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Arteriovenous shunt graft ulceration with sinus and graft epithelialization ☆,☆☆

Pooja Singhal [a], Adriana Luk [b], Amar Ibrahim [a], Minhui Laura Kim [a], Jagdish Butany [a,*]

[a] *Department of Pathology, Toronto General Hospital, University Health Network, Toronto, Ontario, M5G 2C4*
[b] *Division of Cardiology, Department of Medicine, University of Toronto, Toronto, Ontario, M5G 2C4*

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Abstract Arteriovenous fistula and grafts are used as access sites for patients with chronic kidney disease and are prone for complications. Stent grafts are used to treat access site complications. We report a rare and unusual finding of epithelialization of the sinus tract and the lumen of a polytetrafluoroethylene graft, following ulceration of the overlying skin.

1. Introduction

Arteriovenous fistula (AVF) and grafts (AVG) are used as access sites for patients with chronic kidney disease (CKD). Both are prone to complications, which include thrombosis, graft stenosis, infection, pseudoaneurysm, aneurysm, graft disruption and hemorrhage. Stent grafts are used to treat access site complications such as pseudoaneurysm and stenosis. These stent grafts are also associated with complications which include stent migration, fracture, erosion, hemorrhage, and rupture. We present an unusual finding of epithelialization of a sinus tract as well as the lumen of the stent graft, following ulceration of the overlying skin.

2. Case report

A 48-year-old man with a history of CKD secondary to bilateral childhood hydronephrosis, presented with a malfunctioning left AVG (partial loop graft anastomosed from the brachial artery to the axillary vein) used for hemodialysis. Comorbidities included polyarthritis secondary to calcium pyrophosphate dehydrate disease (CPPD) and cigarette smoking. Five years prior he had an AVG removed from his left arm secondary to pseudomonal infection. His current AVG implanted 4-years prior, was found to have low flow rates during dialysis 3 years post implantation. Over the following year, he had five separate procedures involving the administration of tissue plasminogen activator (tPA), and balloon angioplasty for moderate stenosis and implantation of a Flair® endovascular stent graft 9 months prior to presentation. A linear ulcer measuring 0.5 cm in length occurred on the skin surface, and a tunneled vascular catheter was inserted. Empiric antibiotics were initiated, and medications included calcium, vitamin D and iron replacement. Prior to surgery, he was hemodynamically stable and

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* Corresponding author at: Dept. of Pathology 11th floor, 11E-420, Toronto General Hospital, 200 Elizabeth St., Toronto ON, Canada M5G 2C4. Tel.: +1 416 340 3008; fax: +1 416 340 4213.
E-mail address: jagdish.butany@uhn.on.ca (J. Butany).

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physical examination was unremarkable. Blood work was non-contributory with the exception of a creatinine of 805 μmol/L. He underwent excision of the graft without complications. Subsequent wound and blood cultures were negative. He was discharged home in stable condition.

3. Pathology

The surgically excised specimen was a segment of synthetic graft (expanded polytetrafluoroethylene – ePTFE) with overlying skin. The graft was 9.5 cm long with an outer diameter of 0.7 cm at one end and 1.0 cm at the other end. The overlying skin was dark, 5.6 cm long, had a width of 2.1 cm and showed 5 areas of ulceration. The ulcers were of irregular shape and with a width ranging from 0.3 to 1.2 cm (Fig. 1). Multiple sections showed marked perigraft fibrosis. The adventitial and the intimal surfaces of the ePTFE graft showed a marked chronic inflammatory cell infiltrate characterized by lymphocytes, macrophages, plasma cells and multinucleate giant cells. The graft material showed abundant macrophages and lymphocytes as well as fibrin in the interstices of the graft material. Old puncture sites were seen in the graft on the subcutaneous and deep surfaces. These were associated with defects in the synthetic graft, bridged by mature fibrous tissue (Figs. 2 and 3). The intimal surface of the graft showed marked neointima formation leading to luminal stenosis and areas of old thrombosis (Fig. 4). The skin overlying the graft was ulcerated and showed a sinus tract lined by keratinizing squamous epithelium extending from the overlying skin into the lumen of the synthetic graft. The lumen of this tract showed fragments of ePTFE graft and keratin debris (Fig. 5).

