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Sensory deprivation as a consequence of severe head and neck lymphoedema

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Abstract
We report a case of sensory deprivation that occurred as a consequence of progressive head and neck lymphoedema, following combined surgery and radiotherapy for squamous cell carcinoma. The management of head and neck lymphoedema is discussed and measures are suggested for improving the sensory deprivation experienced by the worst affected patients.

Key words: Lymphoedema; Head and Neck Neoplasms; surgery

Introduction

Lymphoedema is an increase in extra-cellular fluid as a result of lymphatic failure in the presence of a normal capillary filtration pressure.[1] As elsewhere, lymphoedema of the head and neck may be classified as primary or secondary. Primary lymphoedema of the head and neck has been reported on few occasions, it may be subdivided into lymphoedema congenita, praecox and tarda on the basis of the timing of its development. It is most frequently seen to coexist with upper limb lymphoedema suggesting a widespread congenitally determined lymphatic insufficiency.[1] Secondary lymphoedema may be localized to a particular part of the face or it may affect the whole of the head and neck. The condition arises when a critical number of lymphatics become blocked by scarring infection, inflammation or tumour. The stiff 'mushroomed' tissue within a trapdoor scar results from a combination of lymphatic insufficiency and scar contracture.[2] Persistent, localized facial oedema can be the consequence of skin disease. Patients with acne vulgaris and rosecea may develop chronic odema as a result of chronic inflammation and lymphatic fibrosis.[3] Recurrent attacks of erysipelas resulting in lymphatic blockage are responsible for the chronic swelling of the lips seen in elephantiasis nostras.[4]

Following extensive surgery and radiotherapy to the head and neck, particularly in those patients who have undergone bilateral neck dissection, there will inevitably be some lymphatic damage. The resultant oedema may be compounded by co-existing venous insufficiency. The potential for collateral lymphatic flow in the head and neck is enormous and as a result while the swelling is disfiguring it is rarely troublesome or prolonged. Massive facial lymphoedema has been described following extensive surgery and radiotherapy and is most frequently seen when the few remaining collateral lymphatics are blocked by repeated episodes of infection or recurrent tumour. We report the case of a patient in whom the head and neck lymphoedema was so severe as to cause significant sensory deprivation.

Case report

A 46-year-old man presented with right cervical lymphadenopathy and an occult primary tumour. He underwent a right modified radical neck dissection. Fourteen of the 30 nodes removed contained squamous cell carcinoma. A magnetic resonance image (MRI) scan, upper gastrointestinal endoscopy and multiple biopsies failed to identify the tumour. He received post-operative radical radiotherapy covering both neck fields and the likely primary sites in the pharnyx and larynx. One month after completion of radiotherapy a further mass was identified in the right posterior triangle, a subsequent MRI scan suggested bilateral neck disease and three courses of chemotherapy, using vincristine, bleomycin, methotrexate and 5FU, were

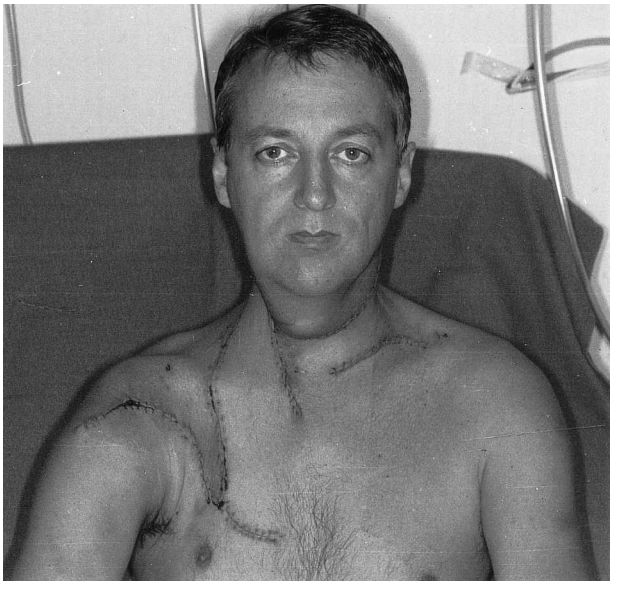

Fig. 1
Patient following surgery and after loading.

