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Case Report

A forgotten difficult entity:Ozena Report of two cases

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Abstract. Atrophic rhinitis or ozena is a chronic nasal disease characterized by progressive atrophy of the nasal mucosa and underlying bone, accompanied by the formation of foul smelling, thick, dry crusts in the greatly enlarged nasal cavities. Although the incidence of ozena is greatly diminished, it is still encountered rarely. The etiology of ozena still remains enigmatic but hereditary, infectious, developmental, endocrine and nutritional factors have been implicated. We describe two patients with endoscopical, clinical evidence and computed tomography findings of atrophic rhinitis along with a review of the current literature.

Key words: Atrophic rhinitis, ozena, computed tomography, *klebsiella ozaenae*

1. Introduction

Primary atrophic rhinitis, or ozena, is a progressive chronic nasal disease characterized by atrophy of the nasal mucosa and with resorption of underlying bone, accompanied by the formation of foul smelling, thick, dry crusts in the greatly enlargement of the nasal space with paradoxical nasal congestion (1-4).

The etiology of ozena still remains enigmatic but hereditary, infectious, developmental, endocrine and nutritional factors have been implicated (2). Some bacteria such as *Klebsiella ozaenae, Proteus, Escherichia coli and Bordatella pertusis* have been isolated from the nasal secretions of patients as causative organisms (5,6).

The diagnosis is made clinically by the presence of characteristic changes inside the nose such as enlargement of the nasal cavities, atrophy of the mucosa and the presence of adherent, thick and green-yellow crusts, or microbiologically by isolation of putative bacteria such as *K. ozaenae* from nasal cultures (5,6). Although the incidence of ozena is greatly diminished, it is still encountered rarely (2). We describe two patients with endoscopical, clinical evidence and CT findings of atrophic rhinitis along with a review of the current literature.

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2. Case report

Case report 1

A girl aged 17 years with a 3-year history of nasal obstruction, fetid odor and anosmia was admitted to Pediatric Endocrinology polyclinic and she was referred to Otorhinolaryngology polyclinic. Diagnostic nasal endoscopy revealed green-yellow crusts in the nasal cavity (Figure 1) and atrophy of the turbinates. A swab from the nasal secretions was cultured and this resulted in the isolation of *K. ozaenae,* which showed susceptibility to ciprofloxacin. Her blood iron level, folate and vitamin B12 blood levels were within the normal range. Seroimmunologic tests were within the normal range. Her VDRL was negative and her blood profile showed no abnormality. Significant enlargement of the nasal cavities and hypoplasia of the maxillary sinuses were described (Figure 2). Coronal computed tomography (CT) scans of the paranasal sinuses revealed extreme atrophy of both inferior turbinates. (Figure 3).

The fetid odor disappeared after the first week of ciprofloxacin therapy (ciprofloxacin oftalmic gutt topically into both nasal cavities two times daily) and parenteral levofloxacin two times daily for three days, following oral levofloxacin tb 500 mg two times daily for ten days and at that time, in the patient all the crusts were cleaned endoscopically (Figure 4). In the one month follow-up visit the fetid odor was improved and crusting was diminished.

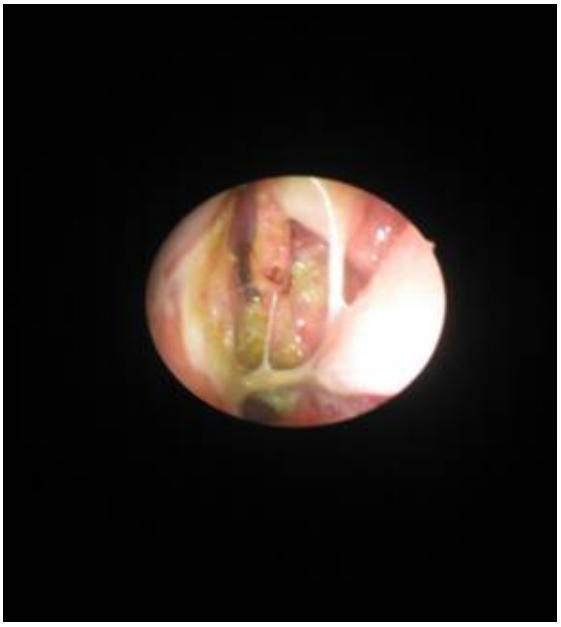

Fig. 1. Diagnostic nasal endoscopy shows green-yellow crusts in the left nasal cavity.(left nasal cavity).