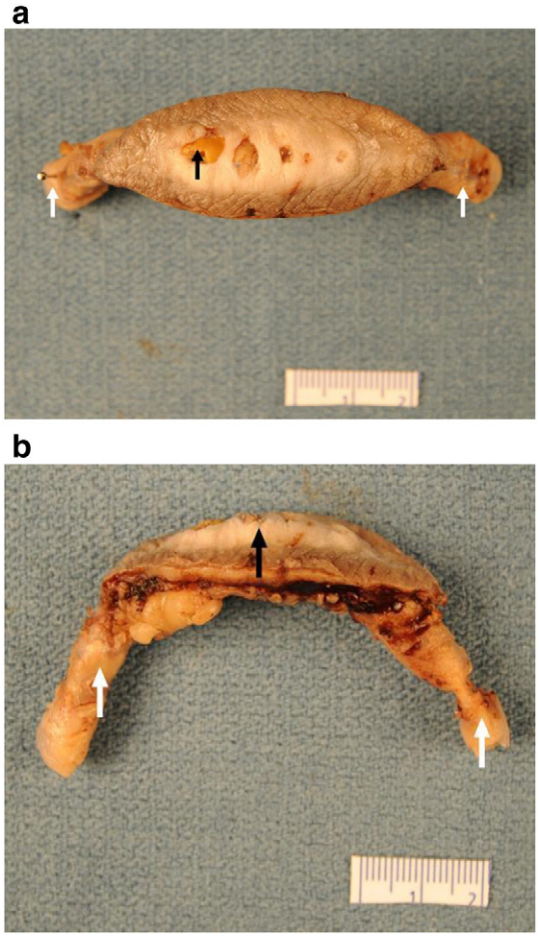

Fig. 1 Gross picture of arteriovenous graft showing PTFE graft (white arrow) with overlying ellipse of skin showing (a) ulceration (black arrow) and (b) pale and edematous cut edge of the skin.

4. Discussion

End stage kidney disease (CKD) is a common problem. Multiple types of access sites have been used to treat CKD which include an AVF, AVG (synthetic PTFE graft, autologous vein grafts) and the tunneled cuffed catheter, but they are prone to complications, which include thrombosis, graft stenosis, infection, pseudoaneurysm, aneurysm, graft disruption and hemorrhage.

Repeated cannulation of an AVG for hemodialysis can cause disruption and damage of graft materials, as well as the subcutaneous tissue. These changes lead to weakening of the wall and subsequent aneurysm or pseudoaneurysm formation, which compromises blood flow and can lead to overlying skin necrosis [1]. Ultimately, spontaneous bleeding and acute graft rupture through the skin can occur [2]. Pseudoaneurysms and stenotic lesions are treated with percutaneous balloon angioplasty (PTCA) and stent

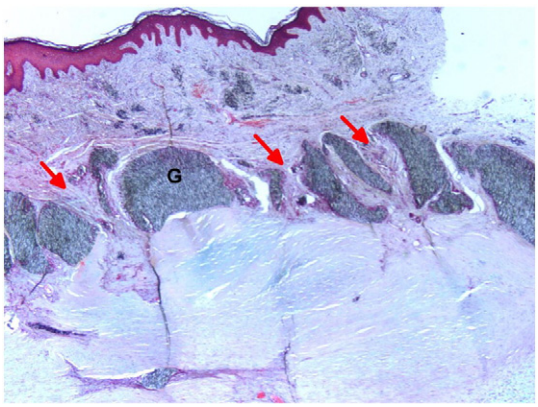

Fig. 2 The PTFE graft (G) below the skin shows multiple old needle puncture sites (red arrow) bridged by fibrous tissue (Movat Pentachrome stain; original magnification ×2.5).

