

Case Report
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Craniofacial Fasciitis Secondary to Odontogenic Infection

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SUMMARY

Craniofacial Necrotizing Fasciitis is an extremely rare entity, whose most frequent location is the pelvis and thoracic limbs, and some cases have been reported in the abdomen. On the other hand, a very low incidence has been reported in the head and neck region, 5% of cases with Necrotizing Fasciitis are mentioned in the literature. The name of this disease is related to the area in which it occurs, when it is located in the face and cervical region it is called Cervicofacial. In the same way when it affects beyond the middle facial third and the scalp it is called Craniofacial. Due to the different anatomical locations, it is essential to perform computed tomography, to provide early diagnosis, management, and a better prognosis to the patient. Craniofacial Necrotizing Fasciitis etiology is diverse, two of the main associated factors are the immunosuppression and comorbidities, which make the patient more susceptible to a possible complication. Some of the Craniofacial Necrotizing Fasciitis complications are sepsis, hemodynamic disorders and death. The most frequent antimicrobial biota found in Craniofacial Necrotizing Fasciitis is anaerobic and includes Gram (+) and Gram (-) bacteria, consequently, it is necessary to provide prompt antimicrobial treatment and aggressive surgical resection. This article present a clinical case of a patient with Craniofacial Necrotizing Fasciitis, who received a multidisciplinary treatment at the Specialty Hospital, National Medical Center, IMSS, located in Mexico City. The object of this article is to demonstrated the rapid spread and complications of Craniofacial Necrotizing Fasciitis.

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Introduction

Craniofacial and Cervical Necrotizing Fasciitis is an extremely rare entity and a rapidly progressing infection, spreading through the planes (skin, fascia, connective, muscular and subcutaneous tissue) of the head and neck. It was described by Joseph Jones as hospital gangrene [1-3]. Likewise, Wilson is one of the pioneers in studying the entity, he is the one who is attributed the term “Necrotizing Fasciitis” The main location of Necrotizing Fasciitis is pelvis and thoracic limbs and only 5% in the head and neck, when it occurs on the head it can be divided into two types Skull (sometimes it affects the eyelids and the entire area covered by the scalp) and on the Face that is closest to the neck, referring to them as Craniofacial and Cervicofacial; both having as main characteristic that it is very aggressive and invasive in a matter of hours, reporting a mortality rate of 15-40% on average, even mentioning it reaches more than 70% mortality. It can be of dental, pharyngeal, surgical origin, due to trauma, radiotherapy, iatrogenesis, and very unlikely due to peritonsillitis. The most vulnerable patients are those who have immune compromise, an uncontrolled disease, elderly patients, or have bad habits such as smoking, alcoholism, obesity, drug addiction, and so on. It is more frequent in the female gender Necrotizing Fasciitis is characterized by presenting a polymicrobial biota that includes Gram (+) and Gram (-) bacteria, anaerobes and anaerobes, in the head, neck and mouth, typical isolated bacteria

are mentioned as [4-6]. *Streptococcus pyogenes*, *Streptococcus viridans*, *Peptostreptococcus* spp., *Staphylococcus aureus*, *Hemophilus influenzae*, *Clostridia* y *Enterobacteriaceae*, mainly [7]. For the diagnosis of Craniofacial Fasciitis, it is necessary to including Computed Tomography to corroborate the extension of the disease, the presence of tissue emphysema, the possible initial septic focus and plan debridement considering the structures involved and those adjacent to the affected area. For treatment, it is important to start broad spectrum intravenous antibiotic therapy with aggressive surgical debridement, resecting and excising the necrotic tissue, when it is accompanied by an abscess-type infection, surgical drainage and lavage should be performed, permeabilizing the spaces with extension of the flaps necessary to allow inspection and comply with drainage principles [8]. In addition to the fact that in the literature, hydroelectrolyte control has been suggested as a complement, in addition to the use of a hyperbaric chamber (since hyperbaric oxygen helps wound healing and decrease the number of bacteria) and few reports mention the use of intravenous immunoglobulin of the type G to improve surgical outcomes and reduce mortality rates. Finally, one of the most serious complications of Craniofacial and Cervicofacial Necrotizing Fasciitis is cavernous sinus and / or internal jugular vein thrombosis, depending on the clinical presentation of Necrotizing Fasciitis, erosion of the carotid artery and disseminated intravascular coagulation. Sepsis, multiple organ failure and death [9, 10, 11].

