4 years. Similar changes were noticed in tarsal and metatarsal bones. Diaphysis appeared to be smaller with very thin cortex (*Fig.*).

Significant positive investigations were ESR-35 mm, alkaline phosphatase 2 KA units, serum calcium 14 mg/dl; serum phosphatase 1.8 mg/dl; and urinary phosphoethanolamine concentration 150 micro ml.

Discussion

Hypophosphatasia is an extremely rare disease. This case appeared to be one of rickets with myopathy but radiological and biochemical findings were characteristic of hypophosphatasia. There are several case reports of hypophosphatasia in world literature(1-4). Seshia *et al.* observed association of nonprogressive myopathy in his two cases of hypophosphatasia as an important early sign(4). In our case there was weakness of muscles of lower limbs and the child was not able to stand by 2 years of age and started crawling and walking with support at age of 4 years (nonprogressive myopathy). They observed that this clinical feature resembles that of osteomalacia-myopathy described by Dastoor *et al.*(5) and concluded that there is no clear explanation for myopathy in few cases of hypophosphatasia. Wolfish *et al.*(2) also observed association of hyperparathyroidism and hypophosphatasia in a few cases. In our case there was no endocrinal disturbance.

We are presenting this case because of its association with nonprogressive myopathy and its extreme rarity in our subcontinent where rickets is common. This case was treated for rickets for 2 years without any response. Vitamin D therapy may have deleterious effects on hypophoshatasia(2). Seshia *et al.*(4) concluded that non-progressive myopathy with osteomalacia syndromes in children may be an important early sign of hypophosphatasia.

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Allergic Bronchopulmonary Aspergillosis

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Aspergillus species are ubiquitous in nature; with large numbers of conidia or spores dispersed in the air inhalation is inescapable. Allergic broncho-pulmonary aspergillosis (ABPA) is one of the hypersensitivity lung disorders caused by *Aspergillus fumigatus* (Af).

Following the first description of three

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cases of ABPA from England in 1952 by Hinson's group(1), there was a spate of papers from England and then other countries including India, describing various aspects of this entity(2-5). However, very few cases of ABPA have been reported in the pediatric age group. Till 1959, the youngest case in world literature was a 15½ years old boy(6). From USA, the youngest patient of ABPA was a 9 year girl reported in 1970(7). The first Indian report of ABPA in 1971(3) described three cases, the youngest of them was 14½ years of age. Later, in two other well-documented Indian series of 46(4) and 50(5) cases of ABPA, the youngest cases were 14 and 13-year-old boys, respectively.

ABPA diagnosed in a 10-year-old child is being reported as the youngest case from India so far.

Case Report

In January 1989, a 10-year-old boy presented with history of intermittent fever, wheezing and cough with mucopurulent expectoration for two weeks. He had been suffering from recurrent attacks of cough and wheezing since birth, and also fever and mucopurulent sputum off and on for the past 2 years. His parents had noticed that he expectorated brownish plugs occasionally. His father and maternal grandfather were asthmatic. His chest *X*-rays done on a number of occasions previously had been essentially normal except during the previous six months, when a large non-segmental homogenous opacity was seen in the right lower zone. He was being managed for bronchial asthma with secondary infection, getting relief with bronchodilators, prednisolone and amoxycillin. Two months prior, he had also received diethylcarbamazine for two weeks for a right lower zone opacity accompanied by high peripheral blood eosinophilia (28%) but without relief.

On physical examination, this moderately built child had a heart rate of 108/min, a respiratory rate of 32/min with dyspnea, low grade fever (38.2°C) and mild anemia. There was no cyanosis, clubbing, lymphadenopathy or other abnormality on general examination and no hepatosplenomegaly. Chest auscultation revealed bilateral rhonchi and bronchial breathing with few crepitations in the left infraclavicular region.

His hematological findings were Hb 10.6 g/dl, total leucocyte count 11,200/mm^3 with 50% neutrophils, 23% lymphocytes, 2% monocytes and 25% eosinophils, absolute eosinophil count (AEC) 2800/mm^3; ESR 46 mm/first h (Westergren). Peripheral blood film was negative for malarial parasite and immature cells. Mantoux test with PPD 1 TU was negative. Urine and stool examination were normal. Sputum smear for acid fast bacilli was repeatedly negative and culture for pyogenic organisms revealed no growth.

His chest skiagram (*Fig. 1*) showed a large homogenous opacity involving the upper half of the left lung field. There was no radiological or clinical improvement either with a 10 days course of ciprofloxacin or when diethylcarbamazine was given earlier for two weeks. To exclude any helminthic infestation responsible for eosinophilia, he also received albendazole for three days, but no change in AEC was observed.

