OLGU SUNUMU CASE REPORT

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Candida parapsilosis Causing Chronic Canaliculitis: A Case Report and Review of Literature

Candida parapsilosis'e Bağlı Kronik Kanalikulit: Olgu Sunumu ve Literatürün Gözden Geçirilmesi

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ABSTRACT *Candida parapsilosis* causes opportunistic and nosocomial infection by creating the formation of biofilm. We present the unilateral chronic canaliculitis caused by *C. parapsilosis* in an immunocompetent patient. A 57-year-old female who underwent external dacryocystorhinostomy with placement of bicanalicular silicone stenting two years ago, was diagnosed with canaliculitis in our practice. *C. parapsilosis* was identified after the canaliculotomy and curettage. Topical fluconazole and oral itraconazole treatment were applied for 6 weeks and no recurrence was observed in 12 months follow-up. This is the first case that *C. parapsilosis* is described as a unique agent causing canaliculitis. It is important to raise awareness about unusual novel pathogens that can cause canaliculitis.

Keywords: Canaliculitis; Candida parapsilosis

ÖZET *Candida parapsilosis,* biyofilm formasyonu oluşturarak nozokomiyal ve fırsatçı enfeksiyonlara neden olmaktadır. Bu olguda, immünokompetan bir hastada tek taraflı kronik kanalikulite sebep olan *C. parapsilosis* olgusu sunulmaktadır. Yaklaşık 2 yıl önce eksternal dakriyosistorinostomi ve bikanaliküler silikon tüp entübasyonu yapılmış olan 57 yaşındaki kadın hasta kliniğimizde kanalikulit tanısı aldı. Yapılan kanalikülotomi sonrasında elde edilen küretaj materyalinden *C. parapsilosis* izole edildi. Hastaya 6 hafta boyunca topikal flukonazol ve oral itrakonazol tedavisi uygulandı ve 12 aylık izlemde herhangi bir rekürrens izlenmedi. Olgumuz, literatürde *C. parapsilosis*'e bağlı kanalikulit bildirilmiş olan ilk olgudur. Kanalikulite sebep olabilecek sıra dışı yeni patojenlerin farkında olunması önemlidir.

Anahtar Kelimeler: Kanalikulit; Candida parapsilosis

Canaliculitis, a rare inflammation of the lacrimal system, typically occurs as primary or secondary and classically presents with epiphora, medial canthal swelling, pouting punctum, and punctal discharge.[1] Diagnosis is often delayed and may be confused with chronic conjunctivitis, dacryocystitis, or inflammatory chalazion. Primary canaliculitis is mainly caused by *Actinomyces*, *Staphylococcus*, and *Streptococcus* species, while secondary canaliculitis is associated with the usage of lacrimal stents and punctal plugs, and most commonly caused by *Pseudomonas aeruginosa*.[1,2] However, several studies have described unusual pathogens related to primary and secondary canaliculitis.[1] Fungal lacrimal canaliculitis has been observed in the previous studies, but to our knowledge, there are no cases associated with *Candida parapsilosis*.[3-5] Herein, we reported chronic canaliculitis associated with *C. parapsilosis* in a patient with a past ocular history of a right external dacryocystorhinostomy (Ext-DCR) with placement of bicanalicular silicone stent.

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CASE REPORT

A 57-year-old female was referred to our practice because of epiphora, redness, intermittent mucopurulent discharge, and a para-canalicular abscess in the right upper lacrimal punctum. Based on the ocular history, she underwent Ext-DCR with bicanalicular silicone stenting because of primary acquired nasolacrimal duct obstruction 2 years ago, and the stent was removed at sixth months. Her symptoms of epiphora and purulent discharge started on the postoperative first month after the DCR surgery and the patient was recommended re-DCR surgery by different specialists. At the presentation, a right ocular examination revealed nasal conjunctival hyperemia with pouting punctum, a para-canalicular abscess resembling a chalazion and mucopurulent secretion from punctum upon pressure over the swelling (Figure 1a). The patient stated that the para-canalicular abscess had been present for 2 months and chalazion surgery was also recommended in another center. Because of the atypical location of a chalazion (medial to lacrimal punctum, no meibomian glands), due to a patent lacrimal irrigation from lower punctum we suspected for an upper lacrimal canaliculitis in the right eye. Canaliculotomy was performed with full curettage under local anesthesia and purulent material was obtained without concretion or canalicular stone. Purulent materials were sent in for a microbiological examination. The upper canaliculus and rest of the lacrimal system were irrigated with fortified cefazolin solution (50 mg/mL) peroperatively. Postoperatively, the patient was advised broad-spectrum topical (moxifloxacin, 4 times a day) and systemic (ciprofloxacin, 500 mg, 2 times daily) antibiotics until the microbiological profile was available. Eight days after the procedure, the microbiological profile was observed as non-albicans *Candida*, but its subtype could not be identified. The specimens were sent to the Public Health Institution of Turkey-National Mycology Reference Laboratory (PHIT-NMRL) to determine the subtype. Until the *Candida* subtype was obtained, the treatment of the patient was changed to topical 0.3% fluconazole drops 4 times a day, which is the only topical antifungal agent commercially available in our country. By the way, oral itraconazole was used 200 mg once daily, and the lacrimal pathway was also irrigated with diluted itraconazole solution (2 mg/mL) for three days.

