Disseminated verrucous hemangioma with subcutaneous hemangioma: a rare association

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Verrucous hemangioma is a rare, localized vascular malformation. The lesions are bluish-red, well demarcated, and compressible. We report a case of a 10-year-old girl with coexistent disseminated verrucous hemangioma and subcutaneous hemangioma over the nape of neck.

Keywords: Disseminated verrucous hemangioma, subcutaneous hemangioma, vascular malformation

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INTRODUCTION

Verrucous hemangioma is a rare, localized vascular malformation [1]. The lesions are bluish-red, well demarcated, and compressible. In time, verrucous hemangiomas gradually enlarge and satellite nodules may arise. The older lesions of verrucous hemangioma acquire a warty surface as a protective phenomena [2]. Herein, we report a case of a girl with coexistent disseminated verrucous hemangioma and subcutaneous hemangioma over the nape of neck.

CASE REPORT

A 10-year-old female child presented with multiple verrucous lesions over various parts of the body and a solitary subcutaneous swelling over the nape of the neck since she was 3 years of age (Figure 1). There was occasional bleeding from the verrucous lesions on trauma but there was no history of gastrointestinal (GI) bleeding or any purpuric lesions over her body.

Cutaneous examination revealed a 4×4 cm soft,

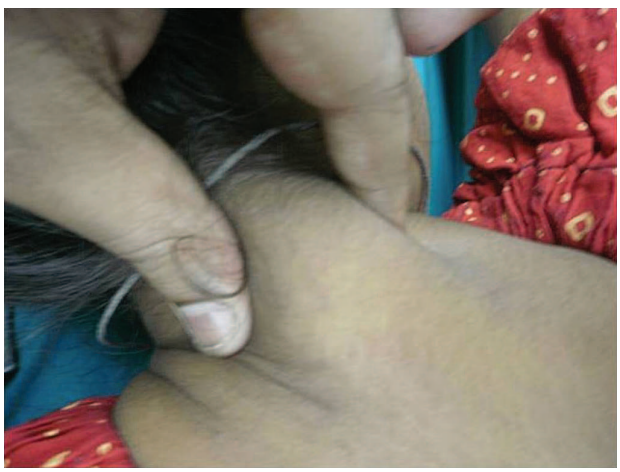

Figure 1. Subcutaneous hemangioma over the nape of the neck.

nontender swelling over the nape of the neck with no overlying skin changes and without any bony or skin attachment. Multiple pigmented, nontender, noncompressible verrucous nodules were present over left palm, dorsum of left hand (Figure 2), left thigh (Figure 3), abdomen and lower extremities. Systemic examination did not reveal any abnormality.

Routine hematological and urine examinations were within normal limits and stool was negative for occult blood. Abdominal screening for hepatic hemangioma was negative.

Histopathologic examination from a verrucous nodule over the dorsum of the hand revealed hyperkeratosis, acanthosis, and elongated rete ridges. Multiple widely dilated thick walled blood vessels containing RBCs were found both in the upper as well as the lower dermis (Figure 4). Upper GI endoscopy revealed no abnormality. Ultrasonography (USG) of lesion over the nape of the neck was suggestive of subcutaneous hemangioma.

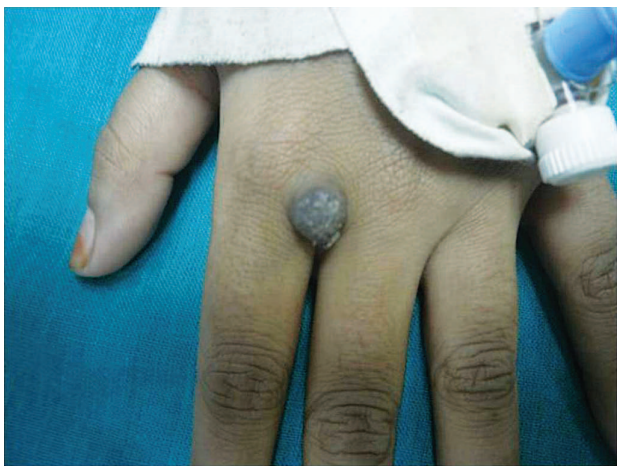

Figure 2. Verrucous hemangioma over the dorsum of the left hand.

