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CASE REPORT

Late Radiographic Findings of Infantile Scurvy in Northern Mali, a Region Facing a Humanitarian and Security Crisis

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ABSTRACT

Scurvy is a disease caused by a deficiency of vitamin C. This disease is rare in developed countries, but can occur in developing countries that face food shortages and humanitarian crises. In these countries, the diagnosis is often missed due to the lack of adequate diagnostic equipment. We report a case of scurvy in a 2-year-old boy who was diagnosed late by radiographic findings, which suggested a diagnosis of scurvy.

Key Words: *Developing countries; Infant; Scurvy*

中文摘要

面對人道主義和安全危機的馬里北部的一個嬰兒壞血病例的晚期影像學結果

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壞血病因缺乏維生素C造成，這病雖然在發達國家中很罕見，但在一些發展中國家由於長期糧食短缺以及面對嚴重的人道主義危機仍會發生。這些國家由於缺乏足夠的診斷設備，壞血病經常會被漏診。本文報告一名兩歲大的男孩在發病較晚期時才進行X線檢查，後被確診為壞血病。

INTRODUCTION

Scurvy was described for the first time in 1536 by Jacques Cartier,[1] as "plague of the Seafarer". Although scurvy is now extremely rare in developed countries,[2] it is more common in developing countries due to humanitarian crises that lead to poor nutritional conditions, especially for children. These conditions can lead to diseases of malnutrition such as scurvy.[3,4] Interruption of food support programmes in these countries reduces the available nutrition. We report a case of a child with late radiologic findings of scurvy in northern Mali, a region facing a humanitarian and security crisis.

CASE REPORT

In June 2014, a 2-year-old boy twin, born to a family

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of eight children, was seen by a non-governmental organisation supporting formal nourishment of children in Mopti, Mali. The child was referred for radiologic investigation of persistent bilateral painful swelling of the thighs and knees 3 weeks after starting management of malnutrition with generalised oedema. The radiographs of both femurs showed osteopenia of the epiphyses, disruption of the alignment of the distal physis of both femurs and their separation from the distal femurs, and large soft tissue swelling with bilateral paradiaphyseal calcifications along the femurs (Figure 1).

In view of the radiographic appearance, differential diagnoses of bilateral periostitis, bilateral chronic osteomyelitis, and bilateral localisation of a bone tumour were considered. The bilateral aspect of the condition led to the decision for further investigations.

Further questioning of the parents who were nomadic shepherds showed that the child was usually fed with milk and meat. He was given very little grain and few cereals, which were not easily obtained because of the ongoing humanitarian crisis in their region of origin (northern Mali). The child, who had started walking at the age of 1 year could no longer walk 9 months previously. The parents also reported an episode of epistaxis, which was treated with herbal medicines.

On examination, the child looked sick, pale, irritable, and bad-tempered. He had delayed weight gain and growth, with a weight below the 5th percentile for age and sex. His height is below the 4th percentile for age and sex. His heart rate was 120 beats per minute, respiratory rate was 28 breaths per minute, temperature was 37.4°C, and blood pressure was 100/60 mm Hg.

Physical examination revealed bilateral painful swelling of the thighs and knees, with limitation of the flexion-extension of the knees (Figure 2). There was no external bleeding noted. There were no abnormal findings on abdominal, cardiovascular, and respiratory examinations.

Laboratory test results 3 days later showed hypochromic microcytic anaemia, neutrophils, and eosinophils. There were no abnormal findings among the biochemical tests performed (Table). Tests for ferritin level and vitamins, especially folate and vitamins C and D, could not be performed because of the lack of adequate technical equipment.

