

A Rare Full-Term Newborn Case of Rib Osteomyelitis with Suspected Preceding Fracture

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Abstract	Acute osteomyelitis is uncommon in full-term neonates and occurs most frequently in those with critical illnesses, often following episodes of sepsis, skin infection, umbilical catheterization, urinary tract anomalies, or a complicated delivery. Here, we report a very rare case of acute rib osteomyelitis due to <i>Staphylococcus aureus</i> in a 13-day-old full-term male neonate. Ultrasonography (US) enabled diagnosis and revealed a coexisting costochondral junction rib fracture, which was not detected on routine chest radiography. Following a 29-day course of intensive parenteral antibiotic therapy, the patient was
Keywords	discharged in good health at 42 days of age without any scar formation. Due to its accessibility and safety, US can be a promising modality for detecting acute osteomyeli-
► rib	tis in neonates with clinically highly suspected conditions in the neonatal intensive care
 rib fractures 	unit setting, particularly those involving thin and mobile bones subject to respiratory
 osteomyelitis 	motion. However, further studies are required to assess the utility of US in these cases
 newborn 	and negative results. In low-risk neonates with osteomyelitis, an accompanying fracture
 ultrasonography 	should be considered.

Case Report

A male neonate weighing 2,875 g with Apgar scores of 9 at 1 minute and 10 at 5 minutes was born to a 36-year-old healthy mother with gravida 0 and para 0 at 40 weeks of gestation by cesarean section due to recurrent episodes of mild-to-moderate variable deceleration with normal baseline heart rate. The pregnancy was otherwise uneventful, and routine vaginal culture for any bacteria at 36 weeks of gestation was negative for significant pathogens. His mother had been afebrile with intact membranes, although her laboratory studies immediately before delivery revealed neutrophilia (white blood cell count [WBC], 10,680/µL; neutrophils, 90.5%). She was treated empirically with an antibiotic (meropenem) after the surgery. Her laboratory studies the day

received October 29, 2015 accepted November 4, 2015 DOI http://dx.doi.org/ 10.1055/s-0035-1570320. ISSN 2157-6998. after delivery revealed a moderate elevation of C-reactive protein (CRP) (12.31 mg/dL).

At birth, the amniotic fluid was clear and no resuscitation was needed. The baby was healthy, predominantly breast-fed and had no history of chest electrodes placed for heart rate monitoring in the nursery. However, the mother's postoperative hematoma obliged him to stay in the hospital. Apart from a venipuncture for routine screening for inborn errors of metabolism, no invasive medical procedures were performed until an attending physician observed a fever of 38.7°C and firm right anterior chest swelling with skin erythema at 13 days of age. After receiving two infusions of cefotaxime in 8 hours, he was referred to our facility for further diagnostic workup and management.

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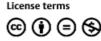




Fig. 1 Photograph of the right side of the chest showing a swelling (measuring \sim 45 × 40 mm) with mild local skin edema and redness; however, no wound, skin laceration, or scar suspected from trauma is evident on the local skin surface.



Fig. 3 Frontal chest radiogram on admission showing no significant findings, except for mild right-sided subcutaneous swelling. The fracture of the right ninth rib is not identified.

On admission, he weighed 3,312 g and showed mild restlessness, and manifested neck stiffness with a fever of 38.7°C, tachycardia of 184 beats/minute, and mild tachypnea of 65 breaths/minute despite normal oxygenation. No redness around or discharge from the umbilicus, or purulent discharge from the eyes was detected. A swelling was observed on his right anterior chest wall which showed mild local skin redness and tenderness. The surface of the skin showed no evidence of trauma or laceration (Fig. 1). The mass measured approximately 24×10 mm under the area of redness. On careful palpation, it felt elastic and soft, had well-defined borders, and appeared to be fixed to a rib. No lymphadenopathy was noted on palpation. He showed no increased discomfort or prolonged episodes of crying as a result of careful passive motion of each extremity. An ultrasound examination (12-MHz linear transducer) revealed that the mass was encapsulated by the periosteum of the ninth rib, adjacent to the costochondral junction, where a fracture was also detected (Fig. 2). Laboratory tests of a blood sample were consistent with moderate inflammation (WBC of 14,410/µL,

