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An Unusual Case of Superficial Lymphangioma of the Right Foot

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SUMMARY Superficial lymphangioma is a microcytic lymphatic malformation that is usually present already at birth. Women are more commonly affected than men are. It occurs because of the deep lymphatic circulation obstruction. Superficial lymphangioma is most commonly found in the mouth, especially the tongue, on the shoulders, neck, limbs, armpits and groin. It appears as a cluster of small firm blisters filled with clear or hemorrhagic lymph fluid, resembling frogspawn. We are presenting an unusual case of superficial lymphangioma of the right foot in an adult female patient.

KEY WORDS: superficial lymphangioma, microcytic lymphatic malformation, lymphatic obstruction.

INTRODUCTION

Superficial lymphangioma is a microcytic malformation of the lymphatic vessels, also known as lymphangioma circumscriptum or cystic lymphangioma circumscriptum (1-3). It is most commonly present already at birth, and is more common in women than in men.

CASE REPORT

A 36-year-old Caucasian female presented to our Department complaining of skin changes on the first and second toes of her right foot. The changes were present for four years. Family history was not significant. The patient was suffering from seasonal allergic conjunctivitis. Otherwise she was healthy. Previously, she had been misdiagnosed as common warts and fungal infection and mistreated with cryotherapy and terbinafine cream. At initial visit she presented with clusters of blisters in a herpetiform arrangement that partially had verrucous surface, on the first and second toes of her right foot (Fig. 1). The skin lesions were painless. She reported occasional swelling of her right foot since her teenage years. The lymphatic stasis test had not been previously performed.

We performed lesional biopsy. Histopathologic examination showed acanthotic, hyperkeratotic

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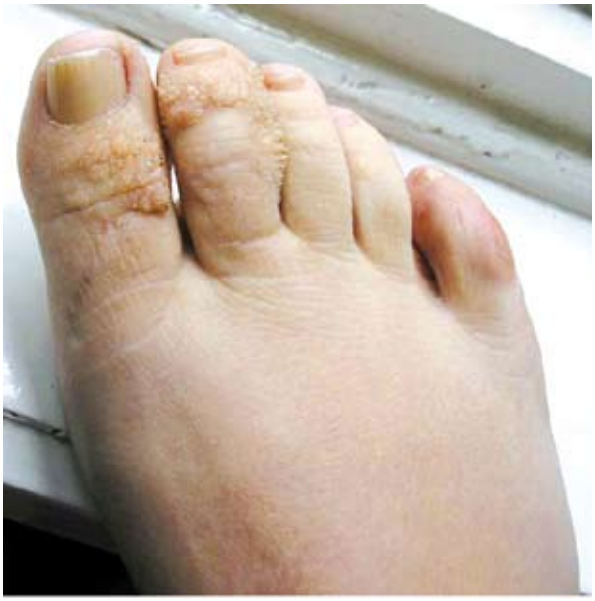

Figure 1. Clusters of blisters in a herpetiform arrangement that partially had verrucous surface on the first and second toe of the patient's right foot.

epidermis and dilated vascular spaces in papillary dermis. Elongated dermal papillae were present (Fig. 2). The diagnosis of superficial lymphangioma was confirmed.

We recommended lymphangiography and testing of lymphatic function. The patient was advised to undergo surgical excision of the skin changes as the treatment of choice for superficial lymphangioma. The patient was informed on the possible recurrence of the disease after surgical excision.

DISCUSSION

Superficial lymphangioma occurs mostly due to lymphatic circulation obstruction and then is also called lymphangiectasis (3). The most common sites are mouth, especially the tongue, gingiva, shoulders, neck, limbs, armpits and groin (3-7). Our patient presented with superficial lymphangioma on the first and second toes of the right foot. This is an unusual site of syperficial lymphangioma, and we found no such case in the literature. Changes on the limbs are mostly associated with more severe vascular malformations and smaller lesions that can be easily infected. In such patients, superficial lymphangioma can be associated with a type of coagulopathy, which complicates their clinical status with painful hemorrhagic episodes, and requires heparin therapy and more frequent check-ups (8). The lymphangiomas of the vulva are associated with cellulitis, Crohn's disease, radiotherapy and lymphedema (3,9,10). Motahhary *et al.* report a case of bilateral lymphangioma of the gingiva in canine area (5).

The authors compare superficial lymphangioma with frogspawn because it looks like a cluster of small firm blisters filled with clear or hemorrhagic fluid (3). Bigger lesions often have erythematic edge and are combined with arterial and vascular lesions, which are then called hematolymphangiomas. Verrucous changes can be found as well (3).

Superficial lymphangioma occurs because of the deep lymphatic circulation obstruction. However, typical lesions may occur spontaneously, without evident signs of lymphatic circulation obstruction. It consists of multiple dilated lymph vessels in reticular and papillary dermis, just beneath the surface epithelium (1).

Typical histopathologic evaluation of superficial lymphangioma, as presented in our case, shows multiple dilated lymph vessels in reticular and papillary dermis, just beneath the surface epithelium (1). There are smooth muscle cells on the lymph vessel surface and very thin endothelial cells, sometimes barely visible. Larger lymph vessels are often split by fibrous septa. Hyperkeratotic epidermis may overlay some clusters of dermal lymphatic vesicles (3,11,12).

