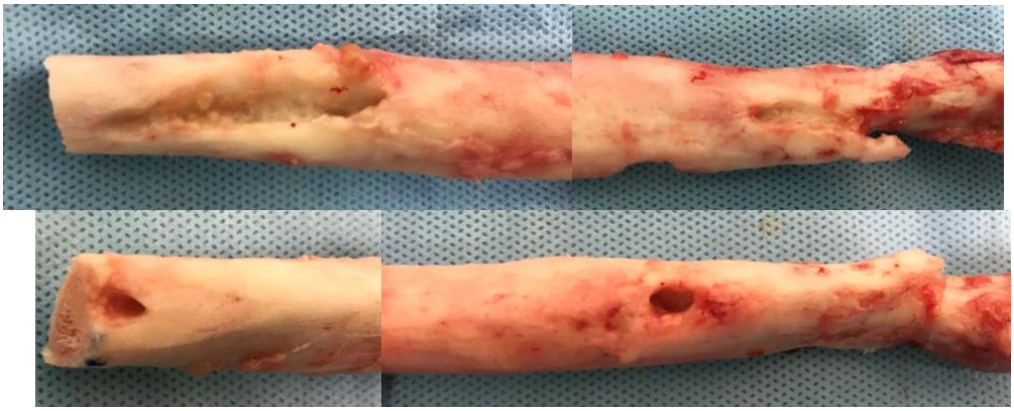


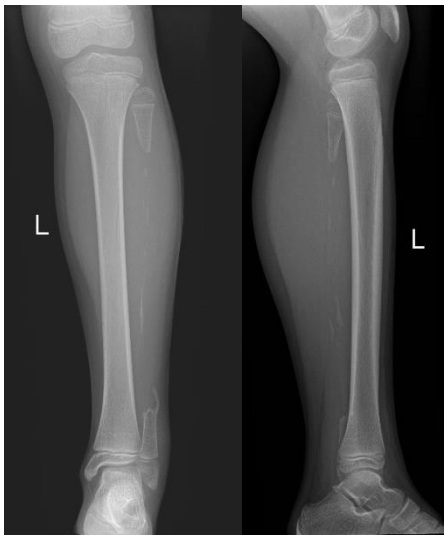
Fig. 5 Intraoperative pictures showing multiple cloaca in the left fibula

Postoperative

Fig. 6a anterior-posterior left leg radiograph
Fig. 6b lateral left leg radiograph

1 month follow-up

Fig. 7a anterior-posterior left leg radiograph
Fig. 7b lateral left leg radiograph



10 month follow-up

Fig. 8a anterior-posterior left leg radiograph
Fig. 8b lateral left leg radiograph

1 year follow-up

Fig. 9a anterior-posterior left leg radiograph
Fig. 9b lateral left leg radiograph

1 year follow-up

Fig. 10a anterior-posterior left ankle radiograph
Fig. 10b lateral left ankle radiograph

several fragments of sequestra (Fig. 5). Specimens were sent for culture and sensitivity. Due to the widespread involvement of the fibula, en bloc resection was carried out, leaving at least five centimeters of fibula, both distally and proximally, to maintain ankle stability and muscular attachments, respectively. The left fibula was sent for histopathology and debridement of soft tissue was performed along with copious irrigation. A Jackson-Pratt drain was placed, postoperative radiographs were taken (Fig. 6) and immobilization was performed with a long leg posterior splint for pain control. Postoperative broad-spectrum antibiotics were started until culture revealed MRSA, for which intravenous clindamycin was given for 7 days, and was shifted to oral for 3 more weeks. Postoperatively, the patient had good pain control without any palsy. Histopathology of the fibula revealed chronic active osteomyelitis.

On follow-up and completion of antibiotics, the patient had no complaints of pain, was ambulatory with normal gait, and had full range of motion. Wound was well coapted, without bleeding nor discharge. Repeat radiographs showed no evidence of osteomyelitis in the remaining fibula, and had three consecutive decreasing ESR values, and three consecutive normal CRP results. Follow-up radiographs were noted to be unremarkable.

Table 1 Fibular Osteomyelitis cases in literature

Authors	Year	Age, Sex/ Number of Cases	Duration of symptoms	Fibular involvement	Treatment	Follow-up	Outcome
Yin et al	2015	8 cases	-	-	Ilizarov bone transport	14-34 months	Healed
Varun et al	2015	8, F	6 months	Pandiaphyseal	Antibiotic therapy and En bloc resection	-	-
Ponio et al	2013	13 cases	-	-	Antibiotic and surgery	-	-
Huang et al	2013	11, M	1 week	Distal 3rd shaft	Debridement with antibiotic therapy	5 years	healed
Elö et al	1994	22 cases	-	-	Surgery	-	-
Ziani et al	1990	18 cases	-	-	Diaphyseal resection, sequestrectomy	-	-

