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Cervical necrotizing fasciitis with thoracic extension after total laryngectomy

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Abstract
Cervical necrotizing fasciitis (CNF) with thoracic extension is rare. It has never been reported in laryngectomized patients. A case of fatal CNF in a laryngectomized patient equipped with a voice prosthesis is presented. Diagnosis and treatment are discussed. CNF with thoracic extension was diagnosed on clinical picture, computed tomography (CT) and biopsies were taken just above the tracheostoma. Antibiotic treatment was started and extensive debridement of the affected tissues performed. A minor extension to the left pleura was considered irresectable. Irradical debridement and the impossibility of administering hyperbaric oxygen therapy caused death within two day after presentation. CNF is a rare disease and to our knowledge, has never been reported after total laryngectomy. This case emphasizes the need for early antibiotic treatment and radical surgical resection of the affected tissues.

Key words: Fasciitis, Necrotizing; Laryngectomy

Introduction

Necrotizing fasciitis (NF) is characterized by extensive necrosis of the superficial fascia with undermining of the subcutaneous tissues and gangrene of the skin. NF of the head and neck is a rare condition. More often the extremities, trunk and perineum are affected. The course of the disease can be rapid and fatal despite intensive treatment. A case of cervical NF with thoracic extension in a laryngectomized patient equipped with a voice prosthesis is reported and the clinical features and management of the disease are discussed.

Case report

A 61-year-old male presented with a progressive, painful swelling above his tracheostoma and of the submental region, that had developed six to eight hours before. Seven months before he had been treated for a $T_3N_{2b}M_0$ squamous cell carcinoma of the left pyriform sinus by a total laryngectomy, partial pharyngectomy, bilateral modified radical neck dissection and primary placement of a Groningen low resistance voice prosthesis.[1] A pharyngocutaneous fistula developed, with an exposed carotid artery for which a myocutaneous pectoralis major flap reconstruction was performed.[2] No further post-operative complications occurred. Oral intake was started on day 10 and the next day the patient enrolled in a speech rehabilitation programme. A course of radiotherapy up to a maximum dose of 6250 cGy was administered to the larynx area and the neck.

On presentation we saw a painful, red-blue, blistering swelling above the tracheostoma. In the midcervical region there was superficial necrosis of the skin, the affected area was an estimated 100 cm^2 (Figure 1). Endoscopy of the newly formed pharynx of the laryngectomized patient using a flexible fibre-optic endoscope showed no abnormalities. Temperature was 39.9°C and, a high pulse pressure and high pulse rate were measured. Laboratory tests showed signs of dehydration, a sedimentation rate of 55 mm/hr and a marked leucopenia of 1.3×10^9/L, indicating septicaemia. Computed tomography (CT) scan of the region showed cellulitis of the submental area and anterior neck, without signs of abscess formation (Figure 2). Biopsies were taken from the centre and edges of the lesion. Frozen-section analysis showed abundant tissue

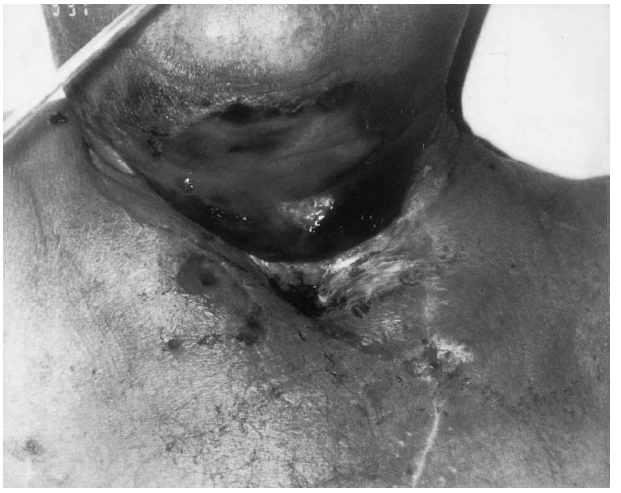

Fig. 1
Clinical picture of the patient on presentation.

