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Case Report A Rare Preterm Newborn Case of Rib Osteomyelitis with Intrathoracic Involvement

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ABSTRACT

Background: Rib osteomyelitis is a very rare form of pediatric osteomyelitis.

Case report: Herein, we reported a very rare case of chest wall abscess with rib osteomyelitis and rib destruction due to Staphylococcus aureus infection in a 14-day-old preterm male neonate. The diagnosis of this uncommon disease requires a high index of suspicion due to its rarity and non-specific clinical presentation. The radiographic findings of osteomyelitis usually require 7-14 days to appear. Ultrasonography enabled us to diagnose a coexisting intra- and extra-thoracic solid mass with calcification. In addition, the chest high resolution computed tomography scan revealed rib osteomyelitis, which was not possible to be detected through the routine chest radiography. Following the administration of intensive parenteral antibiotic therapy for 21 days, the patient was discharged in good health condition at 42 days of age. He continued oral clindamycin consumption for the next three weeks. At the end of the six weeks of treatment, the chest Xray revealed the expansion of the fifth rib end; however, the chest ultrasound showed no obvious fluid collection. *Conclusion:* As indicated in the present case report, the diagnosis of rib osteomyelitis requires a high index of suspicion given its nonspecific clinical manifestations that can easily mimic other diagnoses. The unusual sites of bone infection could be diagnosed by means of HRCT, followed by ultrasound. The prompt treatment leads to a high cure rate with good prognosis.

Keywords: Preterm newborn, Rib osteomyelitis, Staphylococcus aureus

Introduction

Acute osteomyelitis is an uncommon infection in the neonates. Rib osteomyelitis in neonates is an extremely rare condition, accounting for less than 1% of all hematogenous osteomyelitis. This infection occurs most frequently in those with critical illnesses. It is often followed with episodes of sepsis, skin infection, umbilical catheterization, urinary tract anomalies, or a complicated delivery (1).

The clinical presentation of neonatal acute osteomyelitis is usually non-specific, which delays the diagnosis. There is a benign clinical onset in the majority of the neonates with little or no evidence of infection. Most of the cases result from hematogenous spread and typically occur in the metaphysis of long bones, such as the femur and tibia (2).

In a review of more than 300 cases with neonatal osteomyelitis, the male newborns were predominated over the female ones (i.e., a ratio

of 1.6:1). The premature neonates acquire osteomyelitis with relatively greater frequency than their term counterparts. In a series of osteomyelitis, the premature newborns comprised 17 of the 30 diagnosed cases and 4 cases occurred in the term neonates receiving intensive care. In the mentioned study, S. aureus was responsible for 23 of the proven cases of osteomyelitis (i.e., methicillin-sensitive strains in 16 cases and methicillin-resistant S. aureus in 7 cases). Furthermore, *Escherichia coli* and group *B*. streptococci were responsible for the infection of 3 and 2 cases, respectively.

Risk factors for osteomyelitis and septic arthritis in the premature neonates have been mostly iatrogenic, including the use of intravenous or intra-arterial catheters, ventilator support, and bacteremia with nosocomial pathogens. Transplacental bacterial bone infection is the most prominent characteristic of syphilis. The major

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cause of neonatal osteomyelitis is the blood borne dissemination of organisms with metastatic seeding of the skeletal system through nutrient arteries (3).

Case report

Herein, we reported the case of a male preterm newborn with 32 weeks of gestation born to a 28-year-old mother (gravida 3, para 2) through normal vaginal delivery. The amniotic fluid was clear at birth. The pregnancy was uneventful, except for the placental abruption. The patient's mother was afebrile with intact membranes. The 1- and 5-minute Apgar scores were 4-8 and 5, respectively. The neonatal weight, length, and head circumference at birth were 1800 g, 42 cm, and 30 cm, respectively.

The neonate needed cardiopulmonary resuscitation upon birth. The heart, liver, spleen, kidneys, extremities, and skin were normal. Immediately after admission to the Neonatal Intensive Care Unit (NICU), the patient received surfactant by INSURE technique due to respiratory distress. He needed positive pressure ventilation for three days. Immediately after birth, antibiotic therapy with intravenous ampicillin and additional gentamicin was initiated via central venous catheter.

