

SHORT COMMUNICATION

S. Mimasaka · M. Funayama · N. Adachi · M. Nata
M. Morita

A fatal case of infantile scurvy

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Abstract We report a case of infant death due to scurvy, which is very rare in Japan. We initially had little knowledge of the disease and suspected that the bleeding in the body was caused by domestic violence. The case did not fall under the category of the battered child syndrome but the death was caused by ignorance with respect to child care. In addition the parents usually locked the child alone in a room during the day and this is probably a case of neglect.

Key words Scurvy · Periosteal bleeding · Neglect

Introduction

A 6-year-old girl was admitted to an emergency hospital with cardiopulmonary arrest and resuscitation was unsuccessful. The mother stated that the girl's condition suddenly deteriorated while in the bathtub. White foam was present in the nostrils and mouth and clinically the death was diagnosed as drowning. Numerous instances of bleeding on the body surface aroused a suspicion of child abuse and the doctor notified the police.

Autopsy findings

The body was that of a poorly developed child measuring 90 cm in height and weighing 10.6 kg. There were many subcutaneous hemorrhages on the face, the chest, the back and the lower extremities. The gums were swollen and

bleeding and some teeth were absent. The legs appeared swollen and when an incision was made on the lower legs, bloody fluid leaked out from under the periosteum of the tibia. Massive hemorrhages were also seen under the periosteum of the humerus and the femur. These bones were not fractured and the periosteal blood fluid culture was found to be aseptic on bacteriological examination. Visible long bone deformities were not seen, but there was dislocation of the epiphyseal plate in the long bones and furthermore the bones were loosely jointed and easily dislocated. The soft X-ray revealed heavy calcification around the periosteum including the epiphyseal regions (Fig. 1). The periosteal lesions showed calcification, hemorrhage, hyperplasia of cells and a slight degree of fibrosis, but there were no indications of a tumor or periostitis.

The lungs were distended, foam was found in the airways and gastrointestinal bleeding was not observed. The weights of the brain, the heart, the liver, the spleen, and the thymus were 1060 g, 57 g, 400 g, 15 g, and 4 g, respectively.



Fig. 1 Soft X-ray findings showing calcifying periosteum over superiosteal hematoma around epiphysis of femur

S. Mimasaka (✉) · M. Funayama · N. Adachi · M. Nata
Department of Forensic Medicine,
Tohoku University School of Medicine, Seiryomachi 2-1,
Aoba-ku, Sendai 980-8575, Japan
Fax: +81-22-7178112

M. Morita
Department of Legal Medicine,
Sapporo Medical University School of Medicine,
S.1, W.17, Chuo-ku, Sapporo 060-8556, Japan

The autopsy findings led to the tentative diagnosis of drowning. The skin bruises seemed to have been caused by blunt objects but the cause of the periosteal bleeding was unknown. The deceased showed stunted growth and child abuse was suspected.

The mother's statement

The mother stated that she had taken a bath with her daughter (the deceased) beginning at 1100 hours and 1 h later she got out of the tub but the daughter remained in the tub. At 1400 hours the mother heard the sound of struggling and groaning and found her daughter holding on to the edge of the bathtub. According to the mother, the daughter claimed to have almost drowned. She then took the girl from the tub and placed her on the sofa at which time she was still conscious. Moments later, the girl began

gasping for air and frothing at the mouth and soon after she lost consciousness and died.

The daughter had been born prematurely; however, she had rarely received any follow-up medical attention. She had complained about leg pains, difficulty in walking, and loss of appetite for a week prior to death.