CRP of 4.34 mg/dl). Chest radiograph was interpreted as normal (**Fig. 3**), and spinal fluid did not reveal pleocytosis. Urinalysis was normal, and the heart, liver, spleen, kidneys, and adrenal glands were sonographically unremarkable. Subsequent ultrasound-guided fine-needle aspiration revealed purulent material in the lesion, whose smear elucidated gram-positive cocci by bedside Gram staining. The mass was diagnosed as a subperiosteal abscess originating from acute bacterial osteomyelitis of the ninth rib. Immediate chemotherapy with intravenous cefotaxime (200 mg/kg/ day) and additional ampicillin (200 mg/kg/day) was initiated. He became afebrile within a few hours of admission, although the size of the mass was noted to increase rapidly and had doubled by the following day. Neck stiffness resolved 3 days after admission. Thereafter, he was asymptomatic, and the mass gradually diminished in size. Blood inflammatory markers also improved. Blood and urine cultures obtained on the day of admission, as well as the specimen drawn from the mass, grew methicillin-sensitive Staphylococcus aureus (MSSA). All swabs of the body surface (ear canal, axilla, and

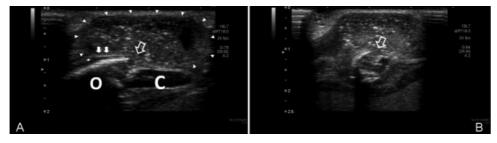


Fig. 2 (A) Longitudinal and (B) transverse ultrasound images of the right ninth rib region on the day of admission, demonstrating a 22×10 mm mushroom-shaped subperiosteal mass (arrowheads) of the right ninth rib, with heterogeneous echotexture including fluid with a relatively hyperechoic sediment. Gentle pressure over the mass with the probe induced motion, confirming the fluid content. (A) The echolucent cartilaginous (C) and shadowing osseous (O) portions are visible. The mass communicates with the medullary cavity of the rib at the costochondral junction, where a prominent discontinuity between the osseous and cartilaginous parts of the rib signifies a fracture (open arrow). Fluid adjacent to the cortex of the rib, elevating the periosteum by approximately 2 mm (narrow arrow), and moderate thickening of the periosteum are also shown (bold arrows).

inguina) and of the conjunctiva, throat, rectum, and umbilical stump were unremarkable, with no growth of pathogens. The cerebrospinal fluid was sterile. MSSA was not detected from the maternal vaginal swab obtained on the day of admission.

Following antibiotic sensitivity testing, treatment was switched to intravenous cefazolin (100 mg/kg/day) from 18 days of age for 3 weeks. At 42 days of age, he was discharged in good health without any scar formation at the local site or any radiologically detectable periosteal reaction following a 29-day course of intensive intravenous antibiotic therapy. Serum immunoglobulin and complement factor levels were within the normal range. Further, medical interviews showed that no family members or relatives had a history of susceptibility to infection.

Discussion

Although uncommon in neonates, acute osteomyelitis most frequently occurs in critically ill newborns, often following episodes of sepsis. Factors that may predispose to the development of acute osteomyelitis in neonates are prematurity, skin infection, umbilical catheterization, urinary tract anomalies, and a complicated delivery.^{1–3}

The distribution of bone involvement in 485 newborns with osteomyelitis has been documented as follows: tubular bones had the highest incidence at 76% (femur 39%, humerus 18%, tibia 14%, and radius 5%), while the maxilla, which uniquely to the newborn period is subject to the presence of predisposing factors such as maternal breast abscess, and delayed diagnosis and treatment compared with cases in children and adults, accounted for 4% of all affected bones.⁴ Rib osteomyelitis in neonates, however, appears to be extremely rare and we could not identify any human reports in the literature apart from a case in a 2-month-old compromised premature infant in an intensive care unit.⁵ Nevertheless, rib involvement has been reported in 57 children (aged 2 months-17 years) in the past 48 years⁶ and is estimated to account for approximately 1% or less of all cases of hematogenous osteomyelitis.⁷ Our patient appeared immunocompetent and otherwise almost healthy with no significant predispositions, apart from the fact that cesarean section was indicated on non-reassuring fetal heart rate monitoring immediately before delivery. To our knowledge, this may be the first report of a full-term neonate with rib osteomyelitis.

