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CASE REPORT

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Spontaneous Fractures in the Differential Diagnosis of Fractures in Children*

ABSTRACT: A four-year-old male with cerebral palsy and spasticity, as a result of a non-accidental head injury sustained when he was two years old, died of pneumonia. Postmortem full body X-rays revealed fractures of varying ages of the left humerus and both femora, tibiae, and fibulae. This led to a thorough investigation of the case by the Office of the Chief Medical Examiner. Child abuse, accidents, metabolic bone disorders, other primary or secondary diseases of the bones, and pathological fractures were ruled out. The final diagnosis was spontaneous fractures secondary to osteopenia. The term spontaneous fractures is used to define fractures that occur without any known external cause, especially in cerebral palsy patients with spasticity.

KEYWORDS: forensic science, spontaneous fractures, cerebral palsy, osteopenia, child abuse

Multiple fractures of varying ages in the long bones of a child are highly suggestive of child abuse. However, a wrong diagnosis can result in untold hardship to the parents; whereas a missed diagnosis can result in further harm to the child. Therefore other causes of fractures in children (i.e., accidents, metabolic bone disorders and diseases) need to be eliminated before a firm diagnosis of child abuse can be made. Spontaneous fractures should also be ruled out. This type of fracture is familiar to orthopedic surgeons and other healthcare workers who are responsible for the management of brain-damaged, long-term bedridden patients, such as those with cerebral palsy with spasticity. However, to the authors' knowledge, spontaneous fractures have not been described in the forensic literature and are therefore less familiar to forensic pathologists.

Case History

The death of this four-year-old child was reported to the Office of the Chief Medical Examiner as deaths of all children in the Province of Manitoba, Canada, are reportable under Section 7(9) of *The Fatality Inquiries Act*. Preliminary investigations revealed that when the child was two years old he sustained what was considered to be a severe, non-accidental head injury while in the care of a relative. Although the police investigated the incident, there was insufficient evidence to lay charges. The child was left in a vegetative state with spastic quadriparesis and was totally dependent on caregivers for daily activities of living. The child showed little response to external stimuli. However, he did display some agitation (i.e., teeth grinding, eye blinking, and hand clenching) during his daily care and while being moved. Medically he was not recognized as having a seizure disorder, but he was prescribed baclofen, an anti-spastic medication. Following discharge from the tertiary care centre after his head injury was treated, he was cared for in a rural hospital and then by his parents who subsequently placed him in another rural health care facility. The last 16 months of the child's life were spent in a chronic care centre for severely disabled children and adults. In the days preceding his death he developed respiratory distress and fever. Although he was transferred from the chronic care facility to a tertiary children's hospital where he was treated for pneumonia, he died the following day.

As part of the investigation, the medical examiner ordered a postmortem examination. Full body X-rays taken before the autopsy revealed recent and remote fractures of the left humerus (Fig. 1*a*, Fig. 1*b*), distal ends of the femora, and proximal ends of the right tibia and fibula (Fig. 2). According to the pediatric radiologist, these fractures ranged in age from four weeks to nine months. He also noted the bones were "somewhat osteopenic."

At autopsy, the pathologist found no evidence of external bruising. However, he noted several older fractures of the left tibia and fibula that had not been discernible radiographically. With the exception of the lungs and brain, all the child's internal organs were normal. The complete autopsy, which included microscopy and neuropathologic examination, failed to reveal any signs of child abuse (i.e., contusions, bruises, burns, bite marks, retinal hemorrhages, etc.); primary or secondary bone diseases; or any bone ma-

