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Severe Fungal Cervicofacial Cellulitis from Otogenic Mucuromycosis Origin: A Case Report

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Abstract:- Cervicofacial fungal cellulitis are rare disease, it's an extensive and serious opportunistic infection with often late diagnosis and fatal progression, especially in immunocompromised or patients with co-morbidities. Their treatment is medico surgical. Only an early and adequate management guarantees a better prognosis. We report the case of cervicofacial fungal cellulitis of otogenic origin for a 39 years male patient, diabetic type I poorly balanced and whose evolution was unfortunately fatal.

Keywords:- *Cervical Cellulitis, Fungal Infection, Otitis, Neuromeningeal Complications, Septic Shock.*

I. INTRODUCTION

Cervicofacial cellulitis is a serious, rapidly spreading and potentially fatal infection. They are defined as infections of the deep fascial spaces and celluloid tissues of the face and neck. The development of antibiotics has radically modified the evolution of these infections, once an early and adapted treatment is implemented without forgetting the etiological treatment. The absence of an anatomical barrier allows the infection to spread rapidly from the base of the skull to the diaphragm **(1)**.

We report through our work the case of a young diabetic patient with fungal cervicofacial cellulitis complicating chronic otitis media in order to emphasize this exceptional location of fungal infections.

II. PATIENT AND OBSERVATION

We report our detailed care activities of a 39-year-old male, diabetic type I for 20 years, on insulin therapy, unbalanced and poorly monitored, who was admitted to the emergency room for severe right ear otalgia, that has been evolving for 10 days, associated with hypoacusis and purulent otorrhea, complicated five days later by a right peripheral facial palsy. The patient received a self-medication of a nonsteroidal anti-inflammatory drug (diclofenac sodium 50 mg) and fluoroquinolone ear drops. The evolution was marked by the appearance of ipsilateral cervical swelling and the deterioration of the general health condition.

On admission, the patient was disoriented with a Glasgow score of 14/15, hemodynamically and respiratory stable. The otoscopic examination showed a hematopurulent otorrhea, with a macerated, inflamed external auditory canal, not allowing seeing the tympanic membrane. The Cervical examination noted a retroauricular swelling facing the mastoid with suppression of the retroauricular groove in the direction of the parotid region. The skin of this region was infiltrated, inflamed without palpable cervical adenopathy.

The contralateral ear was normal. The facial examination revealed a peripheral facial palsy grade III according to House-Brackman. The examination of the other cranial nerves found no notable anomalies. The rest of the clinical examination was without anomalies.

A Cervicofacial cellulitis complicating chronic otitis media occurring in a field of immunodeficiency was suspected, and the patient was immediately put on broad-spectrum antibiotic therapy (Amoxicillin Clavulanic acid 1g * 3 / day + metronidazol 500mg * 3 / day + Genta 160mg / day) associated with an adapted blood sugar correction**)**.

On the paraclinical level, the biological assessment showed a hyperleukocytosis at 32500/mm3 with PNN, a CRP at 253, a sedimention rate at 57 1st hour and a negative HIV serology.

The radiological assessment included a temporal bone computerized tomography, which revealed an aggressive otitis media and otitis externa with holotympanic filling and air trapping, lysis of the tegmen tympani and facial canal in its second portion. A cervico-facial cellulitis made up of an infiltration of the right laterocervical subcutaneous tissues extended towards the deep spaces of the face (the prevertebral space, the para-pharyngeal space) and towards the parotid compartment with an ipsilateral sub-auricular collection of 44*28*21 mm **(Figure 1)**.

