

## Simultaneous Occurrence of Papulonecrotic Tuberculid and Erythema Induratum in a Child: A Case Report

Ezomike Nkeiruka Elsie<sup>1,2\*</sup>, Machona Sharon Musonda<sup>3,4</sup>, Hlela Carol<sup>1,3</sup>

<sup>1</sup>Division of Dermatology, Red Cross Children's Hospital and University of Cape Town, Cape Town, South Africa; <sup>2</sup>Department of Paediatrics, University of Nigeria Teaching Hospital, Ituku-Ozalla, Enugu, Nigeria; <sup>3</sup>Division of Dermatology, Groote Schuur Hospital and University of Cape Town, Cape Town, South Africa; <sup>4</sup>Dermatology and Venereology Section, University Teaching Hospital, University of Zambia School of Medicine, Lusaka Zambia

### ABSTRACT

Hypersensitivity reactions to Mycobacterium tuberculosis (tuberculids) include Papulonecrotic Tuberculid (PNT), Erythema Induratum (EI), and Lichen Scrofulosorum. These rarely coexist in a child. We, therefore, report the coexistence of papulonecrotic tuberculid and erythema induratum in a four-year-old male.

He presented with a two-week history of skin eruptions involving the arm, abdomen, and ears. Reddish bumps later appeared on the legs. Examination revealed erythematous papules, with central necrosis, on the lateral aspect of the left upper arm, ear helices, and trunk. There were also several symmetrically distributed non-tender erythematous nodules, some with necrosis, on the shins, and calves.

Histology of the biopsied papules (PNT) revealed intense wedge-shaped necrosis with perivascular inflammation and of the leg nodule (EI), extensive dermal fat necrosis with granulomatous lymphocytic infiltration. These findings are compatible with TB.

Chest X-ray revealed hilar and paratracheal adenopathy. Sputum was positive for M. tuberculosis DNA using Polymerase Chain Reaction (PCR). He did well on anti-tuberculosis medications. Erythema Induratum (EI), and L. Scrofulosorum. These rarely coexist in a child. We, therefore, report the coexistence of papulonecrotic tuberculid and Erythema Induratum (EI) in a four-year-old male.

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**Keywords:** Papulonecrotic tuberculid; Erythema induratum; Child; Tuberculosis

**Correspondence to:** Ezomike Nkeiruka Elsie, Division of Dermatology, Red Cross Children's Hospital and University of Cape Town, Cape Town, South Africa, E-mail: nkeirukaeszomike@gmail.com

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## INTRODUCTION

Tuberculosis is a global public health concern especially in developing countries. The World Health Organisation (WHO) estimates noted that South Africa is one of the countries with the highest burden of tuberculosis (TB) in the world with an incidence of about 520 cases of active TB per 100,000 population [1]. Furthermore, children are at higher risk of progressing from TB infection to TB disease but TB of the skin (tuberculids) remains rare [2]. Nevertheless, tuberculids consist of about 1 to 2% of extrapulmonary TB manifestations [2]. Tuberculids occur due to intense necrotising reaction caused by a hypersensitivity reaction to haematogenous spread of *M. tuberculosis* or its antigens. It can be papular (papulonecrotic tuberculid), nodular (erythema induratum), micropapular (*L. scrofulosorum*) and atypical nodular tuberculosis [2,3]. Papulonecrotic tuberculid occurs predominantly in young adults and clinically manifests as a chronic recurrent asymptomatic symmetrical necrotizing papules which appear in crops on the extensor surfaces of extremities, trunk, and buttocks, that heal subsequently with atrophic varioliform scarring [3]. Erythema induratum of Bazin most often occurs in adult women and is characterized by erythematous, tender, variably ulcerated subcutaneous nodules, classically arising on the posterior or lateral lower legs [3]. Synchronous manifestations of these tuberculids in an under-five as well as the distribution of the EI lesions on the pinna and abdomen have not been reported in the literature. Previously published studies [4,5] among the South African population have noted the coexistence of PNT and EI in older children and adults but not in a child aged below five years. Here, we report a case of a 4-year-old boy with coexisting clinical features of papulonecrotic tuberculid and erythema induratum accompanying pulmonary TB.