Case Report

Female, 64-year-old, who came to the emergency department for revision, presenting pain in the right third molars (upper and lower). The anamnesis is performed and the patient refers to being diagnosed with uncontrolled diabetes mellitus with glibenclamide treatment and arterial hypertension medicated with propranolol; It mentions as gynecological-obstetric history: 5 deliveries, 6 pregnancies, 1 abortion; currently menopausal, surgical (+): appendectomy and one curettage. Trauma: a fall from the stairs with conservative management; denies allergies smoking and alcoholism. Relative of the patient (daughter) reports that she presented increased volume in the right oral and temporal region and pain in the third molars (upper and lower) right, two weeks prior to going to the hospital, however the patient and her family did not consider it to be An emergency for which they waited for the pain to cease, later they observed that the inflammation decreased, but it began to emit a foul odor, which is why he went to the emergency room. A multidisciplinary assessment is carried out given the general and local condition of the patient; being in charge of the Internal Medicine service and as interconsultants the services of: Maxillofacial Surgery, Metabolic Unit, Infectology, General Surgery, Neurosurgery, Radiology. Surgical management is performed by the Maxillofacial Surgery service (the origin of the infection: dental), a canalization and drainage of the odontogenic abscess is performed in relation to the aponeurotic spaces: buccal, temporal and infratemporal, making three incisions: one in the temporal space, another in the buccal space and a more Risdon type (right submandibular), a rigid drain was placed to permeabilize and communicate the spaces: temporal, infratemporal and right buccal, likewise the extraction of the third molars (upper and lower) right and of all teeth that represented foci of infection by caries (dental root remains); Antimicrobial treatment is started with intravenous clindamycin and cephalosporin, a secretion and tissue culture is taken from the pathology, subsequently cures are performed every 8 hours with a solution of thirds (0.9% isotonic saline solution, povidone iodine and hydrogen peroxide), however the evolution of the patient on the third day becomes torpid, with metabolic lack of control, and evolution of her condition with the presence of Craniofacial Necrotizing Fasciitis, presenting necrotic tissue in the right temporal and oral region (Figure 1a and 1b) purulent material continues to leak, custard is present, predominantly suffered skin is observed in the right mouth region as well as the presence of violaceous macules on the right and infratemporal cheek, the edges of the temporal approach are observed with necrotic, indurated areas and erythema In adjacent areas, a fistula can also be seen in the right cheek in the center of the necrotic area, with an outlet of purulent material. Therefore, the resection of necrotic tissue of the oral and temporal region is performed and surgical washing and permeabilization of temporary spaces is carried out and the healing is maintained with solution at thirds every 8 hours, in addition the Infectology service adds (to the established antimicrobials) Metronidazole; 48 hours after having been operated on again, abundant custards are observed and areas of necrosis are again observed, so a pre-surgical protocol is carried out and the patient is prepared to carry out extension of the resection and surgical lavage under balanced general anesthesia (Figure 2) Subsequently, a reassessment and review of the case was carried out by the Internal Medicine and Infectology service and the antimicrobial scheme was changed to Piperacillin / Tazobactam. On the eighth day of hospitalization, the patient had a discrete metabolic improvement, the infectious process persists and spreads to the parietal and occipital space, so it is necessary to channel and drain under general anesthesia, an

occipito-parietal Penrose-type drain is placed, there is minimal necrotic tissue In the upper temporal border, after resection, loss of the pericranium and temporal muscle is observed, however, there is granulation tissue and with slight improvement in the right malar and buccal region, the purulent exudate from the right submandibular space disappears and begins to close by second intention the Risdon-type approach, in addition Vancomycin is added to the established antibiotic scheme (Figure 3a, 3b. and 3c.). On the 11th day of hospitalization, another debridement of the right temporal region is performed with surgical washing and healing with surgical soap, it is noted that there is remission of the parieto-occipital infectious process itself that is without pus, so the drain is removed. and is left free for closure by second intention; the buccal region is observed with granulation tissue and presents indurated edges with slight erythema, finally the Risdon approach closed by second intention. (Figura 4a. and 4b.) It is highlighted that from his first surgical time he had a maintenance therapy based on healing every 8 hours daily. The patient remains hemodynamically stable for a week; However, three days later (on day 14), the patient relapsed, with metabolic lack of control, again there was a purulent collection in the right parietal region, a dark area was observed in the center of the oral region with an erythematous halo, erythematous and indurated edges, discharge of purulent exudate (Figure 5), She had a fever peak of 38° Celsius, so it was decided to carry out a pre-surgical protocol and perform a surgical lavage again with debridement and elimination of necrotic areas as well as canalization and drainage of the parietal region again, the procedure is performed without complications, the patient is in her period postoperative with electrolyte imbalance, feverish peaks, delirium, tachycardia and dyspnea, it presents with metabolic ketoacidosis and later falls into cardiac arrest, resuscitation maneuvers are performed by the Infectology staff and intensive therapy; However, despite attempts to save her, she dies.