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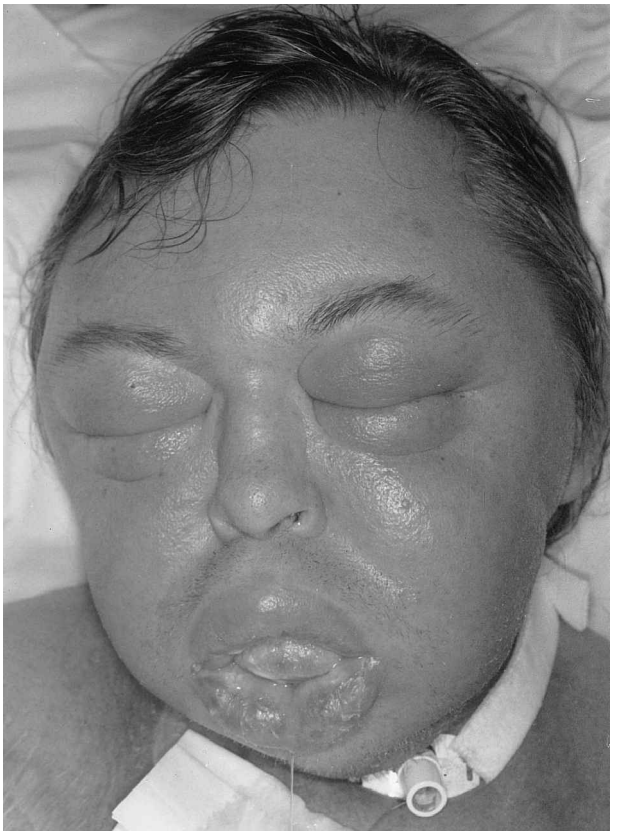

FIG. 2
Patient with severe lymphoedema of the head and neck.

started. Shortly afterward the patient was referred to our unit, several superficial nodules on the right side of the neck and lymphadenopathy on the left were identified, further attempts to identify the primary tumour were unsuccessful. He underwent a right-sided completion neck dissection and a left modified radical neck dissection. The skin of the right side of the neck was excised and nine afterloading catheters were placed down the length of the neck, the defect was covered with a deltopectoral flap. Brachytherapy was administered over an area of 6×10 cm to a dose of 60 Gy using iridium wires, over six days. The post-operative recovery was uneventful (Figure 1).

Eight months later the patient started to complain of facial swelling. The oedema progressed slowly and was initially treated with antibiotics and steroids. However after about four months the swelling was so severe that impending airway obstruction and feeding difficulties resulted in the introduction of both tracheostomy and gastrostomy tubes. Despite a fenestrated tracheostomy tube the severity of the pharyngeal swelling made speech impossible. Shortly afterwards both eyes were closed by extensive lid lymphoedema. The patient then complained of hearing difficulties and inspection revealed completely

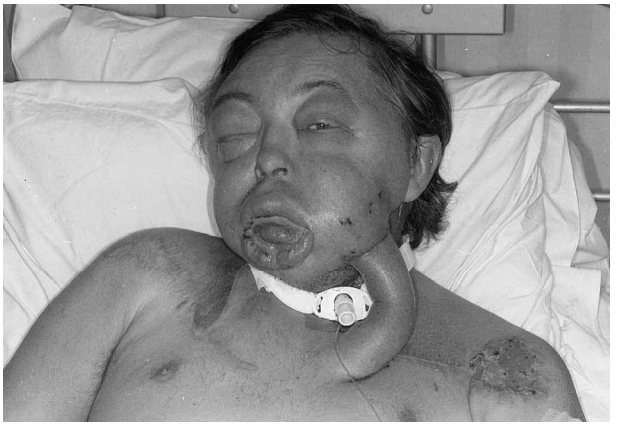

FIG. 3
Patient following treatment with 'lymphatic bridge'.

occluded external ear canals. As a result of the accumulation of fluid and added weight of the tissues the patient found it almost impossible to hold his head straight. Balance became very difficult and he was soon entirely dependent on others. Unable to speak, see or hear he became increasingly isolated (Figure 2).

Further investigations confirmed a patent left internal jugular vein and failed to reveal any recurrent disease. The patient was treated with penicillin V 500 mg daily to prevent recurrent infections. A regime of manual lymphatic drainage was started, in an attempt to encourage collateral drainage. The patient was provided with a bone conduction hearing aid. A dermal flap was introduced into the left cheek in an attempt to open his left eye by draining the cheek via the axial lymphatics of the flap (Figure 3). Three weeks after the flap was inset the lymphoedema of the ipsilateral cheek began to settle and the patient was able to open his eye for the first time in three months.