The most frequently isolated organism in neonates with osteomyelitis is *S. aureus*, which causes more than half of cases. Group B streptococci (GBS) are the second most common causative organisms. Other microorganisms include *Escherichia coli*, which accounts for approximately one-fifth of all infected cases; *Klebsiella*; *Proteus*; *Enterobacter*; and, in rare cases, *Candida albicans*, which is mainly isolated from premature infants with very low birth weight.⁴ Combination therapy with antistaphylococcal penicillin such as oxacillin or vancomycin, if MRSA is suspected, and cefotaxime is theoretically recommended for a neonate with suspected acute bacterial osteomyelitis pending culture and sensitivity results.⁴ In the present case, a more appropriate treatment would have been a single administration of cefotaxime or

additional administration of an antistaphylococcal penicillin instead of ampicillin until sensitivity was demonstrated. This is due to the obvious improvement following two doses of cefotaxime administered in the referring hospital. In addition, MRSA was not prevalent in the nursery. Common clinical manifestations of rib osteomyelitis in 57 children were reported as fever (36%) accompanied by chest, abdominal (13%), or back pain (10%), and sinus discharge (9.3%).⁶ Our case presented with obvious neck stiffness as well as fever and local inflammatory signs on admission. This symptom, which obliged us to initially use additional ampicillin, continued for some time despite the absence of GBS meningitis. In retrospect, this symptom might have been due to back or thoracic pain exacerbated by passive neck flexion, which is described as one of the manifestations of rib osteomyelitis.

The three possible routes for the development of osteomyelitis are hematogenous spread, which is considered the most common route in neonates; direct inoculation; and contiguous spread from nearby infection. Our case appears to be one of hematogenous spread, considering that the abscess developed at the costochondral junction, a common site for hematogenous spread because of its abundant blood supply. The lack of local skin trauma history after birth also points to hematogenous spread.⁶ The entry of the organism into the patient's body and establishment of disease at the unique site of the rib remain unclear due to birth at another institution and the placenta being unavailable for further pathological and bacteriological analyses, as it was discarded soon after delivery. In the present case, postnatal nosocomial infection might be likely, considering the isolated pathogen is MSSA. However, prenatal infection cannot be entirely ruled out due to the absence of skin infections, omphalitis, conjunctivitis, pneumonia, and pyuria on admission, and the fact that S. aureus was not isolated from any site other than urine and blood on admission. The observations of abnormal fetal heart rate, neutrophilia in the mother immediately before delivery, and elevated maternal CRP (12.31 mg/dL) on the day after an uncomplicated cesarean section also support the antenatal entry of the pathogen.

Regarding the establishment of the disease at the unique site of the rib, it is likely that the concurrent rib fracture identified on admission created a suitable environment for bacterial seeding from transient bacteremia in the perinatal period.⁸ The organism's establishment at the fracture site also appears to have been facilitated by the characteristic adherence of MSSA, which is promoted by several its extra- and intracellular virulence factors.

The genesis of the rib fracture has not been identified. In addition, we were unable to identify any reports of neonatal rib fracture at birth via cesarean section, although there are some reports of this fracture at birth via vaginal delivery.⁹ However, the anteroposterior thoracic compression by the operator's hands while pulling a neonate through a small incision in the uterine wall might exert enough force to sustain a costochondral junction rib fracture. It is not surprising that the fracture was not observed using conventional radiography in our case as acute fractures, particularly of the rib, are occasionally occult on initial radiography. This might explain why no neonatal rib fractures following cesarean

section have been reported to date. Although our case report consists of only a single neonatal patient, it might support the hypothesis that ultrasonography (US) is more sensitive than plain radiography in detecting acute rib fractures in adults¹⁰ and children.¹¹ This case also highlights the importance of evaluation for coexisting bone fracture in low-risk neonatal cases with unusual sites of bone infection.

Magnetic resonance imaging (MRI) or bone scintigraphy is standard diagnostic modalities for acute osteomyelitis. In the present case, however, we were able to make a prompt diagnosis and therapeutic decisions based on US alone. US reportedly detects significant changes as early as 48 hours after the onset of infection, manifested by fluid collection adjacent to the bone, elevation of thickened (> 2 mm) periosteum, and heterogeneous hypoechoic/hyperechoic echotexture in the overlying soft tissues.^{7,12–14} Aside from its high accessibility and safety, US has a profound advantage in that immediate subsequent ultrasound-guided needle aspiration permits the confirmation of infection after the survey. We consider that US can be a promising modality for detecting acute osteomyelitis in neonates with clinically highly suspect conditions in the neonatal intensive care unit setting, particularly when thin and mobile bones subject to respiratory motion are involved. Further experience and studies are necessary to assess the utility of US in these cases and negative results.

Conflict of Interest

The authors have no conflict of interest to declare.

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