## CASE REPORT

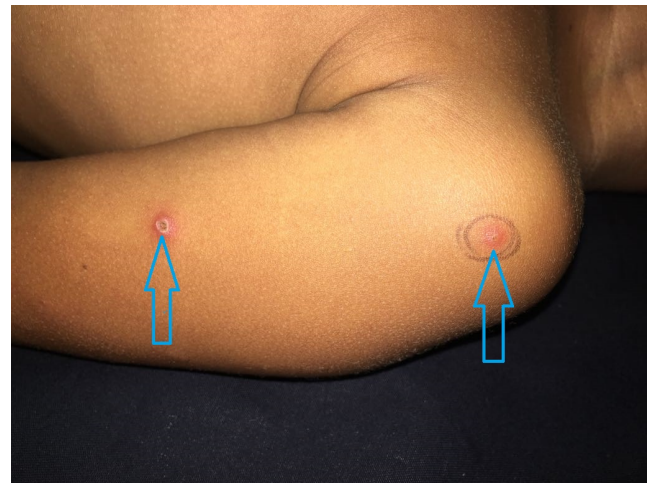
A 4-year-old South African boy was referred to our dermatology clinic for evaluation of skin eruptions on the left upper arm, abdomen, ear helices, and lower limbs of two weeks duration. The eruptions were tender from onset initially on the left arm but gradually spread to the abdomen, ear helices. Then, multiple palpable reddish bumps appeared on the lower limbs. Also, the patient had a cough, loss of appetite, nausea and malaise.

The patient was not on any medication at the time of the presentation. No history of trauma to any other part of his body. The patient's aunt living with the family for 2 years has been diagnosed and on treatment for pulmonary TB.

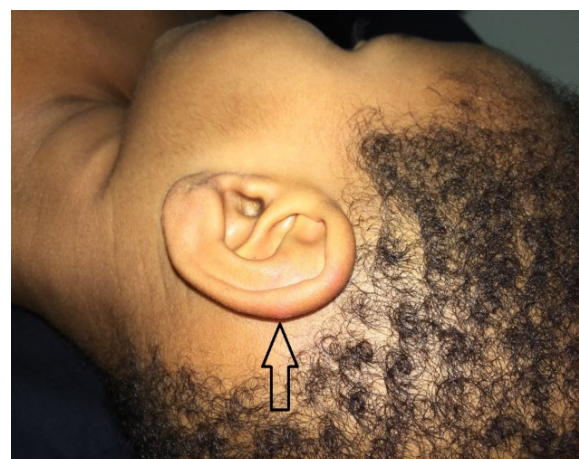
On physical examination, the patient was alert, mildly pale. There was no fever or lymphadenopathy and vital signs were within normal limits. The upper part of the left-arm revealed a BCG scar. However, no significant finding was noted on respiratory and other systemic examinations. The patient's weight was between 50th and 75th centile for age and sex.

Dermatologic examination revealed two distinct types of skin lesions. The first type consisted of few erythematous papules with central necrosis on the lateral aspect of the left upper arm and trunk Figure 1. The second type of lesion consisted of

several symmetrically distributed erythematous nodules about 2 cm-3 cm in diameter, non-tender, on the anterior aspects of the lower legs and calves. Some of these lesions have necrotic centre and have healed with atrophic scars as shown in Figure 2.

