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Congenital bilateral periostitis: Differential diagnosis and a case report

Anna Elia, Egor Kostin, Elena Charalambous, Ioannis Orfanos

ABSTRACT

Introduction: Periostitis, characterized by radiological findings of periosteal new bone formation, can result from various factors including trauma, drugs, infection, and tumors. Among infants up to six months of age, common causes of periostitis include physiologic periostitis of the newborn, Caffey disease, periostitis related to prostaglandin use, and congenital syphilis. The differential diagnosis of bilateral symmetrical periostitis in newborns and infants within this age range is important as it contributes to the possibility for early treatment.

Case Report: A 3-month-old girl, born at 38 weeks by natural birth and with unremarkable prenatal history, presented with multiple episodes of vomiting, loss of appetite, fever, saddle nose, prominent forehead, maculopapular rash with vesicles in both palms, and a palpable liver. Her weight was below the 3rd percentile on growth charts. The infant's syphilis serum screening test was reactive, leading to further testing. Treatment with crystalline penicillin G was administered, and diagnostic investigations revealed symmetrical periostitis affecting the diaphysis of bilateral femorae and tibiae.

Conclusion: This case report highlights the importance of considering congenital syphilis in the differential diagnosis of bilateral symmetrical periostitis in newborns and infants up to six months of age. Early diagnosis and appropriate treatment are crucial. This case report adds to the orthopedic literature by presenting a unique case of congenital syphilis-associated periostitis, emphasizing the significance of considering this etiology in similar clinical scenarios. Increasing awareness of such cases can lead to prompt diagnosis and intervention, ultimately contributing to better clinical outcomes in affected infants.

Keywords: Bilateral periostitis, Caffey disease, Congenital syphilis, Differential diagnosis

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Anna Elia[1], MD, Egor Kostin[2], Elena Charalambous[1], MD, MSc, Ioannis Orfanos[3], MD, MSc, PhD

Affiliations: [1]Pediatrician, Department of Pediatrics, Limassol General Hospital, Cyprus; [2]Medical Student, University of Cyprus, Cyprus; [3]Paediatric Orthopaedic Surgeon, Department of Orthopaedic Surgery, Nicosia General Hospital, Cyprus.

Corresponding Author: Egor Kostin, Nicosia General Hospital, 215, Paleos Dromos Lefkosia-Lemessos str., 2029 Strovolos, Nicosia, Cyprus; Email: egor03kostin@gmail.com

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INTRODUCTION

Periostitis (periosteal new bone formation) is a radiological finding that results when the inner layer of the periosteum reacts to a variety of possible factors. Trauma, drugs, infection, and tumors can cause the elevation of the periosteum from the cortex [1, 2]. Two diseases that are associated with periostitis are physiological periostitis of the newborn and Caffey disease, also known as Caffey–Silverman disease, Roske–Caffey disease, deToni–Caffey disease, or infantile cortical hyperostosis. In early infancy (up to six months of age) [2], these two diseases, along with prostaglandin use and congenital syphilis [3], are the more common causes of periostitis. Notably, there are nine published case reports of congenital syphilis with periostitis (bone involvement). In this case report, we

present a 3-month-old girl who was born with symmetrical periosteal elevation involving bilateral femorae and tibiae due to congenital syphilis, and we discuss the differential diagnosis of bilateral symmetrical periostitis in newborns and infants up to the age of six months.