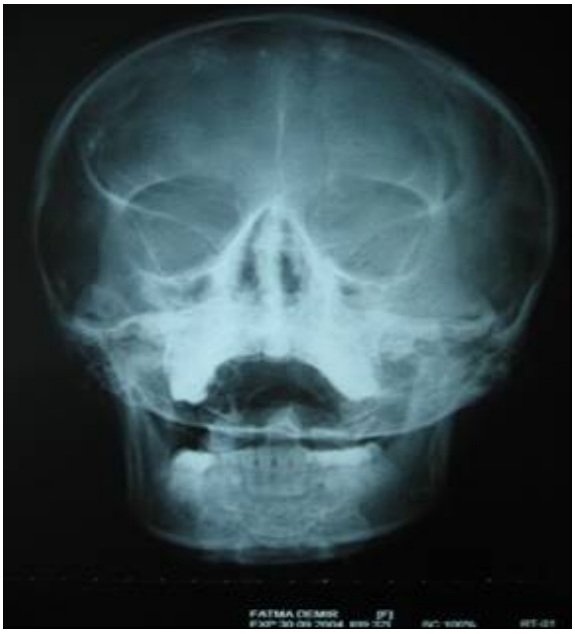

Fig. 2. Significant enlargement of the nasal cavities and hypoplasia of the maxillary sinuses are seen.

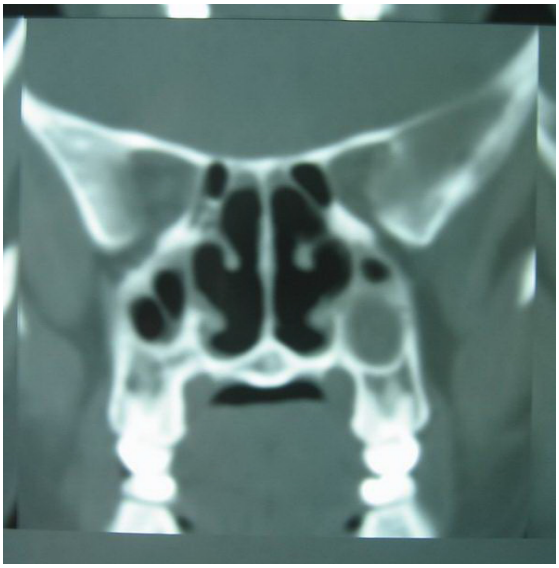

Fig. 3. Coronal CT scans of the paranasal sinuses reveal extreme atrophy of both inferior turbinates.

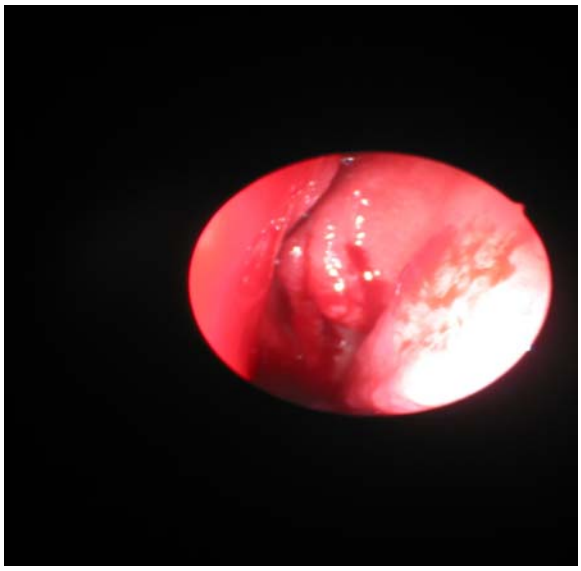

Fig. 4. The crusts were cleaned endoscopically.

But the patient is still complaining from nasal obstruction and anosmia.

