

A rare case of maxillary sinus osteomyelitis with intraorbital, extraconal abscess in a term low birth weight twin

Janampally Ravikiran, Swathi Chacham, Uppin Narayan Reddy, Jillalla Narsing Rao, Ekta Aggarwal, Anumula Soumya, Nori Madhavi

ABSTRACT

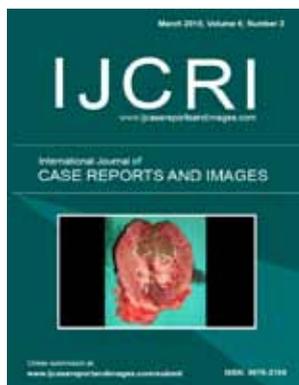
Introduction: Acute osteomyelitis in neonates is a rare infectious disorder of bone leading to diagnostic and therapeutic challenge, more so in neonates. While the long bones are frequently affected sites, maxilla is rarely involved. Physiological immaturity of the immune system coupled with invasive diagnostic and therapeutic procedures render them susceptible for this condition, more so in premature, low birth weight infants. The most common organism causing osteomyelitis in neonates is *Staphylococcus aureus*. Lesion of the orbit as well as contiguous faciomaxillary and sinonasal inflammatory pathology often lead to proptosis in neonates.

Case Report: We report a 15-day-old term female neonate (twin 1) presenting with non-traumatic periorbital swelling and purulent nasal discharge. The neonate also had failure to thrive. On clinical examination, the neonate was sick with sclerema and shock. There was right eye proptosis with edema and erythema. Initial diagnosis of periorbital abscess or orbital tumor was considered and the infant was investigated. Sepsis screen was positive with marked polymorphic leukocytosis. Also, the blood culture and culture from sinus scraping grew *Staphylococcus aureus*. Magnetic resonance imaging scan of the brain including orbit revealed osteomyelitis of maxilla and intraorbital extraconal mass with restricted diffusion, suggesting intraorbital abscess. The neonate responded to intravenous antimicrobials and decompression of the abscess. Although, the source of this major infection is unidentified, low birth weight, failure to thrive and poor socioeconomic status might have contributed.

Conclusion: We report a term, low birth weight, female neonate with culture positive sepsis, maxillary sinus osteomyelitis and orbital abscess. Osteomyelitis of the maxilla is a rare entity and an important etiological factor for orbital abscess.



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INTRODUCTION

Acute osteomyelitis (OM), a rare infectious disorder of bone infrequently manifests in neonates. The reported incidence varies from 5–12 per 100,000 infants [1], with a very high mortality rate (7.3%) [2]. The most frequently affected joints are hip, knee and shoulder, while maxillary sinus is rarely involved [3, 4]. Physiological immaturity of the immune system in neonates renders them susceptible for this condition. Premature and low birth weight (LBW) neonates are more immunocompromised when compared to term and appropriate for gestational age (AGA) infants. Invasive diagnostic and therapeutic procedures add to the risk of OM in premature and LBW neonates [5]. Osteomyelitis of maxillary bone and sinus is a rare entity in pre-antibiotic era, more so in the current era of broad spectrum antibiotics [3, 4]. This resulted from extension of infection from neighboring paranasal sinuses and rarely, it had odontogenic origin [6]. In pre-antibiotic era, it was described as a syndrome of acute OM of superior maxilla in early infancy, manifesting with ophthalmological or sinonasal symptoms, by Lieberman and Brem [6]. While the most common organism causing OM is *Staphylococcus aureus*, *Klebsiella* and other gram negative organisms are rarely reported [7, 8]. Maxillary sinus OM can lead to ocular complications, upper airway obstruction and even death [9]. Ophthalmological complications of sinusitis were reported by Hubert and Chandler in pre-antibiotic era [10, 11]. These include orbital cellulitis, orbital abscess and cavernous sinus thrombosis.

CASE REPORT

We report a 15-day-old term, 1550 grams, female neonate (twin 1) presenting with right periorbital swelling of one week duration. There was no history of injury, administration of injections or application of topical agents to the eye. Also, there was no history of pustular lesions over the eye prior to the onset of periorbital swelling. The infant had fever, purulent nasal, conjunctival discharge, poor feeding, lethargy and poor weight gain. The neonate was fed with dilute formula as the breast milk was insufficient (due to twin gestation). This was born to a second-gravid with twin gestation by spontaneous vaginal delivery and had normal extra-uterine transition. There was no history of maternal pyrexia and rash during the first trimester. Also there was no pregnancy induced hypertension, gestational diabetes and premature rupture of membranes. It was a spontaneous conception. No history of maternal blood transfusion (to suspect retroviral pathology) and maternal hepatitis B surface antigen

(HBsAg), venereal disease research laboratory (VDRL) and human immunodeficiency virus (HIV) serology were negative. Family belonged to poor socio-economic class. However, the other twin was apparently normal.