F female; *M* male

DISCUSSION

The prevalence of osteomyelitis is 239 per 100,000 cases in developing countries, most commonly affecting the femur and tibia. In developed countries, the prevalence is 3 to 14 per 100,000 children [2]. Spread may either occur from direct inoculation from superficial soft tissue infection, or hematogenously from bacteremia [15]. It is more common to find acute hematogenous osteomyelitis in children than in adults [2,4] due to the highly vascular metaphysis, with tortuous vessels, where sluggish blood flow promotes extravascular bacterial seeding [15]. On the other hand, chronic osteomyelitis is due to late presentation with risk factors of poor hygiene, immunocompromise, as well as inadequate healthcare [5]. Other organisms usually seen in immunocompromised children include Mycobacterium tuberculosis, Bartonella henselae, and fungi, such as Histoplasma spp. and Cryptococcus spp. [4].

There are an increasing amount of cases of MRSA positive osteomyelitis, possibly accounting for more than one-third of Staphylococcal positive cultures [4, 15, 16]. In a study by Mantero et al, the prevalence of MRSA culture was at 48% [17]. In a study by Ratnayake et al [16], 38% of the 55 patients developed MRSA infection, while the methicillin-sensitive Staphylococcus aureus (MSSA) was at 47%. In a retrospective study by Ponio et al, there were noted to be 134 cases of chronic osteomyelitis within a 5 year study period in a local hospital in the Philippines. The most common organism isolated is still Staphylococccus aureus at 40%, with MRSA infection at 20%. In a study done in pediatric hospitals in the USA, the prevalence of MRSA infections from 2002 to 2007 increased from 0.3 to 1.4 per 1000 hospital admission, with the pattern of the rate of Staphylococccus aureus remaining constant [2]. This just goes to show that the trend of MRSA infections in osteomyelitis is changing.

In addition to chemotherapy, surgery for osteomyelitis is considered as a mainstay of treatment as antibiotics alone may not be able to penetrate the devitalized tissue [4,5]. Surgical management includes debridement until the observation of the paprika sign, as well as the eradication of dead space to be replaced with healthy, viable tissue. Sequestrectomy must be done adequately, including the removal of all infected bone and soft tissue [1]. Doing so not only reduces the bacterial load, but may also improve the penetration of antimicrobials to the affected areas [5]. Inadequate debridement may result to recurrence of the osteomyelitis years after the initial presentation [2]. Viable biologic samples may also be procured intraoperatively for proper culture guided treatment.

Pediatric fibular osteomyelitis is uncommon, with only several cases reported worldwide (Table 1). In a study by Ziani et al, there was a good outcome in patients who underwent diaphyseal resection of the fibula, despite the fact that there may be a consequential valgus deformity of the ankle and knee instability as a result of a superior advancement of the lateral malleolus and altered ankle kinematics [7, 14, 18]. Another surgical alternative for fibular osteomyelitis is through the use of Ilizarov bone transport, recommended for cases with distal fibular loss as there was no knee or ankle instability, seen in a study by Yin et al with all of the 5 patients who underwent the procedure showed good bony union and absence of recurrence of infection [14]. There is no issue with the simple resection of the fibular diaphysis in adults, as there is no growth concern, in addition to the fact that the fibula only provides a small percentage of weight bearing on the lower extremity. On contrary, if we perform this in children, the repercussions may be hostile for the growing child. It is not enough to simply resort to removing the affected proportion without taking into consideration the affectation of the growth plates, as well as the translational ramification.

In our case, we proceeded with an En bloc resection, due to the widely affected left fibula, wherein partial fibulectomy cannot be an option. A complete resection of an infected bone will create a significant unstable defect but may be employed in widely diseased bone [5]. Postoperatively, until1 year follow-up, the patient was able to function normally and at pre-symptom levels without any undue complications (Figs. 7, 8, and 9). No ankle instability or superior advancement of the lateral malleolus noted on 1 year follow-up (Fig. 10).

In the treatment of fibular osteomyelitis, a study by Elö et al. showed that surgical treatment as the first step in management leads to good outcome [8]. In the local study by Ponio et al, there was a high rate of clinical improvement at 98% with treatment involving antibiotic administration and surgery [2]. In the study of Sierink, the patient was subjected to operative debridement due to the unresponsiveness of the patient to antibiotic

treatment despite the absence of sequestrum formation on plain radiographs and computed tomography. Upon surgical exposure, the patient demonstrated a large sequestrum of the fibula that was assimilated by an involucrum. This finding strengthens the need to perform early surgical debridement in progressive disease [1].

CONCLUSION

In summary, this is a rare case of diffuse chronic osteomyelitis of the fibula in a 9 year-old male, presenting with pain and swelling, with difficulty in ambulation. The en bloc resection of the fibula, along with the treatment of antibiotics, resulted to a favorable outcome, with the patient having a return to normal function of the leg. The use of antibiotic therapy should not be delayed, prudently starting the patient on broad spectrum antibiotics even while awaiting cultures sent from the intraoperative specimen.

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