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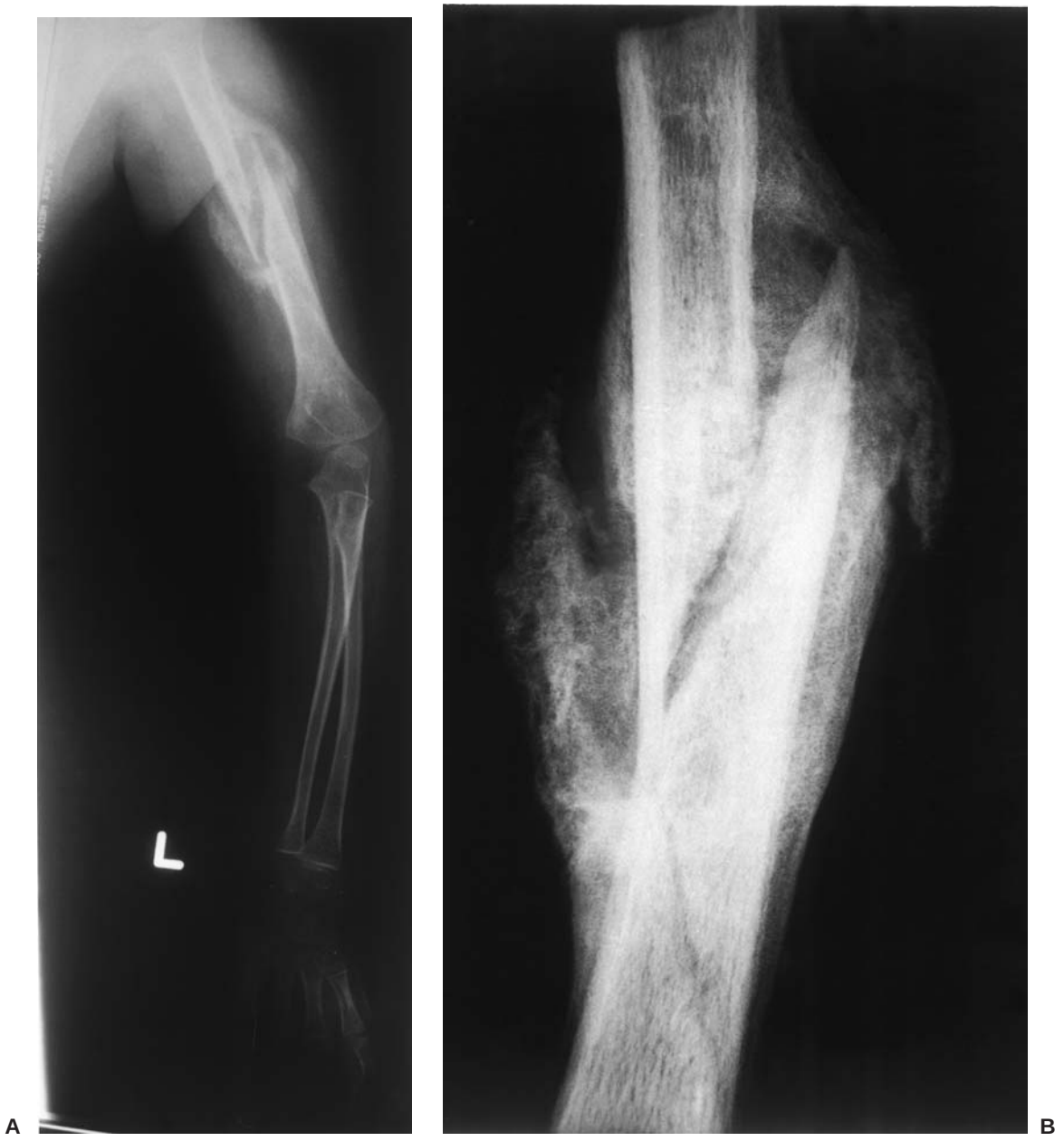

FIG. 1*a* and FIG. 1*b*—*Postmortem radiographs of the left humerus showing oblique fracture of the mid-shaft of the left humerus with extensive well-developed callus of approximately four weeks' duration.*

lignancies. The cause of the child's osteopenia was not explored further. The pathologist concluded that the child's death was due to acute necrotizing bronchopneumonia following neurodevelopmental delay due to traumatic brain injury (remote).

In order to identify the cause of the child's fractures, the medical examiner's office continued its investigation. Investigators learned that the child, while briefly in the care of his mother and stepfather following discharge from the Children's Hospital for the head injury, had sustained a fracture of the right distal femur and had been treated by an orthopedic surgeon. The surgeon suspected from the X-rays that the bones were osteopenic and planned further investigation. However, this medical follow-up never occurred.

