A rare case of nasosinusal actinomycosis with cerebral extension

Un cas rare d'actinomycose rhinosinusienne à extension cérébrale

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ABSTRACT

Objective: Discuss diagnostic and management difficulties of rhinosinusal actinomycosis with brain extension

Observation: A 48-year-old patient was admitted to the emergency for the management of a diabetic ketoacid coma. The patient was feverish. The examination showed a profuse, foul-smelling rhinorrhea, palatal and nasal necrosis with destruction of the nasal cavity soft tissues. The brain and maxillofacial CT scan revealed a partial lysis of the maxillary sinuses, a destruction of the nasal cavities soft tissues and cerebral abcesses. Images and clinical context were strongly suggestive of invasive fungal rhinosinusitis or nocardiosis. Surgical debridement of the maxillary sinuses and the nasal cavities was performed urgently. Biopsies concluded to actinomycosis. Antibiotherapy did not stop the fatal course of the disease.

Conclusion: Rhino-cerebral actinomycosis is a rare, serious and life-threatening disease. It can imitate others mycological or bacterial diseases. In order to not overlook it, the diagnosis must be systematically evoked in case of extensive rhinosinusal necrosis with cerebral extension.

Keywords: Actinomycosis, Actinomycetes, Rhinosinusitis, Brain

RÉSUMÉ

Objectif: Discuter les difficultés diagnostiques et de prise en charge de l'actinomycose rhinosinusienne à extension cérébrale.

Observation: Une patiente âgée de 48 ans a été admise aux urgences pour la prise en charge d'un coma acido-cétosique diabétique. La patiente était fébrile. L'examen objectivait une rhinorhée profuse et malodorante avec une nécrose des muqueuses palatine et nasale. Le scanner cranio-facial mettait en évidence une lyse partielle des parois sinusiennes maxillaires, des parties molles endonasales et des abcès cérébraux frontaux. L'imagerie et le contexte clinique étaient fortement évocateurs de rhinosinusite fongique invasive ou de nocardiose. Un débridement chirurgical des sinus maxillaires et des fosses nasales a été réalisé en urgence. Les biopsies ont conclu à une actinomycose. L'administration d'une bi-antibiothérapie n'a pas permis de freiner l'évolution fatale.

Conclusion: L'actinomycose Rhino-cérébrale est une affection rare, grave et potentiellement mortelle. Elle peut simuler d'autres affections mycologiques ou bactériennes. Afin de ne pas la méconnaître, il faut systématiquement l'évoquer devant une nécrose extensive rhino-sinusienne à extension cérébrale.

Mots-clés: Actinomycose, Actinomycetes, rhinosinusite, cerveau

INTRODUCTION

Actinomycosis is a rare, subacute bacterial disease caused by a gram-positive, sporulated bacillus of the genus Actinomyces. It can simulate other fungal or bacterial diseases. The facial localization of actinomycosis is frequent but bone involvement with extension to the brain is exceptional, difficult to diagnose and has a poor prognosis [1-3]. In order to not overlook this disease, the authors report a rare case of rhinosinus actinomycosis with brain extension and fatal evolution.

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ABSERVATION

A 48-year-old woman was admitted to the emergency for management of a ketoacid coma revealing a putative, fortuitously discovered type II diabetes. In emergency, rehydration, insulin therapy and antibiotic therapy (Amoxicillin - clavulanic acid: 1g three times a day by IVD) were performed. Upon awakening after 48 hours, she presented a necrotic, foul-smelling and profuse rhinorrhea. Anamnesis revealed headache and chronic rhinosinusitis syndrome evolving since 2 months. Obnubilation was noted on examination and the Glasgow Coma Scale was 13/15. The temperature was 38°2C. The blood pressure was 130/80 mmHg and the pulse was 125 beats/mn. Glycemia was 3.2g/l (17.6 mmol/l) with two crosses glycosuria and one cross ketonuria. Examination noted palatal and nasal necrosis with destruction of soft tissues of the nasal cavity and the nasal septum (Figure 1).

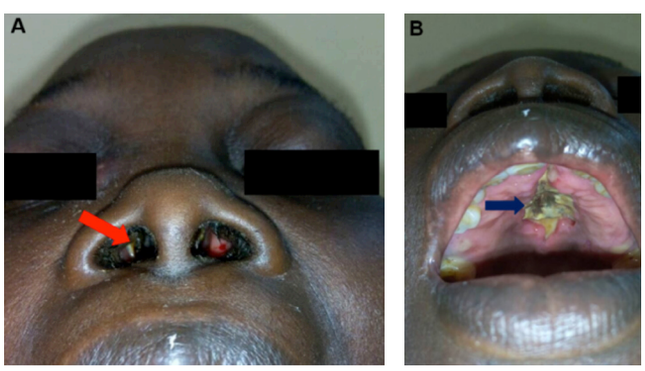

Figure 1:
A: Low angle view showing necrosis of the nasal mucosa (Red arrow);
B: Intraoral view showing medial necrosis of the palatal mucosa (Blue arrow)

We didn't notice tooth decay or dental treatment. Cardiopulmonary auscultation and chest x-ray were normal. Blood examinations revealed hyperleukocytosis (26000 elements with 84% of neutrophils) and anemia (9.4 g/dl). The retroviral serology was negative. The brain and maxillofacial CT scan revealed bilateral pansinusitis, partial maxillary sinuses lysis, destruction of nasal cavities soft tissues and frontal cockade brain lesions, suggestive of brain abscesses (Figure 2).