Intradermal test with 0.02 ml of 1:500 Af antigen (procured from CSIR Center for Biochemicals, Delhi) gave strongly positive type I and type III hypersensitivity reactions. Of the five sputum samples cultured for fungi, growth of AF was obtained in

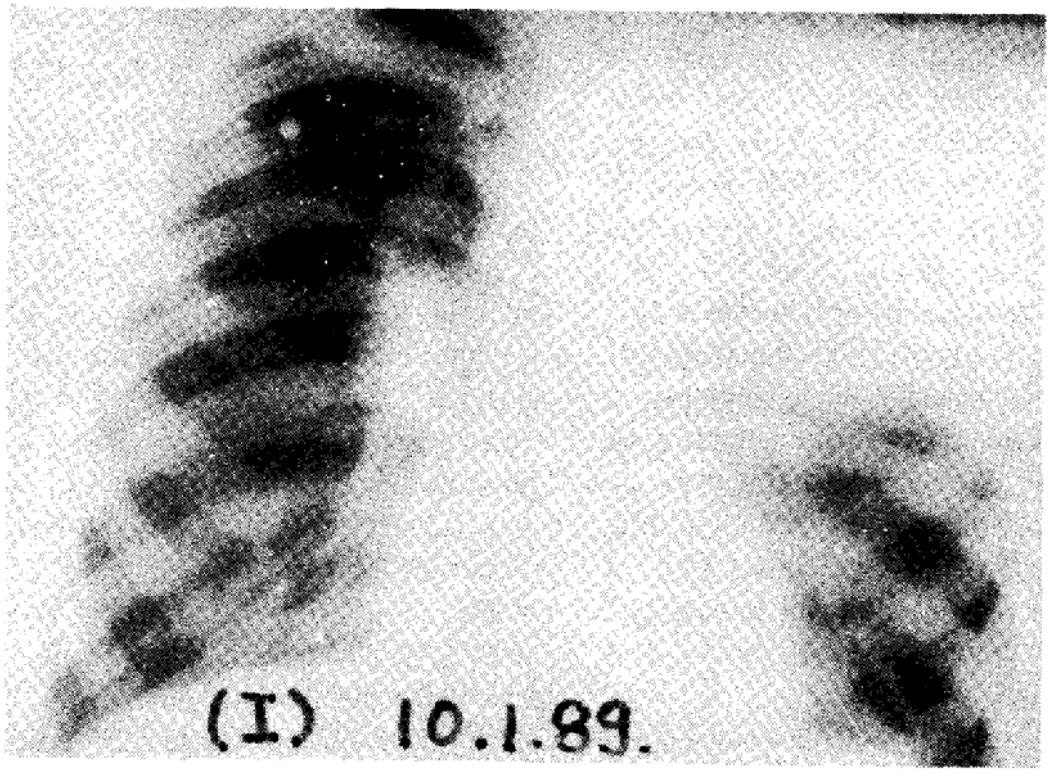

Fig. 1. Chest radiograph showing left upper lobe opacity (10.1.1989).

two. The patient's serum was positive for precipitins against Af. Casoni's test was negative.

On the basis of these reports, he was diagnosed as a case of ABPA and put on 25 mg of prednisolone given daily in a single early morning dose (1.0 mg/kg). There was remarkable clinical and radiological improvement. A repeat skiagram after 4 weeks (*Fig. 2*) showed complete clearance of the left sided opacity. His AEC came down to 360/mm^3.

Prednisolone was gradually reduced to 10 mg daily in the next 8 weeks and then to 5 mg every alternate day to complete a total duration of 6 months. The child remained symptom free and was being managed with bronchodilators alone. However, in September 1989, there was again clinical and radiological worsening with the appearance of a small opacity in the left upper zone and a few small patchy lesions in the right upper zone. These cleared with a repeat course of prednisolone for two weeks. In January, 1990 again fresh non-segmental opacities appeared in right apical and both basal regions, which cleared within two weeks with prednisolone.

For the last 11 months the child is clinically stable off steroids, being managed with oral bronchodilators alone. His chest *X*-rays and blood counts are also normal.

Discussion

This child appears to be the youngest case of ABPA reported from India so far. The diagnosis of ABPA was based on the following criteria (*a*) history of asthma; (*b*) family history of atopy; (*c*) blood eosinophilia; (*d*) shifting pulmonary infiltrates in serial skiagrams; (*e*) type I and type III cutaneous reactivity of Af antigen; (*f*) precipitating antibodies to Af; and (*g*) positive

