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Case Report

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Delayed-onset infection by atypical pathogens 35 years post-frontalis sling surgery

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ABSTRACT

Frontalis sling suspension (FSS) remains a venerable surgical intervention employed in the management of congenital eyelid ptosis, particularly in cases of compromised levator function. Despite its prevalence, the array of complications associated with this procedure is well-explored and meticulously documented. In this report, we delineate the intriguing case of a 57-year-old male patient who underwent FSS three and a half decades prior, only to manifest recurrent preseptal abscess formation at the silicone rod (SR) site a staggering 35 years post-surgery. This instance stands out not only for the unprecedented delay of abscess onset after FSS but also for the novelty of the microbial strains isolated from this anatomical location, previously unexplored in the medical literature.

Keywords: *Citrobacter freundii, Finegoldia magna*, frontalis sling surgery, ptosis, silicon rod

INTRODUCTION

Frontalis sling surgery (FSS), utilizing a silicone rod (SR), is a well-established technique for addressing severe upper eyelid ptosis, particularly in cases where the levator muscle function is poor or absent.[1] This surgical approach leverages the frontalis muscle to elevate the eyelid, thereby compensating for the deficient levator mechanism. The use of a silicone rod offers several advantages, including durability, flexibility, and biocompatibility, making it a preferred material for creating the sling.[1]

This case report aims to detail postoperative considerations associated with the application of silicone rod frontalis sling surgery in patients with significant upper eyelid ptosis. Additionally, it highlights a rare complication, detailing the case of a patient who developed a silicone rod infection 35 years postoperatively, providing insights into the long-term risks and management strategies for such late-onset infections.

CASE

A 57-year-old man with left upper eyelid preseptal cellulitis who had no underlying systemic disease was referred to our oculoplastic clinic. The patient stated that FSS was performed on his left upper eyelid for congenital ptosis 36 years ago. A preseptal abscess was observed at the SR's location, and the skin overlaying it was found to be sensitive by palpation. During ophthalmological examination, no pathological finding was detected on either anterior or posterior segments. Ocular motility was mildly limited in the upper gaze due to the compression of the abscess. The patient stated this was the fourth time in the past twelve months that he had an infection of this kind and had not experienced any infections until last year. To rule out the presence of a systemic disease or condition that could lead to immunosuppression a detailed laboratory workup was performed. Complete blood count, C-reactive protein, erythrocyte sedimentation rate, serological tests including HIV, and fasting blood sugar level with hemoglobin A1C (HbA1c) were evaluated and detected in normal ranges. Also, double-stranded DNA antibody, antinuclear antibody, anti-Ro, anti-La, and anti-Sjögren's-syndrome-related antigen A autoantibodies/anti-Sjögren's-syndrome type B autoantibodies all resulted in negative. During the chest X-ray, no abnormal findings were observed. Systemic antibiotic treatment with ampicillin/sulbactam combination (750 mg, twice a day) and metronidazole (500 mg, once a day) administered with hot compress and topical oxytetracycline hydrochloride ointment for one week. The preseptal cellulitis on the area of SR was significantly improved after 48 hours, however, there was no noticeable decrease in the size of the nodular formation at the end of

the first week (Figure 1). During the physical examination after the antibiotic treatment, the abscess formation was soft, mobile, and had well-demarcated borders. Since the patient had a history of relapses after the cessation of the previous antibiotic treatments, surgical abscess drainage was planned in order to prevent any relapse. During the drainage, an incision was made from the upper eyelid crease, and the orbicular muscle fibers were bluntly dissected to visualize SR. After the dissection, a yellow, fragile tissue covering the SR completely was detected (Figure 2). Although the necrotic tissue overlaying the SR was removed with the greatest care, full extraction of the fibrotic tissue and the SR was not possible due to their stiff attachment to the adjacent tarsal tissue. The removed part of the SR and the necrotic tissue were directly placed in a sterile tube containing 1 ml thioglycolate medium for protection of both aerobic and anaerobic microorganisms. Postoperatively, the patient was treated with amoxicillin and clavulanate, topical chloramphenicol ointment 3% for one week. Swelling and inflammatory signs improved after the treatment. The biopsy resulted in granulation tissue for the necrotic material. *Finegoldia magna (F. magna)* and *Citrobacter freundii (C. freundii)* strains were isolated from the microbiological cultures. During a 6-month follow-up, no recurrence was experienced. Eyelid elevation was achieved spontaneously after the treatment and the margin reflex distance of the upper eyelid was measured approximately 1.5 millimeters (Figure 3).