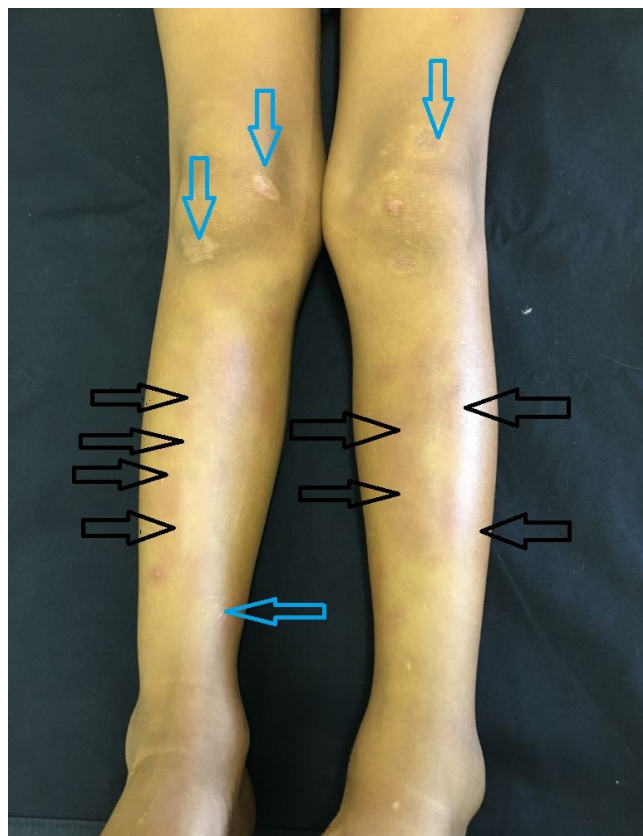


**Figure 1:** Left upper arm of the patient showing erythematous papules with central necrosis (blue arrows).



**Figure 2:** Ear helices of the patient showing erythematous papules with central necrosis (black arrows).

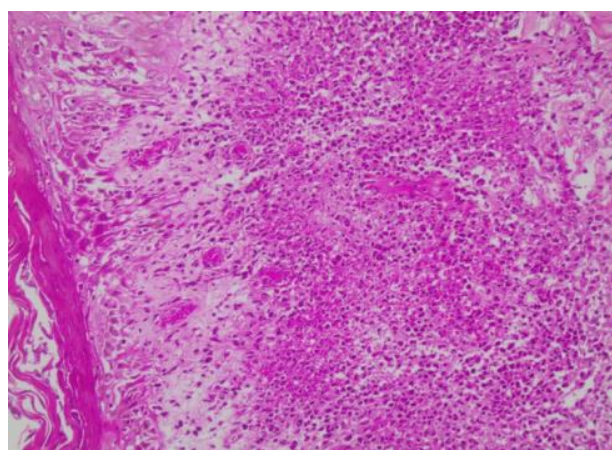
The second type of lesion consisted of several symmetrically distributed erythematous nodules about 2 cm-3 cm in diameter, non-tender, on the anterior aspects of the lower legs and calves. Some of these lesions have necrotic centre and have healed with atrophic scars as shown in Figure 3 below.



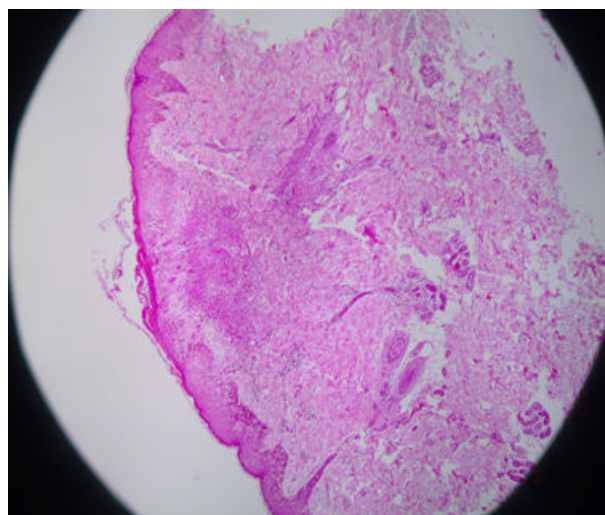
**Figure 3:** Several erythematous nodules symmetrically distributed on the anterior aspects of the legs (black arrows) with some healed atrophic scars (blue arrows).

A biopsy specimen from the papulonecrotic lesions on the left arm revealed a wedge-shaped area of necrotizing inflammation associated with vasculitis in the dermis (Figure 4A and B).

(A)

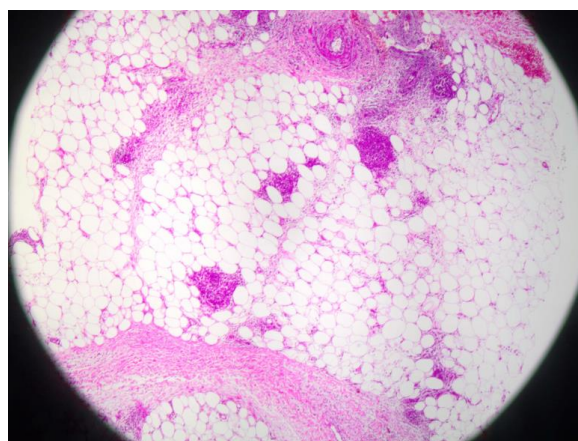


(B)



**Figure 4:** (A) Extensive necrosis with perivascular inflammatory cell infiltrations in the subcutaneous tissue (Haematoxylin and eosin: x40); (B) Wedge-shaped dermal necrosis (black arrow) surrounded by granulomatous inflammation extending into the subcutis (H & E; x15).

