Case Report / Olgu Sunumu

Atrophic rhinitis caused by *Cedecea davisae* with accompanying mucocele

Mukoselin eşlik ettiği *Cedecea davisae* nedeniyle gelişen atrofik rinit

Ömer Bayır, MD.,[1] Gökçe Aksoy Yıldırım, MD.,[1] Güleser Saylam, MD.,[1] Elvan Yüksel, MD.,[1] Ali Özdek, MD.,[2] Mehmet Hakan Korkmaz, MD.[3]

[1]*Department of Otolaryngology, Dışkapı Yıldırım Beyazıt Training and Research Hospital, Ankara, Turkey*
[2]*Department of Otolaryngology, Medical Faculty of Karabük University, Karabük, Turkey*
[3]*Department of Otolaryngology, Medical Faculty of Yıldırım Beyazıt University, Ankara, Turkey*

ABSTRACT

Atrophic rhinitis is a chronic inflammatory disease characterized by progressive atrophy of nasal mucosa. *Cedecea davisae*, a rare pathogen, is a new member of Enterobacteriaceae family. In this article, we report a patient with atrophic rhinitis whose culture test revealed *Cedecea davisae*. The patient was operated due to accompanying posterior ethmoid mucocele. Levofloxacin and nasal irrigation were administered for two months. Significant improvement was observed in patient's complaints and nasal signs at postoperative sixth month. In conclusion, *Cedecea davisae* has been thought to cause atrophic rhinitis and mucocele in this patient. Patient recovered with simple treatment. These bacteria should be kept in mind as a causative agent for atrophic rhinitis.

Keywords: Atrophic rhinitis; *Cedecea davisae*; endoscopic sinus surgery; mucocele.

ÖZ

Atrofik rinit, ilerleyici nazal mukozal atrofi ile karakterize kronik enflamatuvar bir hastalıktır. Nadir bir patojen olan *Cedecea davisae*, Enterobacteriaceae ailesinin yeni üyesidir. Bu yazıda, kültür testi *Cedecea davisae* gösteren bir atrofik rinit hastası sunuldu. Hasta eşlik eden posterior etmoid mukosel nedeniyle ameliyat edildi. İki ay levofloksasin ve nazal irigasyon uygulandı. Ameliyat sonrası altıncı ayda hastanın yakınma ve nazal bulgularında belirgin düzelme görüldü. Sonuç olarak, bu hastada *Cedecea davisae*'nin atrofik rinite ve mukosele neden olduğu düşünüldü. Hasta basit tedavi ile iyileşti. Bu bakteriler atrofik rinitin nedensel bir ajanı olarak dikkate alınmalıdır.

Anahtar Sözcükler: Atrofik rinit; *Cedecea davisae*; endoskopik sinüs cerrahisi; mukosel.

Atrophic rhinitis is a rare chronic infection characterized by atrophy of the nasal mucosa and concha with a sticky and malodorous secretion and crust that results in enlargement of the nasal cavity and paradoxical nasal congestion.[1] Mucocele, caused by impaired drainage of mucus through paranasal sinus ostium due to chronic sinusitis, polyps or tumoral masses in general, are benign cystic structures covered with sinus mucosa.[2] Although they have benign characteristics, they can cause erosions in neighboring bony structures, can pass away from

Available online at
www.kbbihtisas.org
doi: 10.5606/kbbihtisas.2015.93265
QR (Quick Response) Code

Received / *Geliş tarihi:* October 11, 2014 Accepted / *Kabul tarihi:* January 11, 2015

Correspondence / İletişim adresi: Ömer Bayır, MD. Dışkapı Yıldırım Beyazıt Eğitim ve Araştırma Hastanesi, Kulak Burun Boğaz Hastalıkları Kliniği, 06330 Altındağ, Ankara, Turkey.
Tel: +90 506 - 672 62 87 e-mail *(e-posta):* bayiromer@hotmail.com

sinus borders and extend through the orbita and sometimes the intracranial region. They become symptomatic due to the mass effect on neighbor structures.[2,3]

Cedecea davisae (C. davisae) is a gram negative, oxidase negative, rarely reported new member of the enterobacteriaceae family. There are few cases reported about this bacteria in the literature.[4-7]

In this case report, we discuss a patient operated on for posterior ethmoid mucocele due to atrophic rhinitis caused by a new bacterial agent *C. davisae* with the review of the literature.