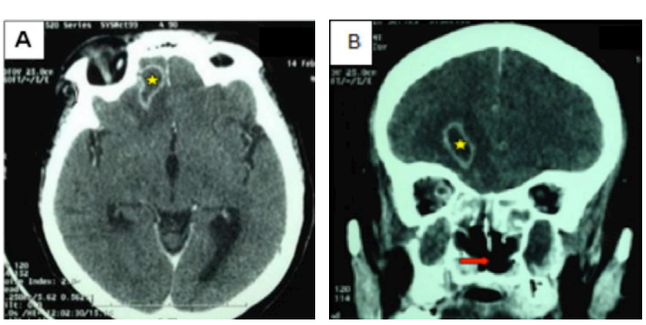

Figure 2: Brain and maxillofacial CT Scan. A: Cerebral abscess viewed as frontal cockade image (yellow star) in axial section. B: In coronal section, note maxillary and ethmoïdal sinusitis besides the cerebral abscess, note destruction of nasal cavities soft tissues (Red arrow).

Surgical debridement of nasal and maxillary sinuses lesions was performed urgently through a trans-collumelar approach (Figure 3).

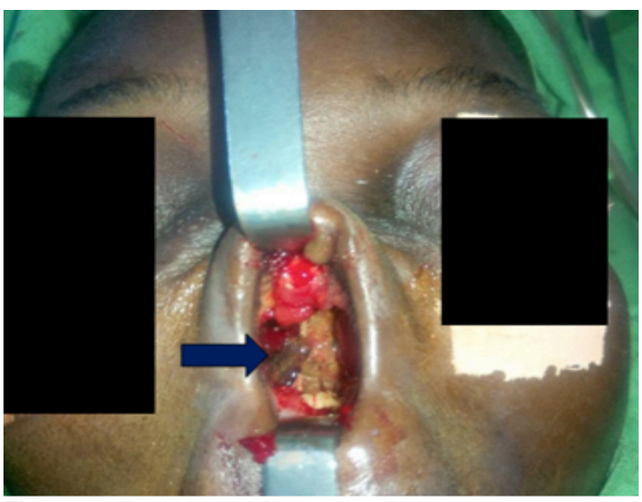

Figure 3: Total destruction of nasal septum (mucosa and cartilaginous septum), vomer, medial face of the maxillary sinuses and ethmoïdal cells with presence of necrosis.

Many samples were realized intraoperatively (necrotic tissue, bone, inflammatory tissue) and sent for bacteriological, parasitological, mycological and anatomopathological examinations. Diagnoses of invasive fungal rhinosinusitis with brain extension (mucormycosis or aspergillosis) or invasive nocardiosis were initially evoked, due to diabetes, sinusitis, lysis of maxillary sinuses and destruction of nasal soft tissues. Presumptive treatment was administered (Itraconazole tablets 100 mg two times per day). Bacteriological, mycological and parasitological examinations did not reveal any germs. Anatomoathological examination concluded in actinomycosis (Figure 4).

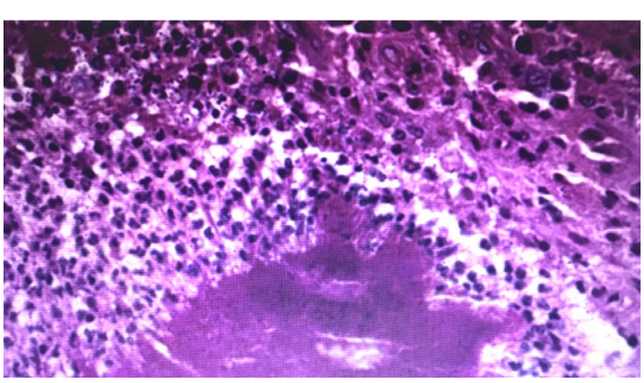

Figure 4: Suppurative granulomatous inflammation around an actinomycotic granule.

Death occurred after 8 days despite administration of Ceftriaxone and Metronidazole intravenously.

Discussion

This observation reports a brain complication of rhinosinusal actinomycosis in a diabetic woman. This extremely rare complication, with nonspecific signs, is often cause of misdiagnosis [2,4,5]. It evokes an invasive nocardiosis due to almost similar clinical manifestations of these two germs, belonging to the

order of Actinomycetales [3,6]. The brain localization is exceptional with high mortality (around 30%). Its localization is most often frontal or temporal and represents about 1% of actinomycotic localizations [5]. Extension to the brain from the nasal cavities and maxillary sinuses could result from either contiguous or hematogenous spread of the infection [4,8,9].

The classic Bergmann triad is not always observed and the signs may be less suggestive of brain abscess, only represented by headache or slight disturbance of consciousness as in this case report. Radiologically, brain lesions are polymorphic and nonspecific, most often unique [4,7,8]. Bone lesions in actinomycosis could be explained by the presence of type I fimbriae on certain species of Actinomycetes, making them able to bind collagen and lead to osteomyelitis [3,8].

There is no consensus for the management of actinomycotic brain abscesses. Some authors considered that treatment should be based on extensive surgical debridement while others recommended medical treatment alone. In case of actinomycotic brain abscess complicating initial nasal and sinusal actinomycosis, prolonged bi-antibiotic therapy crossing the blood-brain barrier, active on Actinomycetes and on commensal germs of the nasopharyngeal tract should be administered [2,3]. The combination of Ceftriaxone and Metronidazole may be recommended. In this observation, the atypical clinical presentation was responsible of misdiagnosis and delay in administration of appropriate medical treatment. Despite surgical debridement and adaptation of antibiotic therapy after histological examination, the outcome was unfavourable.

CONCLUSION

The extension of an abscess in the brain following necrotizing nasal and maxillary sinus pathology should not suggest only invasive mycosis or nocardiosis. This clinical presentation should also suggest cerebral actinomycosis so as not to overlook this particularly serious clinical form, although rare.

Compliance with ethical standards

Conflict of interest: The authors stated that there is no conflict of interest.

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