An infant with unexplained multiple rib fractures occurring during treatment in a neonatal intensive care unit

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It is generally believed that trauma from child abuse or bone fragility from diseases such as osteogenesis imperfecta accounts for most cases of multiple rib fractures. We report an infant with unexplained fractures from the right 3rd rib through to the 8th rib who had undergone resection of a large cervical tumor and had been admitted to a neonatal intensive care unit. Neither trauma nor bone fragility disease was found in the patient during the detailed investigation conducted by the Malpractice Investigative Committee.

Key words: rib fracture, neonate, intensive care unit.

Rib fractures in children are much less frequent than in adults due to the inherent elasticity of children's rib cages. Great force is required to fracture ribs in infants; in children, even cardiovascular resuscitation seldom causes rib fractures[1]. Thus, the discovery of rib fractures on a chest radiograph of a child should always raise the suspicion of abuse. However, several previous studies have described rib fractures that occurred without any episodes of major trauma or other causative factors[2-6]. Thus, considerable care should be exercised when dealing with infants presenting with unexplained rib fractures.

We present the case of an infant with a large cervical tumor who had unexplained rib fractures identified during postoperative care in the neonatal intensive care unit (NICU), where family members could not even touch the patient without supervision by medical staff.

Case Report

A female infant weighing 3,463 g was delivered by cesarean section at a gestational age of 36 weeks. Since the baby had a large cervical tumor that was suspected of potentially causing impending airway obstruction, she was immediately intubated endotracheally for respiratory assistance (Fig. 1). At 10 days of life, the pediatric surgeons resected the tumor (8 cm×8 cm×7 cm) under general

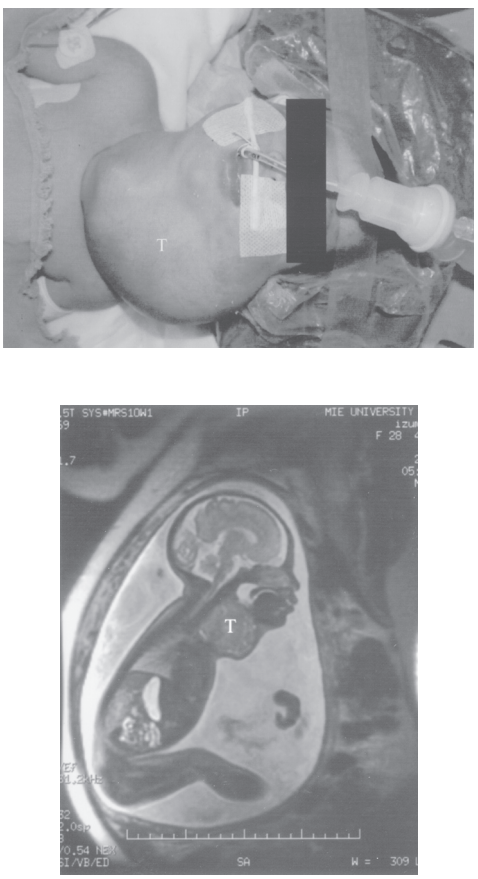

Fig. 1. The cervical tumor was suspected of compressing the trachea: A) macro finding at birth; B) magnetic resonance imaging at a gestational age of 32 weeks. T: tumor.

anesthesia. The right recurrent laryngeal nerve was sacrificed. Postoperatively, the baby was carefully treated in the NICU. Her condition was uneventful, and no anemia or hypoxia developed. On the 33rd day of life, the infant was extubated. The patient's respiratory condition was relatively stable except for occasional hypoxia during crying. Nutrition was delivered through a nasal tube. The baby had been fed with glucose or high caloric infusion up to the 5th day after the operation and with mother's milk thereafter. However, the baby's body weight decreased to 3,330 g by the 42nd day of life. On the 48th day, a nurse noted abnormal motion of the right chest on respiration while bathing the baby. Since the baby looked cheerful and did not cry at all during bathing, the nurse did nothing but record the finding in the chart. A routine chest X-ray taken the next day revealed rib fractures from the right 3rd to 8th ribs (Fig. 2b). In response to the report, the Malpractice Investigative Committee immediately started a thorough investigation, consisting of an inquiry regarding the baby's parents and all nine hospital staff who were involved in the care of the infant, as well as a review of the medical chart and X-rays.

On physical examination, there were no superficial findings, such as subcutaneous bleeding or swelling, on the right chest over the rib fractures. The infant had normal sclera and no familial history of bone dysplasia. Blood tests were normal, including serum calcium and phosphate levels; therefore, metabolic bone disease was excluded.