CASE REPORT

A 57-year-old female was admitted to our outpatient clinic with a chief complaint of headache and symptoms of nasal congestion, bad odor from the nose and intermittent bleeding. Her nasal symptoms were present since childhood. Her headache existed for one year, localized in the frontal and retroorbital regions, aggravated by leaning forward, throbbing in character and improving with pain-killers. She did not have a history of trauma, toxin exposure, surgery or radiotherapy. Although she was prescribed sinusitis treatment, her symptoms were still continuing. On otorhinolaryngologic examination, her oral hygiene was bad, and intense yellow-green crusts with bad odor were bilaterally present in the nasal cavity on anterior rhinoscopy and nasal endoscopy (Figure 1). After cleaning crusts from both inferior and middle atrophic conchae, an intensive enlargement in both nasal cavities were observed. Other otorhinolaryngologic examinations were normal. She had no iron deficiency anemia, hypovitaminosis A or D or other abnormalities.

Paranasal sinus computed tomography (CT) and magnetic resonance imaging (MRI) showed both nasal passages were severely atrophic, with an appearance compatible with mucocele in the right posterior ethmoid cell, and this cell was narrowing the sphenoid sinus with expansion (Figure 2).

Endoscopic sinus surgery was performed under general anesthesia with the diagnosis of atrophic rhinitis and accompanying mucocele. The dense yellow-green material was cultured for microbiological evaluation. After cleaning this material, the ostium of the right sphenoid sinus was found enlarged. A posterior ethmoid cell just anterior to this sinus was opened, and dense mucoid drainage was aspirated (Figure 3). This cell was also enlarged and joined with the sphenoid sinus. Biopsies for histological evaluation were taken from the areas with cobblestone appearance of middle concha mucosa. Antibiotic treatment was arranged according to antibiogram (levofloxacin 400 mg/day per oral) and continued for two months. On her sixth month endoscopic

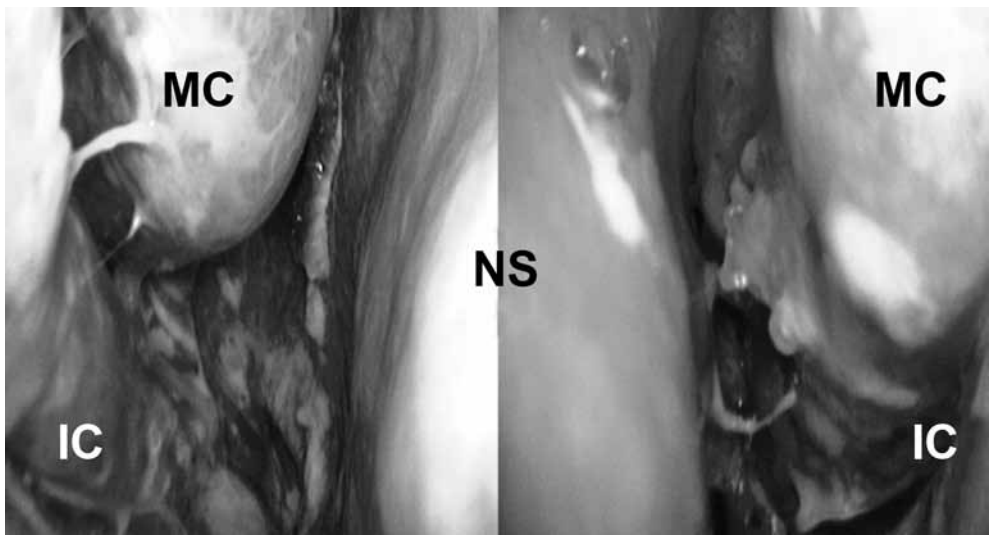

Figure 1. *Preoperative nasal endoscopic examination of the patient (images of both nasal cavities combined). Crusting and enlargement of the nasal cavities is shown in this figure.* *NS: Nasal septum; MC: Middle concha; IC: Inferior concha.*

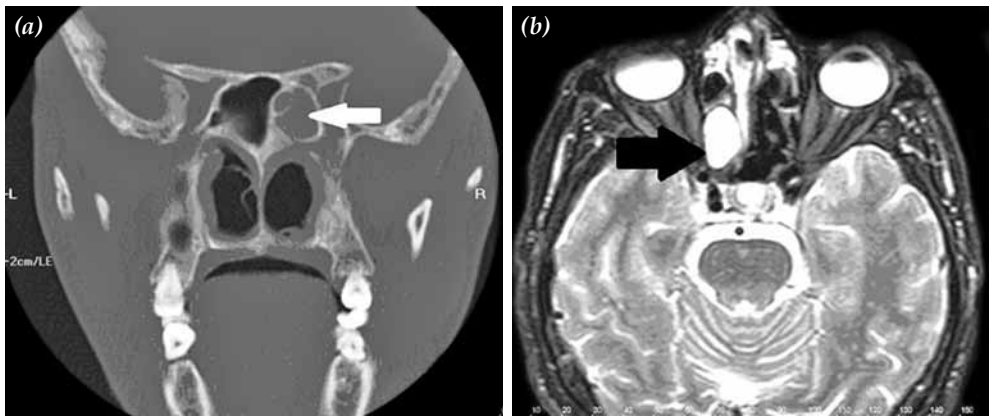

Figure 2. ***(a)*** *Computed tomography of the paranasal sinuses in axial section shows a lesion with soft tissue density in the right posterior ethmoid sinus (white arrow).* ***(b)*** *T_2-weighted magnetic resonance imaging of paranasal sinuses shows hiperintensity in the right posterior ethmoid cell (black arrow).*

