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NECROTIZING FASCIITIS OF THE POSTERIOR CERVICAL COMPARTMENT: AN ATYPICAL CASE DUE TO *STREPTOCOCCUS AGALACTIAE*

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ABSTRACT

Necrotizing fasciitis is a fulminant infection that affects the deep and superficial fascia while initially sparing the overlying skin and underlying muscle. The involvement of the cervical compartment is associated with a high morbidity and mortality rate. Immunocompromised patients are prone to this infection. We present a case of a 80 year-old diabetic woman suffering from streptococcal cervical NF (probably secondary to a arm injury) with an uncommon involvement of the posterior cervical compartment, highlighting the atypical responsible microorganism (*Streptococcus Agalactiae*), the role of imaging for early diagnosis and the timely surgical and medical treatment for a successful outcome.

KEY-WORDS: Necrotizing fasciitis, deep neck infection, streptococcus agalactiae, diabetes mellitus

INTRODUCTION

Necrotizing Fasciitis (NF) was first described in 1871 by Joseph Jones, a confederate army surgeon in the American civil war (1). Wilson coined the term “necrotizing fasciitis” in 1952 and found this infection to be primarily caused by staphylococcal species (2).

NF is characterized by fulminating, devastating and rapidly progressing soft tissue infection characterized by progressive destruction of fascia, subcutaneous tissue, muscles and other adjacent structures. The most frequently involved areas are the thorax, limbs, perineum, abdomen and groin (3,4). Increased numbers of cases of cervical NF have been reported in recent years; the involvement of the cervical compartment is associated with a high morbidity and mortality rate. The disease is caused by a polymicrobial or mixed aerobic-anaerobic infection (5,6,7,8,9). Patients with immunocompromised status such as diabetes mellitus,

cancer, alcoholism, vascular insufficiencies, organ transplants, HIV, or neutropenia are prone to this type of infection (9,10,11). Given the rapid, fulminant progression of cervical NF (characterized by cutaneous necrosis, suppurative fasciitis, thrombosis of small blood vessels of the subcutaneous tissue and extreme systemic toxicity with development of mediastinitis and septic shock), prompt recognition, adequate antimicrobial therapy and early radical surgical debridement are the keys to successful outcome (12).

We present a case of a 80 year-old diabetic woman suffering from streptococcal cervical NF (probably secondary to a arm injury) with an atypical involvement of the posterior cervical compartment, highlighting the atypical responsible microorganism (*Streptococcus Agalactiae*), the role of imaging for early diagnosis and the timely surgical and medical treatment for a successful outcome.

CASE-REPORT

An 80-year-old diabetic woman patient was admitted to the our ENT Department with a three-days history of a non-fluctuating, painful left posterior tumefaction of the neck and the left shoulder, fever (39.5°), moderate level of breathlessness, initial impairment of senses. His past medical history was significant for a slight cardiac insufficiency, an atrial fibrillation treated for approximately 6 years with anticoagulants, hypertension, a left mastectomy 20 years previously and a pace maker fitted 3 years ago. On physical examination, a left, erythematous neck tumefaction spreading to the nape and the homolateral hemishoulder was palpable, without signs of colliquation or emphysema. No evidence of laryngeal, pharyngeal, odontological and nasal phlogosis was detected. There was an inflamed area on the left arm due to an insect bite which occurred three days before (fig. 1).

Due to a suspicion of deep cervical infection, a CT of the neck and chest was carried out, which showed a remarkable, disomogenoeus, diffuse swelling, air and enhancement of the cervical posterior musculature, with edema and necrosis of the prevertebral soft tissues and subcutaneous tissues, extending caudally to the supraclavicular region and posteriorly to the prevertebral plane (fig. 2). Laboratory tests showed hyperglycemia (497 mg/dl), leukocytosis (18.300/ μ l), a high C-reactive protein (CRP) (240 mg/L). Following examination and tests the patient immediately started broad spectrum antibiotic treatment (ampicillin-sulbactam,

