Reconstructive complications of thrombophilia and keloid scarring: free flap surgery in a patient with mild protein S deficiency

Sir,

We wish to discuss the complications of free flap reconstructive surgery of the head and neck in a patient with mild protein S deficiency.

Protein S is one of the major inhibitors of coagulation, which results in the excessive generation of thrombin[1] and the condition can be autosomal dominant (inherited or acquired).[2] Symptoms vary, and the homozygous condition manifests at an earlier age.[3] Coagulation studies often show no abnormalities, and definitive diagnosis is made by assays for free and total Protein S.

A 38-year-old Nigerian woman presented to the oral and maxillofacial department at the Queen Elizabeth University Hospital in Glasgow in 2014. She was diagnosed with an oronasal/oroantral fistula and trismus secondary to operation and radiotherapy for adenoid cystic carcinoma of the palate, which she had had in 2013.

Two free flap operations to repair the fistula were unsuccessful and an obturator was provided. Subsequently, her main issue was trismus with a mouth opening of 3 mm, which obstructed maintenance of the obturator and oral hygiene.

In December 2018 she was diagnosed with extra-articular ankylosis of the left temporomandibular joint secondary to substantial formation of new bone in the pterygoid/ posterior maxillary region and a pathological fracture with viable underlying bone.

She was diagnosed with a mild protein S deficiency, showing low antithrombin activity and free Protein S of 48 IU/dL (compared with a normal range of 65-137). This caused a dilemma between the risk of flap failure and the risk of haemorrhage. The advice from Haematology was to "treat as aggressively as you are able to with low molecular weight heparin prophylaxis".

Further operation and its risks were discussed extensively, and a second opinion was sought with a proposal to release the bony ankylosis, excise the buccal scar, internally fix the pathological fracture, extract the molars, and reconstruct with a soft tissue free flap (including a small island of skin to be used as a monitoring buoy over the access wound to facilitate observation and prevent compression of the vessels).

It was also agreed with the paediatric department that extracorporeal membrane oxygenation would be available - a technique that been used previously to rescue free flaps.[4]

The operation took place in May, 2019.

Unfractionated heparin was given intravenously for 72 hours postoperatively - aiming for an activated partial thromboplastin time of 1.5 seconds - at a rate of 1000 units/hour. It was then switched to twice daily 40 mg low-molecular-weight heparin subcutaneously. Evacuation of a haematoma was necessary four days later, but the flap remained viable. Low-molecular-weight heparin was continued subcutaneously once daily for four weeks after discharge. Mouth opening has since improved appreciably to 1.5 cm.

While protein S deficiency is a rare condition, we hope that the report of this case may help people in future who are faced with similar problems.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patient's permission

Not applicable.

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Calcification of the fibular free flap pedicle, an unusual cause of neck and throat pain

Sir,

A 58-year-old man presented with painful swallowing and pain in his left submental area that hurt when turning his neck to the left, and was more noticeable during daily cycling. He had become aware of a bony lump in the area over the course of six months.

He had first presented with a T2 N0 M0 squamous cell carcinoma in the left tongue base and had concurrent chemoradiotherapy. He subsequently developed a left tonsillar squamous cell carcinoma T1 N2a MO and was treated with excision and left neck dissection in 2009-2010, which was followed by radiotherapy. He later developed Grade 3 Notani osteoradionecrosis of the mandible with trismus of