Case report 2

A woman aged 16 years with a 5-year history of nasal obstruction, headache, fetid odor and anosmia was admitted. The fetid odor was detectable as soon as she entered the room. She described that some complaints were presant in her sibling who was fifteen years old.

Diagnostic nasal endoscopy revealed green-yellow crusts in the nasal cavity, posterior pharyngeal wall and atrophy of the turbinates (Figure 5). A swab from the nasal secretions was cultured and this resulted in the isolation of *K. ozaenae*, which showed susceptibility to ciprofloxacin.

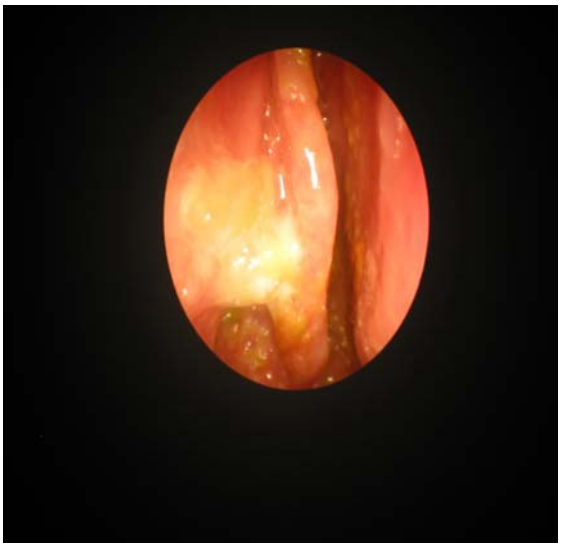

Fig. 5. Diagnostic nasal endoscopy reveals green-yellow crusts in the nasal cavity.

Other otolaryngological investigations of the patient were within normal limits. Biochemical investigations, which included peripheral blood smear, renal and hepatic functions, urine analysis and chest X-ray, were normal. Serum samples for HIV and VDRL/TPHA were non-reactive.

Coronal CT scans of the paranasal sinuses revealed extreme atrophy of both inferior turbinates (Figure 6).

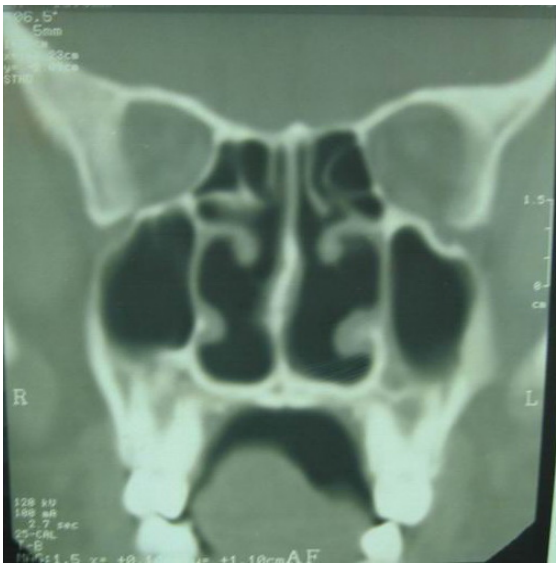

Fig. 6. Coronal CT scans of the paranasal sinuses reveals atrophy of both inferior turbinates.

Following repeated endoscopic removal of crusts, daily nasal irrigations, and antibiotic therapy (ciprofloxacin oftalmic gutt topically into nasal cavities two times daily for ten days, and oral ciprofloxacin two times daily for ten days). In the next visit, asal endoscopy revealed that, her nasal passages had become clearer. But unfortunately the patient could not be followed-up because she did not come to visits.

3. Discussion

Most authors classify atrophic rhinitis into two categories: primary and secondary (1,3). The primary form is of spontaneous onset, slowly progressive, and occurs in a previously healthy nose. Secondary atrophic rhinitis results from chronic sinusitis, trauma, sinonasal surgery, granulomatous diseases, radiation exposure, (1-3) and/or intranasal cocaine use (4). Secondary atrophic rhinitis accounts for most cases encountered today (1-3). Excessive turbinate surgery has been both implicated and acquitted in the literature as an etiology for secondary atrophic rhinitis (1). Our two cases were accepted primary because of spontaneous onset and no chronic sinusitis, trauma, sinonasal surgery, granulomatous disease, radiation exposure and intranasal cocaine use.

