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Severe osteomyelitis caused by *Proteus mirabilis* and *Escherichia coli* in sickle cell disease: a case report

Osteomielite grave por *Proteus mirabilis* e *Escherichia coli* na doença falciforme: estudo de caso

Osteomielitis grave causada por *Proteus mirabilis* y *Escherichia coli* en la enfermedad de células falciformes: estudio de caso

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This is a case study about an infant, a year and six-months old, with sickle-cell disease, who stayed for a prolonged period of time in the hospital to treat osteomyelitis in many upper and lower limb bones at the General Hospital of the Federal University of the Triângulo Mineiro (UFTM), Uberaba, Brazil. The case was conducted through an analysis of medical records. The patient described undergoes follow-up treatments in the Pediatric Hematology Service of the Regional Blood Bank at Uberaba, the Hemominas Foundation, MG/Brazil. The findings of this study were consistent with previous results in the literature and show that early diagnoses and timely treatments collaborate to reduce the sequelae of Osteomyelitis.
Descriptors: Anemia, Sickle cell; Infant; Osteomyelitis.

Este é um estudo de caso que tem como objetivo descrever o caso de um lactente de um ano e seis meses portador de doença falciforme com internação prolongada para o tratamento de osteomielite em diversos ossos de membros superiores e inferiores no Hospital de Clínicas da Universidade Federal do Triângulo Mineiro (UFTM). O caso foi relatado baseado na análise retrógrada em prontuário médico. O paciente descrito realiza acompanhamento no Serviço de Hematologia Pediátrica do Hemocentro Regional de Uberaba – Fundação Hemominas. Os achados são compatíveis com os encontrados em literatura e evidenciam que o diagnóstico precoce e a terapêutica oportuna colaboram para redução nas sequelas da osteomielite.
Descritores: Anemia falciforme; Lactente; Osteomielite.

Este es un estudio de caso que tiene como objetivo describir el caso de un lactante de un año y seis meses portador de la enfermedad de células falciformes con internación prolongada para el tratamiento de osteomielitis en diversos huesos de los miembros superiores e inferiores en el Hospital de Clínicas de la Universidad Federal do Triângulo Mineiro (UFTM), Uberaba, MG, Brasil. El caso fue relatado a partir del análisis retrógrado a partir de la historia clínica. El paciente descripto realiza acompañamiento en el Servicio de Hematología Pediátrica del Hemocentro Regional de Uberaba – Fundación Hemominas. Los hallazgos son compatibles con los encontrados en literatura y evidencian que el diagnóstico precoz y la terapia oportuna colaboran para la reducción de las secuelas de la osteomielitis.
Descriptores: Anemia de células falciformes; Lactante; Osteomielitis.

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INTRODUCTION

Sickle-cell disease (SCD) is the most frequent haemoglobinopathy in Brazil, and is considered a public health problem. It is one of the most common hereditary diseases in the world, originated from a mutation in Africa, and more common among afro-descendents in the country. It can be early diagnosed through the neonatal triage/heel prick[1]. From the diagnosis on, the patient with the disease is directed to a specialized service, to follow up treatment.

In sickle-cell disease, the glutamic acid is replaced by valine in the HBB chain, generating a structural hemoglobin deformity associated with the presence of hemoglobin S. It can present itself in the following forms: homozygous (SS) and heterozygous (genotypes AS, SC and S-thalassemia)[2].

The red blood cells, which mostly contain hemoglobin S, assume, in hypoxia, a form similar to that of a sickle, that results from the polymerization of hemoglobin S. Sickle-shaped red blood cells do not flow properly during microcirculation, blocking the capillary blood flow and eventually being destroyed[2].

Therefore, many complications may arise, such as: algic crises, acute chest syndrome, fever caused by infection, functional aplasia, aplastic crisis, splenic sequestration crisis, biliary lithiasis and strokes.

Infections are frequent complications of sickle-cell disease. One of them is osteomyelitis. It is a bone inflammation, usually caused by a bacterial or fungal infection, which can remain localized or spread, compromising the medulla, the parte cortical, the parte esponjosa and the periosteum. Depending on how advanced the disease is, it can affect several bones, leading to a long treatment, usually at a hospital and possibly leading to long-term bone sequelae, possibly prejudicing bone growth and the stature of affected patients.

