CASE REPORT

Doxycycline sclerotherapy for post-traumatic inguinal lymphocele in a child

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SUMMARY

Lymphoceles are abnormal collections of lymphatic fluid caused by a disruption in the lymphatic channels and leakage of lymph. This most commonly occurs after surgical procedures, but occasionally lymphoceles may be the result of trauma, more commonly penetrating trauma. Lymphoceles resulting from blunt trauma are rare in both adults and children. In the adult population, there are few published case reports, and management principles vary. To date, there are no reports of traumatic lymphoceles in the paediatric population, and therefore there is no precedent for treatment. Here, we report the case of a young boy who developed an inguinal lymphocele from a bicycle handle bar injury which was successfully treated with doxycycline sclerotherapy.

BACKGROUND

A lymphocele is defined as an abnormal lymphatic fluid collection in a cavity that is not lined by epithelium. It is typically the result of lymph leakage caused by disruption of lymphatic vessels by trauma or surgery. Lymphoceles as complications from radical pelvic node dissection, saphenous vein harvest or kidney transplantation have been well described. Traumatic lymphoceles appear to be rare, and specifically those caused by blunt trauma are even less common. Because lymphatic fluid contains no platelets, once injured, lymphatic vessels are prone to leak. It can take up to 48 hours for lymphatic vessels to seal and even weeks to regenerate. More often, these occur extraperitoneally, as intraperitoneal collections can be absorbed more readily by the peritoneal surfaces.

Several adult cases of post-traumatic lymphocele have been reported, but there are no published paediatric case reports. The first case report was published in 1993. A 73-year-old woman developed medial thigh swelling after undergoing knee arthroscopy, which involved the use of an upper thigh tourniquet. She had 300 cc of clear fluid aspirated, which was consistent with lymph, and ultimately required multiple aspirations over a 12-week period prior to resolution of the collection.¹ A case report published in 1994 described a 39-year-old man who was crushed against a pillar by a forklift. He was asymptomatic and therefore no intervention was pursued. There were no long-term outcomes reported.²

In 1995, a 35-year-old man sustained blunt trauma to his medial left thigh from the butt of a rifle. Over the course of the next few weeks, his

thigh swelled and became painful, with no evidence of penetrating wound or fluctuance. This occurred during a war in Central Africa, and therefore imaging was very limited given lack of resources. A parachute surgical team elected to explore the area of swelling, and a thick-walled sac was found under the vastus medialis, which was drained and partially excised. His recovery was uneventful.³

Another report published in 1995 describes a 52-year-old man with alcoholic liver disease, chronic renal failure, congestive heart disease and diabetes mellitus, who was kicked in the groin. He presented 2 weeks later with a 20×12 cm non-pulsatile inguinal mass. Ultrasound revealed a loculated, fluid-filled cystic structure, which was confirmed to be lymphatic in origin by technetium sulfur colloid lymphoscintigraphy. Surgical resection and drainage was attempted, but the collection recurred. It was then treated with tetracycline sclerotherapy with resolution of the collection and without further complications.⁴

Finally, in 2004, a 23-year-old woman presented with a right subclavicular lymphocele, which she already had for 4 years and had occurred after a domestic dispute with her husband. On presentation, she was found to have a 6.9 cm collection between the pectoralis major and minor muscles, close to the subclavian vessels. She underwent percutaneous drainage twice but ultimately required surgical drainage due to recurrence.⁵

These case reports demonstrate the significant variability in aetiology of lymphoceles occurring after blunt trauma in the adult population, with similar disparity in management. In the paediatric population, there have been no reports of traumatic lymphoceles. However, lymphoceles do occur in the paediatric population, as a complication from paediatric kidney transplant or those associated with excision of congenital lesions such as lymphatic malformations, and management principles may be inferred from these aetiologies.