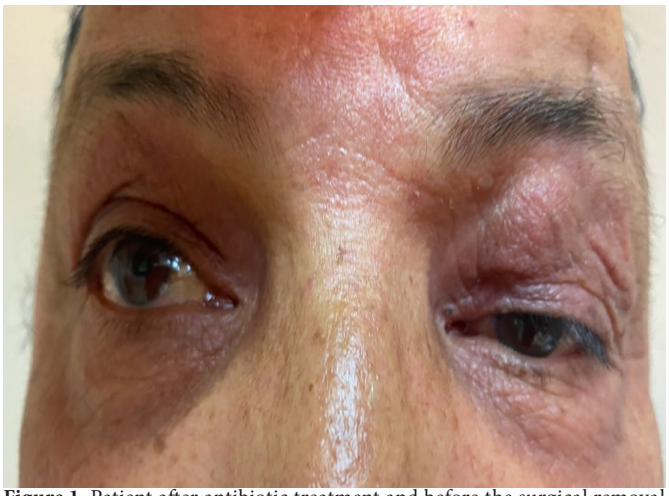

Figure 1. Patient after antibiotic treatment and before the surgical removal of the silicone rod

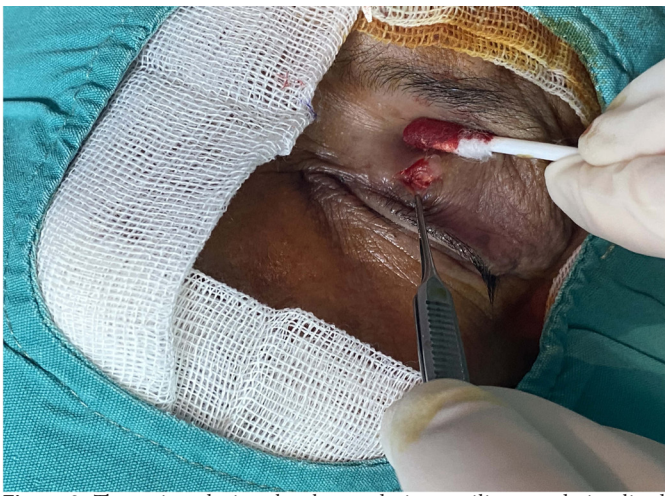

Figure 2. The patient during the abscess drainage, silicone rod visualized and observed covered by a yellow, fragile tissue

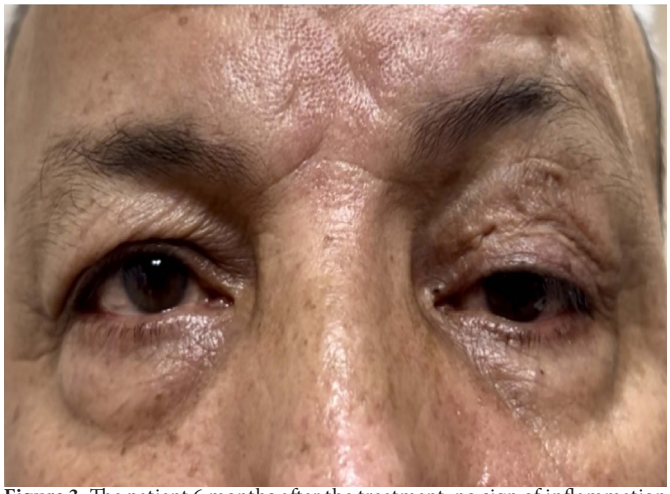

Figure 3. The patient 6 months after the treatment, no sign of inflammation and upper eyelid elevation was observed