examination there was a marked improvement in her complaints and nasal signs (Figure 4).

DISCUSSION

Atrophic rhinitis is classified as primary or secondary. Primary atrophic rhinitis is more commonly reported in under-developed countries and due to hygiene defects; secondary atrophic rhinitis is determined in patients with a history of nasal surgery, trauma, radiation therapy, chronic sinus infections, chronic granulomatous diseases or in advanced ages.[8] The patients have symptoms of nasal congestion, anosmia, bad odor, epistaxis and headache. Nasal cavity enlargement and crusting are two major findings on examination. Concha atrophy and bleeding mucosal ulcers are observed. Progressive atrophy of the nasal mucosa and underlying concha results in nasal crust formation. The nasal cavity enlarges and squamous metaplasia is dominant in the epithelium under microscopy.[9] In our patient, bilateral concha atrophy, crusting, odor, and cobblestone mucosa were observed on endoscopic nasal examination. In etiology, bacterial infections, *(Klebsiella ozaenae, Perez-Hofer bacil),* hypovitaminosis A or D, iron deficiency, excessive sympathetic nervous system activity, enlarged nasal cavity, toxin exposure, hygiene defects, history of nasal surgery or trauma, radiation treatment, or exposure to chronic

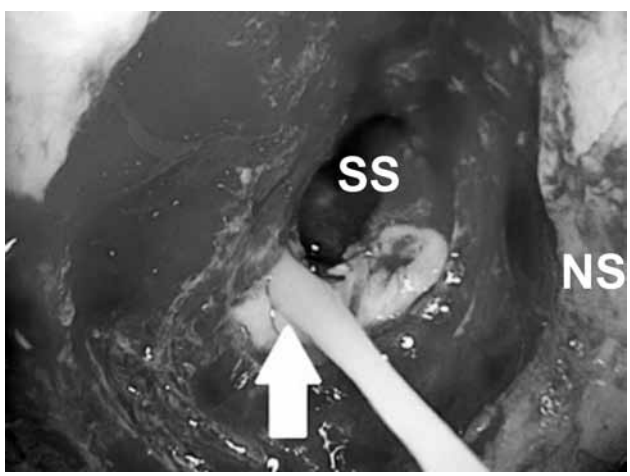

Figure 3. *Peroperative image of the patient.* *NS: Nasal septum; SS: Sphenoid sinus, white arrow; mucocele.*

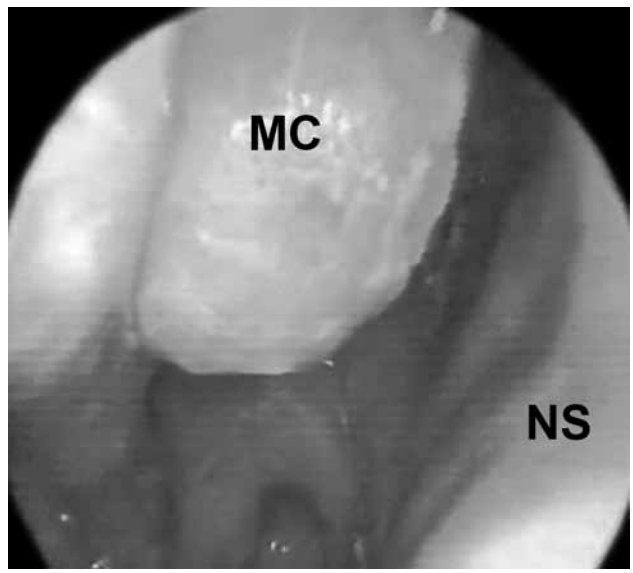

Figure 4. *Postoperative sixth month image of the patient's right nasal cavity. This image shows marked improving of the atrophic rhinitis.* *MC: Middle chonca; NS: Nasal septum.*

sinus infection very commonly are the causative factors.[10] Our patient had none of these etiologies in her medical history.