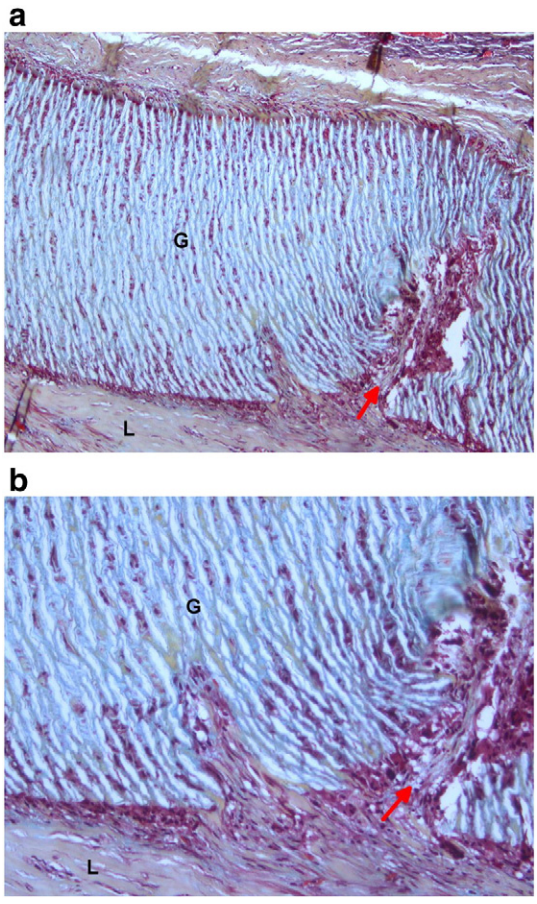

Fig. 3 (a) Section from deep surface of the PTFE graft (G) shows needle puncture site with the growth of fibrous tissue (red arrow). (b) Higher magnification of (a). L: lumen of the graft. Movat Pentachrome stain; original magnification (a) ×10; (b) ×20.

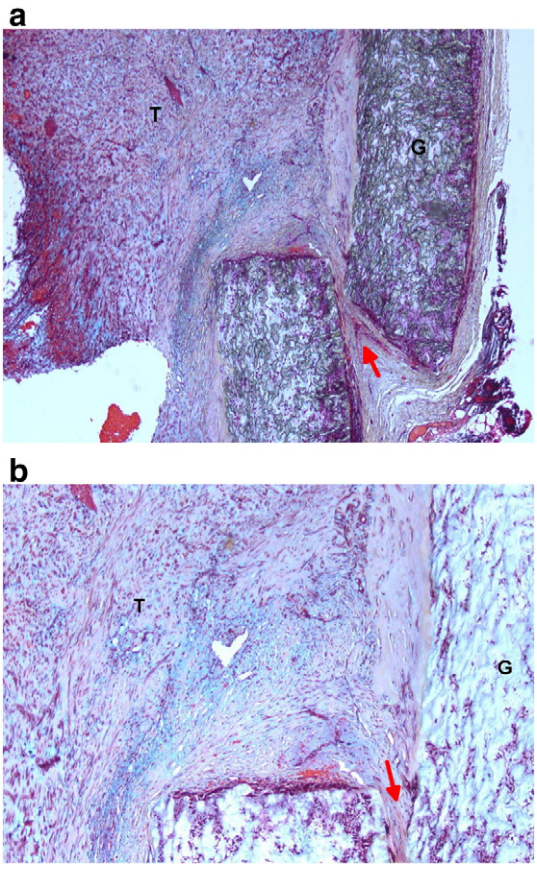

Fig. 4 (a) The PTFE graft shows needle puncture wound with fibrous tissue ingrowth (red arrow) and organizing thrombus (T) in the lumen of the graft. (b) Higher magnification of (a). Movat Pentachrome stain; original magnification (a) ×10; (b) ×20.

placement. The ultimate treatment is a revision of the shunt or placement of a new AV shunt.

Various studies have demonstrated that stent grafts maintain access patency more effectively than angioplasty [3]. Four self expanding stent grafts (Wallgraft®, Viabahn® stent graft, Fluency Plus® stent graft and Flair® stent graft) are available for the treatment of access related issues, though only the latter has been approved by the FDA. The Flair® stent graft is composed of a nitinol stent, covered by ePTFE. These stent grafts are covered with a relatively inert polymeric material, which acts as a barrier to migration of smooth muscle cells, restricts intimal hyperplasia and protects the diseased and thrombogenic wall from the luminal blood flow.

Stent grafts are also associated with complications such as failure and restenosis (due to intimal hyperplasia and chronic inflammation). Bleeding may occur due to migration, erosion and stent fracture. Migration of the stent occurs due to continuous growth and increasingly tortuous anatomy of pseudoaneurysms, improper fixation, early puncture, chronic inflammation and low-grade infection. Lastly, stent fracture, skin erosion and infection can occur as multiple punctures from each dialysis session increase the risk of these complications [4]. When stent fracture has occurred, the stent may protrude through the skin, and lead to chronic infection [2]. Studies show that patients with stent grafts are at three fold higher risk of infection, compared to bare metal stents or angioplasty alone [5,6].

