Verrucous Hemangioma

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Abstract: Verrucous Hemangioma (VH) is a rare congenital vascular malformation consisted of capillary or cavernous hemangioma affecting dermis and subcutaneous tissue. VH usually presents at birth or early childhood, often unilateral and localized on the lower extremity. The clinical presentation shows hyperkeratotic plaques and nodules, bluish-purple, and partly confluent. Early diagnosis and treatment are pivotal for a satisfactory cosmetic result. A 28-year-old male presented with an asymptomatic rough lump on the left leg which gradually enlarged, thickened and became rougher through time. The lesion appeared since birth and was flat with red-bluish color, Dermatologic examination showed hyperkeratotic plaques and nodules, black-grey colored, confluent with defined margins, measured 10 x 4 cm on lower left leg. Histopathologic examination revealed epidermis hyperkeratosis, verrucous growth, acanthosis, elongated rete ridges, and blood vessels proliferation in the dermis. The patient was treated with cryosurgery, and propranolol tablets 2 x 10 mg and subsequently referred for surgical excision. Diagnosis of VH is hallmarked by hyperkeratotic nodules and confirmed by skin biopsy. Excision is one of the recommended treatment whenever possible. The patient was treated with broad and deep excision due to the high recurrence of VH. After four months of evaluation, the lesion showed satisfactory healing without signs of recurrences. The prognosis was *quo ad vitam ad bonam*, *ad sanam* and *ad cosmetikam dubia ad bonam*. We have reported a patient with VH treated by surgical excision, which showed satisfactory healing without signs of recurrences.

1 INTRODUCTION

Verrucous hemangioma (VH) is a rare congenital vascular malformation, consisting of a proliferation of dilated blood vessels of different sizes that occupy the dermis and subcutaneous tissue. The epidermis of the affected area shows a robust proliferative reaction that presents as a warty appearance (fatani et al, 2016).

Verrucous hemangioma is rare, and only a few cases have been reported (Nupur et al, 2014). The exact incidence is difficult to determine as it has been referred to by many different names in the past (Laun et al, 2019). Verrucous hemangioma has been reported under various names in the literature until 1967, including unilateral verrucous hemangioma, hemangioma unilateralis neviforme, nevus vascularis unius lateris, nevus angiokeratoticus, keratotic hemangioma, nevus keratoangiomatosus, and papular angiokeratoma (Fatani et al, 2016; Laun et al, 2019).

Verrucous hemangioma usually presents at birth or in early childhood and then gradually progresses in size with age (Laun et al, 2019; Dhanta et al, 2018). It often unilateral and localized on the lower extremity (Fatani et al, 2016; Dhanta et al, 2018 ; Sandhu et al, 2016). The clinical presentations are hyperkeratotic plaques and nodules, bluish-purple, and partly confluent. The initial lesions present as flat red or bluish lesions that slowly enlarge and become verrucous (Dhanta et al, 2018 ; Sandhu et al, 2016; Oppermann et al, 2018). The lesions are usually scattered but linear, serpiginous and reticular patterns can be seen rarely. The linear arrangement of these lesions usually reflects genetic mosaicism or dermatomal distribution (Dhanta et al, 2018). Verrucous hemangioma linear is a rarer presentation, according to our literature search, only 10 cases have been reported until 2016 (Sandhu et al, 2016).

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A skin biopsy is needed to confirm the clinical diagnosis (Fatani et al, 2016). Verrucous hemangioma, histologically characterized by dilated capillaries and large cavernous spaces, lined by endothelium. These dilated spaces extend into the reticular dermis and subcutaneous fat. The overlying epidermis shows reactive hyperplasia with marked acanthosis, hyperkeratosis, and papillomatosis (Bindhuja et al, 2013).

Early diagnosis and treatment are pivotal for a satisfactory cosmetic result. The treatment of choice for VH is surgical excision. Various therapeutic options such as cryotherapy, ultrasonography, electrocautery, NdYAG laser, and laser *pulse-dye* can be considered as additional therapy, especially for smaller lesions and when excision is not possible (Laun et al, 2019; Sandhu et al, 2016; Prabhakar et al, 2015). This case is reported to increase our understanding of making an accurate diagnosis of VH and choosing an appropriate treatment.

2 CASE

A 28-year-old Indonesian man presented to the outpatient Dermatovenereology Departement Dr. Kariadi Hospital Semarang with complaints of asymptomatic rough lump on the left leg, which gradually enlarged, thickened and became rougher through time. These lesions had been present since birth, and the initial lesion was flat with red-bluish color. There was no history of any trauma or bleeding from these lesions. The patient gave a history of about 14 years ago that he had been taken by his mother for treatment, and some of the lesions had been excised. There was a recurrence of the lesion after several years. The clinical notes and histopathological reports of the previous excision were not available.

