

Bilateral axillary lymph node uptake of radiotracer during lower extremity and scrotal lymphoscintigraphy in a case of primary scrotal lymphoedema

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ABSTRACT Lymphoscintigraphy is a useful technique for the evaluation of lymphatic function in the presence of limb swelling. The authors report a case where genital swelling in a 20-year-old man was investigated by lymphoscintigraphy. We performed lower limb lymphoscintigraphy and scrotal lymphoscintigraphy in the patient on two different days. Lower limb revealed dermal backflow pattern in lower limbs, inguinoscrotal reflux of the lymph and unexpected avid radiotracer uptake in the axillae bilaterally. Scrotal lymphoscintigraphy revealed slow movement of the lymph from the scrotal skin and again unexpected avid radiotracer uptake in the axillae bilaterally. Findings were concluded as congenital hypoplasia of lymphatics in lower limbs, congenital lymphectasia/compensatory megalymphatics in scrotum and aberrant lymphatic pathway, possibly due to malfunctioning/nonfunctioning thoracic duct.

Keywords: Genital lymphedema, inguinoscrotal reflux, lymphoscintigraphy

INTRODUCTION

Most descriptions of lymphatic circulatory disorder rely entirely on clinical impression; that is, on the history and physical examination. Peripheral lymphoscintigraphy is essentially non invasive, easy to perform repeatedly and harmless to lymphatic vascular endothelium. All patients with the provisional diagnosis of peripheral lymphatic dysfunction or idiopathic edema should undergo diagnostic lymphoscintigraphy to verify the accuracy of diagnosis and to provide a blueprint for subsequent therapy.^[1] Interpretation of peripheral lymphoscintigraphy can be difficult due to anatomical and physiological variations between individuals, pathological changes which may vary enormously depending on the chronicity of the disease process and technical factors relating to injection site, radiotracer and scanning technique.^[2]

Lymphedema of the genital region is relatively uncommon, but is extremely uncomfortable and distressing for the patients who suffer with this condition. The cause of genital lymphedema is either primary or secondary.^[3] We report a case of scrotal edema where abnormal lymphatic drainage was demonstrated on lower extremity and scrotal lymphoscintigraphy.

CASE REPORT

An otherwise well immunocompetent 20-year-old single male with no relevant family history was investigated for scrotal oedema and oozing of fluid from the scrotal skin for last 5 years. He had history of circumcision at the age of 1 year with no other relevant past history viz. malignancy, radiation, filariasis or sexually transmitted disease. On physical examination, scrotal skin was diffusely thickened with multiple small vesicles on it, some of which discharging clear fluid. Scrotal ultrasonography revealed mild right hydrocoele with bilateral edematous and thickened scrotal skin. A clinical diagnosis of primary Lymphoedema with lymphorrhoea was made and was referred for lymphoscintigraphy for evaluation of lymphatic system.

We performed lymphoscintigraphy by using ^{99m}Tc sulfur colloid, prepared by aseptic filtration through a 100 nm filter.^[4]

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After skin cleansing using an aseptic technique, a volume of 0.1 ml and activity of 20 MBq (0.5 mCi) ^{99m}Tc filtered sulfur colloid was injected intradermally near the second web space of each foot [Figure 1a]. After the injection patient was made to exercise by brisk walking. Imaging was done using dual-headed gamma camera (Millennium VG Hawkeye, GE Healthcare, USA) with patient in supine position under the camera. Serial static images of popliteal, groin, abdominal and chest area were acquired after 30 min, 1 h, 2 h and 4 h of injection using low energy, all purpose collimator and analyzed using Entegra workstation.

Lower limb lymphoscintigraphy revealed mild dermal backflow in the both legs (right more than left) [Figure 1b] and avid tracer uptake in bilateral inguinal region at 30 min. Reflux of tracer is seen in the scrotal skin at 1 h [Figure 1c]. 2 h images revealed faintly visualized tracer in the iliac and para-aortic lymph nodes [Figures 1d and e]. The 4 h images demonstrated tracer uptake in the liver and bilateral axillary lymph nodes (left more than right) [Figure 1f]. Dermal backflow pattern in the scrotal skin was also increased.

