Journal of Plastic, Reconstructive & Aesthetic Surgery (2013) 66, e321—e324

JPRAS
An International Journal of Surgical Reconstruction
www.JPRASurg.com

CASE REPORT

Particular catastrophic antiphospholipid syndrome, on the sole surgical site after breast reduction☆

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Received 2 April 2013; accepted 6 April 2013

MESH KEYWORDS
Mammaplasty;
Antiphospholipid syndrome;
Skin necrosis;
Negative-pressure wound therapy;
Skin transplantation;
Skin artificial

Summary A 20-year-old woman treated with vitamin K antagonist for antiphospholipid syndrome (APS) (pulmonary embolisms at age 15) was admitted for breast reduction after bridging therapy. At 2 days post-surgery haematomas appeared on the surgical site and anticoagulant therapy was withheld. She developed a skin and breast necrosis leading to the diagnosis of catastrophic APS. Despite medical treatment (anticoagulant therapy, corticosteroids and intravenous immunoglobulins) and surgery, necrosis continued. After 2 weeks of negative-pressure wound therapy (V.A.C.® Therapy™) the patient improved, mammary tissues were alive, well vascularised and budding. Breast reconstruction was then initiated. Artificial dermis graft (MatriDerm® 2 mm) was applied, and 3 weeks later the apposition of split-thickness skin graft on it. Six months later, results of the surgery were good and the patient was satisfied.

☆ **Meeting**: Communication: "Journée nationale des jeunes plasticiens" 9 March 2013, Paris, France.

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1748-6815/$ - see front matter
http://dx.doi.org/10.1016/j.bjps.2013.04.037

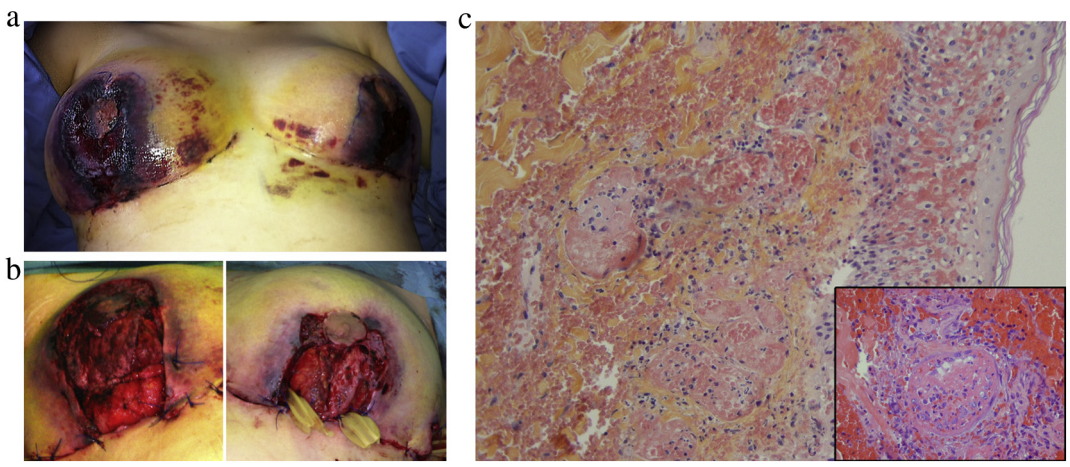

Figure 1 Panel a (left, above): Bilateral breast necrosis at day-10; Panel b (left, below): Breast Preoperative; Panel c (right): Histology: microvascular thromboses.

Antiphospholipid syndrome (APS) is characterised by venous, arterial, small-vessel thromboses or obstetric morbidity associated with persistent antiphospholipid antibodies (aPLs). The catastrophic APS (CAPS) affects <1% of patients with APS and is characterised by multiple small-vessel thromboses in multiple organs that can lead to death.[1]

In this case the occurrence of uncommon CAPS, with severe uncontrolled necrotic skin involvement, originated from the surgical site. Its management required classical treatments and for the first time the use of negative-pressure wound therapy for controlling the self-sustaining process, and an adaptation of reconstruction techniques to avoid further triggering the thrombotic and auto-immune processes.

A 20-year-old woman, with symptomatic macromastia (back pain and psychological consequences), consulted a surgeon for breast reduction. Her past medical history was significant for primary APS revealed by pulmonary embolisms at age 15. She was treated with long-term anticoagulation therapy by vitamin K antagonists (VKAs) since then.

Three days before surgery, she was admitted for bridging therapy for the perioperative period. VKAs were switched to low-molecular-weight heparin (Enoxaparin 7000 UI bid) with the preoperative injection 12 h before surgery. Breast reduction with 'dermal vault' technique was uneventful. The weight of mammary gland removed was 306 and 302 g for the right and the left breast, respectively. Anti-coagulation was reinitiated 12 h after surgery. Because of the occurrence of a clinically symptomatic haematoma of the left breast and the presence of significant bleeding (800 and 200 ml in the left and the right drain, respectively), anticoagulant therapy was withheld at day-2. The presence of an anaemia (haemoglobin 5.7 g dl^{-1}) required the transfusion of three units of packed red blood cells.

At day-5, the patient's clinical condition had markedly improved and bleeding was controlled (haemoglobin at 9 g dl^{-1}). Anticoagulation was carefully reintroduced initially with prophylactic dose of Enoxaparin (4000 UI qd), then intermediate doses at day-7 (4000 UI bid).

At day-10, the patient complained of pain in her breasts and bilateral and symmetrical areas of necrosis appeared, centred by the areola and the vertical scar (Figure 1a). Despite full-therapeutic-dose anticoagulation (Enoxaparin 7000 UI bid) rapid and severe necrosis occurred with a new area of necrosis surrounding the left drain. Laboratory parameters revealed an inflammatory syndrome and a thrombocytopaenia (86 g l^{-1}). Based on these data, a treatment by intravenous immunoglobulin (IVIg) was initiated (2 g kg^{-1}).

Because of unfavourable evolution, surgery revision was decided. Necrotic tissues were debrided, left breast haematoma evacuated and surgical haemostasis was performed (Figure 1b). Postoperative bleeding with severe anaemia (haemoglobin at 5.4 g dl^{-1}) required to withhold anticoagulation and to transfuse five units of packed red blood cells. At day-13, a second surgical revision was required to achieve haemostasis and debridement of remaining necrotic tissues.

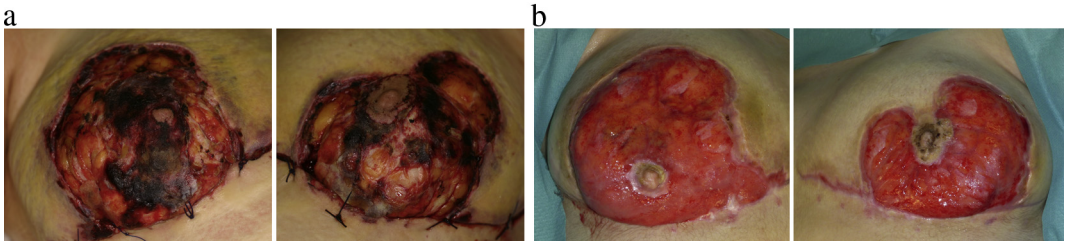

Figure 2 Panel a (left): Necrosis evolution, after second surgery revision; Panel b (right): Breast after two weeks of negative-pressure wound therapy.