On clinical examination, the neonate was sick with sclerema and mottling. The patient had septic shock manifesting with tachycardia (heart rate 180/min), hypotension (mean arterial pressure 36 mmHg) and poor tissue perfusion (capillary filling time >3 seconds). The neonate required dobutamine infusion and shock dose hydrocortisone. Local examination revealed right eye proptosis with inflammatory edema and bulbar conjunctival erythema along with purulent conjunctival and nasal discharge (Figure 1A–B). Initial differential diagnosis of periorbital abscess, orbital tumor and cavernous sinus thrombosis was considered and the child was investigated. Sepsis screen was positive (C-reactive protein 96 mg/L) with marked polymorphic leukocytosis (white blood cell count 2.9×10^4 cells/mm³, neutrophils 80%) and the blood culture has grown *Staphylococcus aureus*, sensitive to augmentin and ciprofloxacin, confirming the infectious nature of the lesion.

The neonate was further evaluated to detect the nature of orbital swelling. Magnetic resonance imaging (MRI) scan of the orbit and brain revealed thick walled fluid collection with rim enhancement (Figure 2A–B). This was present over the right maxilla extending along the nasal, ethmoid sinuses to the medial wall of orbit, suggesting intraorbital extraconal mass. Further, imaging with diffusion weighted MRI scan depicted restricted diffusion (Figure 3A–B) suggesting orbital abscess. However, the brain parenchyma and other structures were normal. The neonate responded to intravenous antimicrobials as per the sensitivity pattern. Decompression of the abscess revealed, copious amounts of purulent material, which grew *Staphylococcus aureus* with similar sensitivity pattern as the blood culture. The antimicrobials were continued for a period of six weeks as recommended. Maxillary sinus OM has been implicated for the orbital abscess. Magnetic resonances imaging scan revealed OM of maxillary sinus and orbital abscess. Also, the pus from



Figure 1: (A, B) Clinical photographs showing right eye proptosis with conjunctival edema (big arrow) and purulent nasal discharge (small arrow).

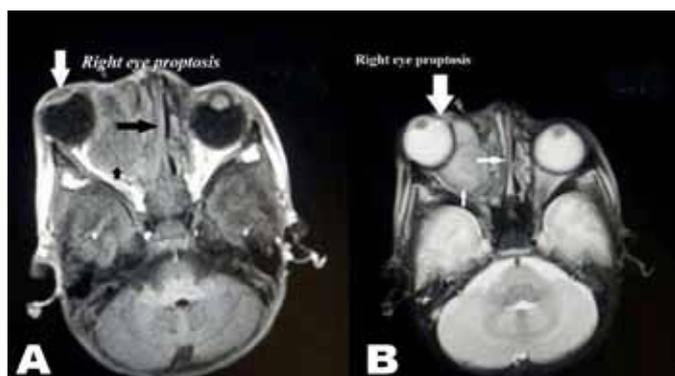


Figure 2: (A) Magnetic resonance imaging T1-weighted (axial image) scan of orbit and brain showing right eye proptosis with opacification of the right maxillary sinus (big black arrow) and a thick walled collection with rim enhancement extending from the right maxilla along the nasal, ethmoid sinuses to the medial wall of orbit, intraorbital extraconal mass (small arrow). Brain parenchyma and other structures is normal, (B) Magnetic resonance imaging T2-weighted (axial image) of orbit including brain showing similar findings (arrows).

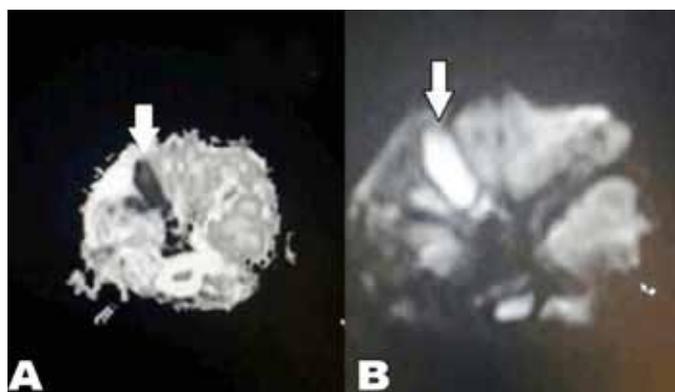


Figure 3: (A, B) Diffusion weighted magnetic resonance imaging orbit and brain revealing restricted diffusion in the right intraorbital extraconal mass suggesting orbital abscess (arrows).

orbital abscess grew *Staphylococcus aureus*. Hence, the coexistence of maxillary sinus OM and orbital abscess in a given patient can implicate the maxillary sinus OM to be the etiological factors for orbital abscess. Although, the exact cause of this major infection was unidentified, low birth weight, growth failure and poor socioeconomic status might have contributed.