In our case, the patient developed five linear ulcers 9 months after the placement of the Flair® endovascular stent graft. Skin ulceration and erosion due to stent grafts are not a usual finding and has been reported with various types of stent grafts. Silas et al., found one case of infected skin ulcer, which developed one month after the placement of a Wallgraft® stent graft [7], and Asif et al., reported a case where the stent graft was found protruding through the skin and three cases of sepsis and cellulitis of skin overlying the stent graft [5]. In addition, other studies have reported cases of soft tissue infection associated with exposed stent graft or stent fracture [3,4]. Unlike these

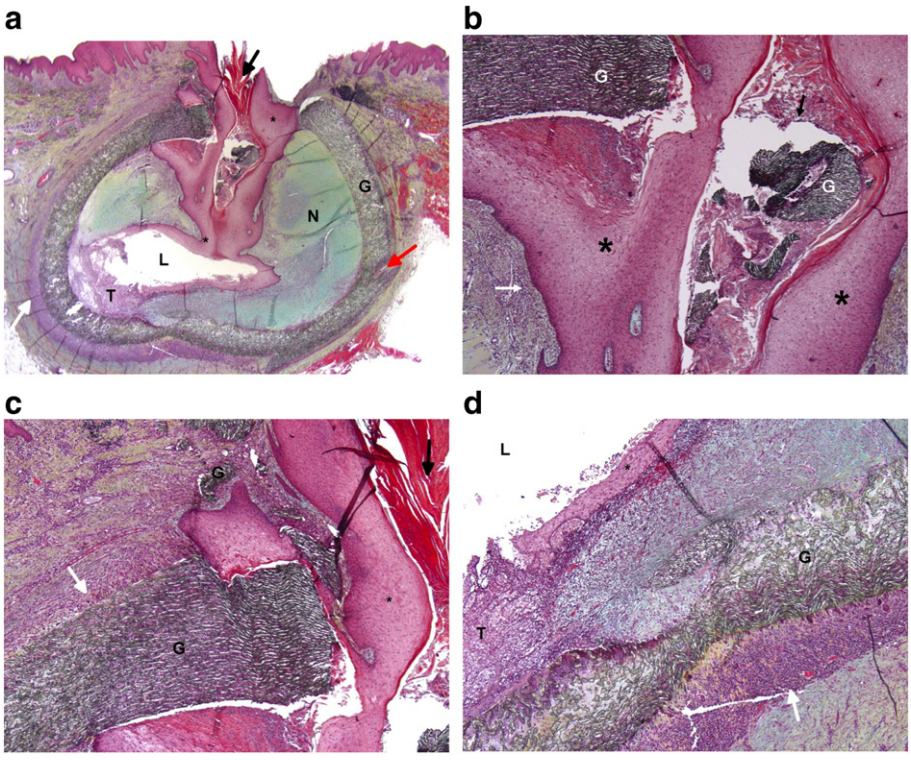

Fig. 5 (a) The microscopic section of PTFE graft (G) shows chronic inflammation on the adventitial surface of the graft (white arrow) with fibrous tract (red arrow), neointimal proliferation (N) and area of thrombosis (T). The sinus tract into the graft (black arrow) and the lumen of the graft (L) is lined by stratified squamous epithelium (asterisk) which is continuous with that of the skin. The mouth of the sinus tract is filled with keratin debris. (b) and (c): Higher magnification of (a) shows disrupted fragment of PTFE graft (G) in the sinus tract (black arrow) with epithelialization of the sinus tract (asterisk) and perigraft chronic inflammation (white arrow). (d) Epithelialization of the lumen of the graft (asterisk). a–d: Stain: Movat Pentachrome; (a) ×1.2; (b–d) ×5.

studies, we did not find an exposed stent or stent fracture, in fact our specimen did not include the stented segment. We did find marked chronic inflammation on both the intimal and adventitial surfaces of the graft along with a lymphocytic infiltrate into the graft. There was an unusual finding of sinus tract formation between the skin and the lumen of the stent graft and exceptionally rare, the sinus tract and the lumen of the stent graft were also epithelialized.

5. Summary

We report a rare and unusual finding of epithelialization of the sinus tract and the lumen of a PTFE graft, following ulceration of the overlying skin. We believe that epithelialization of the sinus tract resulted from the presence of chronic inflammation surrounding the graft and the presence of fragments of the ePTFE graft in the tract, which promoted chronic irritation and ongoing inflammation. In addition, ongoing inflammation may have occurred due to infection of the graft, likely due to the presence of multiple skin ulcerations, the presence of significant synthetic materials and the repeated trauma to these materials. It is not possible to blame any of these factors.

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