CASE REPORTS

HOST-REACTION FOLLOWING SILICONE TUBE IMPLANTATION FOR LOWER LIMB LYMPHOEDEMA MIMICKING IMPLANT INFECTION

Eduardo Silva[1*], Celso Nunes[1], Vânia Constâncio Oliveira[1], Joana Silva[1], Pedro Lima[1]

[1] Department of Angiology and Vascular Surgery, Centro Hospitalar e Universitário de Coimbra (CHUC), Coimbra, Portugal

* Corresponding author: ?????????????

Abstract

Introduction: *Silicone tube implantation in lymphoedema reduces symptoms by improving fluid drainage. Although there are descriptions of implant host reaction that can be misdiagnosed as graft infections these are scarce.*

Report: *A 34 year old female with lymphoedema of the lower limb, underwent silicone tube implantation. Ten months after surgery, the patient presented with fever and dermatolymphangioadenitis of the limb. Ultrasound suggested an abscess surrounding the tubes. Clinical improvement was achieved after a 6-day cycle of meropenem. She was discharged under oral cefuroxime and clindamycin for one week. After 1 month, CT-angiography was performed showing only residual inflammation surrounding the tubes, the patient was asymptomatic and limb diameter was normal.*

Conclusion: *Sudden onset and improvement of the patient's condition after a short cycle of antibiotics without the need of tube removal supports a host-like reaction rather than an actual infection. Doctors should be aware of such complications avoiding unnecessary procedures.*

Keywords: *Lymphoedem, Silicone Tube Implantation, Host-reaction, Dermatolymphangioadenitis, Graft Infection*

INTRODUCTION

Lymphoedema is a chronic condition that causes oedema and progressive skin changes due to accumulated fluid within the interstitium and fibro-adipose tissue, which would normally be drained by the lymphatic system, caused by its obstruction, injury or congenital malformation[1]. Despite its high prevalence, there is a lack of surgical options, as well as a lack of information in the literature about the management of complications arising from such procedures.

In the last century there were descriptions of the use of tubes to help drain the accumulated fluid in the limbs[2-4], although its use was abandoned. More recently, Olszewski et al. have published a series of papers regarding the use of microperfurated hydrophobic silicone tubes tunneled through the subcutaneous tissue of the limbs, for the treatment of chronic lymphoedema with promising results, helping reduce oedema and skin changes on the affected limb[5-7]. The same authors have suggested that a host reaction (dermatolymphangioadenitis (DLA)) may occur in some patients which should not be considered an implant infection[5-7].

CLINCAL CASE

A 30 year old, female patient was referred to our hospital in 2017, due to chronic oedema of the left lower limb, that had started 10 years prior, worsening after her second pregnancy (G2P2). She worked as a factory worker and did

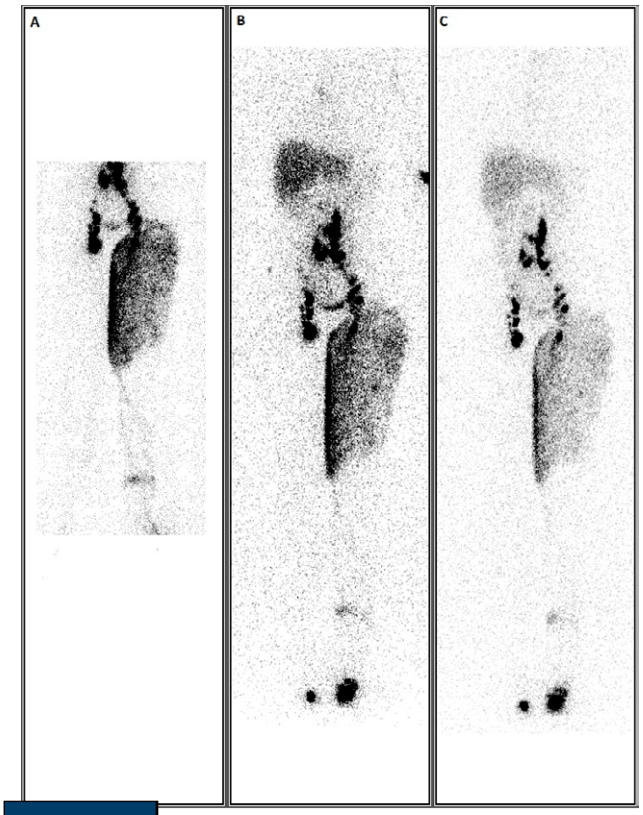

Figure 1 *Lymphoscintigraphy with 99mTc showing accumulation on the left thigh at (A) 2 hours, (B) 5 hours and (C) 6 and a half hours, after administration of the radiolabel.*

not have relevant medical or family history. The workup study (2017) showed neither signs of venous thrombosis on the limb nor signs of venous pelvic compression (CT-scan) and ultrasound showed competence of both great and small saphenous veins, without signs of insufficiency or reflux, yet there was a diffuse subcutaneous infiltrate throughout the entire limb compatible with lymphoedema which was confirmed by lymphoscintigraphy (Figure 1), hence she was diagnosed with lymphoedema praecox.