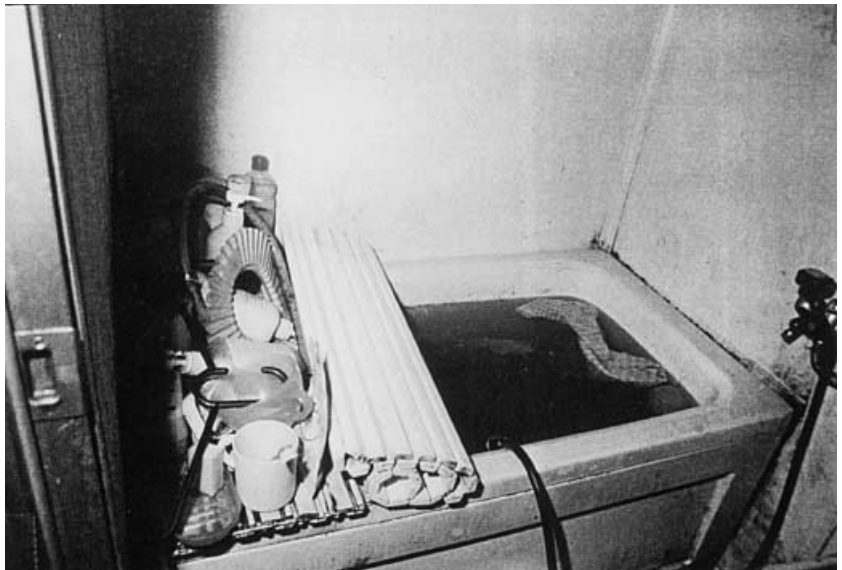
Death scene

The family, the decedent, the father (32-year-old sales clerk) and the mother (26-year-old part-time laborer) lived in an apartment in the northern Japanese city of Sapporo (population of 1.5 million). The living room, the kitchen, and the bathroom were in a state of disorder (Fig. 2) and there was brown muddy water in the bathtub (Fig. 3). Neither parent had a known clinical history or mental disorder. The child's room was comparatively tidy. The

Fig. 2 Very disorderly living room



Fig. 3 Brown muddy water filled the bathtub and according to the mother's statement the child had been playing in the tub for ca. 3 h



child's death happened before the mother's eyes, but the parents usually locked her alone in the room during the day.

Discussion

Initially, the bleeding under the periosteum appeared to have been caused by trauma but this was very difficult to explain. Why was the bleeding mainly limited to under the periosteum? Why were there no muscular injuries? Why was the periosteal bleeding symmetrical? Several months after autopsy, we turned our attention to the bleeding of the gums, which has many possible causes, such as trauma, inflammation, leukemia, hemophilia, thrombocytopenic purpura and deficiency diseases. The oral lips and the labial mucosa were intact but the child was poorly developed so that scurvy, a deficiency disease, was taken into consideration.

Clinically, bleeding into the skin, the gum and the periosteum of the long bones suggested scurvy. Periosteal bleeding of the tibia, the femur and the humerus are also characteristic signs of infantile scurvy [1]. Alveolar bone resorption and hemorrhages into the periodontal membrane cause loosening and eventual loss of teeth [2]. Infantile scurvy may cause visible bone deformities resembling rickets [2]; however, the bones seemed to be intact on macroscopic examination. Radiographic changes of scurvy are typical [1, 3] and the dominant X-ray findings in infantile scurvy are [2] as follows:

1. A broadened zone of divisional calcification as manifested by a thickened white line at the ends of the long bones.
2. A zone of rarefaction beneath or on the shaft side of the thickened "white line"
3. Separation of the epiphysis
4. Spur formation
5. A "ground-glass" appearance of the shaft
6. Thinning of the cortex
7. A "halo" appearance of the epiphysis

Because our autopsy room was not equipped with X-ray equipment, we removed some bones and the epiphyseal lesions and X-rayed them using soft X-ray radiography.

The periosteum around the epiphysis of the femur was heavily calcified and this finding resembled the halo effect around the epiphyses of knee joints which is one of the dominant X-ray findings of scurvy. The abnormal findings on the shaft, e.g., the thinning of the cortex, were not clear, and on laboratory examination no ascorbic acid was detected in the body serum. There have been some reports about a causal relationship between sudden cardiac death and severe scurvy [4, 5, 6]. In this case, no cardiac abnormalities were found at autopsy; however, the cause of death seemed to be cardiorespiratory failure due to scurvy and not from drowning.

Scurvy is a rare disease in developed countries. In 1975, Sauberlich [7] reported that manifest clinical scurvy seldom occurred in the United States. Berant et al. [8] stated that many cases of scurvy were seen in religious Jews who did not eat fruit or vegetables during the Shemittah year (which means letting the land lie fallow, hence the lack of fruit). In present-day Japan, there are very few reports of scurvy-related deaths and it would seem to be a forgotten disease. When considering the state of the room, it was suspected that the parents suffered from some sort of mental disorder but the case did not fall under the category of "willful abuse". However, it is safe to say that the death was caused by ignorance with respect to child care, and was possibly a form of neglect.

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