Further diagnostic examinations are not necessary since histologic evaluation and clinical picture are sufficient criteria for the correct diagnosis. However, examination of the lymph stasis should be done, as it was recommended to our patient.

Differential diagnosis includes angiokeratomas, verrucous hemangiomas, herpes simplex and herpes zoster (3).

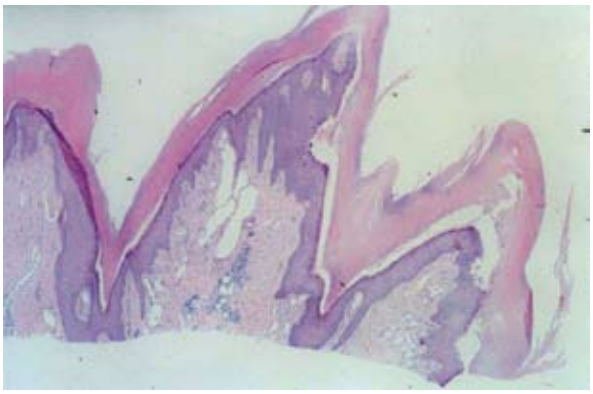

Figure 2. Acanthotic, hyperkeratotic epidermis. Dilated vascular spaces in papillary dermis. Elongated dermal papillae are present (H&E staining, original magnification X40).

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Treatment is not necessary but surgical excision can be considered. Recurrence of the disease is quite common (1,3). Larger lesions require either split-thickness grafts or inflation of skin expanders before resection, to allow an adequate covering of the surgical wound (8). There are successful attempts in the treatment of superficial vulvar lymphangioma with CO_2 laser therapy (13), and with ablation of vulvar lymphangioma with YAG laser (14). Besides surgical treatment, after which recurrence of the disease is possible, laser treatments, especially with CO_2 and YAG lasers, can be considered in adults (13,14).

CONCLUSION

We present a case of superficial lymphangioma at an unusual location on the right foot. Superficial lymphangioma should be considered in patients with prolonged skin changes forming clusters of herpetiform vesicles that might appear anywhere on the skin and show lymph stasis.

References

1. Lončarić D. Benigni vaskularni tumori kože. In: Lipozenčić J *et al*., eds. Dermatovenerologija. Zagreb: Naklada Zadro;1999. pp. 316-8.
2. Pašić A, Dobrić I. Benigni vaskularni tumori kože. In: Dobrić I *et al*., eds. Dermatovenerologija. Third Completely Changed and Revised Edition. Zagreb: Grafoplast; 2005. pp. 387-90.
3. Braun-Falco O, Plewig G, Wolff HH, Burgdorf WHC. Mesenchymal and neural tumors. In: Braun-Falco O, Plewig G, Wolff HH, Burgdorf WHC, eds. Dermatology. 2nd Completely Revised Edition. Berlin: Springer; 2000: pp. 1553-601.
4. Brennan TD, Miller AS, Chen SY. Lymphangiomas of the oral cavity: a clinicopathologic, immunohistochemical and electron-microscopic study. J Oral Maxillofac Surg 1997;55:932-5.
5. Motahhary P, Sarrafpour B, Abdirad A. Bilateral symmetrical lymphangiomas of the gingiva: case report. Diagn Pathol 2006;1:9.
6. Josephson P, van Wyk CW. Bilateral symmetrical lymphangiomas of the gingiva. A case report. J Periodontol 1984;55:47-8.
7. Mc Daniel RK, Adcock JE. Bilateral symmetrical lymphangiomas of the gingiva. Oral Surg Oral Med Oral Pathol 1987;63:224-7.
8. Bolognia JL, Jorizzo JL, Rapini RP. Lymphatic malformation. In: Bolognia JL, Jorizzo JL, Rapini RP, eds. Dermatology. Volume Two. Edinburgh: Mosby; 2003. pp. 1624-8.
9. Johnson T, Kennedy A, Segal G. Lymphangioma circumscriptum of the vulva. A report of two cases. J Reprod Med 1991;36:808-12.
10. Bartels U, Krauss T, Sattler B, Maghsudi M, Kuhn W. Therapy of extensive lymphangioma of the vulva. Zentralbl Gynaekol 1995;117:220-3.
11. Wassef M, Enjolras O. Les malformations vasculaires superficelles: classification et histopathologie. Ann Pathol 1999;19:253-64.
12. Calonje E, Wilson-Jones E. Vascular tumors and tumor-like conditions of blood vessels and lymphatics. In: Elder D, ed. Lever's Histopathology of the Skin. Eighth Edition. Philadelphia: Lippincott-Raven Publishers; 1997. pp. 889-932.
13. Smith H, Genesen M, Feddersen R. Dermal lymphangiomata of the vulva and laser therapy: a case report and literature review. Eur J Gynaecol Oncol 1999;20:373-8.
14. Oshsendorf FR, Kaufmann R, Runne U. YAG laser ablation of acquired vulval lymphangioma. Br J Dermatol 2001;144:442-5.