teicoplanin, metronidazole) and correction of the hyperglycemia. It was suspected that the patient was suffering from necrotizing fasciitis and surgery was carried out under general anaesthetic. On the basis of the CT scans a left cervicotomy was performed at the level of the sternocleidomastoid extended to the tumefaction in the supraclavicular area. This incision is preferred to the customary cervicotomy as it gives easier access to the posterior space of the neck. Once the flap was freed, the border of the sternocleidomastoid muscle was identified, the anterior jugular vessels were dissected from the following left ligature and the dense, fetid pus was collected, then the surgery proceeded further back, laterally, towards the large vessels which seemed to have shifted towards the surface. The surgical approach proceeded deeper from the scalene plane and the brachial plexus to the cervical fossa until reaching another purulent sac which extended back to the splenius along the head and neck. After carefully draining the area it was then thoroughly cleaned of the fascial and muscular necrosis. An "in and out" drainage tube was used to rinse the area with antibiotics. The patient was transferred to the ENT clinic after two days in intensive care where she was treated for diabetes and initial renal insufficiency, no deficits or neurovascular damage was noted on recovering consciousness. *Streptococcus agalactiae* was identified from culture of the purulent material (fig. 3). The strain is sensitive to sulbactam/ampicillina and imipenem which was administered as soon as the patient was recovered in hospital. The visiting dermatologist confirmed there was an infection on the left arm and that this might have been the origin of the infection. Broad spectrum antibiotics (ampicillin-sulbactam 6 g/d, imipenem-cilastatin 2 g/d, metronidazole 400 mg/d) were administered for the following 13 days and medication was carried out twice a day with antibiotic and antiseptic washing through the drainage and constant removal of the necrotic tissue and skin until the wound had healed by secondary intention. The patient remained in a medical ward for a further 14 days for general pain relief, cardiac and nephrological treatment. Thereafter she was discharged with medication for pain relief in generally good condition, underweight by approximately 8 kg.

DISCUSSION

Necrotizing fasciitis (NF) is a fulminant infection that affects the deep and superficial fascia while initially sparing the overlying skin and underlying muscle. It is uncommon in the cervicofacial area and the usual nidus of infection in these cases is the teeth (10,11,12).

However, various other causes of cervical NF were also reported such as trauma, tonsillar and pharyngeal abscess, cervical adenitis, sinusitis, salivary gland infections and tumor infections (5,8,9,13,14,15).

The predisposing factors for the development of cervical NF are diabetes, hypertension, obesity, malnutrition, peripheral vascular diseases, severe liver disease, alcoholism and AIDS. With regard to diabetes, the hyperglycemic state impairs leukocyte function and thereby suppresses the host's immune system. However, high glucose levels form a good culture media for bacteria and predispose to an environment of low oxygen tension (10,16,17). Even the administration of steroid and non-steroid anti-inflammatory drugs appears to support NF, masking the clinical manifestations and leading to delayed diagnosis (18,19).

NF of the head and neck usually results from polymorphic flora including numerous bacilli and cocci, with a mixture of anaerobic (*Bacteroides*, *Fusobacterium*, *Clostridium*, *Propionibacterium*, *Peptostreptococcus*, *Prevotella*) and aerobic (*Alpha and Beta-Hemolytic Streptococcus*, *Streptococcus viridans*, *Staphylococcus aureus*, *Staphylococcus epidermidis* and *Pseudomonas aeruginosa*) organisms; it is microbiologically divided into two types: type 1 is polymicrobial and type 2 is monobacterial. Type 1 infection is most common in cervical NF. Skitarelic et al. however reported that type 2 NF was more common in their study, where the majority of cervical NF originated from a peritonsillar abscesses. The prevalence of type 1 in cervical NF is often correlated to diabetes and/or to multiorgan dysfunction (20,21). In our case microbiological examination identified *Streptococcus Agalactiae* as responsible of the infection. *Streptococcus agalactiae*, often referred as Group B Streptococcus, is a diplococcal gram-positive, non acid-fast bacterium (~2.0µm) that does not form spores, is not motile, and is catalase-free. It occurs in pairs or short chains and has group B Lancefield antigen present. *S. agalactiae*, originally discovered as a cause of bovine mastitis, is part of the normal bacterial flora colonizing the gastrointestinal and genitourinary tracts of a significant proportion of the human population. However, it occasionally becomes an infectious pathogen colonizing the uterus, blood, brain, and meninges.