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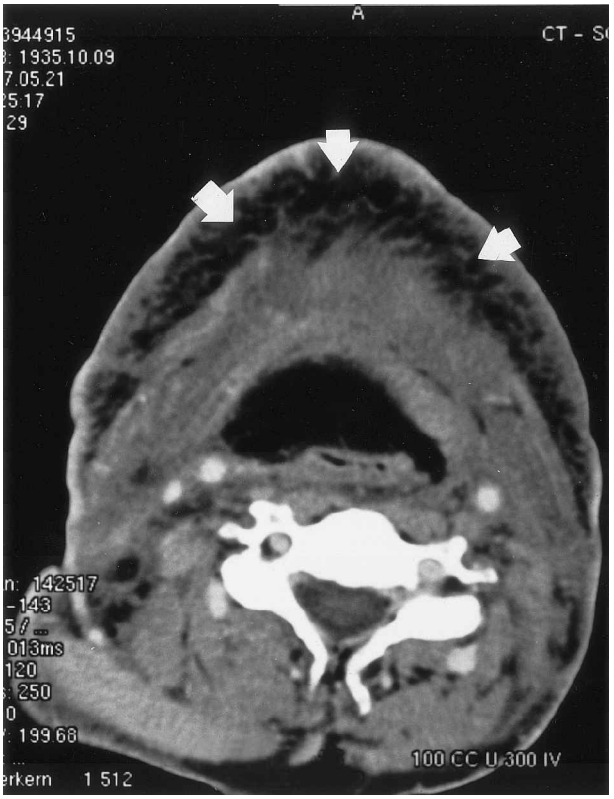

FIG. 2

Axial computed tomography of the submental region. Arrows mark the soft tissue swelling suggestive of cellulitis.

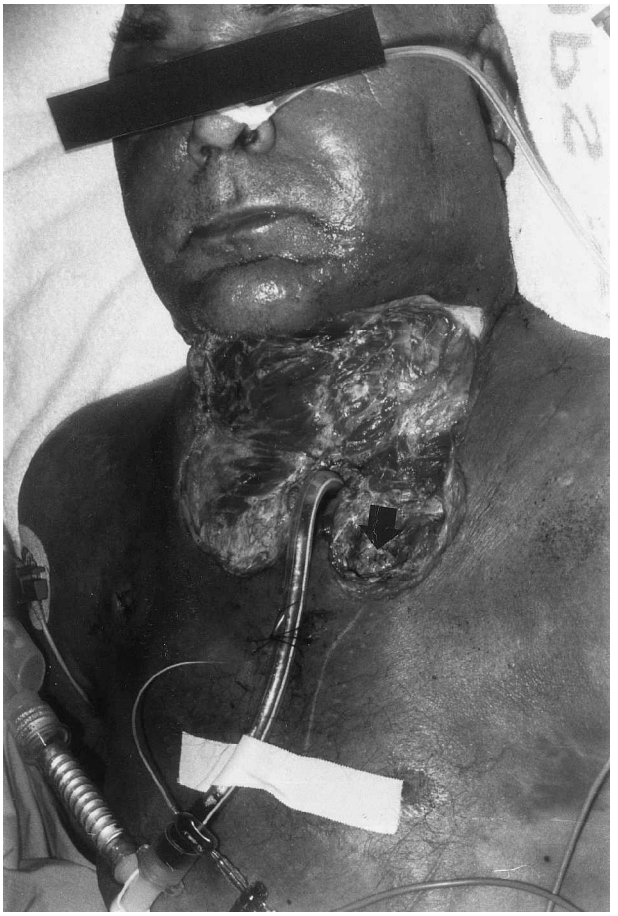

FIG. 3

Clinical picture immediately post-operative, after extensive debridement of avascular- and necrotic skin and subcutaneous tissues. Note pleura necrosis left behind (arrow).

necrosis, vasculitis and bacteria. Gram-stains showed Gram-positive cocci. The diagnosis of cervical necrotizing fasciitis was made a few hours after presentation of the patient. Treatment was started immediately, with intravenous antibiotic treatment consisting of amoxycillin/clavulanate potassium, clindamycin and intravenous rehydration. On the day of admission, immediate debridement of the affected tissues was carried out under general anaesthesia (Figure 3). Despite partial excision of the clavicle, an extension to the left pleura was considered irresectable by the general surgeon. Thus part of the affected tissue could not be removed. The operation wound was covered with saline soaked dressings. The patient was mechanically ventilated, received circulatory support and antibiotic therapy was continued. Twelve hours later, the infection spread to the chest wall. Shortly thereafter, the patient developed septic shock and went into fatal ventricular fibrillation within 24 hours after presentation. Cultures later showed group A β-haemolytic streptococci. Autopsy showed a clean neck wound with extension to the chest wall, where part of the left pleura was necrotic, bilateral pneumonia, tracheitis, septic spleen, shocked kidneys and an enlarged liver.