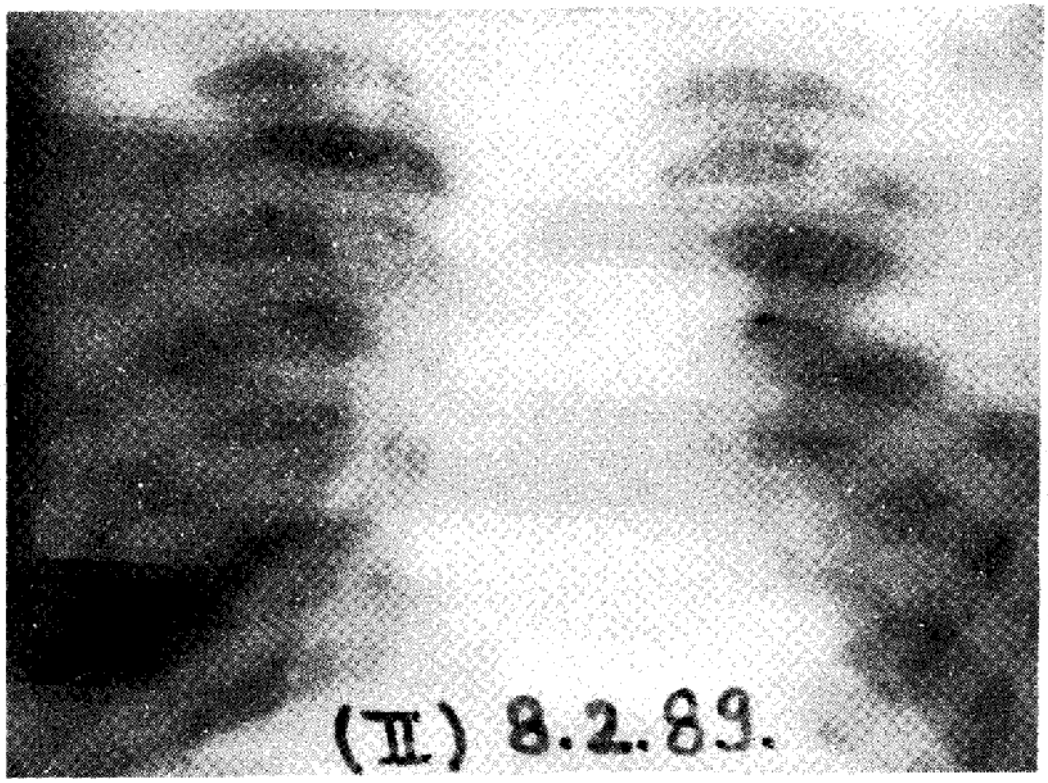

Fig. 2. Complete clearance after four weeks of steroid therapy (8.2.1989).

results of sputum culture of Af. Total serum IgE and Af-specific IgE levels could not be estimated due to lack of facilities.

McCarthy and Pepys(2) have commented on the importance of age of onset of asthma prior to the age of onset of ABPA. According to them the time gap between the onset of asthma and of ABPA was much longer in those patients where asthma started at a younger age. Of 111 patients, 63 had onset of asthma before the age of 10 years and developed ABPA after an average gap of 24 years; whereas in 28 cases, where asthma started after 31 years of age, ABPA developed within 3.5 years. In the present case, ABPA developed within 10 years of onset of asthma.

A large number of conditions like bacterial pneumonia, tuberculosis, pulmonary eosinophilia, *etc.* require differentiation radiologically. At present, prednisolone is the drug of choice in the therapy of ABPA; the dosage schedule has to be individualized, depending upon the extent and stage of the disease(8).

Even in the pediatric age group, ABPA may not be that uncommon as is usually considered. Inappropriate use of steroids in asthmatics may delay diagnosis of ABPA. A high index of suspicion is required, as early diagnosis and institution of proper therapy can alter the course of the disease, decrease morbidity and may prevent irreversible lung damage.

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Ocular Myiasis

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Myiasis is essentially a disease of the tropics like India. It is most commonly caused by a fly *Chrysomyia bazzina* whose larvae are creamy white in color(1), the other flies which may be responsible include–*Oestrus ovis, Hypoderma bovis, Wohlfahrtia magnifica, Cochliomyia hominivorax, Dermatobia hominis(2), Rhino-estrusbovis, Hypoderma lineata* and *Gastrophilus intestinalis(3)*. This had predilection for the nose, ear and the trachea in that order. Other sites including the eye are rarely affected(4). We report a case of maggots affecting the upper eye lid.

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Case Report

A 1½-year-old female child belonging to a poor farmer's family presented in the month of October with infected wound and maggots on the right upper lid. Child had a small boil and fever of moderate grade 20 days back. The parents, however, took no treatment. Three days prior to admission the maggots made their appearance. On clinical examination there was marked edema and a small ulcer was seen over the upper eye lid. Creamy white maggots were seen crawling in the ulcer area. The skin around it was red, swollen and tender. The palpebral fissure was reduced but the eye movements were normal. *X*-ray orbit revealed no abnormality. The child was put on amoxicillin and ibuprofen. Twenty five maggots were manually removed with the help of an aural forceps spread over 3 sittings. The wound healed in 10 days. At subsequent follow up one month later, the child was absolutely normal.

Discussion

Myiasis has a seasonal variation, the peak being from September to November(1) as in the present case. All cases come from the low socio-economic class of society with grossly unhygienic living conditions. Nearly 85% of them belong to the