On physical examination, the patient was composmentis. Height 168 cm, weight 67 kg, blood pressure: 120/70 mmHg, heart rate: 84 beats/minute, respiratory rate: 22 breaths/minute, and axillar temperature: 36,7ºC. There were no enlarged lymph nodes in the inguinal or in the area around the lesion. On clinical examination showed hyperkeratotic plaques and nodules in a linear pattern, black-grey colored, confluent with defined margins, measured 10 x 4 cm on the lower left leg (Figure 1.A).

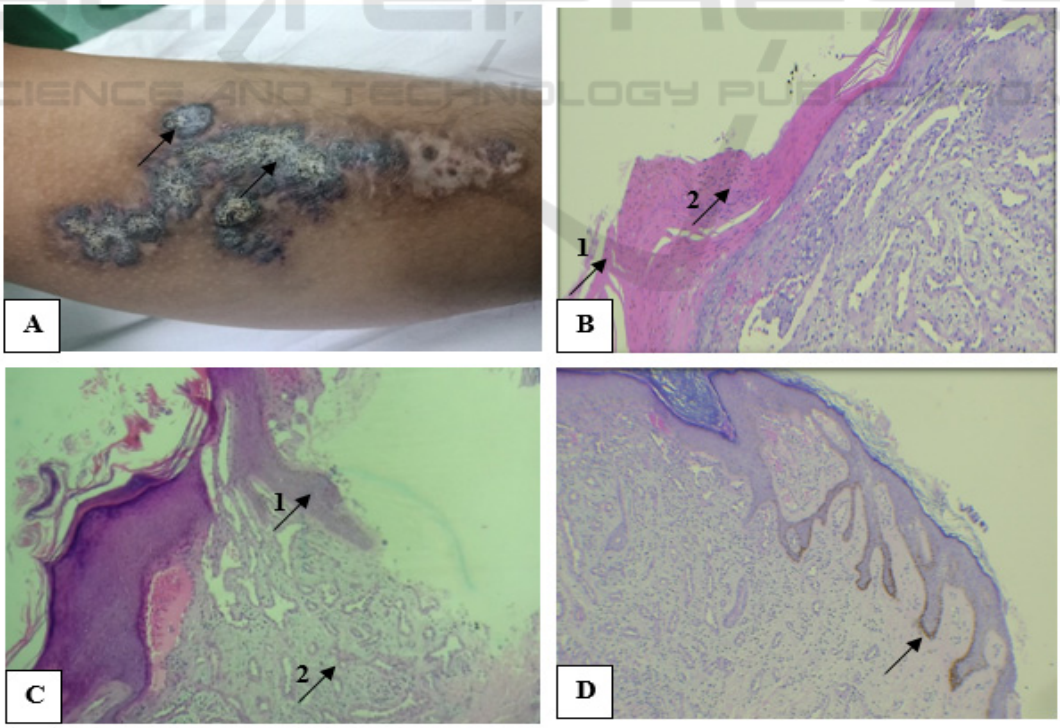

Figure. 1. **A.** Hyperkeratotic plaques and nodules in a linear pattern, black-grey colored, confluent with defined margins on the lower left leg. **B. 1.** Hyperkeratosis, **2**. Parakeratosis (Hematoxylin & eosin, x40). **C. 1.** Acanthosi**s**, **2.** The proliferation of blood vessels in the superficial dermis (Hematoxylin & eosin, x10). **D.** Elongated rete ridge (Hematoxylin & eosin, x10).

The routine laboratory and coagulation factors examination result were standard. The lesion was biopsied and confirmed to be a verrucous hemangioma. Histopathological examination showed the epidermis in the form of a stratified squamous cell epithelium, keratinized, hyperkeratosis, parakeratosis, verrucous growth, elongated rete ridge, acanthosis containing a proliferation of partially dilated blood vessels lined with endothelial cells, with lumen containing erythrocytes surrounded by epidermal papillae. In the superficial dermis, there was the varying size of blood vessel proliferation, skin adnexa accompanied by a mild distribution of interstitial lymphohistiocytic. No malignancy sign found (Figure 1. B-E).

Previously this case had been treated with cryotherapy, propranolol tablets 2x10 mg, and retinoid acid 0,1% cream applied twice daily. After ten days of evaluation post, cryotherapy showed that the lesion had a mild regression. Propanolol tablets and retinoid acid 0,1% cream were still continued. However, after three months of evaluation, the lesions recurred (Figure 2. A-B). Due to recurrence, the patient was referred to the surgery department for excision. All the lesions were excised with a 1 cm margin (Figure 2.C-F).