Discussion

Cervical necrotizing fasciitis (CNF) is an infection caused by micro-organisms that are found in the upper aerodigestive tract, mostly group A β-haemolytic streptococci (*Streptococcus pyogenes*), *Staphylococcus aureus* and anaerobes.[3] The infection occurs in children as well as in adults of all ages. The mean age of patients with CNF is 44 years. CNF can be caused by any trauma. About 68 per cent of the patients are male, the mortality is approximately 19 per cent[3] and ranges from 0–32 per cent in different series.[3–5]

The prognosis seems to depend on the localization and extension of the infection. If the infection has an intrathoracic extension, mortality rises to 50 per cent.[6,7] Poor prognosis is associated with involvement of the lower part of the face and cervical region, delay in surgical treatment and immunocompromised subjects[4,6,8,9] (e.g. postpartum state, alcoholism, intravenous drug abuse, malnutrition, malignancy, polymyositis, age above 50 years, peripheral vascular disease, chronic renal failure and post-radiation status).

The differential diagnosis consists of gas gangrene, myositis, erysipelas, cellulitis, pyoderma gangrenosum, thyroiditis and trauma.[10]

The infection spreads along the deep fasciae and causes thrombosis of the dermal and subdermal perforating vessels, resulting in necrosis of the overlying tissues.[10] Within one to two days following the incubation time (usually two to four days), the initially oedematous pale-red skin rapidly turns grey as necrosis spreads along the facial planes. Blisters containing yellow-coloured fluid appear, followed by gangrene. The skin becomes anaesthetised and loses contact with the underlying tissues as a result of suppuration.[4,8] Depending on the primary site of the infection there are several ways by which thoracic involvement can develop. The infection might spread via the carotid sheath, the prevertebral space, or the deep cervical fascia that fuses with the parietal pericardium and great vessels and adheres closely to the mediastinal parietal pleura.[7] The latter route would have been the

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way the infection spread in the patient presented in this report. Thoracic involvement can lead to necrosis of the fascia of the chest wall, mediastinitis, pleural effusion, pericardial effusion and empyema.[7]

CT scanning and magnetic resonance imaging (MRI) can be important diagnostic tools. They show thickening and infiltration of the cutis and subcutis; diffuse enhancement and thickening of fascia and muscles, always extending beyond the necrosis of the overlying skin. Fluid collections in multiple neck compartments can be seen as well.[11] MRI makes it possible to differentiate between non-necrotizing cellulitis and severe, necrotizing infections.[12]

The diagnosis can be suspected by frozen section biopsies showing necrosis, inflammatory granulocyte infiltration and bacterial invasion, but is mainly a clinical diagnosis.

Initial treatment consists of broad spectrum parenteral antibiotics–penicillin and clindamycin or metronidazole; gentamicin or ceftazidime can be added when indicated.[13] Eventually the antibiotics are selected on the basis of Gram stains and cultures obtained at the edges of the involved area.

Surgical debridement has to be performed as soon as possible.[14] Treatment with hyperbaric oxygen has been advocated as an adjunctive treatment.[6,13] According to literature, post-operative complications and long-term complications occur in 15 per cent, and 60 per cent respectively of the patients that were equipped with a voice prosthesis after laryngectomy. These complications include: haemorrhage, peristomal cellulitis and aspiration of saliva.[1,2,15,16] It is well known that a *porte d'entré* for the infection cannot be found in a number of patients suffering from NF since only a minor trauma, blunt or sharp, is sufficient to provide one.[14]

Although NF has not been reported as a late complication in laryngectomized patients equipped with a voice prosthesis before, in the case that is reported here, the primary site of the infection was the skin of the previously irradiated neck, just above the tracheostoma. The outcome of this case was fatal despite the early diagnosis and the promptly started antibiotic and surgical treatment. The impossibility of radical debridement of the patient's left pleura was an important negative prognostic factor, as was the poor post-operative condition of the patient that ruled out transportation and hyperbaric oxygen treatment, that might have influenced the final outcome.

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