This study aims to describe the case of a one year and six month old infant afflicted by sickle-cell disease, hospitalized for a long period for the treatment of osteomyelitis in various upper and lower limb bones in the General Hospital of the Federal University of the Triângulo Mineiro (UFTM).

METHOD

This case study analyzed patient records through the clinical reports registered at the patient's medical chart and the regional hemotherapy service.

The General Hospital of the Federal University of the Triângulo Mineiro (GH/UFTM) stores all medical records of registered patients who have already received care in there.

The research also used the records from the Regional Blood Bank at Uberaba, where the patient performs regular follow-up tests due to the sickle-cell disease. The case being discussed is from 2016.

Clinical data will be presented as well as analyses of the evolution of osteomyelitis, through exams stored in softwares in the hospital and made available by the radiology team.

For the purposes of patient and family protection, no names were mentioned and the family authorized publication, as did the hospital administration regarding the description and publication of results.

RESULTS

The patient was an infant, one year and six months old, formerly afflicted by sickle-cell disease, with prior admissions due to algic crises. Admitted to the ER of the GH-UFTM in February, 2016, due to splenic sequestration crisis, the patient had his condition properly taken care of. However, febrile syndrome arose, and antibiotics were necessary.

In the same hospitalization, the patient presented painful edemas in the hands and feet, accompanied by hyperthermia and possible dactylitis. There was no improvement, and the patient remained feverish, with edemas on the feet.

An ultrasound of inferior limbs revealed abscesses in the feet. The osteomyelitis was diagnosed. It was affecting the humerus, the tibia, as well as bones in the feet and hands, in both sides. Continuous

drainage of purulent secretion in the region of the tibias was conducted.

The diagnosis of osteomyelitis was confirmed through radiographies (Figures 1 to 5). In cultures of tibial secretion, *Escherichia coli ESBL* and *Proteus mirabilis* were isolated, both susceptible to imipenem and amikacin. Thus, a treatment with parenteral antibiotics was chosen, to be conducted for an extended period of hospitalization.

A complementary hyperbaric oxygen treatment was suggested, thirty sessions of which were performed. The treatment with antibiotics continued for eight weeks. After hospital discharge, amoxicillin and clavulanic acid were prescribed for four months, and the patient wore a plastered cast for 60-90 days, according to an orthopedic evaluation. The treatment continued for five months, in the outpatient clinic.

Image 1. Lower limbs affected by osteomyelitis. Tibial region on both sides with lytic lesions and bone density reduction. Uberaba, MG, Brazil, 2016.