Figure 1a. and 1b. Persistent craniofacial necrosis is observed, oral and temporal areas (yellow arrows), and drainage of the temporal space that communicates with the right oral space, Risdon-type approach (right submandibular space, blue arrow)



Figure 2. Photograph of 5 days of hospitalization, with necrotic areas in the center and edge of the oral and temporal approaches (yellow arrows), presence of some custard. And lysis of the fat bag of Bichat. (blue arrow)



Figure 3a. Right occipital and parietal abscess drainage with pen rose drain (yellow arrow), persistent necrotic borders (blue arrow) **Figure 3b.** Exposure of the temporal region of the right skull, temporal necrotic border (yellow arrow) with areas of granulation to clinical improvement (green arrow) **Figure 3c.** Clinical photograph with the presence of facial asymmetry at the expense of surgical resections of Craniofacial Necrotizing Fasciitis.



Figure 4a. Anterior and posterior enlargement of temporal fasciotomy, right retroauricular and parietal edema (red arrow) **Figure 4b.** Right Risdon-type approach in the healing phase (yellow arrow), granulation tissue in the buccal area. (green.Arrow)



Figure 5. Drain in the right parietal region (green arrow), necrosis is observed in the center of the buccal region (yellow arrow), erythematous edges and purulent exudate (red arrow)

Discussion

With respect to the higher prevalence of Craniofacial Necrotizing Fasciitis, which globally has been reported a greater number of cases in women compared to the male gender, the case presented is an example of a female patient with uncontrolled comorbidities of these diseases coupled with an infectious process of dental origin, as mentioned Endorf, Yoder et al., among others [5,6]. In short, like various authors, we confirm that Craniofacial Necrotizing Fasciitis is a very rare disease in the craniofacial region, as mentioned Banerjee, It is a serious infection, with a high risk of complications, mainly for immunocompromised patients as reported in the present case and as reported by authors such as Wilson, Fairbanks, Among many others, in this case it is emphasized that the progression is rapid and fulminant despite having the joint management of several medical specialties, the general physical condition and the organic response of the patients cannot be predicted [3,7,12]. Therefore, it is important to maintain constant monitoring of the haemodynamic and haematological status as well as renal function parameters should be considered essential In the Maxillofacial Surgery service of the Specialty Hospital of National Medical Center, There are not many reported cases of Craniofacial Fasciitis, in necrotizing fasciitis within its very low frequency produced by dental infections, most are located in the Cervicofacial region, covering the middle third of the face to the neck, with Mediastinitis

and the most frequent complication in some cases the patients die and the survivors continue the multidisciplinary management, and the defect reconstruction is performed by surgery in conjunction with the plastic surgeons.

Conclusions

Necrotizing Craniofacial Fasciitis has a rapid advance through the tissues, when it occurs in an immunocompromised and / or imbalanced patient, it worsens the prognosis and evolution of the disease, and can even compromise the patient's life. The present case is a clear example that this pathology is unpredictable and that despite the adequate management indicated by the norms and clinical practice guidelines (pharmacological and surgical guidelines in a timely manner) the patient did not progress to improvement. Some patients with Necrotizing Fasciitis show improvement peaks and decline, it is not possible to know exactly if the patient is going to get complicated. Unfortunately, the factors associated with comorbidities, quality of life, habits, hygiene, nutrition, education and life expectancy, etc., as mentioned in the literature, are decisive for a patient to follow the indications and care and with it the susceptibility and risk of the disease. Generally the clinic of Craniofacial Necrotizing Fasciitis is cumbersome, since they present great loss of tissue in different planes and depth due to the extension of the necrosis, it is mentioned in the literature microspaces in the tissue that could be a starting point for persistence or recurrence of necrosis. Radical management plays an important role as well as maintenance measures (as was done in the case presented, performing cures every 8 hours daily) and medical / metabolic control. Multidisciplinary management must always be carried out jointly, as was done in this case and as mentioned in their articles (respectively) Yadav, Lazow y Hakki Among many other authors, in order to provide a better treatment and prognosis to the patient, however there are cases that are very advanced since their hospital admission and are difficult to maintain or bring to borderline parameters and very rarely to normal values in their laboratories and cabinet studies; It is then that in these cases the prognosis becomes worse for the patient and a greater possibility of a fatal outcome [9-11]. In the case presented, the general condition of the patient did not favor her conditions, presenting severe systemic complications quickly, delirium and septic shock are examples of the severity of the disease. Complications associated with Necrotizing Fasciitis (including Craniofacial Fasciitis) can be: sepsis, septicemia, pericardial effusion, Mediastinitis, pneumonia, spread of infection or necrosis to the chest, abdominal or pelvic cavity and / or neuroinfection. It can progress to septic shock, thrombophlebitis, disseminated intravascular coagulation, organ failure and death. That is why it is necessary to act quickly between all medical and surgical specialties.

Conflict of Interest

Neither the authors nor any member has a financial or interest relationship (currently or in the last 12 months) with any entity producing, marketing, reselling or distributing health care products or services consumed by, or used in, the patients.

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