CASE REPORT

The authors have obtained the patient's guardian informed consent for print and electronic publication of the case report. A 3-month-old girl born by natural birth on 38 weeks, appropriate for gestational age and with unremarkable prenatal history, was admitted to Pediatric Clinic of Limassol General Hospital, Cyprus, because of multiple episodes of vomiting and loss of appetite. At the time of admission, she was febrile. During the clinical examination we noticed some special face characteristics like saddle nose, prominent forehead, and maculopapular rash with vesicles in both palms and palpable liver. Her weight was below the 3rd percentile on growth charts. Except from blood analysis and routine cultures for sepsis, we set up an investigation of congenital infections (TORCH), as well as human immunodeficiency virus (HIV) and hepatitis A, B, and C. The mother had an inadequate gynecological observation during the first two trimesters of pregnancy. The screening on the 3rd trimester was negative for infectious diseases like syphilis, but she admitted that she and the father of the baby were drug users. Syphilis serum screening test of the infant was reactive, so we sent treponemal serum antibodies to the mother and child and we have performed a lumbar puncture of the baby to check the cerebrospinal fluid (CSF) for the possibility of neurosyphilis. The titer of rapid plasma reagin (RPR) was 1:32 and Treponema pallidum hemagglutination assay > 1:2560. The mother's titer of RPR was 1:16. Cerebrospinal fluid culture was negative and Western Blot on CSF for Treponema pallidum was positive. As soon as we had the suspicion of congenital syphilis, according to the distinct facial characteristics and palpable liver we have started a 10-day intravenous treatment with crystalline penicillin G and before discharge we gave a single dose of intramuscular penicillin G benzathine for central nervous system (CNS) involvement. An extended investigation of the systems affected in the case of congenital syphilis was performed. X-ray of the long bones showed symmetrical periostitis affecting the diaphysis of bilateral femorae and tibiae (Figure 1). No pathological fractures were found. Auditory and ophthalmological examination was normal. A follow-up was set up for three months after treatment to check the titer of the antibodies both in serum and CSF.

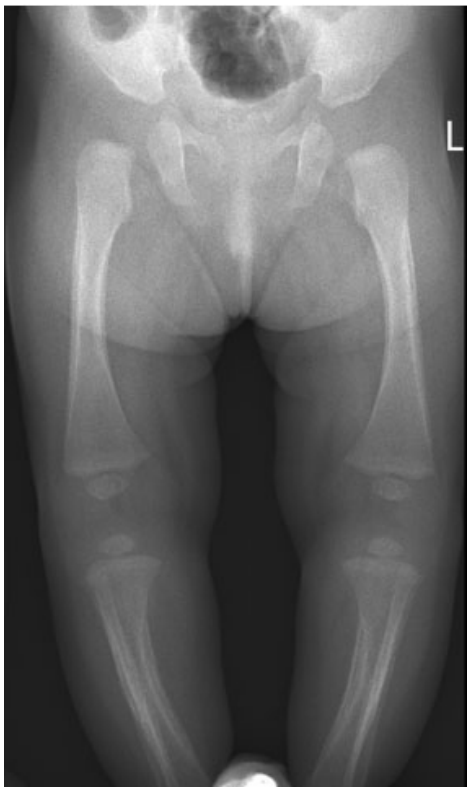

Figure 1: The anteroposterior (AP) X-ray was performed with the infant lying supine on the radiographic table, ensuring that both legs were fully extended and positioned parallel to each other for symmetrical imaging. A low-dose pediatric technique was utilized to minimize radiation exposure, 60 kVp (kilovolt peak) and 2 mAs (milliampere-seconds). The X-ray beam was directed perpendicular to the film, centered at the midpoint of the lower limbs, ensuring even distribution across both femurs and tibias. A high-resolution digital detector was used to capture the image, which clearly demonstrates the characteristic bilateral periostitis. The periosteal reaction is visible along the diaphysis of both femurs and tibias, consistent with findings in congenital syphilis.

DISCUSSION

In the differential diagnosis of bilateral symmetrical periostitis in newborns and infants up to six months of age, one should consider congenital syphilis, physiologic periostitis of the newborn, Caffey disease, and prostaglandin administration. Physiological periostitis in the newborn is found in both preterm and term infants aged from one to six months (commonly one to four months) [2, 4, 5] and the exact etiology is unknown. It is commonly seen in long bones [2] and is invariably symmetrical in distribution, although occasionally more prominent on one side than the other. The femorae (thigh bones), humeri, and tibiae (shin bones) are almost equally affected, but may initially be limited to one pair of bones. It is always on the diaphysis [6] and usually asymptomatic and in the differential diagnosis one should consider child abuse, syphilis, osteomyelitis, trauma, bone tumor, and tumor-like lesions [7, 8]. Caffey disease also appears at a very young age. The classic form appears at the age of two months and regresses spontaneously at the age of two years. The most valuable diagnostic test is radiography. One can observe cortical