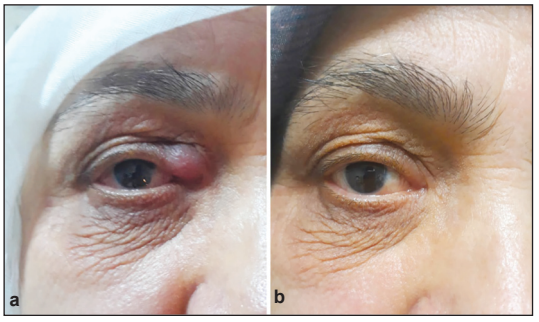

FIGURE 1: a) Preoperative and **b)** 12th month postoperative appearance of the patient.

YEAST IDENTIFICATION

The species identification of the strains was performed at the PHIT-NMRL. The mucoid-appearing yeast colonies on Sabouraud dextrose agar at 30 °C, the morphological evaluation in the Corn Meal-Tween 80 agar, observed ovoid blastoconidia, singly or in small clusters are seen along the curved appearance, short pseudohyphae at 25 °C for 72 hours (Figure 2) determination of negative urease activity at 25 °C for four days, assimilation features detected by API ID 32C (bioMérieux, France) kit, and On selective and differential medium for the isolation of fungi (BBLTM CHROMagarTM Candida Medium CHROMagar Microbiology, Paris, France) at 35 °C for 24 h are seen light rose to pink, large flat colonies with a whitish border (Figure 3) evaluated together with conventional mycological methods identified the species as *C. parapsilosis*.[6]

IN VITRO SUSCEPTIBILITY TEST

Susceptibility testing was performed for amphotericin B, fluconazole, itraconazole, posaconazole, voriconazole, and anidilofungin with the broth microdilution method according to CLSI M27-A3. Resistance to all agents was determined using the new species-specific CLSI CBPs (M27-S4). *Candida krusei* ATCC 6258 and *C. parapsilosis* ATCC 22019 reference quality control strains were used. The antifungal agents used in the study were amphotericin B, itraconazole, posaconazole, voriconazole, anidulafungin flucona-

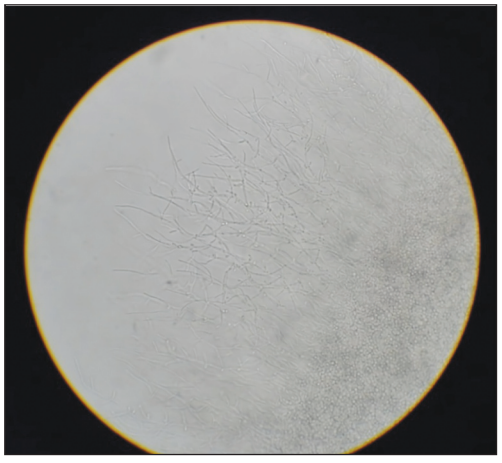

FIGURE 2: On cornmeal-Tween 80 agar at 25 °C for 72 h blastoconidia, singly or in small clusters are seen along the curved appearance, short pseudohyphae.

FIGURE 3: On selective and differential medium for the isolation of fungi (BBLTM CHROMagarTM Candida Medium) at 35 °C for 24 h are seen light rose to pink, large flat colonies with a whitish border.

zole, and caspofungin. According to the newly determined CLSI CBPs for fluconazole, voriconazole, anidilofungin, itraconazole, *C. parapsilosis* was susceptible.

In infectious diseases consultation, it was suggested that the current treatment should take at least 6 weeks. Her systemic examination, history, and blood testing including tests for HIV did not show immunosuppression. Two weeks after the treatment, her symptoms and clinical findings were resolved, and current anti-fungal treatment was continued for 6 weeks. On the evaluation in the 12th month, the patient's symptoms and clinical findings of canaliculitis did not recur (Figure 1b). Written informed consent was obtained from the patient.

DISCUSSION

Mycotic flora is not normal components of the lacrimal drainage system, but there are cases demonstrating atypical fungal presentation of canaliculitis such as *Fusarium*, *Aspergillus*, and *Candida* in the previous studies.[3-5] *Candida albicans* has been reported as the only *Candida* species to cause canaliculitis in the previous studies.[4,5] Ocular diseases linked to *C. parapsilosis* include keratitis and endophthalmitis, however there is no case of canaliculitis.[7,8] *C. parapsilosis* is a biofilm-forming yeast that causes opportunistic and nosocomial infections. It has an affinity for foreign material with infectious being related to dialysis catheter, prosthetic heart valve and other indwelling access devise.[9] Also the use of topical and systemic corticosteroid are known as a possible predisposing factor for *C. parapsilosis*.[5,7,8] Since the symptoms of our patient who was immunocompetent started after Ext-DCR with bicanalicular silicone stenting, obtaining of *C. parapsilosis* in our case can be explained by the affinity of the agent to the stents. The fact that our patient used corticosteroids for a long time due to misdiagnosis is also a possible predisposing factor.