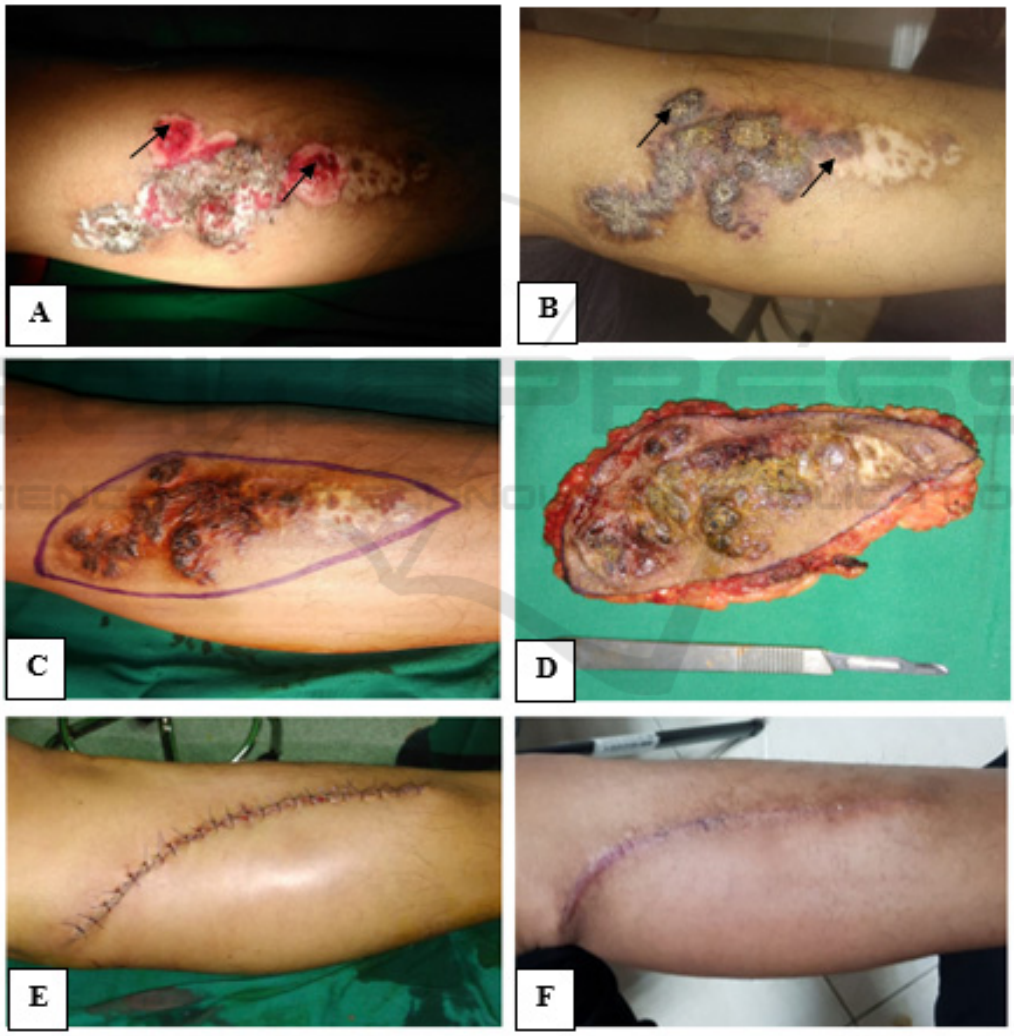

Figure. 2. **A.** The lesions immediately following the cryotherapy procedure. **B.** Recurrence of the lesion after three months post cryotherapy (black arrow). **C.** The resected margin was 1 cm away from the lesions. **D**. The lesions were completely excised. **E**. Postoperative photography following surgical excision. **F**. After four months of evaluation, the lesion showed satisfactory healing without signs of recurrences.

3 DISCUSSION

The term VH is defined by Imperial and Helwig in 1967, which means that congenital localized vascular malformations. (fatani et al, 2016; (Dhanta et al, 2018; Prabhakar et al, 2015). In 1996, The International Society for the Study of Vascular Anomalies classified vascular anomalies into vascular malformations, and vascular tumors. (J.Bindhuja et al,2013 Diagnosis of VH is established based on clinical features and histopathological examination. Prabhakar et al, 2015;Nargis et al,2017).

Verrucous hemangioma presents on the lower extremity in 95% of cases and typically is unilateral (Laun et al, 2019;Singh et al,2017) It may also involve unusual anatomic locations such as the abdomen, arm, and glans penis. Although VH almost invariably presents at birth or in early childhood, it may appear later on, even in adult life (Sandhu et al, 2016; Singh et al,2017) In our case, the lesion appeared from birth and located on the lower left leg.