hyperostosis, usually involving more than one bone, most commonly the mandible, clavicles, scapulae, ribs, and long bones. The diaphyses are affected with sparing of the metaphyses and epiphyses. Laboratory findings are non-specific [9]. Prostaglandins should also be considered during differential diagnosis of bilateral symmetrical periostitis as they have become essential agents for infants with ductal-dependent congenital heart disease. Their ability to maintain patency of the ductus arteriosus is critical in these lesions. Although infusion of prostaglandins is usually limited to a short period of time required to stabilize a neonate before surgical palliation or correction, sometimes long-term infusion is needed when surgical intervention is not immediately feasible or is unsuccessful. In such cases, an unusual side effect of the treatment is the development of symmetrical periostitis of the long bones. Both clinical and radiological signs are improved after cessation of the drug [10]. Finally, congenital syphilis is an intra-uterine infection which usually manifests shortly after birth. Spirochaetes cross the placental barrier after the fourth month of pregnancy, although clinical symptoms may not appear for several weeks after birth. Prematurity, low birth weight, anemia, hepatosplenomegaly, skin lesions, and rhinitis are the predominant clinical features. Less common symptoms are jaundice, lymphadenopathy, cutaneous lesions, and pseudoparalysis. The most common radiographic finding is bilateral, symmetrical periostitis concerning the long bones (usually tibiae and femorae) [4, 5, 11]. Osteitis and metaphysitis can also be found [12]. Skeletal manifestations of congenital syphilis occur in 60–80% of infants with clinical symptoms, but also in 20% of asymptomatic cases. Diagnosis may be difficult when the clinical presentation of bilaterally symmetrical periostitis is absent [12].

CONCLUSION

This case report highlights the significance of considering congenital syphilis as well as other diseases in the differential diagnosis of bilateral symmetrical periostitis in newborns and infants up to six months of age. This unique case adds valuable knowledge to the medical literature, emphasizing the importance of recognizing atypical presentations to facilitate more timely and accurate diagnoses, as an early diagnosis and appropriate treatment are crucial for the improvement of clinical outcomes in affected infants.

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Author Contributions

Anna Elia – Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Egor Kostin – Conception of the work, Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Elena Charalambous – Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related

to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ioannis Orfanos – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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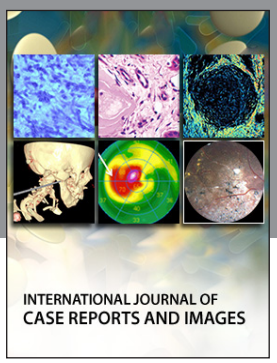
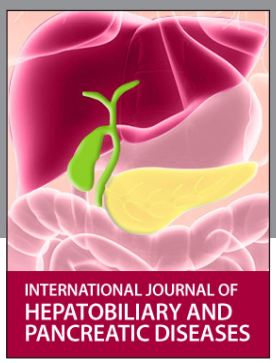
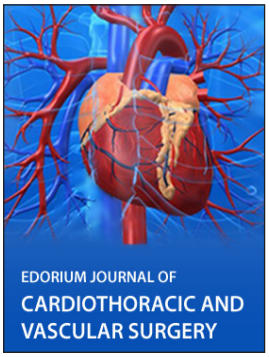
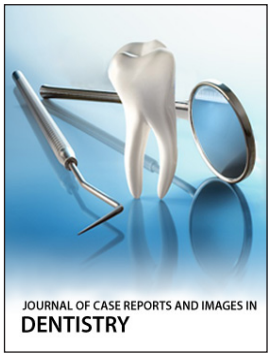
Authors declare no conflict of interest.

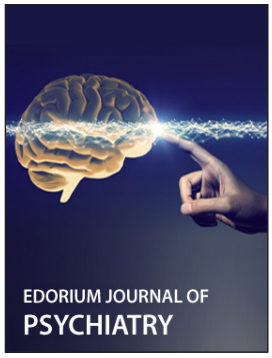
Data Availability
All relevant data are within the paper and its Supporting Information files.

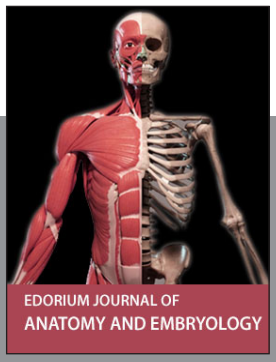


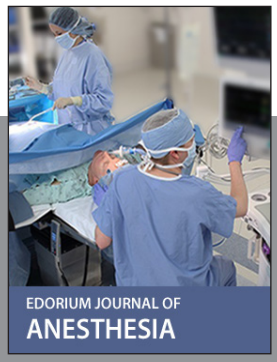












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