CASE PRESENTATION

A 14-year-old boy presented to his paediatrician 4 days after a bicycle accident in which the child's bicycle hit a rock causing him to fall forward onto his handlebars. He and his mother initially noted extensive bruising from his right groin down to his knee. There was initially a 3 cm swelling noted over his right groin that was tender to palpation, but ultrasound evaluation demonstrated only soft-tissue oedema without vascular injury or pseudoaneurysm. Over the ensuing weeks, the pain and

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Figure 1 Right inguinal lymphocele. Appearance at initial presentation.

bruising resolved but the mass-like swelling persisted and began to increase in size. A month after the injury, the boy returned to his paediatrician for follow-up, where an obvious fluid-filled inguinal mass was present, and he was referred to paediatric surgery for further evaluation.

On initial examination, a 9 cm non-pulsatile, non-tender, compressible mass was present in the right groin ([figure 1](#)). There was no erythema or drainage. The mass was not reducible. The remainder of the physical exam was normal, specifically the abdominal, urological and vascular examinations. An MRI study including arterial and venous phase postcontrast images was obtained to further evaluate the mass and its relationship to nearby structures. This MRI demonstrated a T2 hyperintense, T1 hypointense cystic mass within the right groin measuring 5.5cm anterior-posterior $\times$ 6.6cm medial-lateral $\times$ 10.7cm cranial caudal ([figure 2A–E](#)). The mass had minimal peripheral enhancement, and its posterolateral margin was in contiguity with the boy's right superficial femoral neurovascular bundle. The appearance was felt to be consistent with a lymphocele. Placement of a pressure garment and a period of observation were suggested.

TREATMENT

There was minimal improvement with compression alone, and 10 weeks after the injury, the patient underwent ultrasound-guided drainage of the cystic mass. Approximately 140 mL of fluid ([figure 3](#)) was aspirated through an 18-gauge Angiocath until the entire fluid-filled mass had completely collapsed. His thigh was wrapped with a compression dressing which was continuously worn. Analysis of the aspirated fluid

Figure 2 (A–E) MRI demonstrating a 5.5 cm AP $\times$ 6.6 cm ML $\times$ 10.7 cm CC, simple fluid-filled structure with no internal architecture and a thin-enhancing border, consistent with a lymphocele (arrows). T2-weighted image (fat suppressed, simple fluid is bright) (A), T1-weighted image (fluid is isointense to muscle) (B), fat suppressed T1 precontrast (C) and postcontrast images (D). Note the relationship of the lesion to the femoral neurovascular bundle (arrowhead, D). Coronal postcontrast image of lymphocele (arrow) demonstrating relationship to medial thigh (E).

Figure 3 Straw-coloured fluid aspirated, grossly consistent with lymphatic fluid. Fluid analysis revealed 1620 leucocytes, and 97% of these were lymphocytes.

demonstrated 1.62×10^9 /L leucocytes per microlitre of fluid, and 97% of these cells were lymphocytes, confirming this fluid to be lymph.

Three weeks later, the right inguinal swelling returned and therefore repeat aspiration and doxycycline sclerotherapy was recommended. In the operating room, an 18-gauge Angiocath was used to aspirate the lymphocele cavity, and a mixture of doxycycline (10 mg/mL) and bupivacaine (0.25%) was injected into the cavity. The doxycycline solution was left in place for approximately 40 min and then aspirated before a compression dressing was again placed.

OUTCOME AND FOLLOW-UP

He underwent aspiration with sclerotherapy, in similar fashion, every 2 to 4 weeks, to complete four subsequent treatments, with a smaller lymphocele collection noted each time, until there was no evidence of recurrence (figure 4).

DISCUSSION

Lymphocele formation after paediatric kidney transplantation has decreased in frequency during the past decade. Since 2006, the incidence has dropped from greater than 10% to between 3% and 5%.^{6,7} This is theorised to be due to limited and more accurate retroperitoneal dissection. One retrospective review identified older age, transplant prior to 2006, male gender and body mass index $>95\%$ as independent risk factors for developing a post-transplant lymphocele, which was found to reduce 1-year graft survival by 10%.⁶ There is no consensus on the best treatment, but typically patients undergo aspiration

Figure 4 Appearance of right inguinal region after doxycycline sclerotherapy shows resolution of the lymphocele with minor atrophic changes of the overlying skin.

with or without sclerotherapy and rarely surgical intervention. Management tends to be simply surgeon preference, as there is no accepted standard of care.