In view of the clinico-radiological investigations, a diagnosis of calcified subperiosteal haematoma as a consequence of vitamin C deficiency (scurvy) was made. The child received a substitutive treatment with oral vitamin C 200 mg daily, combined with

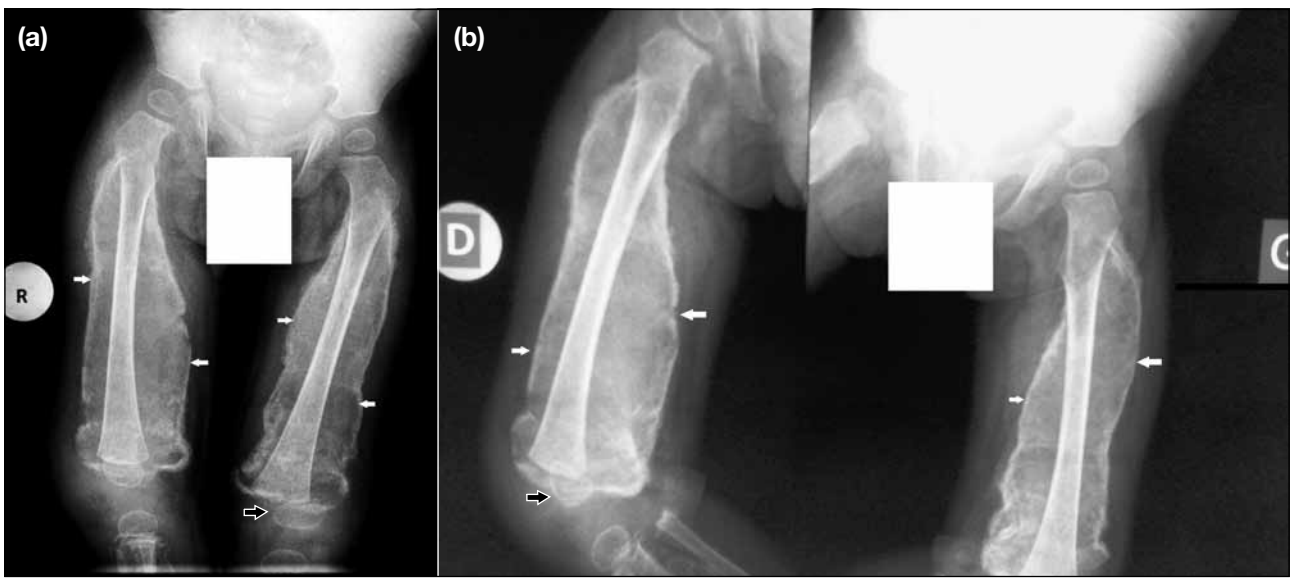

Figure 1. Radiographs of both femurs of a child with scurvy. (a) Anteroposterior and (b) oblique images show osteopenia of the epiphyses, disruption of the alignment of the distal physis and their separation from the distal femurs (black arrows), and large soft tissue swelling with bilateral paradiaphyseal calcifications along the femurs (white arrows).

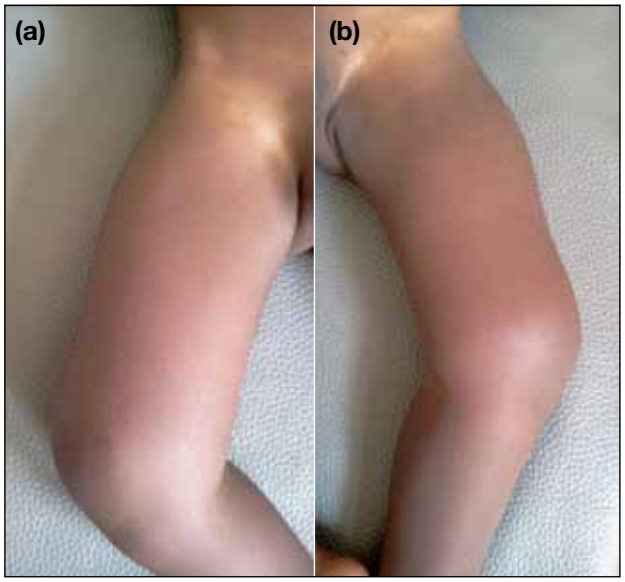

Figure 2. Photographs of both thighs showing swelling of the (a) right and (b) left thighs and knees.

continuation of nutritional management of the malnutrition. This resulted in complete regression of pain and gradual recovery of walking ability after 6 weeks.