There is some debate in literature as to the utility of microbiological testing on material from abscess. Many Authors do not consider this testing fundamental since results are often negative, doubtful or there are bacteria from many different origins. Symbiotic or saprophytic bacteria are often identified. The fact that gram positive, gram negative and anaerobic

polymicrobes are involved probably gives protection from phagocytic processes, intracellular “killing” and antibiotic treatment thereby fostering the necrotizing development of the pathology making it impossible to identify the species involved. Authors confirm that microbiological testing is useful when an individual strain is identified, a strong microbic charge as in our case, which directs (or confirms) the correct antibiotic treatment (14,18,21).

The most common clinical presentations of cervical NF are painful edema, erythema, warmth, tenderness, crepitation, and submandibular abscess. The divisions and subdivisions of the deep cervical fasciae confine the spread of the process of infection. Nevertheless, it is difficult to detect this infection before it has spread to the deep fasciae, when complications are more frequent. The synergistic action of anaerobic and facultative bacteria in a hypoxic environment promotes the production of enzymes and endotoxins that suppress host defenses, resulting in the dissemination of infection. Complications include airway obstruction, pneumonia, pulmonary abscess, septic shock, jugular venous thrombophlebitis, and mediastinitis (3,9). CT scan has been advocated for identifying the spread of infection in vascular sheaths, and detecting gas and the extension of infection to remote areas (mediastinitis and pleural or pericardial effusions). Constant CT features of NF are diffuse thickening and infiltration of the cutis and subcutis (cellulitis), diffuse enhancement and/or thickening of the superficial and deep cervical fasciae (fasciitis), enhancement and thickening of the platysma, sternocleidomastoid muscle, or strap muscles (myositis) and fluid collections in multiple neck compartments. Inconstant CT features included gas collections, mediastinitis, and pleural or pericardial effusions. A normal CT does not rule out NF. Not every case will present with frank abscess formation or subcutaneous air, and in some cases initial findings are nonspecific (22). Although there has been no well-controlled, clinical trial published comparing the efficacies of various diagnostic imaging modalities in the diagnosis of necrotizing infections, magnetic resonance imaging (MRI) is the preferred technique to detect soft tissue infection. MRI provides unsurpassed soft tissue contrast and spatial resolution, has high sensitivity in detecting soft tissue fluid, and has multiplanar capabilities (23).

Effective treatment and management of cervical NF is based on early recognition, aggressive surgical intervention, use of broad-spectrum antibiotics, and supportive therapy. Although antibiotic agents are not a substitute for radical surgical debridement, high-dose intravenous broad-spectrum antibiotic agents initially should be prescribed to help eradicate these mixed

soft-tissue infections. Hyperbaric oxygen continues to receive much attention as an effective adjuvant therapy to antibiotics and surgery in the treatment of NF. Successful use of intravenous immunoglobulin has been reported in the treatment of streptococcal toxic shock syndrome (10,23,24).

Historically, early and sometimes multiple, radical surgical debridement has remained the cornerstone in the management of this condition. After the incision through the skin and platysma to the level of the deep cervical fascia, the lack of bleeding or production of dishwater-coloured fluid are highly suspicious for NF. An easy separation of tissue planes under gentle finger dissection is also highly suspicious. Once NF is confirmed intra-operatively, the surgeon must be prepared to widely debride the affected area, including all grossly necrotic skin and muscle. Often, apparently normal tissues may become involved soon after surgery, and repeated debridements are the rule rather than the exception. A delay in appropriately aggressive surgery has been associated with a high mortality rate (23,24).

The need for immediate surgery is confirmed by the fact that improvement is rapid following once surgery has been carried out. A wide, mid-posterior cervicotomic surgical approach has proved to be suited to managing deep, posterior collections even if this approach gives a greater risk of lesion to the trapezium spinal nerve. It is mandatory to identify the branches of the brachial plexus below the deep cervical fascia, which is not easy in the presence of acute phlogosis, and to drain safely the deep posterior sacs of purulent/gaseous liquid. By positioning large "in & out" drainage it was possible to rinse the cavity thereby reducing the risk of requiring further surgery under general anaesthetic. We confirm that it is better not to close the surgical wound in order to carry out "nursing" which, in our case, consisted of medication, washing and curettage several times a day until the wound healed completely by second intention approximately 30 days after surgery.