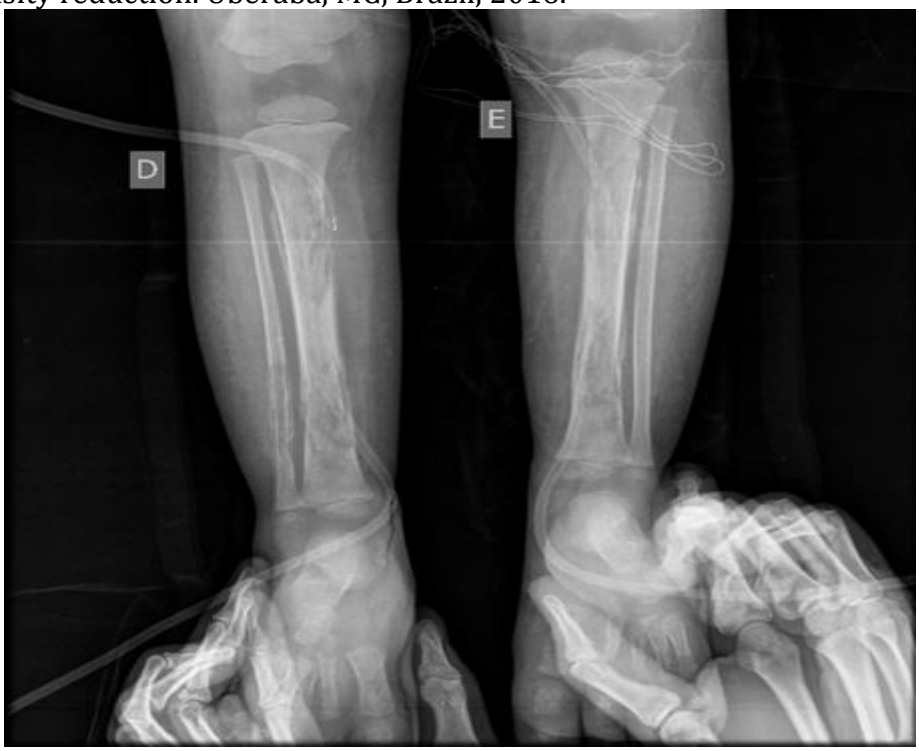


Image 2. Dactyl bones affected by osteomyelitis: lytic lesions in the phalanges with decreased bone density. Uberaba, MG, Brazil, 2016.

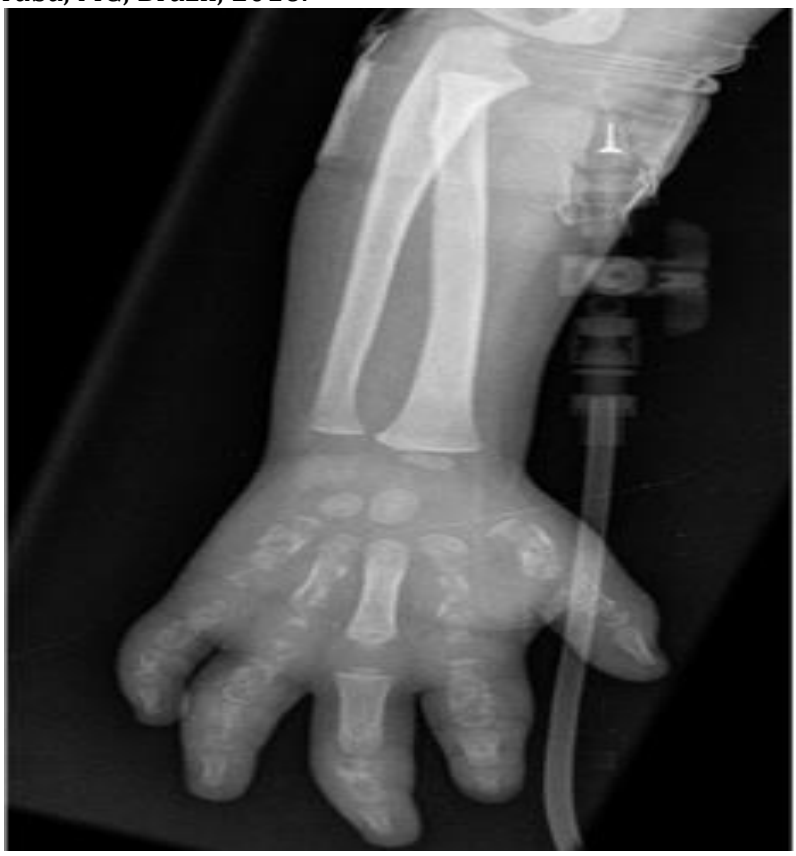


Image 3. Feet phalanges of both sides affected by osteomyelitis: lytic lesions with decreased bone density. Uberaba, MG, Brazil, 2016.