DISCUSSION

Radiographic Findings

Musculoskeletal manifestations are present in 80% of patients with scurvy.[5] Our patient had undergone radiography 9 months after the onset of his illness. Scurvy could explain the radiographic findings in this patient, especially the calcifications of the periosteum. Indeed, periosteal calcifications, which seem to result from a subperiosteal haematoma, are commonly seen during the healing phase of the disease.[6,7] The other radiographic findings such as osteopenia, metaphysis abnormalities, and disruption of the alignment of the distal physis were suggestive of vitamin C deficiency (scurvy). Due to the rarity of the disease, the radiographic findings of scurvy are not well-known, which can lead to misdiagnosis or diagnostic delay. In many cases, the radiologic diagnosis has been confused with conditions such as osteomyelitis, septic arthritis, acute rheumatic fever, juvenile rheumatoid arthritis, domestic violence / child abuse, complex regional pain syndrome, vasculitis, neuroblastoma, bone tumour.[8,9] Even with the new imaging modalities that are sensitive to bones and joint imaging such as magnetic resonance imaging (MRI), diagnostic confusion persists.[7,10] Indeed, subperiosteal haematoma associated with bone signal abnormalities on MRI may falsely suggest periostitis, subperiosteal abscess, osteomyelitis, or a tumour process, as for the radiographic images.

Laboratory Findings

The diagnosis of scurvy is made by clinical and radiographic findings, and may be supported by additional findings such as reduced levels of vitamin C in the serum or a buffy coat of leukocytes.[7] In this patient, we did not measure the vitamin C level because of the lack of adequate technical equipment. A low vitamin C level in the plasma is specific for the diagnosis of scurvy. This is not always a reliable indicator, however, because plasma levels may be

Table. Laboratory test results.

	Value	Reference range
White blood cell count (x 10^9 /l)	13	4-11
Lymphocytes (x 10^9 /l)	4	3.5-4.5
Neutrophils (x 10^9 /l)	7.2	3.5-6
Eosinophils (x 10^9 /l)	0.7	0.1-0.5
Basophils (x 10^9 /l)	0.03	0-0.1
Monocytes (x 10^9 /l)	1	0.1-1.5
Red blood cells (x 10^{12} /l)	4.66	3.8-6.5
Haemoglobin (g/l)	94	120-170
Reticulocytes (x 10^9 /l)	45	40-80
Haematocrit (%)	28	37-54
Mean corpuscular volume (fl)	70	80-100
Mean corpuscular haemoglobin concentration (g/l)	260	320-360
Platelets (x 10^9 /l)	228	150-400
Glucose (g/l)	1	0.74-1.10
Urea nitrogen (mmol/l)	13	5-16
Creatinine (μmol/l)	106	50-120

normal after recent intake of ascorbic acid.[7] Measuring vitamin C levels in the buffy coat of leukocytes better reflects the body's stores, but this test is technically more difficult.[7] Additionally, accurate laboratory measurement of vitamin C levels is difficult because of the instability of vitamin C, and serum plasma vitamin C measurements do not correlate well with tissue levels.[11]

Treatment

In this patient, the pain had disappeared after 6 weeks and the child had gradually started walking, confirming the diagnosis of scurvy by the adequate response to treatment. This confirms that the best evidence of scurvy is resolution of the manifestations of the disease after treatment with ascorbic acid.[7,8,12,13]

CONCLUSION

In conditions of humanitarian crises, scurvy can be observed in children due to nutritional deficiencies. Its diagnosis and treatment are delayed due to the combination of lack of security leading to movement of populations, and lack of health infrastructure and food. Radiography can be an interesting method of diagnosis, especially when laboratory tests are not available.

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