Recently two large studies have reported that percutaneous catheter drainage as a novel treatment for cervical NF is less invasive than conventional surgical drainage and gives a similar outcome. Moreover, percutaneous catheter drainage areas are less likely to become secondarily infected by antibiotic-resistant bacteria, and it seems superior to surgical drainage in pain control and in preventing protein leakage from the wound (25,26). On the other hand, more research is needed to fully evaluate the

effectiveness and suitability of less invasive treatment strategies in this life-threatening disease.

Morbidity and mortality seem to be related to the promptness of medical and surgical intervention. The mortality rate reported in literature ranges from 19% to 40%. The higher mortality rate is related to pre-existing systemic illness, late surgical intervention, septicemia within 24 hours, old age and mediastinal and thoracic extension of the infection (5,8,14,15,27).

CONCLUSION

Cervical NF is characterized by widespread necrosis of the subcutaneous tissue and the fascia. Although the pathogenesis of necrotizing fasciitis is still open to speculation, the rapid and destructive clinical course of NF is thought to be due to multibacterial symbiosis. Early recognition and prompt intervention are the keys to successful treatment. MRI and TC are fundamental for the diagnosis. Mortality is directly proportional to the time to intervention. More research is needed to fully evaluate the effectiveness and suitability of less invasive treatment strategies in this life-threatening disease. What is particular about our case is its manifestation in the posterior part of the neck and the identification of *Streptococcus Agalactiae*, which is known to be a cutaneous saprophyte; in our opinion this is linked to the atypical origin of the infection (on the skin of the arm) in a patient suffering from serious comorbidity.



Figure 1



Figure 2

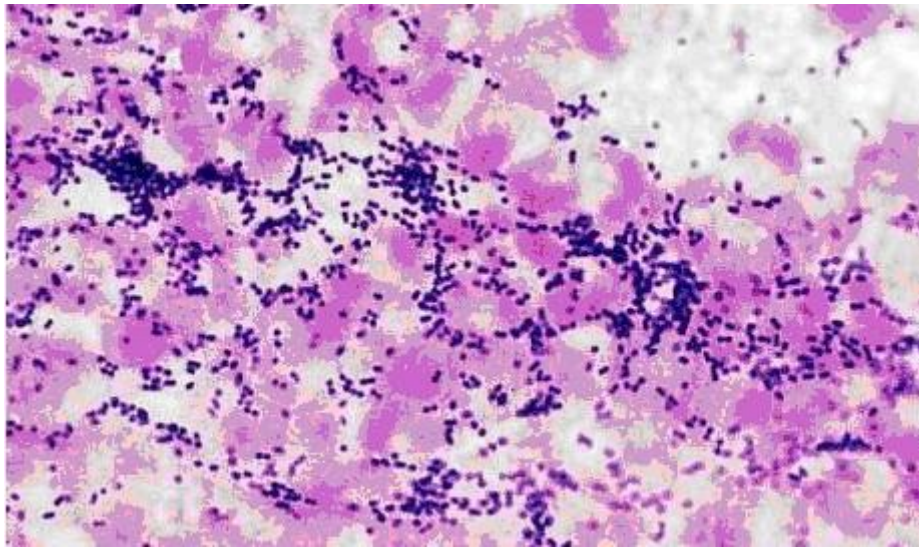


Figure 3

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LEGENDS OF FIGURES

Figure 1: Inflamed area on the left arm due to an insect bite which occurred three days before the admission to our Department.

Figure 2: Axial CT scan of the neck showed a remarkable, disomogeneous, diffuse swelling, air and enhancement of the cervical posterior musculature (arrow), with edema and necrosis of the prevertebral soft tissues and subcutaneous tissues, extending caudally to the supraclavicular region and posteriorly to the prevertebral plane.

Figure 3: *Streptococcus agalactiae*, Gram stain, Resolution magnification X1000.