Discussion

Following treatment of head and neck cancers with surgery and radiotherapy some lymphoedema is common. This is usually transient, improving as collateral channels open and inflammation subsides. In rare cases lymphoedema is progressive causing severe swelling, endangering the airway, and occasionally blocking the pharynx. The potential for collateral lymphatic flow within the head and neck is so extensive that the severe progressive lymphoedema is rarely due to surgery and radiotherapy alone, however extensive such treatment may have been. The majority of severe cases follow blockage of collateral vessels by recurent tumour or fibrosis following recurrent episodes of infection.

Management of cases of massive head and neck lymphoedema (Table I), or so called 'pumpkin head' oedema, should focus on gaining control of the airway and digestive tract and avoiding further blockage of any remaining collateral lymphatics. It will almost certainly be necessary to insert both tracheostomy and gastrostomy

TABLE I
TREATMENTS TO BE CONSIDERED FOR DIFFERENT GRADES OF HEAD AND NECK LYMPHOEDEMA

Minor	Moderate/severe	'Pumpkin head'
Simple massage	Education/hygiene MLD Prophylactic antibiotics Investigate for recurrent disease – if persistent and severe	Supportive PEG tracheostomy Benzopyrones Diuretics Steroids Chemotherapy Surgical 'drainage'

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feeding tubes at some stage in the management. Patients should be fully informed about the nature of the condition, and must be aware of the imperative for rigorous skin care and hygiene in an attempt to avoid further infection. Prophylactic antibiotics should be prescribed.

All possible efforts should be made to identify recurrent tumour. This may be difficult because of the distortion of the normal anatomy following surgery, and the signal intensity overlap that is frequently seen between recurrent disease and post-treatment scarring. Positron emission tomography (PET) scanning has shown some potential in identifying recurrent tumour in these cases.[5]

Manual lymphatic drainage is an effective method of reducing limb lymphoedema, and success has been reported in cases of severe head and neck lymphoedema[6] However, care must be taken with the technique as excessive vigour can cause damage to the remaining normal lymphatics.[7]

Although well intentioned, attempts to treat pure lymphoedema with diuretics are both physiologically unsound and unhelpful. However, in most cases of post-surgical head and neck lymphoedema the picture is mixed, with a combination of lymphatic and venous insufficiency. In these cases diuretics may help the venous component of the oedema by reducing the capillary filtration pressure.[8] Benzo-pyrones are thought to act by increasing macrophages proteolysis and improving the contractile properties of the collecting lymphatics, their exact role in the management of lymphoedema remains controversial because of doubts over effectiveness and reports of liver damage. It is suggested these drugs may even be effective where the lymphatics are completely occluded, by allowing lysed proteins to escape the tissues via the blood stream, and so, despite concerns, they may be useful in such devastating cases.[7] Where recurrent disease is responsible for blockage of lymphatic collaterals chemotherapy may palliate the symptoms of the swelling. Although the administration of chemotherapy to a patient with a pharynx blocked by oedema and a mouth filled by a swollen tongue may result in disastrous consequences.

In this case no relief was experienced following the administration of manual lymphatic drainage. Diuretics and steroids were prescribed without any significant effect. Progressive isolation made it imperative that attempts were made to improve the patient's sensory input. A bone-conduction hearing aid was successfully fitted and significantly improved his hearing. Care had to be taken to avoid pressure necrosis of the swollen post-auricular skin and the patient was limited in the time that he could use the aid.

Gilles reported the successful drainage of lower limb lymphoedema using a skin flap raised from the medial side of the arm.[9] This flap, containing axial lymphatics running toward the axilla, was inset into the upper thigh. After three months the proximal end of the flap was divided and inset into the chest below the axilla, thereby creating a conduit for lymphatic flow from the leg into the axillary lymphatics. In an attempt to reduce the lymphoedema of the left eye we tubed a deltopectoral flap and inset a five by three cm de-epithelialized segment beneath the dermis of the left cheek. After three weeks the swelling on the left side of the face began to settle and the eye opened. Had this technique been unsuccessful we had planned to remove the subcutaneous tissues of the upper and lower lids, laying the thinned skin flaps back onto the orbicularis occuli, in an attempt to open the palpebral fissures.[10]

We report the management regime of a patient with severe head and neck lymphoedema in whom the goals of treatment were to improve the quality of life by reducing the degree of sensory deprivation and resultant social isolation.

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