Chest X-rays showed no rib fractures before the 46th day of the patient's life (Fig. 2a). Rib fractures were first noted on X-rays taken on the 49th day (Fig. 2b). The callus formation in this area became visible on the X-ray taken on the 54th day (Fig. 2c). These findings suggested that the rib fractures occurred within a few days of the observation of respiratory abnormalities. The X-ray of the long bones showed no hypoplasia, thinning of the cortex, or scant spongiosis, characteristic features of osteogenesis imperfecta, and the skull X-ray showed no irregular mineralization.

An inquiry by the Malpractice Investigative Committee confirmed that all medical staff handled the baby appropriately. Tapping during respiratory physical therapy had been prohibited and had not been done. Medical procedures, including suctioning of the septum, were performed only by staff with technical expertise. Many of these staff members noticed that the baby bent back repeatedly when being held for the procedure. Following a detailed investigation of the chart and lengthy discussion, the committee concluded that slight trauma, in addition to transient brittle bone caused by prematurity or long-term bottle feeding, might have caused the rib fractures, and that this required close follow-up by her physicians.

During three years of follow-up, no new fractures were noted, and a chest X-ray taken at three years of age showed no abnormalities (Fig. 2d). The patient's growth and development have been uneventful, although the child had a hoarse voice due to recurrent nerve injury.

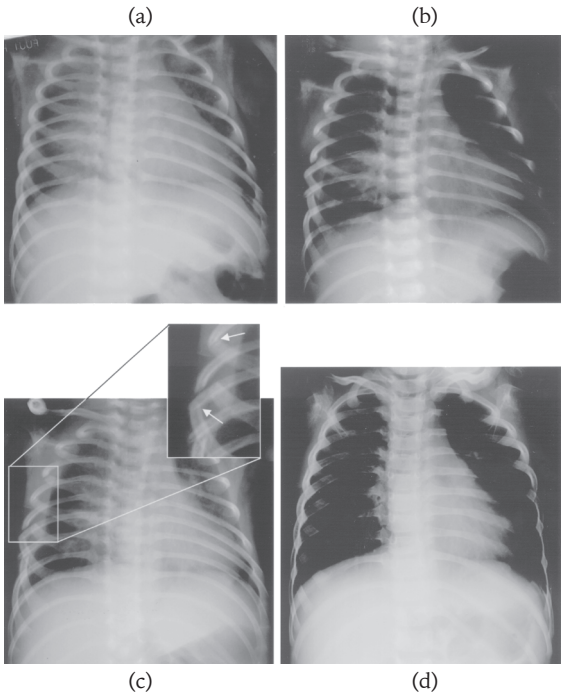

Fig. 2. Radiological findings at: A) 46 days; B) 49 days; C) 54 days; and D) 3 years. White arrow indicates callus formation at the injury site.

Discussion

In children younger than three years of age, the positive predictive value of a rib fracture as an indicator of non-accidental trauma is 95% as described by Barsness et al.[7]. Furthermore, Bulloch et al.[8] reported that 32 of 39 infants with rib fractures were victims of child abuse. Thus, rib fractures in infants are a strong

indication of child abuse. In contrast, Paterson[6] in 1990 proposed the entity of temporary brittle bone disease (TBBD), which consists of transient bone weakness in the first year of life with unexplained fractures and can be confused with child abuse. The clinical features of TBBD include a discrepancy between the radiological findings of fractures and the lack of superficial evidence of trauma, and the fact that unexplained fractures occur in later childhood[9].

In the case reported here, child abuse and an iatrogenic accident were ruled out since: 1) there were no superficial findings at the involved site; 2) the rib fractures occurred in the NICU under strict observation; 3) in spite of a thorough investigation by the Malpractice Investigative Committee, no evidence of iatrogenic injury or child abuse was found; and 4) the baby had no additional unexplained fractures for the next three years. In addition, the patient had no findings suggestive of osteogenesis imperfecta. Thus, the clinical findings in the present case seem to be similar to that of TBBD. However, child abuse and pediatric radiology communities have, for the most part, been unwilling to accept this as a real condition because none of the postulates have been substantiated with sound scientific data and it is also unclear as to how patients with TBBD differ from abused children[9,10]. In fact, extreme caution should be used when making the diagnosis of TBBD in infants with unexplained fractures, as misdiagnosis of an abused child could result in serious negative consequences for the child.

However, neonatologists have stated that prematurity is an important risk factor for fractures that occur during early infancy, since bone accretion is greater in the intrauterine environment than in the extrauterine one[11]. Amir et al.[12] reported that prolonged parenteral nutrition in the NICU and bronchopulmonary dysplasia were the two most frequently associated features in 11 premature infants with unexplained fractures. These data indicate that prematurity or long-term bottle feeding, as in this patient, could cause transient bone brittleness, rather than Paterson's TBBD. Furthermore, since the baby bent backward repeatedly during sputum suctioning, and this movement could cause excessive levering of the posterior ribs at the costotransverse process articulation, as demonstrated by Kleinman et al.[13], this could have resulted in the fractures.

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