Scrotal lymphoscintigraphy was performed on a separate day. After skin cleansing using an aseptic technique, a volume of 0.1 ml and activity of 20 MBq (0.5 mCi) ^{99m}Tc filtered sulfur colloid was injected subcutaneously in the most dependent part of scrotal skin on each side [Figure 2a]. Imaging was done with patient in supine position under the gamma camera. Serial static images of groin, abdominal and chest area were acquired after 30 min, 1 h, 2 h, 3 h, 4 h and 5 h of injection.

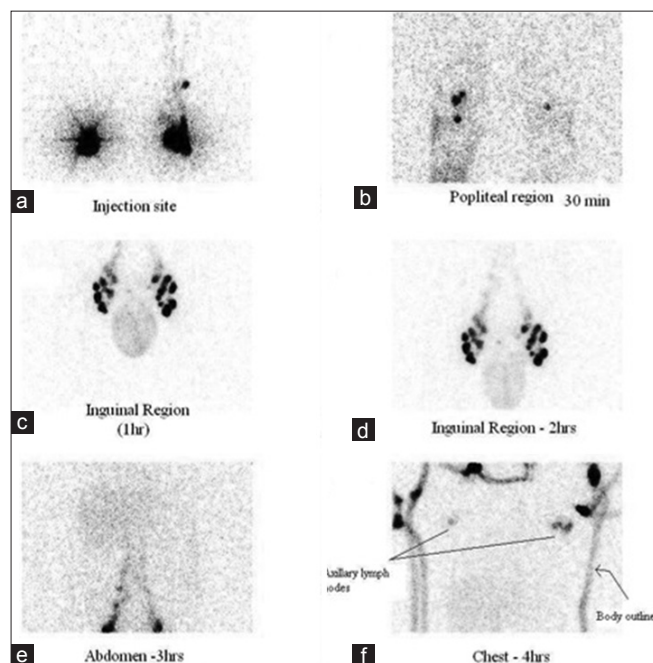


Figure 1: (a) Injection site. (b) Popliteal lymph nodes and mild dermal backflow in the both legs (right more than left). (c and d) Bilateral inguinal and iliac lymph nodes with reflux of tracer in the scrotal skin. (e) Iliac and retroperitoneal lymph nodes. Faint activity is also seen in the liver. (f) Tracer uptake in bilateral axillary lymph nodes

Scrotal lymphoscintigraphy revealed faint tracer activity in bilateral inguinal lymph node at 1 h [Figure 2b]. Next image at two hours [Figure 2c] shows slightly more activity in inguinal nodes. Tracer activity in the axillae noted at 3 h image [Figure 2d]. Tracer activity is increased in the inguinal lymph nodes and faint tracer activity in the iliac and para-aortic lymph nodes at 4 h image [Figure 2e]. Liver is not visualized till 5 h [Figure 2f].

DISCUSSION

Lymphedema has been classified into two major groups: Primary or secondary lymphedema. Primary lymphedema can be further divided into three groups: Lymphedema congenita-onset before the 1 year of age, Lymphedema praecox-onset 1-25 years of age and thereafter Lymphedema tarda – after 25 years of age. The main reasons for primary genital lymphedema are that the lymph vessels are absent or reduced in number or simply don't work as well as they should, i.e., functional failure.^[3,5] As the affected individual grows, the involved lymphatic system has to face more pressure to drain the tissue fluid, and the swelling becomes far more obvious. Primary Lymphedema affecting only genitals is rare. Chronic genital lymphedema secondary to lymphangiectasia and chylous reflux, in patient with Noonan syndrome has also been reported.^[6] Secondary lymphoedema more commonly affects the genital region than primary lymphoedema. In Africa, India and other tropical countries, genital swelling is frequently seen due to filariasis. This can lead to gross elephantiasis of the penis and scrotum. Secondary lymphedema in North America and Europe is most