The laboratory tests of the blood sample were unremarkable showing no growth of pathogens, clear spinal fluid, and sterile urinalysis. On the 10th day of life, he showed tachypnea. Although we found grade 3/6 systolic murmur, the chest X-ray was normal. He received packed red blood cell for anemia, and serial brain sonography showed normal results.

Echocardiography detected patent ductus arteriosus, left-right shunt, systolic pressure gradient of 36 mmHg, diastolic pressure gradient of 80 mmHg, peripheral capillary oxygen saturation of 92%, and mild left atrial and ventricular enlargement with pulmonary hypertension. Nevertheless, no cardiac vegetation was detected. The neonate was treated by fluid restriction, diuretics, and ibuprofen for three days.

On the 14th day of life, a focal soft tissue nonmobile non-erythematous mass gradually grew on the right side of the chest, and then changed to a soft tissue mass (30×40 mm) with skin edema and local redness (Figure 1). The patient's general condition was good, while he was on enteral feeding.

The chest radiography was repeated (Figure 2). Accordingly, a chest wall sonogram, and high



Figure 1. Photograph of the right side of the chest showing a swelling (measuring $\sim 30 \times 40$ mm) with local skin edema and redness without wound, skin laceration, or scar suspected of trauma on the local skin



Figure 2. Frontal chest radiogram on admission showing no significant findings

resolution computed tomography scan (HRCT) were performed. The HRCT revealed a heterogeneous soft tissue density (23×17 mm) in the anterolateral of the chest and destruction of the fifth rib (Figure 3). The sonogram showed rib



Figure 3. Chest high resolution computed tomography scan revealing heterogeneous soft tissue mass and rib destruction



Figure 4. Chest X-ray after three weeks of treatment showing fifth rib expansion

lytic lesion and focal chest wall solid mass with calcification and a lesion expanded to the intrathoracic area exerting pressure on the surrounding tissues.

The patient underwent incision and drainage of the right chest wall lesion. The fluid obtained from the abscess grew *S. aureus* sensitive to vancomycin and clindamycin. The mass was gradually diminished in size. The blood inflammatory markers (i.e., white blood cell count of 12,500 cell/µl, C-reactive protein [CRP] of 12.5 mg/dL, and erythrocyte sedimentation rate of 30 mm) were also improved. The serum immunoglobulin and complement factor levels were within the normal range. The patient received vitamin D due to having the serum level deficiency (9.2 ng/mL).

The patient was treated with intravenous vancomycin for three weeks. At this time, the chest ultrasound showed an intrathoracic opacity lesion of less than 2 mm. The chest X-ray revealed the expansion of the involved fifth rib end without obvious fluid collection (Figure 4). The newborn was discharged in good health condition (Figure 5),



Figure 5. Patient after three weeks of treatment

while the treatment was switched to the use of oral clindamycin for three weeks. The follow-up visit showed health progression with a normal CRP. At the end of the sixth weeks of the treatment, the chest ultrasound showed no obvious fluid collection.

Discussion

Acute osteomyelitis is an uncommon infection in the neonates. *Staphylococcus aureus* is the most common organism isolated in the patients diagnosed with this condition. The predisposing factors for the development of acute osteomyelitis in the neonates include prematurity, omphalitis, umbilical catheterization, prolonged hospitallization, pneumonia or meningitis, osteopenia of prematurity (4).

In a study investigating the distribution of the bone involvement in 485 newborns with osteomyelitis, tubular bones (76%) had the highest incidence rate (i.e., 39%, 18%, 14%, and 5% of the cases were observed in the femur, humerus, tibia, and radius, respectively) while the maxilla accounted for 4% of all the affected bones (5). Maxillary osteomyelitis is unique in the neonatal period, which may be caused by such factors as maternal breast abscess as well as delayed diagnosis and treatment.

Rib osteomyelitis in the neonates is an extremely rare condition. There is no evidence in the literature reporting the incidence of this disease in a human, except for a 2-month-old compromised premature neonate admitted to the NICU (4). The three possible routes for the development of osteomyelitis are hematogenous spread (as the most common route in the neonates), direct inoculation, and contiguous spread from a nearby infection.