DISCUSSION

In neonates, acute osteomyelitis (OM) is not only an infrequent complication, but also a diagnostic dilemma. The principal risk factors for OM in preterm and LBW infants include iatrogenic procedures like central veins lines, umbilical and intraarterial catheters. Similarly, the treatment modalities like total parental nutrition, artificial ventilation and their consequences like hospital acquired infection also predispose to OM in this high

risk group [12, 13]. Being a LBW neonate, the index case was at risk for OM. However, there was no invasive monitoring and no history of umbilical catheterization in this case. The prime causes for periorbital swelling in infants and children include adjoining sinonasal and faciomaxillary infective lesions and their complications such as cavernous sinus thrombosis. These often manifest with orbital congestion and proptosis [6, 14]. The index neonate presented with fever, nasal discharge followed by periorbital swelling. Hence, the upper respiratory tract infection complicated by maxillary sinus OM might have contributed to the orbital abscess in this case. The anatomy and embryological development of the maxilla per se, predisposes it to infection from adjacent structures.

Maxillary antrum is present at birth with greater volume and high vascularity. Also, the lower border of the orbit forms the upper boundary of the maxilla, which extends down up till dental ridge with primordial follicles and embryonic teeth. Hence, this superior maxilla can acquire infection from maxillary antrum (an area highly prone for air borne infections in infants), embryonic teeth follicle and lachrymal duct or sac infection [14]. In the pre-antibiotic era, OM of maxilla as been reported in the infantile age group which had hematogenous origin.

Even in the current era, maxillary sinus OM is more prevalent in the infantile age group when compared to other age groups [14, 15]. The predominant route of neonatal OM is hematogenous. However, direct transmission of bacteria due to breach in the protecting skin and mucosae coupled with invasive procedures can occur in preterm and LBW infants [16]. But these were absent in this neonate. The commonly reported organisms for sinusitis complicated by infantile orbital cellulitis are *Staphylococcus aureus* and *Streptococcus anginosus* [17] which were present in the index neonate case. The C-reactive protein (CRP), a rapid indicator of systemic inflammation and tissue damage, is useful as an acute phase reactant, but not specific for skeletal infection. Elevated values of CRP and erythrocyte sedimentation rates could be used to monitor response to therapy or identify complications. Likewise, the index infant also had elevated CRP along with significant polymorphic leukocytosis. Radiological investigations confirm the suspicion of neonatal OM, define the infection site, differentiate between unifocal and multifocal disease patterns and identify secondary complications. Radiographs should be the first diagnostic assessment to be performed in patients with suspected OM. The first line of investigation in neonates with suspected OM is plain radiograph. This reveals involvement of the soft tissue with obliteration of fascial plains within a week of inception of OM [18]. However, deep seated OM as in the index case, might be missed in plain radiography, requiring other superior imaging modalities. Correspondingly, ultrasonography can detect features of acute OM several days earlier, than radiographs in children and infants with good tissue penetration [18]. Studies have reported

that, MRI scan is associated with good sensitivity (97%) and specificity (94%) in detecting OM, when compared to other diagnostic modalities. Also, it can demonstrate changes within 3–5 days after the commencement of infection. Likewise, it is the diagnostic modality of choice for OM in deep seated areas and rare sites like maxillary sinus [19]. In the index infant, MRI scan of orbit and brain was done to delineate the exact pathophysiology of the orbital mass and to note the route and the extension of the lesion. MRI scan showed thick walled fluid collection with rim enhancement. This was present over the right maxilla extending along the nasal, ethmoid sinuses to the medial wall of orbit, suggesting intraorbital extraconal mass. An abscess can be differentiated from necrotic neoplasm by its restricted diffusion in diffusion weighted MRI, which was noted in this case. As the brain parenchyma and other structures were normal in this case, the orbital abscess is likely to result from the adjoining maxillary sinus OM. Usually, the response to the antimicrobials is satisfactory in the pediatric age group [14]. Likewise, the orbital swelling resolved and the general condition improved with appropriate anti-microbial agents in the present case. Hence, the coexistence of maxillary sinus OM and orbital abscess in this infant implicate the maxillary sinus OM to be the etiological factors for orbital abscess.

CONCLUSION

We report a term low birth weight female (twin 1) neonate presenting with proptosis of the right eye and *Staphylococcus aureus* sepsis. Imaging revealed osteomyelitis of maxilla and intraorbital extraconal mass with restricted diffusion, suggesting orbital abscess secondary to maxillary sinus osteomyelitis. This maxillary osteomyelitis is a rare entity in infancy, and is the prime differential diagnosis for unilateral proptosis.

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Author Contributions

Janampally Ravikiran – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published

Swathi Chacham – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Uppin Narayan Reddy – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Jillalla Narsing Rao – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Ekta Aggarwal – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Anumula Soumya – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Nori Madhavi – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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