Another biopsy specimen from the nodular lesions on the lower limbs showed extensive dermal and fat necrosis with granulomatous lymphocytic infiltration consistent with erythema induratum as shown in Figure 5.



**Figure 5:** Focal fat necrosis, perivascular mononuclear cell infiltration and haemorrhage in the dermis.

His laboratory studies showed haemoglobin level of 9.4g/dl, total white blood count of  $13 \times 10^9$  cells/l, erythrocyte sedimentation rate was 85 mm/hr, HIV test was negative. The chest radiograph revealed right perihilar lymphadenopathy suggestive of pulmonary TB. The Ziehl-Neelsen staining was negative and AFB culture of the skin biopsy did not yield the bacilli. Tuberculin test was positive. The PCR assay for *M. tuberculosis* from the sputum was positive.

The positive findings from the clinical, histopathologic, laboratory, and radiologic were supportive of the diagnosis of papulonecrotic tuberculids and erythema induratum. However, the anteriorly located lower limb lesions of erythema induratum were unusual.

*Mycobacterium tuberculosis* was found to be Isoniazid resistant therefore, he was placed on Rifampicin and Pyrazinamide at

doses of 15 mg/kg (240 mg/day) and 15 mg/kg (240 mg/day) respectively. On the first visit six weeks later, the lesions improved completely, with few atrophic scars.

## DISCUSSION

Although papulonecrotic tuberculid is known to be a rare, chronic, recurrent, and symmetric eruption of necrotizing skin papules arising in crops, involving primarily the buttocks and extensor surfaces of the arms and legs [3] our patient had atypical features. The distribution of the papules was asymmetrical involving only the left upper arm and the trunk. This may be attributed to the patient's presentation to the clinic when the onset of the disease is early compared to that obtainable in patients in the previously published literature. Kumar B, et al. [6] the duration between the onset of lesions and detection in the hospital has widely ranged from 2 months to up to 10 years. The possibility of dissemination of the disease with a high carrier rate is likely in these children.

The presence of erythematous nodules with the necrotic centre and some atrophic scars on the anterior aspect of the lower limbs was uncommon though the histologic finding was supportive of EI.

The coexistence of PNT and EI is very rare and only eleven cases have been reported in the english literature [2]. The patients involved were adults and adolescents. Among these reports, three were South Africans, the earliest was documented two decades ago. Our patient is the first report of an under-five with this entity in South Africa. This low reports in the literature could be explained by the rising prevalence of TB among this age group with poor immunity whose immune response is usually impaired. Therefore, the clinical manifestation may be different and atypical with difficult identification on dermatological examination. This study highlights the fact that this entity can easily be unidentified or misdiagnosed in younger age groups. The clinical data are summarised in Table 1 [5-11].

**Table 1:** Previous report of patients with concurrent Occurrence of Papulonecrotic Tuberculid (PNT) and Erythema Induratum (EI) (Adopted and Modified [11-15]).

Patient	Year/country (ethnic origin)	Age (years)/sex	PNT distribution	EI distribution	Focus of TB Infection	Medication/ period	Outcome
1	1985/ Japan	43/F	Upper and Lower limbs	Lower limbs	Cervical and axillary lymph nodes, lung calcifications	PNT: H, S/40 days EI: H, R, S/30days	Improvement in PNT but relapse in EI
2	1986/England	11/F	Hands and Feet	Calves	Negative finding	Combination	Abrupt resolution
3	1989/Mexico	61/F	Lower limbs	Lower limbs	Peritoneum	Not available	Not available
4	1910/U.K	61/F	Feet	Lower limbs	Calcifications on the lung apex	H, E, R/4 weeks then H, R/8 months	Resolved completely
5	1993/Germany	59/F	Face, forearms, buttocks, thighs, calves, feet	Posterior aspect of Lower limbs	Supraclavicular, hilar, axillary, abdominal lymph nodes	Not available	Abrupt resolution
6	1994/U.K	31/F	Forearms, hands, lower limbs, feet, soles	Posterior aspects of calves	Paratracheal lymph nodes	H, R, P/6 months	Invariable resolution
7	1994/South Africa	28/F	Upper and lower limbs	Lower limbs	Inguinal lymph nodes with overlying Scrofuloderma	H, E, R/6 months	Resolved completely without recurrence
8	1994/South Africa	25/F	Upper and lower limbs	Lower limbs	Negative finding	H, E, R/6 months	Resolved completely without recurrence