Traumatic lymphoceles can mimic lymphatic malformations, which are congenitally dilated lymphatic channels and cysts that do not communicate with the normal lymphatic or venous system. Classified as macrocystic or microcystic, these congenital lesions occur in 1/2000–4000 births, with no gender bias. While there is no consensus definition, macrocystic lesions are usually greater than 2 cm and are amenable to aspiration. Most (up to 75%) are found in the cervicofacial region, and the rest in the extremities. They typically present before age two as solitary or multifocal fluctuant masses with normal overlying skin. At ultrasound, macrocystic lesions are compressible, anechoic cysts with thin septations, while microcystic lesions consist of tiny cavities that might produce a hyperechoic, solid appearance. At MRI, these lesions should follow fluid signal on all sequences, with notably no intraluminal enhancement, filling defect or thrombus to suggest a vascular malformation (though venous and vascular

malformations can coexist in the same lesion, which is termed a 'venolymphatic' malformation).^{8,9}

Historically, operative drainage or resection was the gold standard for treatment of lymphatic malformations, but recurrence rates and complications were frequent: incomplete resection, scarring and local nerve damage occurred in 12.5%–44%.⁸ Sclerotherapy, however, has demonstrated similar outcomes at 1 year, with reduced morbidity.¹⁰ Sclerotherapy is typically performed using one of a variety of sclerosing agents by instilling 50% of the volume of aspirated lymph under ultrasound guidance. A catheter may be left for larger lesions or if repeated rounds of sclerotherapy are anticipated. One retrospective review noted reduced rate of recurrence or need for additional procedures if a drainage catheter is left to suction for 3 days.¹¹

No optimal sclerosant has been recommended. The most frequently used agents include pingamycin, ethanol, OK-432, bleomycin, ethanolamine oleate, polidocanol, sodium tetradecyl sulfate and doxycycline. Instillation of a sclerosant is thought to cause endothelial damage, followed by inflammation, thrombotic vascular occlusion and ultimately regression and resolution of the vascular malformation or other lesion requiring sclerotherapy. Complications include pain and the sequelae of sclerosant extravasation including skin necrosis, cellulitis or scarring. Extensive treatment of lesions or large volume extravasation can lead to compartment syndrome or permanent nerve damage. Phrenic or recurrent laryngeal nerve injury may occur when these agents are used in the cervicofacial region. Anecdotal complications include hypoglycaemia and metabolic acidosis in neonates.¹²

Learning points

- ▶ Lymphoceles resulting from blunt trauma are rare in both adults and children.
- ▶ In the adult population, there are few published case reports, and management principles vary.
- ▶ There are no reports of traumatic lymphoceles in the paediatric population, and therefore there is no precedent for treatment.
- ▶ Based on the presented case and the available literature, sclerotherapy with doxycycline appears to be an attractive option for the treatment of persistent post-traumatic or postoperative lymphoceles in children.

Doxycycline is more frequently used in the paediatric population due to its accessibility and safety profile. There have been no significant complications reported. The exact mechanism is unknown, but it likely causes collagen and fibrin deposition, leading to adhesions and fibrosis. Interestingly, macrocystic lymphatic malformations, similar to the traumatic lymphocele described in the current case, respond very well to sclerotherapy, with resolution rates approaching 100%.^{8,12}

Contributors AWH contributed through conception, drafting and revising the work and is accountable for all aspects related to the accuracy or integrity of the work. JJT contributed through acquisition of data. BLG contributed through conception, acquisition and analysis of images and figures. RAC, senior author, contributed through conception, acquisition, drafting, and final revisions of the work and is accountable for the accuracy and integrity of the final manuscript.

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