Diagnosis of atrophic rhinitis can usually be made on the basis of a thorough history, physical examination, biopsy, and imaging studies (1). Patients are diagnosed with primary atrophic rhinitis if their condition developed in a previously healthy nose and secondary atrophic rhinitis if their condition developed after sinonasal surgery, trauma, or chronic granulomatous disease (1). In our two cases diagnosis of atrophic rhinitis was made on the basis of history, physical examination, nasal endoscopy and CT and isolation of *K. ozaenae.* Patients can manifest with nasal obstruction, crusting, epistaxis, anosmia, and/or headache. A strong permeating odor may dominate the clinical picture. Nasal endoscopy usually reveals the presence of thick, adherent crusts that are yellow-green to gray-brown on the turbinates. The use of an endoscope (4 mm, 0 degrees) is critical for examining the posterior ends of the nasal cavities. Dried crusts may obstruct the small openings. Because there is a significant incidence of concurrent sinusitis, CT is frequently included in the diagnostic evaluation (4). In our two cases, there was strong odor and nasal endoscopy reveal-ed the presence of thick, adherent crusts that were yellowgreen to gray-brown on the turbinates and posterior pharyngeal wall.

CT is frequently included in the diagnostic evaluation of atrophic rhinitis. Pace-Balzan et al. (7) list characteristic changes identified by CT as the following:1) Mucosal thickening of the paranasal sinuses, 2) Loss of definition of the ostiomeatal complex secondary to resorption of

the ethmoid bulla and uncinate process, 3) Hypoplasia of the maxillary sinuses, 4) Enlargement of the nasal cavities with erosion and bowing of the lateral nasal wall, 5) Bony resorption and mucosal atrophy of the inferior and middle turbinates (7).

The disease appears to be endemic in subtropical and temperate regions like South Asia, Africa, Eastern Europe and the Mediterranean, and the patients are usually poor and live in unhygienic conditions (8). There is a slight female predominance (1.4 to 1) (2). Our two patients in the present study were females and living in dormitory.

K. ozaenae is the most commonly found pathogen in cultures as a causative organism (7). In our two cases swab from the nasal secretions was cultured and this resulted in the isolation of *K. ozaenae* and was given ciprofloxacin. The fetid odor disappeared and crusting diminished after antibiotherapy, indicating that *K. ozaenae* was responsible for the infection and odor.

Treatment is aimed at clearing secondary bacterial infections, reducing the amount of crusting, and alleviating the foul odor. Medical management usually suffices for most patients. The mainstay of treatment is continuous nasal hygiene, which can improve the patient's quality of life. Standard hygiene therapy entails vigorous and regular intranasal irrigations with a saline or sodium bicarbonate solution. In the event of purulent secretions or evidence of sinusitis, systemic antibiotic therapy is indicated and is guided by endoscopically obtained sinus cultures. In addition, periodic and regular debridement of the crusts may be helpful (4). Various surgical procedures, ranging from closure of the nostrils as suggested by Young to endonasal microplasty, have been employed (2).Treatment of this disease is conservative in the first place. Surgery is indicated if the medical treatment fails. The aim of surgery is either to narrow the nasal cavity or in special cases to close the nostril. Closure of the nostril (Young's operation), is achieved by raising a circular skin flap. Raising the skin flap is difficult, the suture line may break down and an excessive scar tissue may form resulting in vestibular stenosis (9). Lobo and their friends described a method for closing the nasal vestibule in cases of secondarily-acquired atrophic rhinitis. This involves occlusion of the nasal vestibule with an obturator made from dimethylpolysiloxane. Being a non-invasive method,it is specifically indicated in the management of cases of secondarily-acquired atrophic rhinitis where any surgical treatment is contra-indicated (10).

In conclusion, the exact pathophysiologic mechanism is still unknown in atrophic rhinitis. CT is very useful for the diagnostic evaluation of atrophic rhinitis. In our opinion, ciprofloxacin therapy, endoscopic removal of the crusts and long-term nasal irrigation are successful in the management of these patients.

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