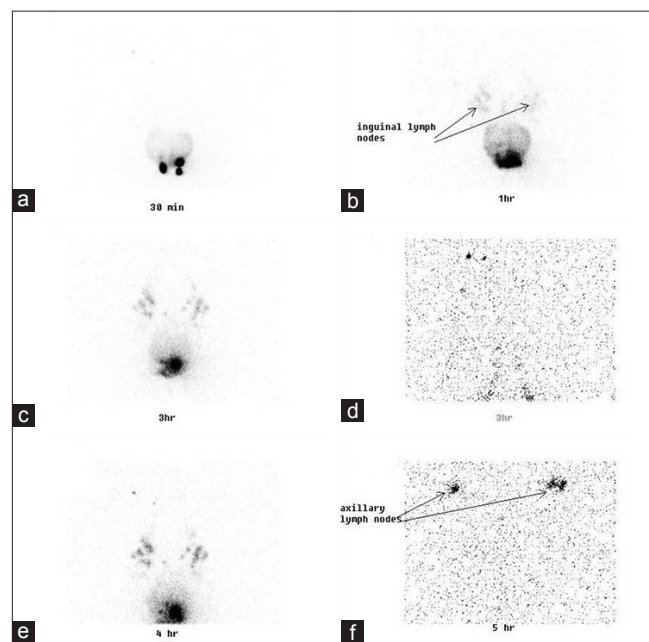


Figure 2: (a) Injection sites in scrotal skin. (b) Diffuse tracer distribution in the scrotal skin and faint uptake in bilateral inguinal lymph nodes. (c) Slightly more increased uptake in bilateral inguinal lymph nodes. (d) Faint uptake in bilateral iliac and retroperitoneal lymph nodes and also right axillary lymph nodes. (e) Tracer activity is slightly increased in the inguinal lymph nodes and faint tracer activity in the iliac and para-aortic lymph nodes. (f) Bilateral axillary lymph nodes

frequently caused by surgical excision and radiotherapy of the axillary or inguinal lymph nodes. Other common causes are infection, trauma or invasion by tumors. The incidence also increases if there has been surgery and radiotherapy plus episodes of cellulitis.^[3,7,8]

Lower limb scintigraphy in this case shows dermal backflow pattern in both lower limbs (right more than left) suggests congenital hypoplasia of lymphatics, however clinically lower limb edema has not been precipitated by now in this patient. Inguino scrotal reflux of tracer in lower limb lymphoscintigraphy and slow movement of tracer from scrotal skin in scrotal lymphoscintigraphy is possibly due to compensatory megalymphatics/congenital lymphectasia.

Visualization of bilateral axillary lymph nodes in lower limb lymphoscintigraphy and in scrotal lymphoscintigraphy without visualization of liver suggests aberrant lymphatic pathway, possibly secondary to malfunctioning/nonfunctioning thoracic duct.

A case of almost similar lymphoscintigraphic findings has been previously reported which presented as lymphedema of scrotum and right proximal thigh precipitated by a series of insults, i.e. trauma, surgery and infection. In this case subsequent surgery resulted in exacerbation of lymphedema. The author emphasizes that identification of primary lymphedema is important when operative intervention is contemplated to minimize disruption of existing lymphatics.^[9]

The inguinoscrotal reflux of lymph in the cases of primary as well as secondary scrotal oedema has also been previously reported as lymphangiographic^[10] as well as lymphoscintigraphic finding.^[11,12] Various type of successful reconstructive surgery has also been reported in cases of primary scrotal lymphedema but there is need of selecting and individualizing appropriate surgical procedure according to the individual's need and pathogenesis of lymphedema.^[6,12-14]

CONCLUSION

Lymphoscintigraphy is a simple, cost effective and repeatable technique to understand associated pathophysiology of Lymphedema. The case we present demonstrates congenital

variation and adaptability of lymphatic system, it also emphasizes the utility of lymphoscintigraphy in diagnosing primary lymphedema and understanding its pathogenesis, which direct the further management plan. It is important to carefully interpret lymphoscintigraphy as it can give clues of pathogenesis of lymphedema which helps in differentiating primary and secondary lymphedema, planning their treatment and predicting outcome.

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