The investigators also learned that the nurses at the chronic care facility had noted a swelling, but no external bruising, on the child's left upper arm 11 days prior to his death. The physician who examined the child did not feel there was an underlying fracture. However an X-ray taken three days later showed an oblique fracture of the mid-shaft of the left humerus with extensive, well-developed callus (Fig. 3). Although the child's medical records had been thoroughly reviewed and all caregivers at the facility with access to the child interviewed, no evidence of child abuse was found. It was noted as well that the mother had visited the child occasionally and that the staff at the facility, who were in the vicinity of the child during those visits, had never reported the occurrence of any unusual incidents.

As there continued to be no adequate explanation for the cause of the child's fractures, other potential causes were pursued by the medical examiner's office. This involved another in-depth review of the child's health records. Metabolic defects (e.g., osteogenesis imperfecta) were eliminated following a review of his past medical

history. Nutritional deficiencies such as abnormally low Vitamin D levels, which can lead to osteomalacia, were also discounted after it was determined that the child had been under the care of a nutritionist during his entire stay at the chronic care centre. After these possibilities were eliminated the medical examiner's office considered spontaneous fractures related to his osteopenia. From a personal communication with the director of the chronic care facility (which cares for approximately 230 residents), the investigators learned that the facility diagnoses four to five, non-fatal cases of spontaneous fractures yearly.

Discussion

In a normally active four-year-old child one might expect fractured long bones as a result of accidents even though multiple fractures of different ages would be rare. However, the child in this case was bedridden and investigators originally thought it unlikely that he could accidentally sustain such fractures during daily activities.

The cause of long bone fractures in children has been studied by many (1–6). Terms such as pathological fractures, stress fractures, and spontaneous fractures are frequently interchanged in the English medical literature. The term pathological fracture is commonly used to describe fractures secondary to distinct pathological lesions at fracture sites (i.e., metastatic tumor in the bone, and primary malignant or benign conditions of the bone) (7–10). Stress fracture is used to describe fractures that are the result of repetitive stress on normal bones (e.g., a march fracture) (8,9,11). On the other hand "spontaneous fracture," as defined by Miller and Glazer (12) in 1976, is used to describe a fracture "that occurs without any known, external cause." While this term may erroneously imply that bones fracture spontaneously without physical stress, to the authors' knowledge this term is well established in the medical literature. Its usage is widespread within the medical community when describing non-accidental fractures common to long-term bedridden patients. Although other more specific terms such as handling or insufficiency fractures may currently be in use, in the authors' experience they are not as prevalent.

While many articles have been written on the prevention and treatment of fractures among bedridden neurologically impaired patients (11–20), one of the first studies done on fractures among cerebral palsy patients was by McIvor and Samilson (13) in 1966.