In our case, the nosocomial hematogenous spread seemed to be the responsible route, considering that an abscess was developed at the costochondral junction, a common site for hematogenous spread, due to its abundant blood supply. The lack of local skin trauma history after birth is also indicative of the hematogenous spread as reported by Nascimento et al. (1). It is likely that the concurrent rib fracture identified on admission created a suitable environment for bacterial seeding caused by transient bacteremia in the perinatal period (6). However, we did not detect any rib fracture in the first chest X-ray.

The clinical presentation of neonatal acute osteomyelitis is usually non-specific and causes delay in diagnosis (2). The common clinical features include soft tissue swelling, tenderness, and decreased motion. Neonatal osteomyelitis most frequently involves the humerus and femur. In both sites, septic arthritis often accompanies the osteomyelitis, and the neonate frequently has swelling and remarkably decreased motion, referred to as pseudoparalysis in 95% of the cases (4).

Although our patient presented with common symptoms, there were other symptoms that made the diagnosis less obvious. In this regard, at the initial phase, the ultrasound findings revealed a mass with calcifications (may be sequestrum), which obliged us to think about soft tissue tumors rather than osteomyelitis.

Microbiology and imaging are equally important in confirming the diagnosis of acute osteomyelitis. A bacteriological diagnosis can be made in 50-80% of the cases if the blood and bone cultures are obtained. The radiographic findings of osteomyelitis usually require 7-14 days to appear and are readily confirmed by the presence of expansion and destruction of the rib, patchy necrosis, and extrapleural soft tissue swelling. In addition, a sequestrum may occasionally be present.

Magnetic resonance imaging is the most sensitive and specific diagnostic test for rib osteomyelitis (1). This technique facilitates the identification of abscesses and differentiation soft-tissue between bone and infection. Radioisotope bone scanning, ultrasonography of the bone, and CT scan are also useful in making an early diagnosis (2).

The literature review showed no reports regarding the description of a case with neonatal osteomyelitis after the administration of ibuprofen used for the treatment of patent ductus arteriosus. The neonates receiving ibuprofen treatment may show a higher decrease in the IL-6 and CRP levels, compared to those who are not subjected to such a therapy (7). Indomethacin has also been shown to inhibit neutrophil activation in animal studies; nonetheless, it is not well studied as ibuprofen. It is still unknown whether indomethacin has a dichotomous effect on neutrophil migration into lungs at different indomethacin concentrations as shown for ibuprofen (8).

According to Ono, ultrasound can be a promising modality for the detection of acute osteomyelitis in the neonates with highly clinical suspected conditions due to its accessibility and safety (6). This might support the hypothesis that ultrasound is more sensitive than plain radiography in detecting acute rib fractures in the

adults and children (9, 10). The osteopathy of prematurity is a multietiologic metabolic bone disease resulting in reduced bone mineralization and fractures.

The most common risk factors are inadequate supply of calcium and phosphorus, immobility, parenteral nutrition, and diuretic or steroid therapy. The treatment of this condition involves the initiation of enteral feedings as soon as possible to allow the efficient absorption of phosphate and adequate doses of vitamin D (4). Our patient and his mother had both vitamin D deficiencies.

The successful treatment of osteomyelitis depends on the appropriate selection and administration of antibiotics and surgical intervention as needed (2). Fungal osteomyelitis occurs as part of multisystem disseminated disease in the immunocompromised patients. Mycobacterial infection also plays an important role in rib osteomyelitis, which is responsible for 7% of the reported cases (1).

Conclusion

As indicated in the present case report, the diagnosis of rib osteomyelitis requires a high index of suspicion given its nonspecific clinical manifestations that can easily mimic other diagnoses, such as tumor. Our case also highlighted the importance of highly clinical suspected conditions in a preterm neonate admitted to the NICU with osteopathy and vitamin D deficiency. The unusual sites of bone infection could be diagnosed by means of HRCT, followed by ultrasound. The prompt treatment leads to a high cure rate with good prognosis.

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Conflicts of interests

The authors of this manuscript certify that they have no financial or other competing interest concerning this article.

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