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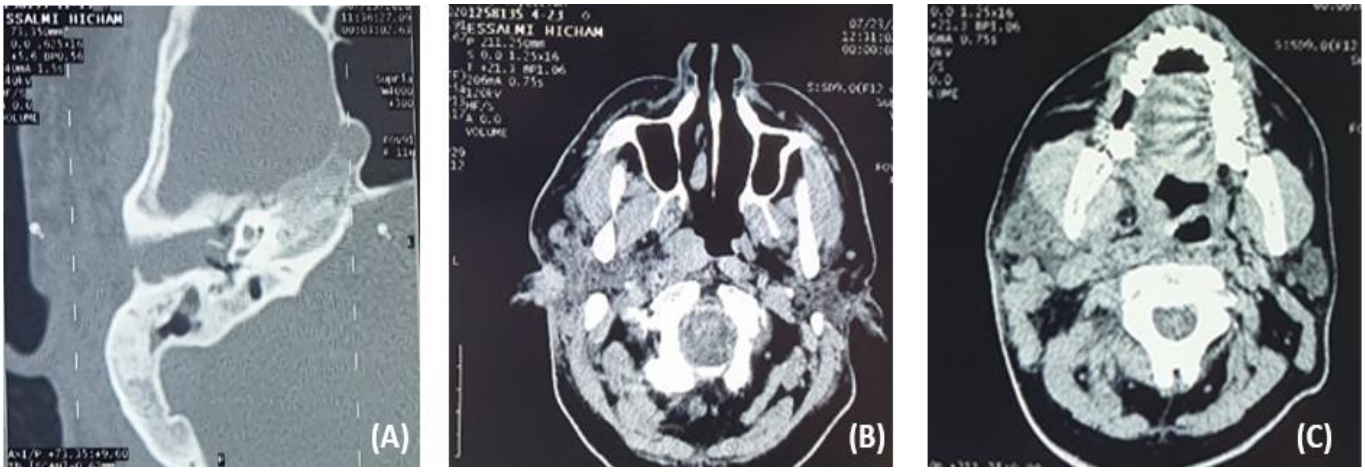

Figure 1: Radiological assessment (iconography of our department).
(A) - Computerized tomography of the right petrous temporal bone in axial section showing the filling of the external auditory canal and middle ear.
(B) & (C) - Cervical computerized tomography in axial section showing the sub-auricular collection and infiltration of the soft tissues opposite the parotid region.

A Surgical drainage with a necrosectomy of the subcutaneous tissues was performed urgently bringing 40ml of frankly purulent fetid fluid as well as an opening of micrologette that was filled with air bubbles. We finished the intervention by the insertion of Delbet blade **(Figure 2)**.

An intraoperative cytobacteriological and histological sample was taken. A twice-daily dressing with abundant washing with betadine and oxygenated serum was performed.

Figure 2: The pictures showing the evolution of the patient: from left to right D1 of drainage, D2, D3, D4 (Iconography of our department).

During the evolution, the patient developed a consciousness disorders with acid ketosis decompensation of his diabetes and local aggravation made of an extension of the lesions, a necrosis of the tragus and the sides of the drainage incision as well as the appearance of cottony filaments at the external auditory canal and at the cervical region. The patient was transferred to an intensive care unit with expanding of the antibiotic spectrum to ceftazidime **(Figure 2)**.

Cytobacteriological examination with histological study performed intraoperatively revealed large, branched spores and mycelial filaments, visible on periodic acid Schiff (PAS) and Grocott stains, suggestive of mucormycosis of the external auditory canal. Direct mycological examination was negative and culture isolated **Rhizopusnigricans**.

Consequently, parenteral treatment with amphotericin B (1 mg / kg / day) was started gradually. The subsequent evolution was unfavorable by installation of multiple organ failure with septic shock. CT angiography revealed thrombophlebitis of the right lateral sinus extending to the ipsilateral internal jugular vein complicated by right hemispherical infarction **(Figure 3)**. The patient died two days later.

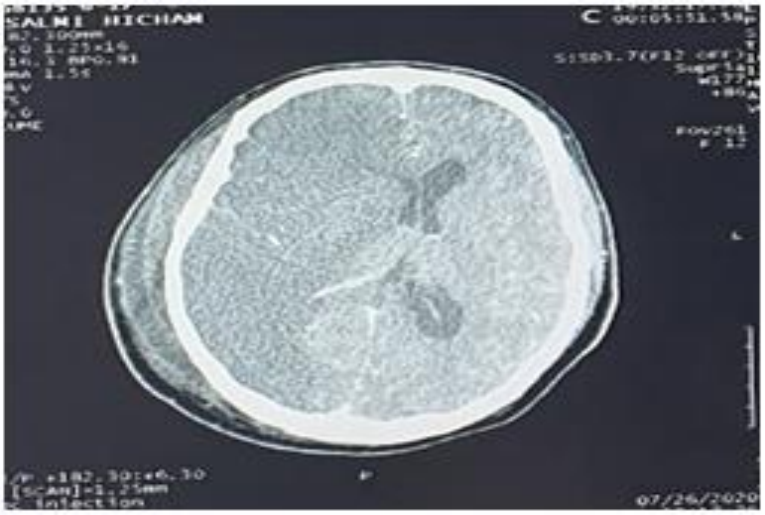

Figure 3: Brain computerized tomography showing right hemispherical infarction (iconography of our department).

