Sir,

An 8-year-old boy presented to us with a raised asymptomatic cutaneous lesion over his left leg for the past one and half years. It started as a bluish, slightly elevated spot which gradually increased to attain its present size and shape. There was no preceding history of trauma or use of any medication. There was no overt history of external bleeding or oozing. Cutaneous examination revealed an ill-defined, oblong-shaped plaque (4 cm × 3 cm size) with a bluish hue situated over the anterior aspect of the left thigh. It was studded with a few dark-blue and reddish papules. Focal crusting over the surface was also noted [Figure 1]. The lesion was non-tender and soft in consistency, and there was no lesional thrill or bruit. No other cutaneous lesion was present elsewhere in his body. Hair, nail, and mucosae were normal. General and systemic examinations were noncontributory. Routine blood investigations were within normal limits. Assuming the case to be a vascular malformation, the lesion was completely excised a year back, and regular follow-up showed no recurrence until date. Histopathological examination (hematoxylin and eosin stain) [Figures 2-4] of the excised specimen revealed compact hyperkeratosis with irregular acanthosis. Dilated thin-walled spaces lined by intermittent flat endothelial cells resembling lymphatic channels were seen in the upper and mid-dermis. Similar, but more slit-like, structures were seen in the deeper dermis along with dilated blood vessels containing red blood cells.

The presence of a continuous space lined by endothelium containing hemosiderin on one side and eosinophilic material on another in the upper dermis was a striking feature. There was almost no inflammatory infiltrate. Hemosiderin was seen scattered within a somewhat disorganized collagen in the rest of the dermis. Based on the clinical and histopathological findings, a diagnosis of benign lymphangioendothelioma (BLAE) was made.

BLAE also known as acquired progressive lymphangiomma is a locally infiltrative vascular proliferative lesion of lymphatic origin. In fact, it is a complex vascular hamartoma with three components, including lymphatic vessels, blood vessels, and smooth muscle.\(^1\) The condition was originally described by Jones et al. in 1964. It is characterized by slowly enlarging erythematous to brownish patch or plaque.\(^2\) Lesions are usually asymptomatic, but occasionally present with pain, itching, and stinging sensation.\(^2\) Most common sites of involvement are lower limb and head and neck region followed by the wrist, shoulder, upper limb, trunk, and mucosa. Both genders are equally affected.\(^2,3\) Trauma, surgery, radiation therapy, femoral arteriography, tick bites, and hormonal stimulus may act as precipitating factors for the development of BLAE.\(^1,2\) Most of the cases have been reported in adults.\(^3\) In a large series, Messeguer et al. found the average age of presentation was 43 years.\(^2\) However, children may rarely be affected\(^4\) as in the present case. Histopathologically,
Correspondences

BLAE is characterized by thin-walled interconnecting vascular channels with dilated vascular spaces in the upper dermis and smaller and irregular vessels in the lower dermis.

Vessels are lined by endothelial cells, and they are interspersed between strands of collagen. Cellular atypia or mitotic figures are usually absent. The endothelial cells stain positively for lymphatic markers such as podoplanin (D2-40), LYVE-1 and variably positive for factor VIII, Ulex europaeus–I lectin, CD31, and CD34. Although we were unable to perform these tests due to institutional unavailability, the clinical presentations along with the HPE findings were characteristic in our case. BLAE should be differentiated from Kaposi’s sarcoma, angiosarcoma, and targetoid hemosiderotic hemangioma both clinically and histopathologically. The absence of promontory sign, atypical cells, mitotic figure, inflammatory infiltrate, extravasated erythrocytes helps to differentiate BLAE from Kaposi’s sarcoma and angiosarcoma as in our case. Targetoid hemosiderotic hemangioma presents as central violaceous papule with surrounding pallor and ecchymosis. Histopathological examination shows vascular channels lined by hobnail endothelial cells in some cases with a variable amount of inflammatory infiltrate and extravasations of erythrocyte and hemosiderin around vessels. Old lesions show collapsed thin-walled anastomosing vascular channel with hemosiderin. These features were absent in the present case. Complete surgical excision is the treatment of choice. We report this case for the rarity of the condition. Furthermore, the condition is still rarer in children. In spite of best of our effort we could find only one previous case report of cutaneous BLAE in Indian population. At the same time, we would like to stress on the importance of histopathological examination in the accurate diagnosis of such vascular lesions.

Financial support and sponsorship
 Nil.

Conflicts of interest
 There are no conflicts of interest.

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How to cite this article: Rudra O, Ghosh A, Ghosh SK, Bhunia D, Mandal P. Benign lymphangioendothelioma: A report of a rare vascular hamartoma in a young Indian child. Indian J Dermatol 2017;62:528-9.

Received: July, 2016. Accepted: August, 2017.