DISCUSSION

The current patient presents an unusual case of an FSS resulting in a local inflammatory response with purulent discharge 35 years after a congenital ptosis correction with an SR. This is the first case reported in the literature in which late-onset inflammation was observed for the longest period of time following the FSS. The other published case with a late-onset infection on the surgical site was reported by Hostovsky et al.[2] years after the FSS. In the current case, segmental removal of the necrotic material that overlays the SR adequately managed to cessation of recurrence of the previous infection. The eventual outcome was favorable. Complete SR removal must be taken into account in FSS to prevent problems like infection and extrusion; however, in the present case, it was not possible due to potential tissue damage from the extensive extraction.[3] Our case indicates that it might be beneficial to prevent a localized infection by removing a certain amount of infected tissues.

In the current literature, cases of postoperative wound infections after FSS caused by *nontuberculous mycobacterial* infections are documented.[4] In the current case the isolated strains were in contrast with the literature findings. *F. magna* is an anaerobic, gram-positive *coccus* that rarely causes implant-related infections of bone and joints.[5] It also has been isolated from soft tissue abscesses and acute as well as chronic wound infections.[6,7] *F. magna* is a member of the human gut microbiome and is also frequently found on the skin.[6] It adheres to inert surfaces and has the ability of to produce biofilms but it is responsive to the majority of antibiotics.[6] *F. magna*'s incidence of clinical infections is likely underreported due to difficulty cultivating it and obtaining high-quality anaerobic specimens.[7] About 90% of *F. magna* types possess special enzymes and proteins that facilitate adhesion to specific layers of the skin, enabling it to get through the patient's protective barrier and induce rapid and persistent infections.[8]

Citrobacter species are members of the *Enterobacteriaceae* family of facultative, anaerobic, gram-negative bacilli which are typically detected in water, soil, food, and human intestines.[9] Previously identified as pollutants with low virulence, they are directly linked to a broad range of diseases involving soft tissue.[9,10] *C. freundii* represents an important pathogen, particularly in immunocompromised

people however sporadic infections can occur, and strains could cause wound infections.[9] There is only one case of an immunocompetent patient who developed a spontaneous skin infection caused by *C. freundii*.[10] The isolation of this bacteria is surprising since it has the tendency to infect mostly immunocompromised patients and there is no published case of isolation this strain from periocular area. Since our patient is immunocompetent and the wound infection has been going on for a long time and has not been treated adequately, these could all lead to the reproduction of *C. freundii*. This finding should be supported by other comprehensive studies with materials obtained from other wound infections and abscesses.

The current case stands as an extraordinary rarity, characterized by the unprecedented timing of abscess formation and the isolation of remarkably unusual strains. As far as our extensive exploration into medical literature extends, this case marks the first instance of soft tissue abscess emergence a remarkable 35 years post-FSS. Moreover, the occurrence of a periocular infection attributable to these strains, coupled with such a protracted latency period culminating in abscess formation, remains unparalleled in documented medical records. However, comprehending the mechanism through which these strains induce delayed-onset infections necessitates further comprehensive data gathering. Furthermore, the imperative recognition of these specific strains bears significant importance in the context of late-onset wound infections following surgeries necessitating the use of foreign body implants.

ETHICAL DECLARATIONS

Informed Consent

Written informed consent was obtained from the patient for the publication of this case report and any accompanying images. The patient has reviewed the final draft of the manuscript and consents to its publication. The authors are grateful to the patient for his cooperation and contribution to medical knowledge.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

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Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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