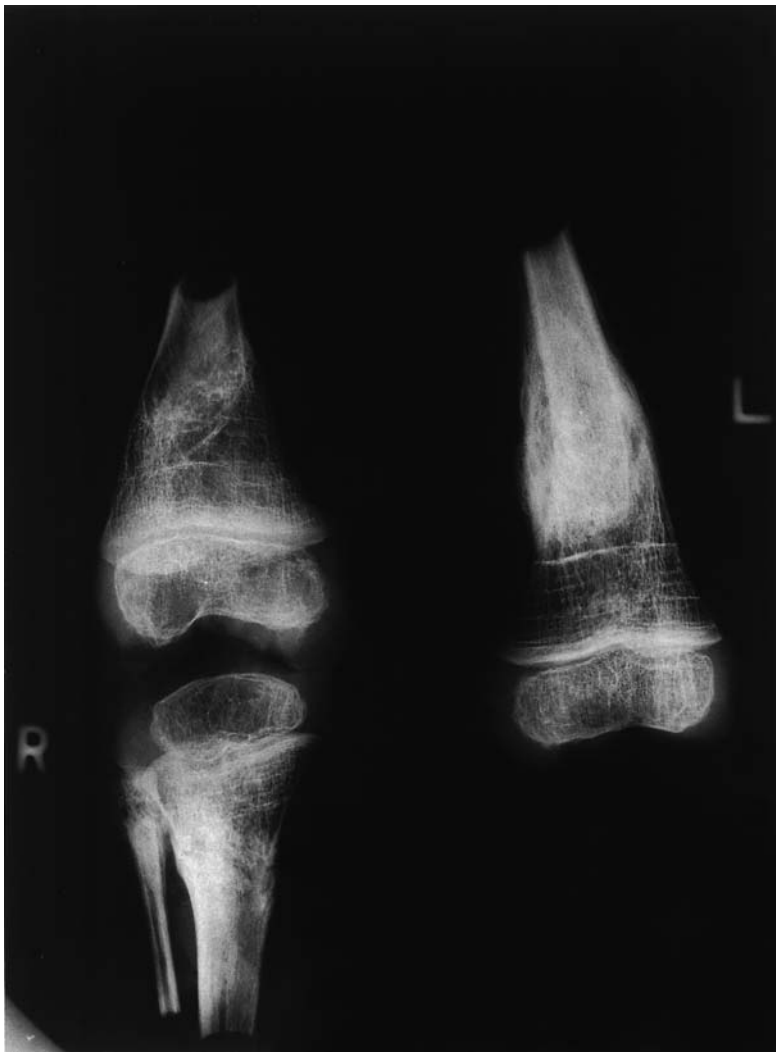

FIG. 2—*Postmortem radiograph showing right distal femoral fracture of approximately nine months duration. It has healed and largely remodeled. Also visible are healing undisplaced fractures of distal left femoral shaft, proximal right tibial shaft, and proximal right fibular metaphysis of approximately six weeks' duration.*

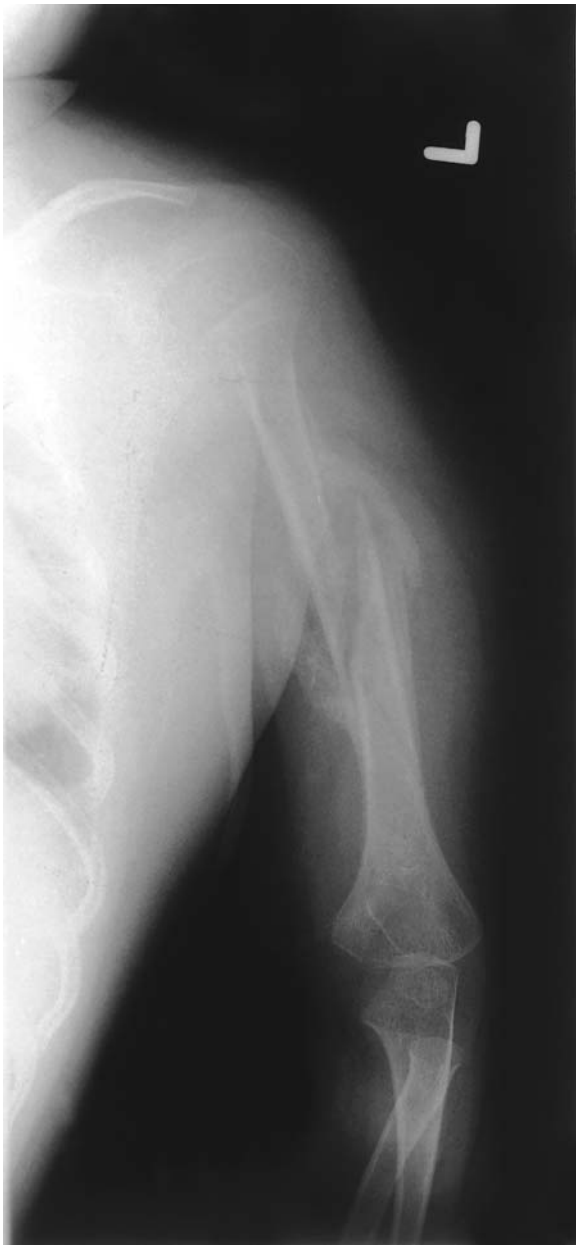

FIG. 3—*Antemortem radiograph of left humerus oblique mid-shaft fracture taken seven days prior to death.*

They studied 134 cases of fractures, which occurred between 1953 and 1963 in 92 cerebral palsy patients, with the intention of creating a program for the prevention and management of fractures in cerebral palsy patients. Their study showed that 39 of the fractures were due to accidental falls or blows, 16 were the result of normal handling by caregivers; five occurred when patients caught a limb on the side of a crib; and four were classified as miscellaneous. Although the sources of the remaining 70 fractures were undetermined, McIvor and Samilson did not suggest they were due to abuse. They recognized that pre-existent contractures and disuse osteopenia in long-term bedridden patients are major etiological factors in these fractures. Although the fractures in their study correspond to the spontaneous fractures described in this case study, McIvor and Samilson do not refer to them as such. Their work focused, not on the cause of spontaneous fractures, but rather on the prevention and therapeutic aspects of such fractures.