Mucosal infection caused by gram negative bacteria, especially *Klebsiella ozaenae* as a causative factor, has been demonstrated to result in primary atrophic rhinitis. Klebsiella is a gram (-), marked encapsulated, dormant, urease (+) bacillus from the enterobacteriaceae family. *Cedecea* is a new and rarely reported member of this family. It is a gram (-), oxidase (-), fermentative bacillus. It has many similarities with *Serratia* from the same family. Although first reported in the early 1980s, very few *C. davisae* cases have been reported in literature. This bacteria is isolated from blood, sputum, skin, urine, cutaneous and mucous membrane ulcers, gastrointestinal system and peritonea dialysis fluid.[4] *C. davisae* is reported as an opportunistic pathogen in immunosuppressed or advanced aged patients with many comorbid diseases. Mawardi et al.[6] reported the first case in the head and neck of *C. davisae* isolated from the oral ulcer of a 42-year-old patient with renal transplantation and under sirolimus treatment, and treated with ciprofloxacin for 21 days. This is the first case *C. davisae* isolated from the nasal cavity of a patient who was diagnosed with atrophic rhinitis. Since it is a member of the same family as *Klebsiella ozaenae*, an etiologic agent in atrophic rhinitis, and was cultured from crusts in the nasal cavity, the etiologic agent of atrophic rhinitis in this case is thought to be *C. davisae*.

A mucocele is caused by the chronic obstruction of sinus ostium that results in filling of the sinus cavity with mucous secretion and enlargement of bony walls. Mucoceles are most commonly reported in the frontal sinus but may also be seen in ethmoid, maxillary and sphenoid sinuses less frequently.[2] Although mucoceles are benign in structure, by forcing adjacent bony structures with pressure or destroying with the effects of enzymes, they may enlarge through important structures like orbita and intracranial structures.[2] Posterior ethmoid and sphenoid sinus mucoceles are rarely described however, since these sinuses are in close contact with important structures. The invasion of mucocele into these structures may cause severe symptoms.

Atrophic rhinitis and accompanying mucocele has not been reported before. The dense crusts and inflammatory process in atrophic rhinitis may have resulted in obstruction to the drainage pathways of the paranasal sinus. We conclude that mucocele developed secondary to the obstruction of posterior ethmoid sinus drainage in this case.

To the best of our knowledge, *C. davisae* isolation in the nasal cavity or paranasal sinuses has not been reported yet in literature. *C. davisae* has been reported in nasal culture of our patient and levofloxacin was given that was susceptible in antibiogram.

Conclusion

Herein we have presented a case of *C. davisae* isolated from the nasal cavity for the first time. In this case, this bacteria has been thought to cause atrophic rhinitis and mucocele. Cure was obtained with sinus surgery and oral levofloxacin treatment. *C. davisae* should be kept in mind as a causative agent among patients with atrophic rhinitis.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

REFERENCES

1. Druce HM. Chronic rhinitis. Allergy Proc 1990;11:295-8.
2. Arrué P, Kany MT, Serrano E, Lacroix F, Percodani J, Yardeni E, et al. Mucoceles of the paranasal sinuses: uncommon location. J Laryngol Otol 1998;112:840-4.
3. Kennedy DW, Josephson JS, Zinreich SJ, Mattox DE, Goldsmith MM. Endoscopic sinus surgery for mucoceles: a viable alternative. Laryngoscope 1989;99:885-95.
4. Akinosoglou K, Perperis A, Siagris D, Goutou P, Spiliopoulou I, Gogos CA, et al. Bacteraemia due to Cedecea davisae in a patient with sigmoid colon cancer: a case report and brief review of the literature. Diagn Microbiol Infect Dis 2012;74:303-6.
5. Ismaael TG, Zamora EM, Khasawneh FA. Cedecea davisae's Role in a Polymicrobial Lung Infection in a Cystic Fibrosis Patient. Case Rep Infect Dis 2012;2012:176864.
6. Mawardi H, Pavlakis M, Mandelbrot D, Woo SB. Sirolimus oral ulcer with Cedecea davisae superinfection.Transpl Infect Dis 2010;12:446-50.

7. Peretz A, Simsolo C, Farber E, Roth A, Brodsky D, Nakhoul F. A rare bacteremia caused by Cedecea davisae in patient with chronic renal disease. Am J Case Rep 2013;14:216-8.
8. Shehata MA. Atrophic rhinitis. Am J Otolaryngol 1996;17:81-6.
9. Chen HS. Desquamation and squamotransformation of rhinomucosa as a prodromal sign of atrophic rhinitis. ORL J Otorhinolaryngol Relat Spec 1984;46:327-8.
10. Dutt SN, Kameswaran M. The aetiology and management of atrophic rhinitis. J Laryngol Otol 2005;119:843-52.