The patient was treated with conservative measures (compression stockings and physiotherapy to increase lymph flow), and was to be evaluated 6 months later, yet follow-up was lost due to missing multiple appointments.

In 2021, at 34 years of age, the patient showed to our emergency department with fever (38,6ºC) and inflammatory signs of the left lower limb that started three days before. She stated that the conservative measures prescribed in 2017 did not improve her condition, thus she went to another institution in a different country, ten months prior (2020), having undergone lymphatic surgery with placement of silicone tubes along the length of the limb, which had reduced the perimeter of the leg and improved her symptoms.

At the emergency department blood results showed elevated C-Reactive Protein (CRP) (18,9 mg/dL) with leukocytosis (16,1 x10^9/L) and ultrasound of the affected limb revealed a graft along the medial aspect of the thigh and leg with a collection surrounding it, suggestive of an abscess (Figure 2).

A diagnosis of dermatolymphangioadenitis of the left lower limb was made and the hypothesis of an implant infection as the cause of the problem was raised. The patient was admitted under broad spectrum antibiotics (IV meropenem (1000mg, q8hr)).

We contacted the institution where the surgery was performed to obtain information about the procedure and their experience with post-procedure complications, namely infections and the need for implant removal, yet a more conservative approach was suggested.

After a 6-day course of antibiotics the patient showed clinical improvement, without fever, absence of inflammatory signs on the limb and reduced perimeter on the thigh and leg. Her blood results showed decreased CRP (6,95 mg/dL) without leukocytosis (7,8 x10^9/L) and her

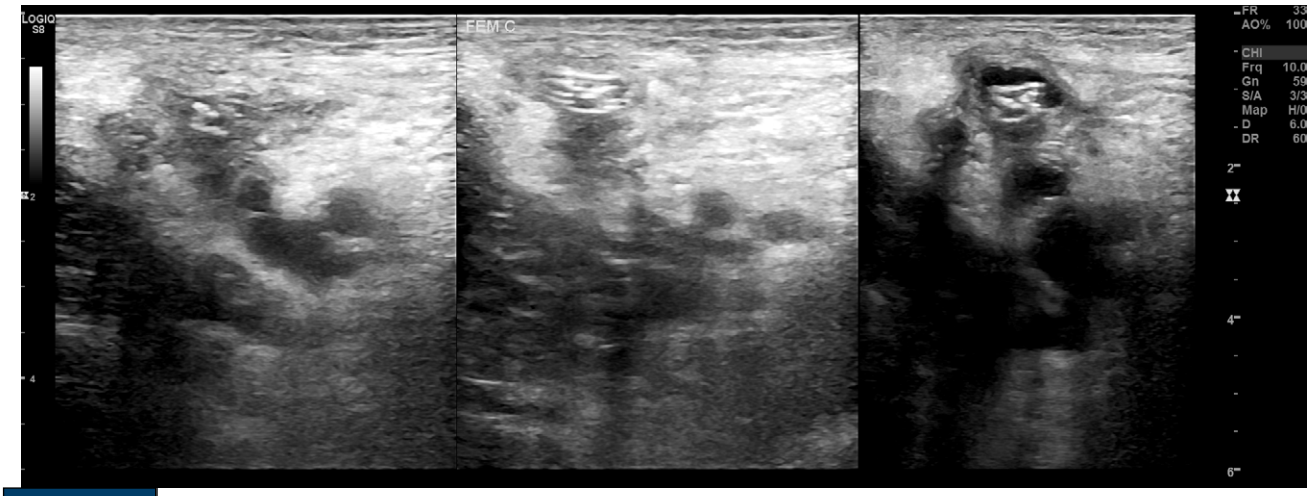

Figure 2 *Ultrasound performed at admission in the emergency department, revealing a collection surrounding the tubes.*