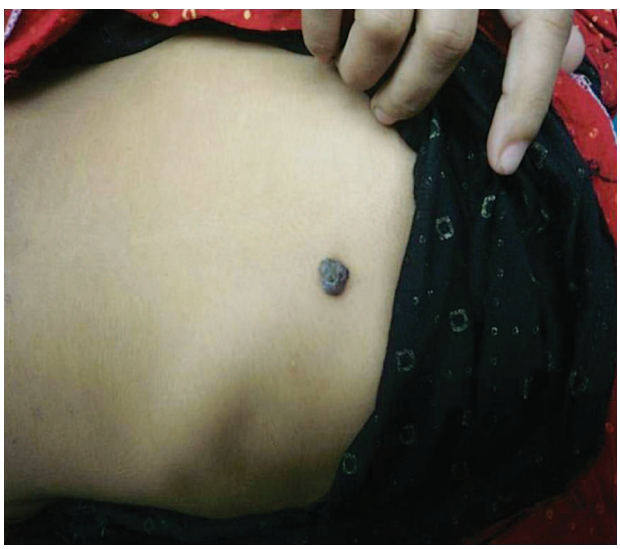

Figure 3. Verrucous hemangioma over the left thigh.

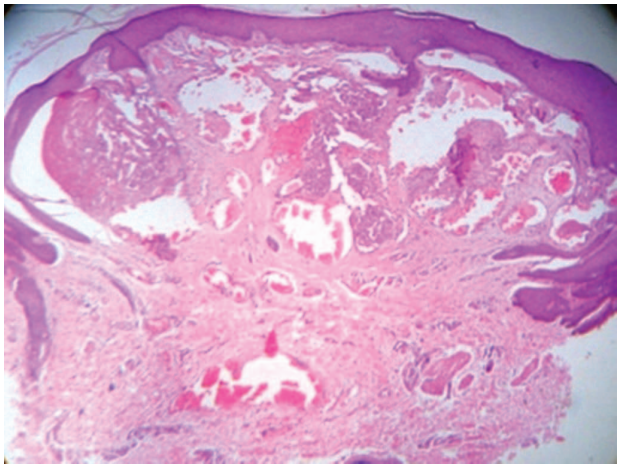

Figure 4. Photomicrograph (*40) of H&E stain showing dilated blood vessels in the dermis.

DISCUSSION

Verrucous hemangioma has been reported in the literature with a variety of names including hemangioma unilateralis neviforme, unilateral verrucous hemangioma, nevus vascularis unius lateralis, keratotic hemangioma, nevus keratoangiomatosus, and papulous angiokeratoma [3]. In 1967, Imperial et al introduced the term "verrucous hemangioma". They described it as a congenital vascular malformation comprising a capillary or cavernous hemangioma in the superficial as well as deep dermis and subcutaneous tissue [4].

Most verrucous hemangiomas are located on the lower extremities in a linear form and involvement is generally unilateral. Lesions are usually present since birth or appear in the early childhood although they may develop later [1,2]. In our case, the lesions appeared within three years of age. The early clinical lesions of verrucous hemangioma are nonkeratotic, soft, and bluish-red in color. Over time, they progressively enlarge and gradually become hyperkeratotic and verrucous as a protective response, usually following trauma or infection [2].

A variant has been described in which multiple lesions occurred with no systemic involvement [5].

In our case, there were also no features suggestive of systemic involvement. A new variant, digital verrucous fibroangioma, is a separate clinical and pathological entity consisting of dome shaped nodules of the dorsum of the fingers [6].

The diagnosis of verrucous hemangioma is primarily based on histopathological examination although clinical correlation is required to confirm the diagnosis. Histologically, verrucous hemangioma shows hyperkeratosis, variable epidermal acanthosis, and papillary telangectasias overlying a deep cavernous or capillary hemangioma. The histological appearance closely resembles an angiokeratoma. However, in contrast to angiokeratoma, the vascular spaces in verrucous hemangioma also affect the lower dermis and may extend into the subcutaneous tissue [3].

Clinically, the differential diagnosis of verrucous hemangioma includes angiokeratoma, Cobb syndrome, angioma serpiginosum, lymphangioma circumscriptum and verrucae [2]. Verrucous hemangiomas do not resolve spontaneously and have a tendency to relapse. Early diagnosis is important to get a better cosmetic result. Verrucous hemangioma requires a large deep excision. In previously reported cases, there has been no association with subcutaneous hemangioma.

The presence of the subcutaneous hemangioma in addition to the disseminated verrucous hemangioma was unique in our case and, to the best of our knowledge, has not been reported so far.

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