In the early phase of evolution, the lesions are non-keratotic, soft, blue/red plaques, and clearly demarcated. Gradually the lesions become increasingly hyperkeratotic and verrucous (Nargis et al,2017;Moss et al,2010). The lesions may vary in size from roughly 0,5 to 8 cm in diameter and maybe single or grouped. (Laun et al, 2019) Verrucous hemangioma in its mature phase presents as hardened, hyperkeratotic plaques or nodules with a brownish to bluish-black appearance. This is often accompanied by a history of tenderness and/or bleeding following minimal trauma.(Vijayan et al,2016) The linear form of VH is rare, and only a few cases have been reported. It is not known whether linear lesions actually follow Blaschko's lines or the linear arrangement represents genetic mosaicism. (Nupur et al, 2014) In our case, the lesion showed in a linear pattern, and there was no history of any trauma or bleeding from these lesions.

Histologically, VH shows with hyperkeratosis, variable epidermal acanthosis, and papillary telangiectasias overlying a deep capillary or cavernous hemangioma. The abnormal proliferating vessels are situated in the dermis and hypodermis. The hemangiomatous component mostly comprises dilated capillaries and wider cavernous, endothelium-lined, blood-filled spaces. Inflammatory cells, fibrosis, and hemosiderin may exist in the upper dermis. (fatani et al, 2016; J.Bindhuja et al,2013; Moss et al,2010). Typical histopathological features were observed in our case also. Immunohistochemical staining with endothelial markers like CD 31, CD 34 and GLUT1 may be done for confirmation, but the diagnosis can be made by light microscopic features alone. (J.Bindhuja et al,2013;(Vijayan et al,2016)

The differential diagnosis with angiokeratoma can be excluded. The histologic appearance closely resembles angiokeratoma, as both lesions show vascular spaces beneath a papillomatous and hyperkeratotic epidermis. However, in contrast to angiokeratoma, the vascular spaces in VH also involve the lower dermis and subcutaneous tissues. (Oppermann et al, 2018;.Naveen et al,2016)

Verrucous epidermal nevus (VEN) also can be excluded because histologically, the hallmark finding of VEN is hyperkeratosis, acanthosis, and papillomatosis. In VEN, there are no abnormal proliferation of blood vessels (Das et al, 2015)

Verrucous hemangioma should be identified, diagnosed, and treated as early as possible to limit the extent of resection. Because of the risk of recurrence, resection should encompass the deep portions of the lesion with usually a 1 cm margin of excision. If the lesion is small (<2 cm), cryosurgery, electrocautery, or laser therapy can be used, but resection is the primary treatment. These additional therapies can be used in combination with resection for extensive lesions to further assist in reducing the risk of recurrence. (Laun et al, 2019) In our case, a propanolol tablet was given for three months, but after that, there was a recurrence. Propranolol is the treatment of choice for troublesome haemangiomas. Other studies that have employed oral propranolol therapy would not recommend using it on other vascular anomalies. Oral propranolol is more effective in hemangioma infantile than in an adult.(Dimaguila et al,2017)

Surgical excision is one of the recommended treatment whenever possible, and Incomplete excision leads to persistence, recurrence, and continued enlargement of the lesion. Due to the deeper vascular infiltration, the recurrence rate of VH is 33%, especially when the lesions are more significant than 2 cm in diameter (Dhanta et al, 2018) The patient was treated with broad and deep excision. After evaluation for four months, the lesion showed satisfactory healing without signs of recurrences.

The prognosis for VH is excellent, with recurrence being low when adequate surgical margins are utilized and if in combination with additional therapies. If inadequate wide excision is performed, recurrence can exceed 30%.(Laun et al, 2019) The prognosis of this case was *quo ad vitam*

ad bonam, quo ad sanam, and *quo ad cosmeticam dubia ad bonam.*

4 CONCLUSION

We have reported a case of verrucous hemangioma in a 28-years old male. The diagnosis was made on the basis of anamnesis, clinical examination, and histopathological examination. On the anamnesis, the patient complained of asymptomatic rough lump on the left leg, which gradually enlarged, thickened, and became rougher through time. The lesion appeared since birth and was flat with red-bluish color. The clinical examination, we found hyperkeratotic plaques and nodules in a linear pattern, black-grey colored, confluent with defined margins, measured 10 x 4 cm on lower left leg. Histopathological examination confirmed the diagnosis of verrucous hemangioma. The patient was treated with broad and deep surgical excision, which showed satisfactory healing without signs of recurrences. The prognosis of this case was *quo ad vitam ad bonam, quo ad sanam dubia ad bonam,* and *quo ad cosmeticam dubia ad bonam.*

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