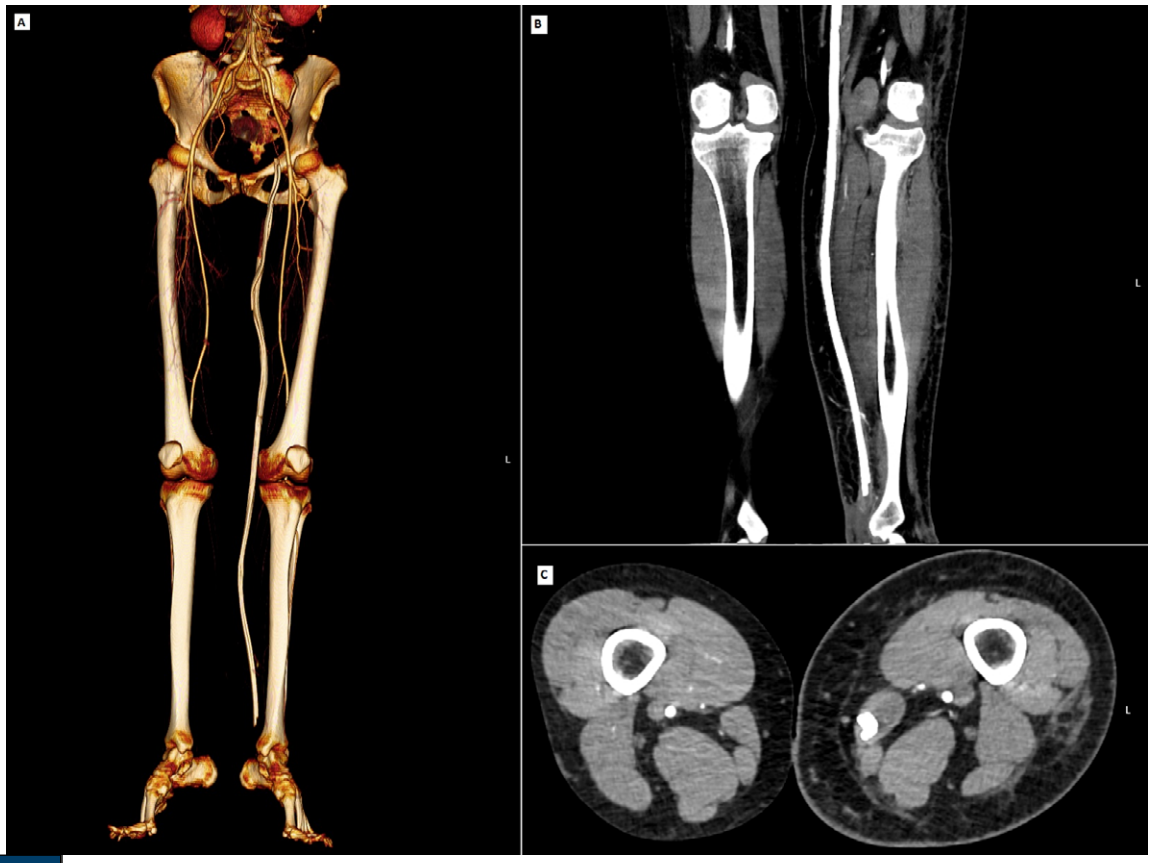

Figure 3

CT angiogram 1 month after the acute event. (A) 3D model reconstruction with silicone tubes implanted along the medial aspect of the left lower limb. (B) and (C) heterogeneity of the tissues surrounding the implant at the thigh level, suggestive of a residual peri-implant inflammation.

blood cultures were negative. The patient was discharged maintaining an additional 8 days of oral cefuroxime (500mg, q12hr) and clindamycin (150mg, q6hr).

At 1 month follow-up, the patient was asymptomatic, despite still presenting mild lymphoedema of the affected limb, which did not interfere on her daily activities. CT-angiography showed residual inflammation of the tissues surrounding the tubes at the thigh level, without signs of organized abscesses or local collections (Figure 3).

DISCUSSION

The use of microperfurated hydrophobic silicone tubes tunneled through the subcutaneous tissue provides a simple surgical alternative to complex procedures, such as vascularized lymph node transfer or lymphovenous bypass, with satisfactory results, by creating artificial pathways to help draining the fluid accumulated at the interstitial space to unobstructed areas where normal lymph absorption can take place[5]. This technique was attempted in the last century but was abandoned due to infections[3], yet nowadays medical grade nontoxic silicone tubes are available, which are considered biologically inert. Not only that but bacteria do not adhere to the silicone surface and their hydrophobic properties prevent obstruction by ingrowing fibrous tissue[5-7].

Olszewski et al., have proposed that along with the drained lymph, bacteria may also be transported through the silicone tubes, just like as under normal conditions bacterial cells are transported to the regional nodes through the lymphatics. As large bacterial loads are transported, it can cause a host reaction to develop in the form of DLA which should not be confused with an actual implant infection[5-7].

On the same series, the authors mention using a prophylactic protocol of long-term penicillin, due to recurrent episodes of dermatolymphangioadenitis in these patients[5-7]. We did not have information regarding the use of pre- and post-operative antibiotics at the time of surgery for this patient, yet she was not under long-term prophylaxis before and after the procedure. The need for chronic antibiotics, with the associated risk of dermatolymphangioadenitis, may pose as a limitation for this technique, yet there are no randomized controlled trials showing that long-term prophylaxis reduces the risk of DLA.

The case reported supports the idea of a host reaction, as 10 months after surgery the patient developed acute dermatolymphangioadenitis of the lower limb with fever and elevated inflammatory markers, yet after a short cycle of antibiotics the clinical condition improved without need of surgery to remove the implants nor invasive procedures to drain the collection initially misdiagnosed as an abscess. Her blood cultures were negative and CT-

angiography showed no signs of collections at 1 month, thus emphasizing the importance of being aware of host reactions as conditions that can mimic implant infections in these types of procedures, avoiding unnecessary and incorrect treatments.

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