9	1994/South Africa	17/M	Face, trunk, upper and lower limbs	Lower limbs	Abnormal CXR findings	H, E, R/6 months	Resolved completely without recurrence
10	1995/Taiwan	32/M	Chest, back, abdomen, limbs, and soles	Buttock, calves, and feet	Negative finding	H, E, R/9 months	Resolved completely
11	2012/Korea	12/F	Limbs and buttocks	Lower limbs	Upper lobe TB	H, E, R, Z/2 months then R/4months	Resolved completely
Present case	2018/South Africa	4/M	Abdomen, ear helices and upper limb	Anterior aspects of lower limbs	Hilar lymph nodes	R, Z/6 weeks	Improved

There are reports in the literature of identification of the causal organism by polymerase chain reaction and resolution of the lesions following antituberculous treatment [12,15]. The presence of a strongly positive Mantoux test, the clinicopathological findings which regressed following antituberculous treatment, were all in support of the diagnosis of PNT in our patient.

The manifestation of cutaneous TB in children has been documented to be atypical and different from those seen in adults [2]. There is a paucity of data on the distributions seen in children. Our 4-year-old patient is the first in English literature with coexisting distinct PNT and atypical presentation of EI lesions. The lesions were localized on the anterior aspects of the lower limbs unlike the typical distribution usually seen on the posterior aspects. We attribute this finding to the possibly impaired immune response in this age group coupled with the high endemic nature of TB in South Africa.

However, the absence of detection of underlying tuberculous foci at the onset of tuberculids has also been reported [2,8,12]. The frequency of locating extracutaneous TB foci in the presence PNT varies from 26% to 67%, [12] therefore, a detailed examination is needed especially of the lymph nodes, lungs, and bones. In our patient the physical examination findings were not revealing but chest radiograph showed abnormalities suggestive of pulmonary TB. This is similar to findings in most studies [2,8,12].

Isolation of tuberculous bacilli from PNT lesion is difficult with consistent negative AFB culture results [8,10] but mycobacterial DNA is demonstrable in both PNT and EI skin lesions [2,11]. Similarly, this was obtainable in our patient. This may be due to low numbers of bacilli, which the local delayed hypersensitivity reaction rapidly destroys [2,8]. The results of tuberculin test in almost all the reported cases in Table 1 were strongly positive as seen also in our patient.

Chuang YH [11] the coexistence of PNT and EI in a patient may represent a morphological continuum and different levels of immune-complex mediated vasculitis. The PNT lesions may involve the smaller, more superficial vessels while for EI, the larger, deeper, subcutaneous vessels [2].

## CONCLUSION

In conclusion, based on the present case and previous reports PNT and EI lesions can coexist with distinct uncommon clinical findings and may affect the entire age spectrum in children even without underlying TB infection especially among people in high TB endemic areas. Therefore, a high index of suspicion is needed for early diagnosis.

Finally, irrespective of the patient's age and clinical presentation, with sufficient circumstantial evidence, tuberculin testing, and PCR for mycobacterial DNA on the lesional skin make the diagnosis more confirmatory. This subsequently, enhances prompt antituberculous treatment which can prevent the progression of the TB disease morbidity and mortality. Certainly, with a timely diagnosis and correct treatment, most people who develop TB disease can be cured.

## CONSENT

Consent was obtained from the mother to use the patient's photographs for publication in research and teaching.

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## CREDIT AUTHOR STATEMENTS

Ezomike Nkeiruka, Machona Musonda, and Hlela Carol: Conceptualization, methodology and investigation.

Ezomike Nkeiruka and Hlela Carol: writing, reviewing, and editing.

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