Handelsman (17) in 1972 studied 77 children with spina bifida cystica over a four-year period and observed that 11 of these children developed multiple recurrent spontaneous fractures in the lower limbs. Osteoporosis due to muscle flaccidity, immobilization, lack of weight bearing, the loss of protective sensation and a deficiency of Vitamin C were contributing factors in these fractures.

Between 1955 and 1974, Miller and Glazer (12) observed 31 spontaneous fractures in 29 of 50 institutionalized patients, the majority of whom were totally bedridden as the result of severe cerebral palsy associated with brain injury. They ranged in age from seven to 64 years, with an average age of 20 years. Miller and Glazer agreed with previous researchers (13, 17) that the cause of spontaneous fractures in these patients was multi-factorial. In their study they concluded that the bone matrix, compromised by a combination of severe motor handicap, spastic cerebral palsy, disuse atrophy of the skeletal system, progressive contractures, and marginal or deficient nutritional status, could fracture under minimal force, thus resulting in a spontaneous fracture.

In 1989 Lee et al. (18) published the results of a study involving 50 children and young adults whose selection from a clinic's population of severely handicapped patients was based on certain criteria (severe handicaps, adequate metabolic evaluation, and osteopenia). Osteopenia was a frequent condition among patients at the clinic, and it was known certain factors (disuse of limbs, nutritional deficiencies, and the interference of drugs, like anticonvulsants, in normal bone mineral metabolism) attributed to it. The findings by Lee et al. showed this group to have a high prevalence of abnormally low vitamin D levels. In a retrospective study published by Lee and Lyne (19) a year later, evaluating the predisposing factors, etiology, location, and treatment of pathological fractures in severely handicapped children and young adults, low Vitamin D levels were again noted.

In 1994 Lingam and Joester (20) examined five cerebral palsy patients who were between ten and 19 years of age. These patients had little or no voluntary movement, displayed bone demineralization on X-ray examination, and developed spontaneous fractures. As described previously by Miller and Glazer (12), they also noted that children with cerebral palsy were fairly immobile, had decreased musculature, tended to develop contractures, and could experience involuntary extensor spasms. All these factors, when combined with poor muscle support and excessive stress to the bone matrix, can result in spontaneous fractures. Lingam and Joester therefore concluded the importance of recognizing spontaneous fractures as such and not confusing them with fractures as a result of non-accidental injuries or trauma. However, Hobbs and Wynne (21) questioned this concept. They were critical of Lingam's and Joester's use of the term spontaneous fracture in cerebral palsy patients and were concerned that child abuse cases might be missed in these patients.

Brunner and Doderlein (22) studied 37 cerebral palsy patients who had sustained 54 fractures without any significant trauma. They identified long and fragile lever arms, stiffness of major joints, and osteoporosis due to immobilization as contributing factors. They referred to these fractures as pathological fractures. However according to the definition used by the authors, based on the criteria established by Miller and Glazer (12), these fractures fall into the category of spontaneous fractures. This may be explained by the focus of Brunner's and Doderlein's work, which was on preventing and treating these fractures, not defining them.

Conclusion

The initial postmortem radiographs of multiple long bone fractures in this four-year-old child raised strong suspicions of child abuse. However abuse was ruled out following a complete and thorough investigation, which included a postmortem examination, review of medical records, and interviews with the child's caregivers. Although the mechanism of the child's fractures was never definitively established, they probably occurred during daily care giving activities. This case created an awareness of the tendency for spastic cerebral palsy patients to experience spontaneous fractures. As previously mentioned, although this condition is recognizable to orthopedic surgeons and to others who care for brain-damaged, long-term bedridden patients, it is not as familiar to the forensic medical community. Forensic pathologists in particular need to be informed about this condition to differentiate between non-accidental fractures and spontaneous fractures of long bones in spastic and/or immobilized individuals.

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