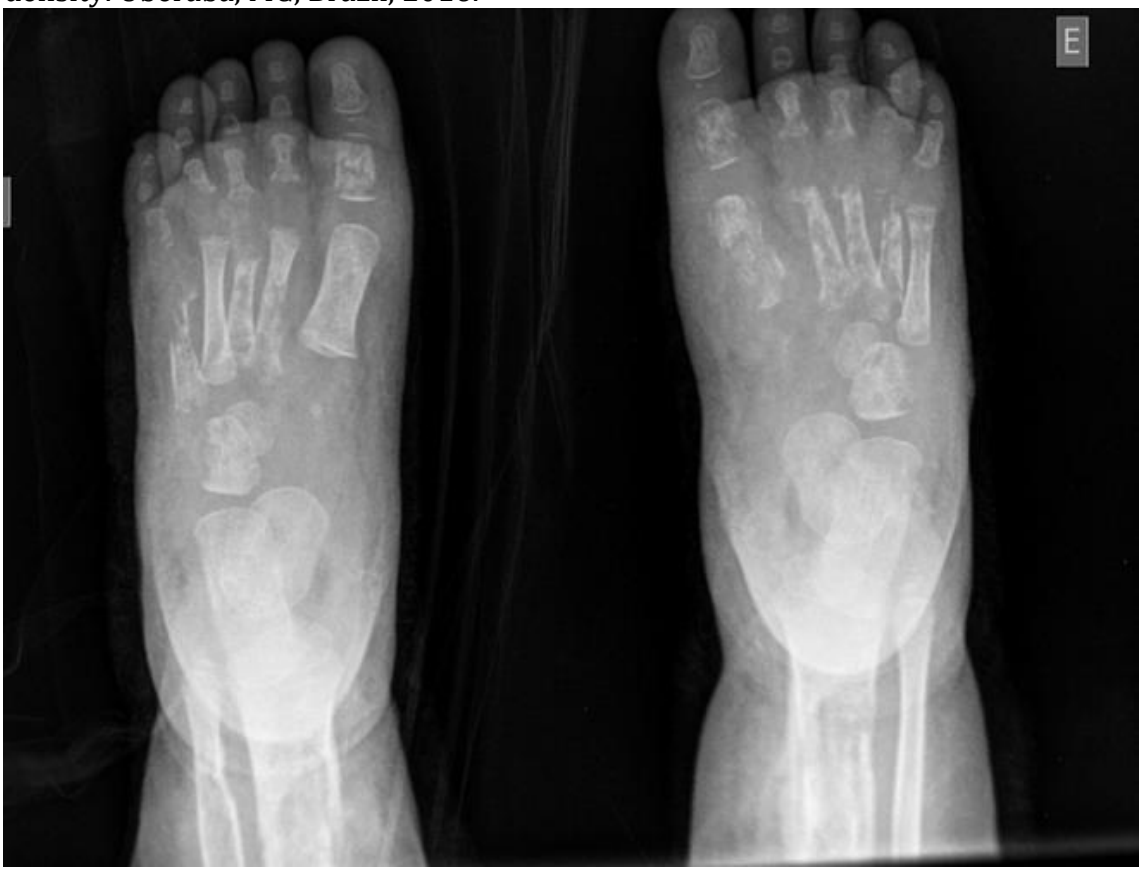


Image 4. At the end of the treatment that took place in hospitalization, the tibial pattern improved in both sides, with apparent increase in bone density. Uberaba, MG, Brazil, 2016.