Our patient was being misdiagnosed with nasolacrimal duct obstruction, conjunctivitis, and even chalazion, which can lead to the recommendation of unnecessary interventions and delay of diagnosis and appropriate treatment. As stated in the literature, there are no specific clinical findings and treatment strategies for mycotic canaliculitis.[3-5] The resolution of canaliculitis depends on the thorough removal of the concretions or discharge from within the canaliculus, and the use of antimicrobial therapy, which is the causative agent.[2] In the current case, after canaliculotomy and curettage, we applied the topical fluconazole and systemic itraconazole treatment which has been detected to be susceptible with *in vitro* susceptibility test. These agents also have been reported as a safe and effective antifungal drug for the treatment of ocular *C. parapsilosis*, including keratitis and endophthalmitis.[7,10] There is no clear evidence for the duration of the treatment in *Candida* canaliculitis in the literature, however in the keratitis and endophthalmitis caused by *Candida*, the recommended du-

ration of treatment has been reported to be 6 to 12 weeks.[10] In our case, symptoms and clinical findings resolved in the second week of treatment, which was continued for four more weeks with the recommendation of infectious diseases, and completed in 6 weeks.

To our knowledge, this case in which we reported canaliculitis due to *C. parapsilosis* is the first reported case of canaliculitis among non-albicans species in the literature. Microbiological evaluation in all cases of canaliculitis is important in identifying novel pathogens to contribute to the definition, etiology, epidemiology, and treatment of canaliculitis.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Fatma Çorak Eroğlu;* ***Design:*** *Fatma Çorak Eroğlu, Nilgün Karabıçak;* ***Control/Supervision:*** *Fatma Çorak Eroğlu, Nilgün Karabıçak, Emine Şen;* ***Data Collection and/or Processing:*** *Fatma Çorak Eroğlu, Nilgün Karabıçak, Mihriban Yücel;* ***Analysis and/or Interpretation:*** *Fatma Çorak Eroğlu, Nilgün Karabıçak;* ***Literature Review:*** *Fatma Çorak Eroğlu, Emine Şen;* ***Writing the Article:*** *Fatma Çorak Eroğlu;* ***Critical Review:*** *Fatma Çorak Eroğlu, Emine Şen, Mihriban Yücel;* ***References and Fundings:*** *Fatma Çorak Eroğlu;* ***Materials:*** *Fatma Çorak Eroğlu, Nilgün Karabıçak, Mihriban Yücel.*

REFERENCES

1. Kaliki S, Ali MJ, Honavar SG, Chandrasekhar G, Naik MN. Primary canaliculitis: clinical features, microbiological profile, and management outcome. Ophthalmic Plast Reconstr Surg. 2012;28(5):355-60. [Crossref] [PubMed]
2. Freedman JR, Markert MS, Cohen AJ. Primary and secondary lacrimal canaliculitis: a review of literature. Surv Ophthalmol. 2011;56(4):336-47. [Crossref] [PubMed]
3. DONAHUE HC. Unusual mycotic infection of the lacrimal canaliculi and conjunctiva. Am J Ophthalmol. 1949;32(2):207-10. [Crossref] [PubMed]
4. Newton JC, Tulevech CB. Lacrimal canaliculitis due to Candida albicans. Report of a case and a discussion of its significance. Am J Ophthalmol. 1962;53:933-6. [Crossref] [PubMed]
5. Rüchel R. Chapter 2: Antifungal Agents. Behrens-Baumann W. Mycosis of the Eye and Its Adnexa. Developments in Ophthalmology. Vol. 32. 1st ed. Basel: Karger; 1999. p.27-107. [Crossref]
6. Larone DH. Yeast and yeastlike organisms. Laboratory technique. Medically Important Fungi: A Guide to Identification. 4th ed. Washington, DC: ASM Press; 2002. p.109-43, 325-49.
7. Li PH, Chen CC, Liou SW. Candida parapsilosis keratitis treated successfully with topical and oral fluconazole. Taiwan J Ophthalmol. 2016;6(3):155-7. [Crossref] [PubMed] [PMC]
8. Scartozzi R, Tom D, Liggett PE, Flynn HW Jr. Postoperative endophthalmitis caused by Candida parapsilosis: clinical features and treatment outcomes. Ophthalmic Surg Lasers Imaging. 2011;42 Online:e37-9. [Crossref] [PubMed]
9. van Asbeck EC, Clemons KV, Stevens DA. Candida parapsilosis: a review of its epidemiology, pathogenesis, clinical aspects, typing and antimicrobial susceptibility. Crit Rev Microbiol. 2009;35(4):283-309. [Crossref] [PubMed]
10. Matsumoto Y, Murat D, Kojima T, Shimazaki J, Tsubota K. The comparison of solitary topical micafungin or fluconazole application in the treatment of Candida fungal keratitis. Br J Ophthalmol. 2011;95(10):1406-9. [Crossref] [PubMed]