Eur J Vasc Endovasc Surg **35**, 373–374 (2008) doi:10.1016/j.ejvs.2007.09.003, available online at http://www.sciencedirect.com on **ScienceDirect**

SHORT REPORT

Unilateral Lower Limb Swelling Secondary to Cavernous Lymphangioma

S.K. Bains^{*} and N.J. London

Department of Vascular and Endovascular Surgery, Leicester Royal Infirmary, Leicester LE1 5WW, UK

Introduction. We report an unusual case of unilateral leg swelling secondary to cavernous lymphangioma (cystic hygroma), which normally affects the head and neck regions.

Case report. A 25 year gentleman presented to our department with a 13-year history of gradually increasing unilateral leg swelling and recurrent infections. Investigations showed appearances consistent with cavernous lymphangioma, and partial excision of the lesion led to resolution of symptoms.

Discussion. The most common sites for cystic hygroma are the head and neck areas, but the extremities can be affected as demonstrated. Complete surgical excision is often difficult, and there is a tendency for recurrence.

This case acts as an illustration of an uncommon yet important cause for unilateral leg swelling.

© 2007 European Society for Vascular Surgery. Published by Elsevier Ltd. All rights reserved.

Keywords: Leg swelling; Cavernous lymphangioma.

Introduction

Unilateral leg swelling is a common presentation to the vascular surgeons. The differential diagnosis includes lymphoedema, venous insufficiency and deep vein thrombosis.

We report an unusual case of unilateral leg swelling secondary to cavernous lymphangioma (cystic hygroma), which normally affects the head and neck regions.

Case Report

A 25 year old man presented to our department with right leg swelling causing disfigurement and functional impairment. The swelling was first noticed when the patient was aged 12 years, and had since been increasing in size.

Recently the groin region had become repeatedly infected with serous discharge from one main area.

*Corresponding author. S. K. Bains Department of Vascular and Endovascular Surgery, Leicester Royal Infirmary, Leicester LE1 5WW, UK.

E-mail address: salenabains@hotmail.com

Extensive oedema extended from the groin into the lower thigh, calf and foot (Fig. 1).

An MRI scan was performed which showed a well demarcated lesion starting in the right thigh, superficial to the femoral vessels, travelling through the femoral ring. It spread retroperitoneally to the inferior border of the kidney into the right side of the pelvis, displacing the rectum and bladder to the right. Appearances were consistent with cystic hygroma (Fig. 2).



Fig. 1. Pre operative photograph.

1078–5884/000373+02 \$34.00/0 © 2007 European Society for Vascular Surgery. Published by Elsevier Ltd. All rights reserved.



Fig. 2. Pre operative MRI scan.

The patient was taken to theatre for excision of the lesion. Intra-operatively the lesion from the groin was excised superficial to the deep fascia of the thigh. The long saphenous vein was preserved. There was no leakage of fluid from the groin and as the abdominal component was asymptomatic this was not excised. Histology confirmed a cavernous lymphangioma and bacterial colonies within an abscess cavity. Postoperatively the patient made a good recovery and the wounds healed well. Compression bandaging was applied to the leg and six weeks post-surgery the swelling resolved completely (Fig. 3).

Discussion

The most common sites for cystic hygroma are the head and neck areas, but the extremities can be affected as demonstrated. Lymphangiomas are congenital malformations of the lymphatic system and usually arise during infancy.

Complete surgical excision is often difficult, and there is a tendency for recurrence. Newer treatments include sclerotherapy as an alternative to excision.



Fig. 3. Post operative photograph.

OK-432 is a lyophilized mixture of Streptococcus pyogenes and benzylpenicillin and is currently being used for reduction and complete obliteration of lesions with varying degrees of success.^{1–3}

This case acts as an illustration of an uncommon yet important cause for unilateral leg swelling.

References

- 1 PETERS DA, COURTEMANCHE DJ, HERAN MK, LUDEMANN JP, PRENDIVILLE JS. Treatment of cystic lymphatic vascular malformations with OK-432 sclerotherapy. *Plast Reconstr Surg* 2006 Nov; 118(6):1441–1446.
- 2 KOBAYASHI D, KUMANGAI H, SATSUMA S. Cavernous lymphangioma of the leg in children treated by the injection of OK-432 after resection. J Bone Joint Surg Br 2003 Aug;85(6):891–894.
- 3 HALL N, ADE-AJAYI N, BREWIS C, ROEBUCK DJ, KIELY EM, DRAKE DP et al. Is intralesional injection of OK-432 effective in the treatment of lymphangioma in children? Surgery 2003 Mar;133(3):238–242.

Accepted 1 September 2007 Available online 26 October 2007