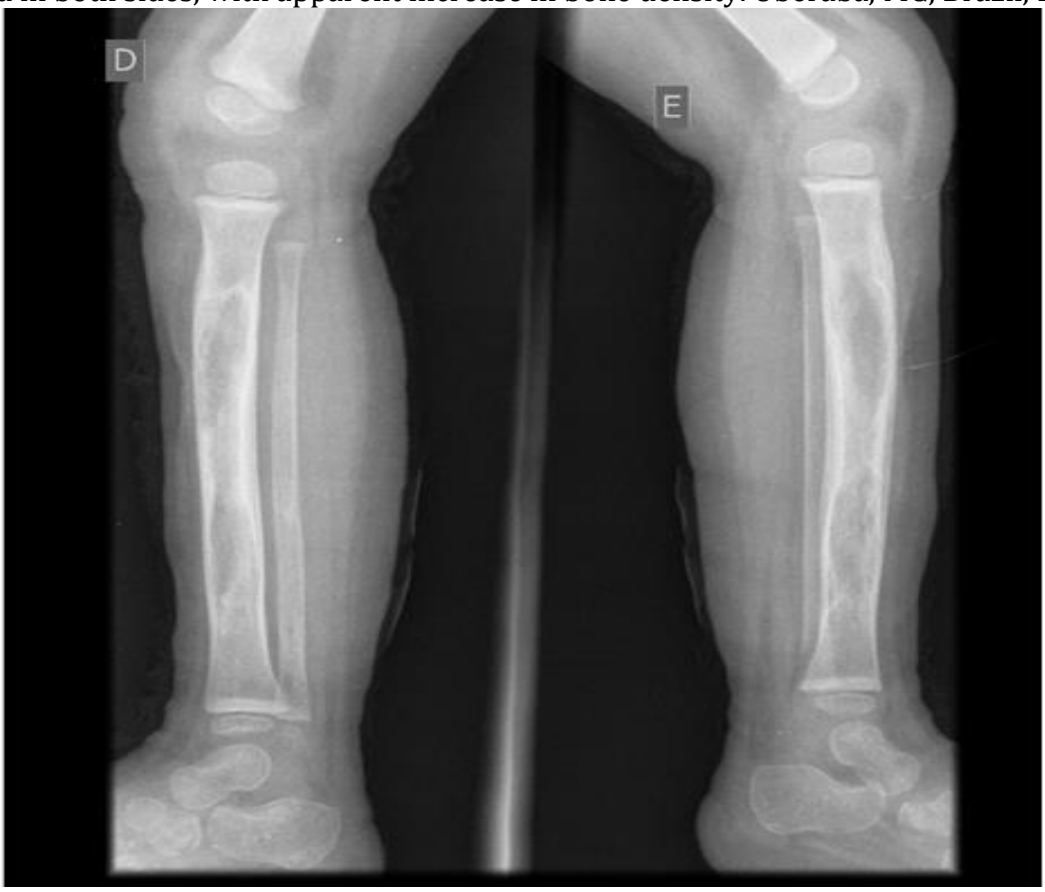
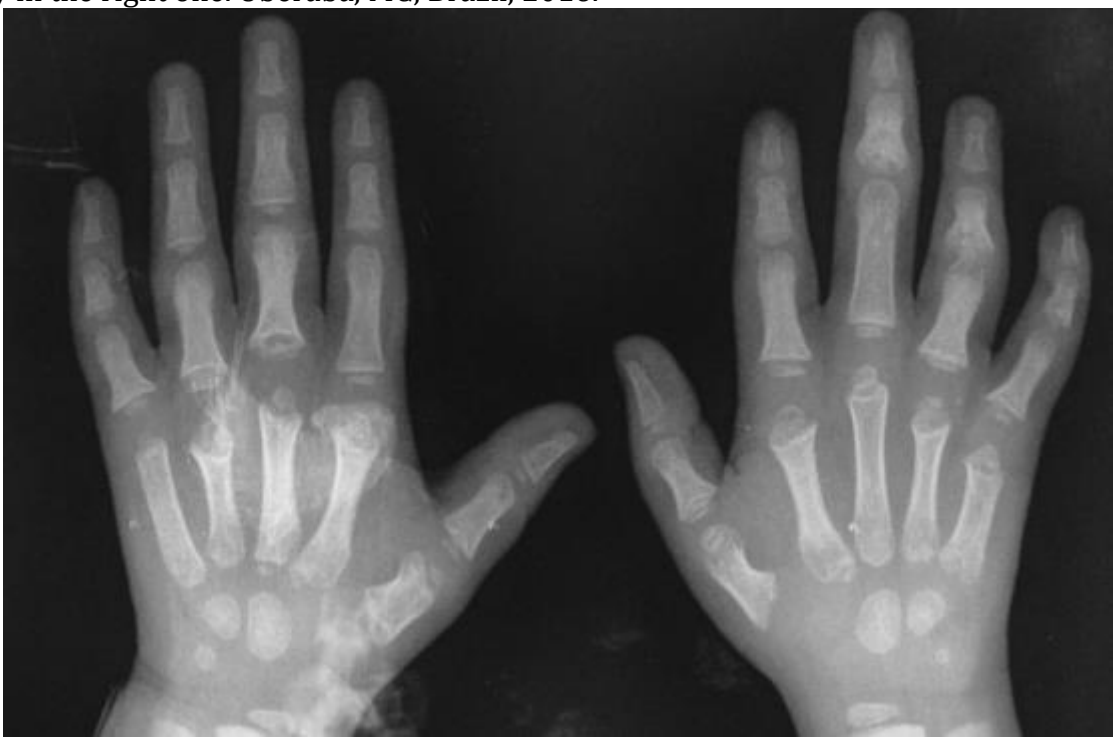


Image 5. Radiography in October 2016, showing osteomyelitis sequelae in distal phalanges, mainly in the right one. Uberaba, MG, Brazil, 2016.



