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Case Report

Scurvy in a six-month-old infant

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ABSTRACT

We report a case of scurvy. Presentation at six months of age is being reported for the first time as only prolonged deficiency of vitamin C leads to florid scurvy. Literature review for last twenty years has revealed that except from Thailand, disease is quite rare now a days and not even 50 cases have been reported in children in this period.

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1. Introduction

Scurvy, caused by Vitamin C (ascorbic acid) deficiency, is a rare nutritional disorder nowadays. Disease was often fatal in sailors in mediaeval period and Lind first reported in 1753 association and curability of the disease by fresh fruits.¹ It is still occasionally found in infants on un-supplemented milk diets or in children as well as adults with restrictive eating habits. We report one such case in a six-month-old infant.

2. Case Report

A six-month-old male baby, was brought to emergency with complaints of difficulty in breathing and inability to move lower limbs for 15 days. Patient was referred from another hospital to surgical side for failure of treatment of suspected osteomyelitis / septic arthritis both knee joints and “need for possible drainage of knee joints”. Pediatric team was called to assess for possible haemarthrosis and exclusion of haemophilia and for stabilization of respiratory status before surgery.

Patient was afebrile, heart rate 96 /min, SpO₂ 98% with oxygen via nasal prongs @ 4L/min, blood pressure 95/70 mmHg, respiration 65/min with substantial suprasternal, substernal and intercostal retractions. Flaring of alae nasi was present and accessory muscle of respiration were prominent. Normal vesicular breath sounds without accompaniments were audible bilaterally.

Cardiovascular as well as nervous systems were normal. As movements of upper limbs were normal but active movements were grossly restricted in both lower limbs and passive movements of both lower limbs were very tender. Child used to stay calm in ‘pithed frog position’. Plantar reflex could not be elicited. There was cutaneous bruising at multiple places over both lower as well as upper limbs, both soles and also face (Figure 1). Both knee joints were swollen and tender. There was Severe wasting with loose skin folds. Weight was 2.86 kg, length 54 cm, head circumference 39 cm and MUAC 10.1 cm. Bipedal edema was also present. Child had never been breast fed and was getting diluted cow’s milk in one part milk two parts water dilution with total daily intake of about 250 ml of it.

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Fig. 1: Severe muscle wasting, bruising at multiple places and swelling of both knee joints

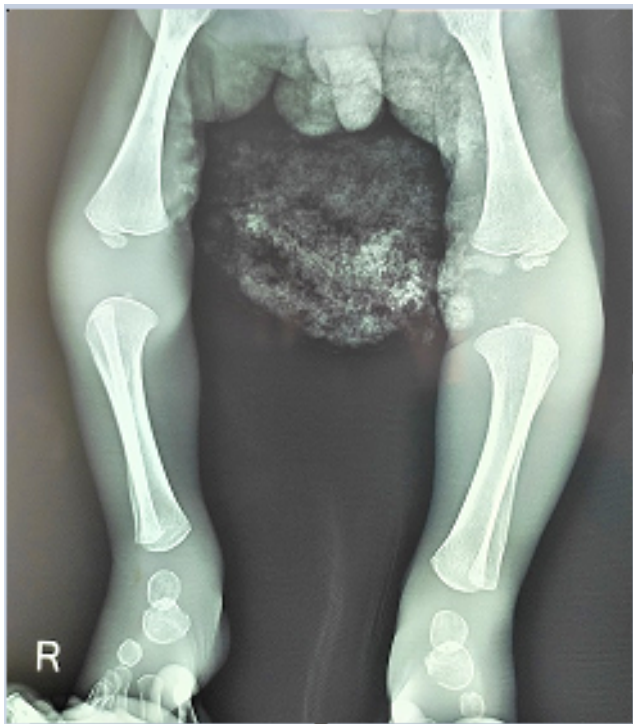


Fig. 2: Subperiosteal hematoma on medial aspect of right femur with white line of Fraenkel, zone of rarefaction proximal to white line, Wimberger ring sign and pelican spur.

Leucocyte counts were normal but C-reactive Protein (CRP) was raised (35.7). Liver and kidney functions were normal. Vitamin D level was 13.8 ng/ml. Serum ascorbic acid levels were not available and could not be done.

X-ray findings of knee and ankle joints were suggestive of scurvy. (Figure 2). There was thin pencil line cortex in long bones and ringings of small bones at ankle (Wimberger ring sign). A large subperiosteal hemorrhage with some calcification could be visualized on medial aspect of right femur. White line of Fraenkel was visible at lower end of

both femurs and extended laterally to form pelican spurs. Trummer's clear zone (Trummerfeld zone or scorbutic zone)—an area of less dense bone and rarefaction—was visible proximal and parallel to the white line at metaphysis and was diagnostic of scurvy. It was more marked at lateral ends to form the corner sign of Park.

History of any trauma was negative and there were no features of coagulopathies, neoplasia, or vasculitis syndromes.

Initial stabilization was done with intravenous fluids, multivitamin infusion and oxygen. Child was then put on nasogastric feeds and vitamin C supplementation orally. With this he improved over three weeks and was discharged after one month.

3. Discussion

Ascorbic acid is essential for formation of collagen, osteoid and intercellular connective tissue. Humans lack L-gulonolactone oxidase which synthesizes it from glucose and require it in diet.

Deficiency of ascorbic acid in diet over prolonged period causes manifestation called as scurvy. Although disease is rare now a days, occasional case reports do occur. Most cases occur without any apparent associated factor.² Common manifestations in children are pseudo-paralysis, swelling of knee joints, bleeding diathesis, poor appetite and lethargy and swollen bleeding gums. Infants may be restless and irritable due to subperiosteal or interfascial hematomas causing musculo-skeletal pain on moving legs. Unusual presentation in adults have also been reported and misdiagnosis is high (86%).³⁻⁵ We could get only two series, both from Thailand^{3,6} and one review series of 166 cases over twenty years.⁷ Patients are often first brought for orthopedic consultation due to joint swelling and painful limbs.⁸

Our case had two unique features—age and association with severe protein energy malnutrition. Cases have been in children above one year of age and no case below 10 months of age has been described previously. This may be due to adequacy of vitamin C in milk diets in first 6 months. Similarly, association specifically with marasmic-kwashiorkor is also not reported as no new bone formation may be happening and latent deficiency gets masked. Hence, a need for critical evaluation of vitamin C intake in vulnerable populations is suggested. A high index of clinical suspicion is needed as most present generation physicians may not have seen even a single case and often lack ability to interpret x-rays correctly.

4. Conclusion

Scurvy is a rare disease now a days and younger generation of physicians or surgeons may not see even a single case in their entire professional career. A great degree of clinical

suspicion with excellent history taking and examination are a must. As vitamin C levels (value < 0.1 mg/dL are considered deficient) or serum pyruvate levels are mostly not available, diagnosis is usually clinical and supported / confirmed by radiological findings.

5. Source of Funding

None.


6. Conflict of Interest


None.

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