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CASE REPORT

Lymphoedema caused by idiopathic lymphatic thrombus



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Received 29 March 2013; accepted 4 April 2013

KEYWORDS

Primary lymphoedema;
Lymphatic thrombus;
Podoplanin;
Lymphatico-venous anastomosis

Summary Primary lymphoedema includes some diseases whose genetic anomaly is detected and others whose pathology is unknown. In this article, we report a lymphatic thrombus found in a limb with lymphoedema during lymphatico-venous anastomosis (LVA). A 32-year-old man was aware of oedema in his left calcar pedis 3 years previously, which appeared without any trigger. Indocyanine green lymphography indicated lymphatic stasis in the left calf and thigh region, and we performed LVA for the patient. During the operation, we found yellow vessels, which were thought to be lymphatic vessels filled with a yellow solid substance, just beneath the superficial fascia at the left ankle. Pathological examination of the thrombi revealed hyaline material mixed with cell components. The cells were categorised as lymphatic endothelial cells, as they were positive for podoplanin. There was no evidence of malignancy. Causes of idiopathic lymphatic thrombus such as this may be one of the causes of so-called primary lymphoedema, and evaluation of such cases may be the first step towards elucidating the mechanisms involved in the development of primary lymphoedema.

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Lymphoedema can be divided into two categories, primary and secondary, and is caused by an accumulation of protein-rich fluid in the subcutis.¹ Secondary lymphoedema occurs after lymphadenectomy or radiotherapy, most often for breast, uterine or ovarian cancer.^{2,3} However, primary

lymphoedema can occur in genetic disorders such as Milroy disease⁴ and Meige disease,⁵ or in others with unknown pathology.^{6,7} There seem to be many different pathological conditions that occur in primary lymphoedema, and elucidation of these conditions is anticipated in the future.

Sometimes, lymphatic thrombus is formed within the lymphatic vessels, and infections such as lymphatic filariasis or chlamydia trachomatis and other conditions such as congestive heart failure, inflammation of the lower limb or amyloidosis can be leading causes in the pathology of

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lymphatic thrombosis⁸; it can also be caused by axillary lymph node dissection or cancer metastasis. Lymphatic fluid is thought to have weaker clotting capability than blood because it contains little thrombokinase and high levels of antithrombin⁹; therefore, the risk of lymphatic thrombus is estimated to be lower than that of blood thrombus. To date, to our knowledge, there have been no reports of lymphoedema caused by lymphatic thrombus and no histopathological examinations of idiopathic lymphatic thrombus.

We have performed lymphatico-venous anastomosis (LVA) for the treatment of both primary and secondary lymphoedema and evaluated the condition of the lymphatic vessels during the procedure.¹⁰ In this article, we report a lymphatic thrombus found in a limb with lymphoedema during LVA. We also evaluated the lymphatic thrombus histopathologically.

Case report

A 32-year-old man had experienced fatigue in his left leg for approximately 10 years. He was aware of oedema in his left *calcar pedis* 3 years previously, which appeared without any triggers, and started compression therapy using compression garments and bandages. He used cold therapy for his left foot, even while at work, because he had severe pain and heat sensation. Heart, kidney and liver function were normal, and no finding indicated malignancy. Venous reflux, varix and thrombus were not found. As the oedema gradually worsened and expanded, he presented to our department complaining of the sensation of burning and pain in his lower leg (Figure 1).