DISCUSSION

In up to 12% of cases sickle-cell anemia cases, osteomyelitis can happen. The most affected bones are femur, tibia, and humerus. The main etiological agents are *Salmonella, Staphylococcus* and enteric gram negative bacilli. In the case being analyzed, the *Proteus mirabilis* and the *Escherichia coli* were the isolated agents[2].

Bone marrow necrosis happens as a consequence of the bone infarction, leading to a predisposition for osteomyelitis and septic arthritis. Complications are more common among males (2:1), and it rarely takes place in children below one year of age; that means that, although the patient was very young, he fits within the most likely statistics[3].

The clinical framework displayed by the patient was indicative of osteomyelitis, including pain, edema, heat, flushing, increased local sensitivity, and fever. After the spleen, the bones are the second organ most affected by sickle-cell disease[4].

A series of radiographies were carried out, followed by a surgical abcess drainage surgical procedure, in which drains were placed. Cintilografias and MRIs were not conducted, since the diagnosis was late, taking place only after the disease was in a mature state. These last exams would contribute to a more precocious diagnosis. There was no bone biopsy of the involved areas, an exam that would be eventually needed[1]. In some cases, radiographies may be normal [4], but in others, as in the case described, they present abnormalities, as seen in the images.

Prolonged antibiotic therapy was required. The patient was put on imipenem, vancomycin, amikacin and clindamycin, due to the sensitivities revealed in an antibiotic sensitivity test, and to the fact that the patient had been previously hospitalized with osteomyelitis. The choice of antibiotics, usually, is done according to the microorganism detected; if there is evidence of fluid buildup in the sites affected by infection, drainage is recommended[5].

Hyperbaric oxygen treatment sessions were used to improve the healing of the drainage and of the lesions caused by the osteomyelitis with good results.

In follow-up at outpatient clinics, the patient presented better radiological patterns, as evidenced in recent radiographies, with no delay in growth, since plates epiphyseal were not affected. He remained in an extended antibiotic therapy for another five months, in addition to the period schedule at hospital discharge. There were no more complications causd by osteomyelitis. Admissions due to algic crises or other types of infection have already been recorded.

A multidisciplinary team with physiotherapists, occupational therapists and psychologists, in addition to physicians and nurses, was asked to address the case in order to improve the speed of the patient's recovery.

During hospitalization, the prognosis of the patient and the possible sequelae were discussed, and even the possibility of death; but after months of treatment, it sequelae were found to be minimal, and the situation of the patient has been developing greatly.

CONCLUSION

Early diagnosis of osteomyelitis allied with brief treatments with antibiotics prevents complications to the patient. However, a clinical treatment alone is not sufficient, and sometimes, irrigating and surgically debriding the affected region may be necessary. In this case, hyperbaric oxygen treatment sessions were required, as one more option to deal with the patients framework.

Prolonged hospital treatments and long-lasting follow-up outside the hospital are essential, since the growth and the stature development of the patient can be compromised.

Antibiotic therapy outside the hospital can be prescribed for long periods of time, depending on the severity and evolution of each patient.

In patients with comorbidities, such as sickle-cell disease, other complications may arise during the treatment, making it longer,

but there is a possibility of satisfactory evolution.

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CONTRIBUTIONS

Nathália Borges de Melo was responsible for reviewing the patient documental, and for the organization of the events of the case. **Valeria Cardoso Alves Cunali** performed a critical revision of the final text. **Jussara Silva Lima** made the survey of similar cases in Brazilian references. **Kellen Cristina Kamimura Barbosa Silva**, **Valquíria Cardoso Alves** and **Vanessa de Paula Tiago** participated in the clinical follow up, and in the critical review of the final essay.

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