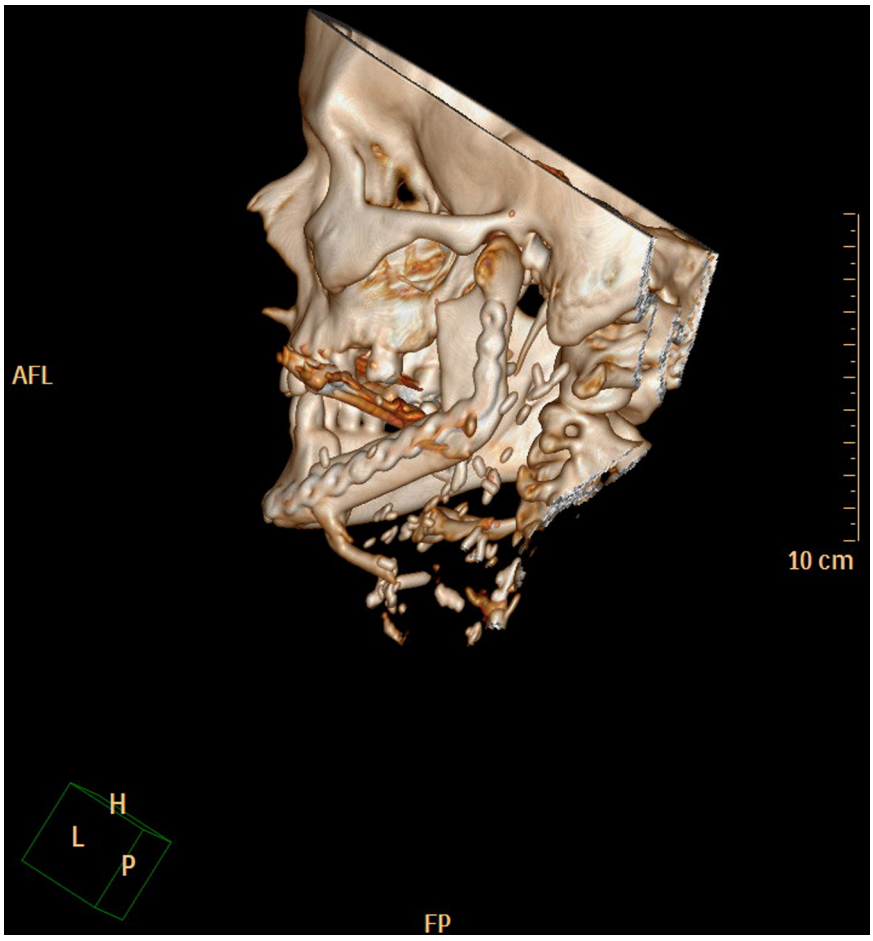

Fig. 1. Three-dimensional computed tomographic scan showing the left inferior pedicle extending towards the hyoid bone.

16 mm. He had been treated with tocopherol, pentoxifylline, topical antibiotics, and chlorhexidine.

He had a terrible quality of life as a result of the osteoradionecrosis, and in 2015, he had a segmental mandibulectomy with reconstruction with a fibular flap. The pain began after two years. He was investigated using computed tomography (Fig. 1), magnetic resonance imaging, and orthopantomogram, which showed an area of ossification under the lower border of the mandible. A barium swallow initially showed some slight pooling in the vallecula, which had improved on subsequent views a month later, after speech and language (SALT) exercises (Fig. 2).

Heterotrophic ossification of the pedicle has been reported between 4%–17% after reconstruction with a fibular flap and may be even higher (27%), particularly in younger patients and those who have had radiotherapy.[1] Taristano et al showed that dissection off the periosteum reduced ossification from 17% in one group using a standard technique (41 flaps) to 0 in the other group (20 flaps).[2] This study was, however, limited to only two-years' follow up. In the large study of fibular flaps by Mays et al, removal of the periosteum from the pedicle during operation increased the likelihood of damaging it.[3]

By the time heterotrophic ossification has begun to show symptoms many years later, as in our patient, the risk of damage to the pedicle is low because the flap has become incorporated into the surrounding tissues. Removal of this bony spicule can be done by reflecting a subplatysmal flap and excising it with standard osteotomy cuts.

This simple approach has eased this patient's symptoms and once again improved his quality of life after the complications of his oncological treatments. This case highlights some of the symptoms this condition can present concerning trismus, neck pain, odynophagia, and limited neck movement.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patient's permission

None required.

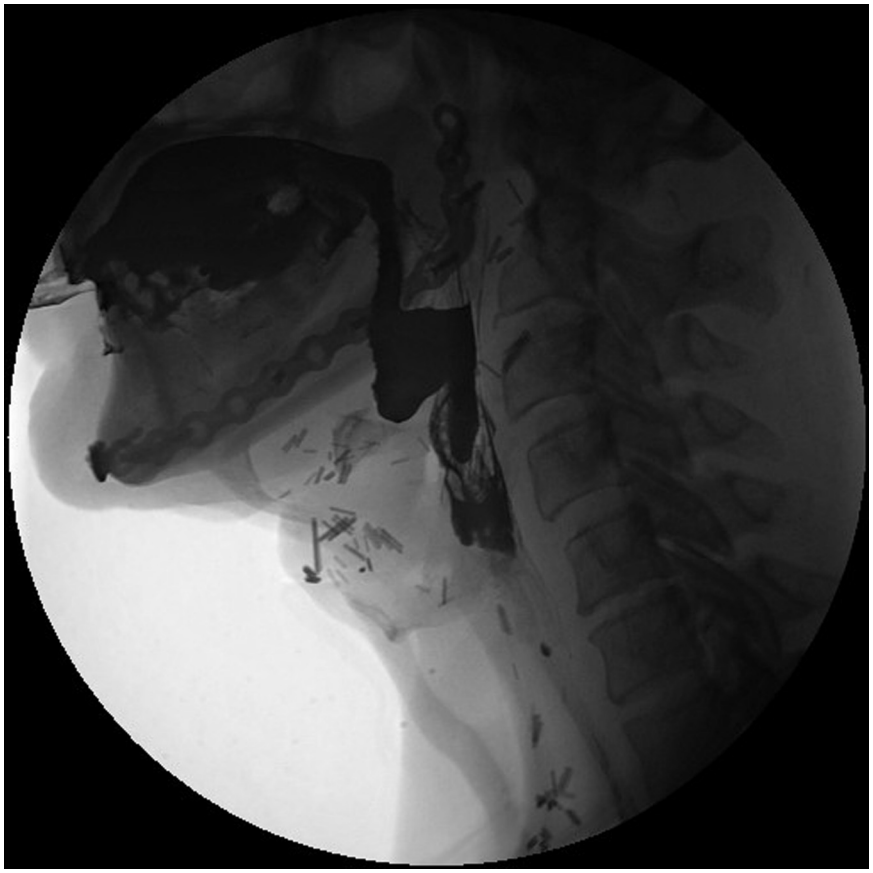

Fig. 2. Barium swallow.

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