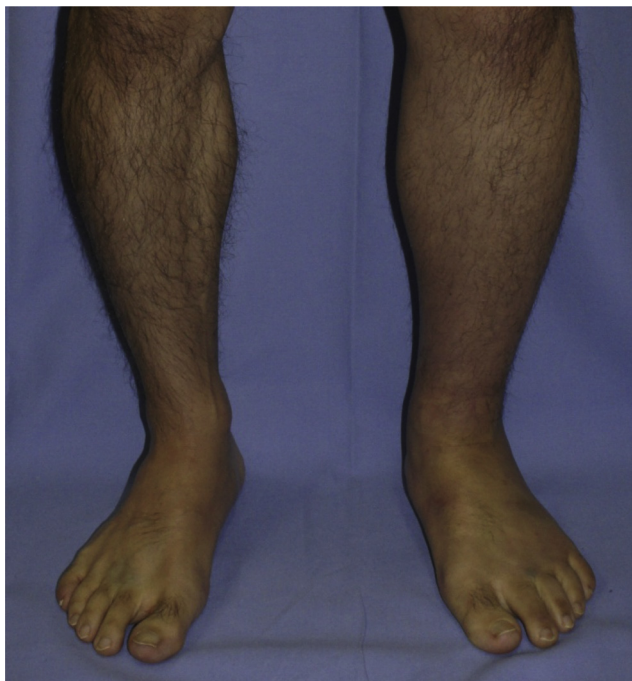


Figure 1 Clinical appearance of bilateral lower legs. Edema and redness can be seen at the left ankle and dorsum of the foot.

Indocyanine green (ICG) lymphography revealed a linear pattern in the dorsum of the left foot and a stardust pattern in the left calf and thigh region. In the right lower limb, a normal linear pattern was observed. He was diagnosed with idiopathic lymphoedema based on the findings from ICG lymphography, and we scheduled the patient for LVA. During the operation, we found yellow vessels (Supplementary Figure 1a), which were thought to be lymphatic vessels filled with a yellow solid substance (Figure 2, Supplementary Figure 1b, yellow arrow), just beneath the superficial fascia at the left ankle. The vessels were approximately 0.5 mm in diameter. The yellow solid had spread along the lumen of the lymphatic vessels for at least the length observable within the incisions. The substance showed no elasticity and was easily split using No. 5 forceps. We were able to push the substance out of the lymphatic vessel and removed as much of it as the procedure allowed. We closed the incision without performing LVA to avoid postoperative venous thrombus. At the left dorsum of the foot, we also found yellow lymphatic vessels (Supplementary Figure 2), which involved a yellow viscous liquid that was almost consolidated. At the left calf, thigh and groin region, sclerotic lymphatic vessels without thrombus were found. Therefore we performed LVA in these three incisions.

The patient's pain and heat sensation gradually decreased within the 3 months after the operation, and lymphoedema was slightly improved.

Pathological examination of the thrombi revealed hyaline material mixed with cell components (Figure 3a; haematoxylin and eosin stain). The cells were categorised as lymphatic endothelial cells, as they were positive for podoplanin (Figure 3b; podoplanin stain). The lymphatic

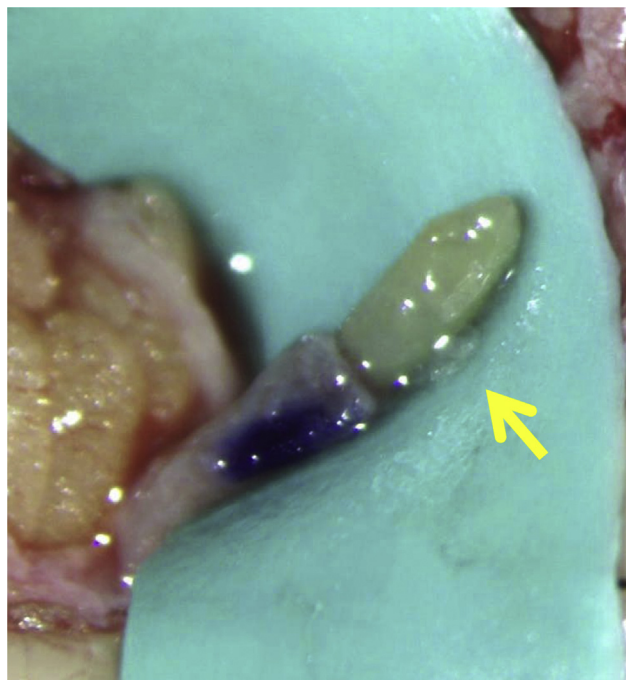


Figure 2 Intraoperative findings at the left ankle. The solid was easily split using No. 5 forceps and exuded from the vessel (arrow). The solid spread along the lumen of the lymphatic vessel for at least the length observable within the incisions.

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