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Unique Case of Postural Acquired Lymphangiectasia of the Scrotum Exacerbated by an Erect Position

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Dear Editor:

Acquired lymphangiectasia (AL) is thought to arise from lymphatic occlusion¹. AL appears as thin-walled, translucent, white, or skin-colored papules and vesicles². AL on the scrotum is rare and may present as a congenital condition or, rarely, develop secondary to radiotherapy, infection, or surgery³. Herein, we report an interesting case of postural AL of the scrotum, which became more prominent with the erect position.

A 10-year-old boy presented with multiple skin-colored papules on the scrotum for 6 months. On physical examination, the skin lesions became more prominent with the erect position (Fig. 1A, B). The size of each lesion had recently increased abruptly without oozing. Eight years before presentation, he had orchiopexy of the right testis due to cryptorchidism. He denied any skin lesions on the scrotum at that time. On ultrasonography at the time of presentation, right testis was found to be orchiopexy state, whereas left one was cryptorchidism in high sacral area, and there was no evidence of lymphatic malformation.

Histopathological examination of a representative lesion revealed multiple dilated channels on the upper dermis. Endothelial cells lining the dilated channels showed positivity in D2-40 immunohistochemistry (Fig. 1C, D). Therefore, we confirmed the dilated channels in the upper dermis to be lymphatics. Based on these clinical and histological findings, he was diagnosed with AL. Because there was little cosmetic concern or complication at presentation, we decided to observe. We recommended magnetic resonance imaging for further evaluation, if symptoms develop later.

AL usually results from lymphatic obstruction caused by a spectrum of scarring processes often due to surgery⁴. Postsurgical fibrosis occludes the normal lymphatics, causing subsequent dilatation of the dermal lymphatics leading to AL². Our patient had a history of orchiopexy due to cryptorchidism, so we suspected that AL occurred due to operation. Our case is unique since the lesions were more prominent with the patient in an erect position. Lymph flow within the skin depends on tissue movement caused

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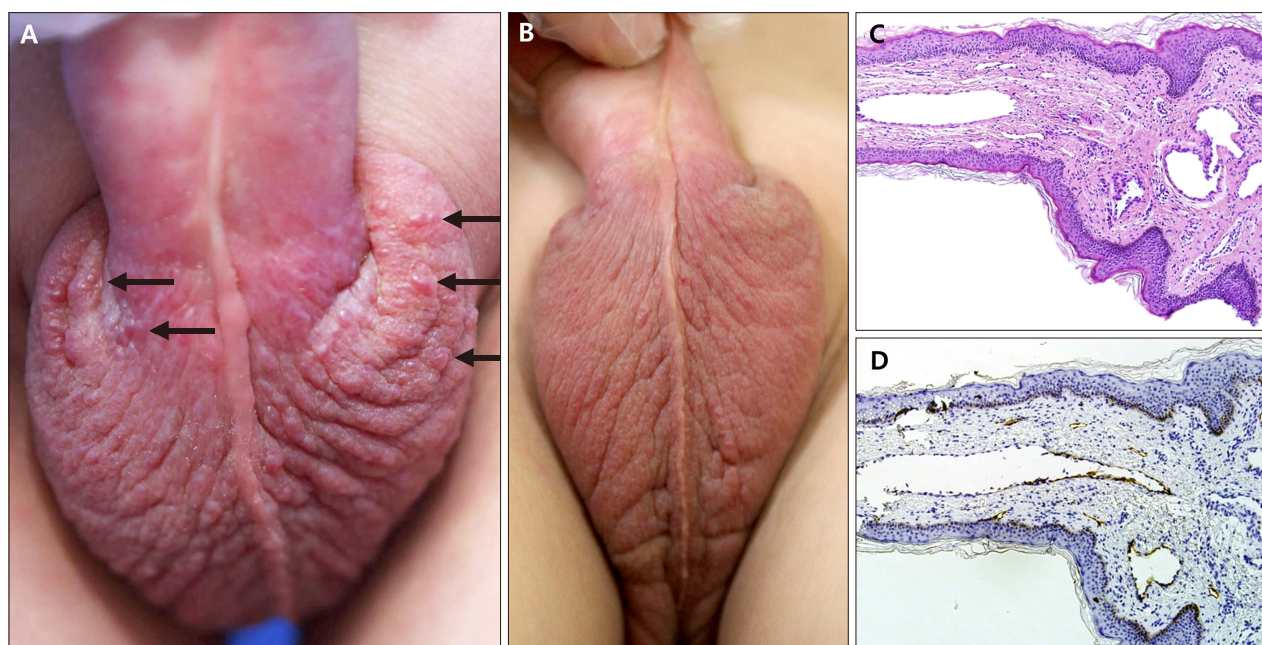


Fig. 1. A 10-year-old boy presented with multiple skin-colored papules (indicated by black arrows) on the scrotum, which interestingly became more prominent with the patient (A) in the erect position, and less prominent with the patient (B) in the supine position. (C) Histopathological examination of a representative papule from the scrotum revealed multiple dilated channels on the upper dermis (H&E, $\times 100$). (D) Endothelial cells of dilated channels showed positivity in D2-40 immunohistochemistry (D2-40, $\times 100$).

by arterial pulsations, muscular contractions, and movement of the body. Therefore, we speculated that the postural change might influence the lymph flow inside an AL, resulting in size changes of the lesions. Two cases of AL developed during pregnancy has been reported⁵. In those cases, postural change was not documented, and AL is thought to occur due to the change of lymph flow secondary to pregnancy. The nomenclature for lymphangiectasia varies and it can be referred to as lymphangiectasis, lymphangioma, or lymphangioma circumscriptum. The suffix ‘-oma’ means tumor, and the suffix ‘-etasia’ means dilatation of a tubular structure². It might be assumed that the postural change of AL could not be rarely observed with attention, but it has been underestimated. Along with the previously reported cases in pregnancy our case indicates a neglectable feature of AL and also supports the concept that ‘lymphangiectasia’ is a more appropriate term than ‘lymphangioma circumscriptum’. Indications for treatment of AL include both its cosmetic appearance and prevention of complications. The definitive treatment is surgical excision, and other palliative modalities includes ablative lasers and sclerotherapy³.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

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