III. DISCUSSION

Cervicofacial fungal cellulitis is a rare and invasive disease that should be considered in any diabetic and/or immunocompromised patient presenting with rapidly progressing necrotizing cellulitis despite broad-spectrum antibiotic treatment. A delay in diagnosis and therapy inevitably exposes the patient to serious or even fatal complications, which is why an early diagnosis is necessary **(1, 2)**.

Several factors are involved in its development. Among others, we find poor oral hygiene, an immunodeficiency ground, the administration of non-steroidal anti-inflammatory drugs...

The responsible germs differ depending on the entry point (cutaneous, tonsil, dental, submaxillary, nasosinus or more rarely otogenic ...) directing the antibiotic therapy, that must be initiated rapidly, initially as a probabilistic treatment, targeting the germs colonizing the ENT sphere, and then adapting it according to the pre- and/or peroperative bacteriological results **(3).** As a general rule, the antibiotics used are beta-lactams, aminoglycosides, imidazole derivatives and macrolides.

In our case, diabetes decompensated on a dragging and neglected fungal otitis in addition to self-medication with non-steroidal anti-inflammatory drugs and the delay of consultation which generated this fatal evolution.

Among the common fungal infections found, we find mucormycosis, which are rare fungal infections, most often developing acutely and mainly on a weakened ground, exceptionally in immunocompetent people **(4, 5)**. They are due to the proliferation of filamentous fungi belonging to the class of zygomycetes and to the order of mucoral. They are ubiquitous, saprophytic from the soil and from several plants **(6, 7)**. Contamination usually occurs by inhalation of spores explaining the preferred nasosinus and pulmonary locations, more rarely by ingestion or transcutaneous inoculation **(6, 8)**.

Their pathogenicity is based on their high affinity for vascular walls, causing thrombosis and ischemic tissue necrosis. The extension then occur step by step or hematogenous way **(4, 8)**. Different descriptions of mucormycosis in ENT can be observed. Rhinofacial or rhino-cerebral localization is the most common **(6, 7, 9)**. Certain ENT localizations of mucormycosis were exceptionally reported, such as the otological localization **(5, 9)**.

Yun al. **(10)** and Macdonell al. **(9)** reported a case of isolated middle ear mucormycosis complicated by facial paralysis in a diabetic patient, diagnosed late and the course of which was unfavorable by the occurrence of cavernous sinus thrombophlebitis.

Hazarika et al. **(5)** reported the observation of an immunocompetent patient who presented with mucormycosis of the middle ear successfully treated with surgery without recourse to amphotericin B. These studies conclude that the probable entry point for otologic mucormycosis may be the nasopharynx or via a pre-existing tympanic perforation **(4, 5, 8)**.

The diagnosis of mucosal fungal cellulitis, whatever its location, is based on pathological evidence from biopsy samples. The examination allows, by special histochemical stains (periodic acid Schiff [PAS], Gomori-Grocott), to find non-septate mycelial filaments growing at right angles. Their fibrils invade the vessel wall wich causes ischemia and necrosis **(11, 12, 13)**.

Treatment should be initiated as soon as clinical suspicion is established after mycological samples are taken. Delay in diagnosis and treatment can lead to serious or even fatal complications **(5, 12, 13)**. Some authors recommend an extemporaneous examination, secondarily confirmed and completed by histological and mycological analyses **(5, 14)**.

The therapeutic care is based on three elements: antifungal treatment, a large iterative surgical debridement (carcinological type) and repeated necrosectomies to reduce the fungal load and increase the bioavailability of the medical treatment in the infected areas **(12, 13, 14)**. It is currently recommended to start treatment with Amphotericin B at a high dose of 1 mg / kg / day parenterally for the first two to three weeks, followed by consolidation with itraconazole at a dose of 300 mg / day. The optimal duration of treatment varies from three to six months depending on the evolution **(5, 7, 8)**. The Hyperbaric oxygen therapy as an adjuvant treatment is under study for its fungistatic role in addition to the neovascularization of ischemic territories **(8, 15)**. The functional and vital prognosis of mucormycosis is reserved and the mortality rate is high, up to 20% to 50% of cases despite the progress in treatment. **(8, 12, 15)**.

IV. CONCLUSION

Cervicofacial Mucormycosis Cellulitis is a very rare fungal infection. Rhinocerebral involvement is the most frequent clinical form. Otological involvement is exceptional. The diagnosis, which is often difficult must be made as soon as possible in order to institute appropriate emergency treatment. The prognosis is still reserved.

Conflicts of Interest
Authors do not declare any conflict of interest.

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