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Spina ventosa-an uncommon presentation of a common infection: A rare case series.

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Abstract

Spina ventosa also called tuberculous dactylitis is an uncommon form of osteo articular tuberculosis which involves the short tubular bones of the hand and foot. Spina means short bones and ventosa means expanded with air. This is generally a rare manifestation of tuberculosis. Herewith we present a case series three children including an adolescent diagnosed with spina ventosa, whom we encountered over a period of two years, highlighting the diagnostic challenges in arriving at a diagnosis in each child.

Keywords: Tuberculous, Dactylitis, Adolescent, Spina ventosa, ATT

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Introduction

Tuberculous dactylitis, is a rare form of skeletal tuberculosis is manifested as tuberculous infection of the metacarpal, metatarsal, phalanges of the hands or feet. The first case of tuberculous dactylitis was reported by Leung in 1978. The rarity of the isolated osseous involvement would be due to small size of the bones of the hand [1]. The exact mechanism of how mycobacterium tuberculosis reaches the musculoskeletal system is not fully understood, although haematogenous dissemination after pulmonary involvement and direct inoculation are viable hypotheses [2]. It has been observed that nearly 85% of the cases of tuberculous dactylitis occur in children less than 6 years of age. This is because the marrow in the short tubular bones of children is still active. Thus, providing a favorable environment for the tubercle bacilli to lodge and multiply. Once the entire marrow space is involved, the marrow is destroyed and replaced with the granulomatous inflammation causing thinning of the overlying cortex. There is a fusiform dilatation of the involved bone due to destruction of the marrow space. This balloon like spindle shaped cystic expansion of the bone together with the thin overlying cortex has been given the name spina ventosa. Spina means short bones and ventosa means expanded with air the diagnosis is established by histology (caseating granulomas) and bacteriology (demonstrating acid fast bacilli in the lesions). The treatment consists of long term anti tubercular therapy.

Case Study

Case 1

A 12 year old adolescent boy presented with a painless swelling over the dorsum of his left foot. 2 weeks later he developed a painful swelling on the dorsum of his left hand which progressively increased in size over the next four months. He consulted a general practitioner and was prescribed short course of oral antibiotics and analgesics however since the symptoms didn't resolve and the swelling was progressively increasing in size he was brought to our pediatric outpatient department. On probing the history there was a contact with open case pulmonary tuberculosis which was his paternal grandfather who was taking ATT for past 4 months. However the child had no history of fever, cough, or loss of weight. There was no history of trauma. On examination he had an oval swelling about 3 cm × 2 cm over the left first metatarsal bone with a discharging sinus and another swelling over the left third metacarpal bone 2 cm × 1 cm. The swellings were non-tender however movements were restricted at the corresponding inter phalangeal joints. The systemic examination was within normal limits. Investigations revealed normal total leucocyte count and elevated ESR, mantoux test strongly positive (20 mm) chest X-rays was unremarkable, open biopsy of the swellings were done and histopathology revealed caeseating granulomas suggestive of tubercular infection. Radiograph of the involved foot and hand showed diffuse thickening of the metatarsal/metacarpal bone with subpersiosteal new bone formation. Based on all these features a diagnosis of spina ventosa-tuberculous dactylitis was made, and the child was initiated on appropriate Anti-Tuberculosis Therapy (ATT). The child is responding well to treatment and is on periodic follow up. There is a substantial reduction in the size of the swelling and the improvement in range of movements.

Case 2

A five year old girl child was brought with complaints of swelling over the right index finger noticed for the past two to three months. Following unsuccessful treatment, with several repeated courses of antibiotics for a possible osteomyelitis, the child was referred to our tertiary care hospital for further management. The swelling was initially small but progressively increased in size. There was no history of trauma. There was history of low grade fever on and off with occasional dull aching pain. There was no history of loss of weight or appetite. However there was history of exposure to pulmonary tuberculosis. On examination there was a non-tender fusiform firm swelling 3 cm × 2 cm over the proximal phalanx of right

index finger. A provisional diagnosis of tuberculous dactylitis was made with chronic pyogenic osteomyelitis being the closest differential diagnosis. Investigations revealed leucocytosis with lymphocytic predominance, an elevated ESR. Radiograph of the right hand revealed a cystic swelling involving the proximal phalanx 9f the right index finger with a thinned out periosteum. Mantoux test was positive, however chest X-ray was unremarkable. Excision biopsy was performed and the specimen was subjected to histopathological and bacteriological examination which revealed caseating granulomas and acid fast bacilli respectively thus confirming the diagnosis of spina ventosa. The child was started on appropriate ATT regimen following which there was good clinical improvement and the child is doing well on follow-up.

Case 3

A four year old male child was referred to us with progressively enlarging swelling of the right hand noticed for the past two months associated with fever, cough, and loss of appetite. There was no history of exposure to pulmonary tuberculosis. On examination the child was malnourished, afebrile; with 4 cm × 3 cm painless firm swelling over right fourth metacarpal bone.

The systemic examination was otherwise unremarkable. Investigations revealed leucocytosis with polymorphonuclear predominance elevated crp and esr. Xray of the right hand revealed a cystic swelling involving the right fourth metacarpal with periosteal thinning.Chest xray revealed normal lung fields. Mantoyx was positive serology for HIV was negative. Incisional bone biopsy was performed. Biopsy specimen showed caseating granulomas on histopathology and AFB on bacteriological examination. A diagnosis of tuberculous dactylitis was made and the child was started on ATT following which his clinical condition improved (Table 1).

Parameter	Case 1	Case 2	Case 3
Age in years	12	5	4
Sex	Male	Female	Male
Site involved	Left first metatarsal and left third metacarpal	Proximal phalanx of right index finger	Right fourth metacarpal
SLab parameters			
Total leucocyte count	Increased 14500	Increased 13700	Increased 14200
Differential count	Lymphocytosis	Lymphocytosis	Lymphocytosis
ESR/CRP	Increased	Increased	Increased
Mantoux	Positive 14 mm	Positive 16 mm	Positive 12 mm
Chest X-ray	Normal	Normal	Normal
Histopathology	Caseating granulomas	Caseating granulomas	Caseating granulomas
Bacteriology	AFB positive	AFB Positive	AFB positive

Table 1. The findings of the three cases are summarised in the following table.

Discussion

Even in a country like India where burden of tuberculosis is high, such rare manifestations of tuberculosis like spina ventosa can be missed unless a high index of suspicion is maintained [3,4].

Chronic pyogenic osteomyelitis can be a close mimicker and needs to be differentiated. In case 1, the presence of simultaneous swelling in both the hand and foot without attendant symptom did pose a diagnostic challenge. But a carefully elicited history revealing the contact of the child with a known pulmonary tuberculosis patient helped us pin the diagnosis in this child. Thus this highlights the importance of a well taken history and through physical examination even in this era of modern diagnosis. Histopathology and bacteriological confirmation is utmost essential to differentiate from other pathologies of short tubular bones like benign or malignant tumors, pyogenic osteomyelitis, sickle cell dactylitis etc. which can mimic tuberculous dactylitis [5-7]. The lesion is usually pauci bacillary and there may not be concomitant pulmonary involvement as we can see in our patients [8-10].

Conclusion

This case is being presented for the rarity of its occurrence. As clinicians we must remember that tuberculosis can present in the most unusual forms and in the sites that we can least expect to manifest. Early diagnosis and early institution of appropriate treatment (ATT) can relieve the patients of their symptoms and help them lead a disease free life.

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