

Case Report

Tungiasis infestation of dermis fat graft in an anophthalmic socket



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Abstract

A patient with an anophthalmic socket with a dermis-fat graft (DFG) developed inflammation and a foul odour in the right socket. The DFG was surgically removed and Tungiasis infestation was detected. This is the first case to report Tungiasis infestation in a DFG in an anophthalmic socket.

Keywords: Dermis fat graft, Anophthalmic cavity, Orbital inflammatory reaction, Tungiasis infestation

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Introduction

Dermis graft (DFG) is a safe and effective option to rehabilitate a contracted anophthalmic cavity, simultaneously replacing volume and enlarging the anterior surface.^{1,2}

However, there are complications of the DFG procedure which mainly occur in severely traumatized contracted sockets with previous surgeries or in patients with abnormal wound healing.^{3–5}

The DFG is composed of dermis and fat and with the epidermis removed, the surface of the socket is “exposed”, with no barrier against external agents.

We present a case report of a patient with a DFG in an anophthalmic socket who developed Tungiasis infestation postoperatively resulting in orbital inflammation.

Case report

A 44-year-old Saudi male who resides on a farm containing livestock (camels and sheep) underwent evisceration of the right eye after trauma. The left socket was eviscerated 2 years prior to presentation due to another trauma to the eye. The left eye did not receive an implant. The right socket underwent evisceration with a four petal technique and received a DFG. This technique has been previously described.⁶ The DFG was harvested from the antero-superior iliac area and was 30x25 mm which was 30% larger than the orbital defect. The skin over the surface of the graft was excised, and the graft was composed of only dermis and fat. The extraocular muscles attached to the sclera were sutured to the dermis of the DFG using 4-0 polyglactin

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(Vicryl, Ethicon Inc., USA) with interrupted sutures. The graft was sutured to the Tenon capsule and conjunctiva using 6-0 polyglactin (Vicryl, Ethicon Inc., USA) with interrupted sutures.

There were no intraoperative complications. Postoperatively, the patient was prescribed a combination antibiotic-steroid drop (Maxitrol, Alcon Inc., Fort Worth, Tx, USA) qid for 2 weeks. One month later, the patient presented with pain and pruritus in the right socket, with discharge and a foul odour. On examination, the socket was injected and a whitish secretion was detected. The fat was not exposed, the dermis appeared blanched, with melted edges and multiple brown dots superficially (Fig. 1). Magnetic resonance imaging (MRI) indicated infectious process within the fat graft with restricted pattern on diffusion-weighted images and peri-graft infiltration suggestive of graft infection with fat necrosis and episcleral infiltration (Fig. 1). Graft swab cultures were positive for Methicillin-resistant *Staphylococcus aureus*. The patient was admitted to underwent intravenous administration of Gentamicin 100 mg (Hospira Inc., Lake Forest, IL, USA) every 8 h, Cefazoline 1gr (Glaxosmithkline, Research Triangle Park, NC, USA) every 8 h and Vancomycin hydrochloride (preservative free) 10 g (Hospira, Inc., Lake Forest, IL, USA) every 8 h. Additionally, the patient was prescribed a topical combination of Neomycin, Polymyxin B and Dexamethasone ointment (Maxitrol; Alcon Inc., Fort Worth, Tx, USA) qid. However, the infection was unresponsive to the antibiotic regimen and the DFG was surgically removed after 2 days.

Microscopic examination of the DFG identified refractile, pigmented, elongated chitinous exoskeleton structures, ova embedded in dense connective and fibrous tissue. Polymorphonuclear leukocytes infiltrated the connective tissue with

surrounding necrosis (Fig. 1). Fragments of insect leg were also embedded in the dermis. Gram stain was negative. The size and structure of the exoskeleton and leg fragments were suggestive of a *Tunga penetrans* infestation.

A family member confirmed the farm where the patient was living and his bed was full of fleas; most of the farm animals had proliferative cutaneous lesions consistent with Tungiasis. The patient had flea bites in other areas of the body. Two weeks after DFG removal the patient still complained of pain and a foul odour in the right socket.

MRI (T2) showed residual heterogeneous signal intensity along the central aspect of the orbital fat, extending posteriorly. A second surgical procedure was performed to remove the remaining orbital tissues surrounding the previous DFG. Histopathology indicated chronic non-granulomatous inflammation with no flea tissue or eggs. Six months after the second surgery, the patient was asymptomatic and the socket was quiet.

Discussion

We present a bilaterally blind patient living in a farm in contact with flea infested animals in southwest Saudi Arabia. This is the first report in the literature of flea infestation in an anophthalmic socket. The patient developed an intense inflammatory reaction in his right socket after a DFG. In a DFG, the dermis is exposed providing no protection against infectious agents or parasites. Hence fleas can easily deposit eggs over the graft. As the patient was blind, he could not detect the fleas in the surrounding environment. The inflammatory reaction included pruritus, inflammation and pain and resulted in extensive DFG necrosis, with secretion and a foul odour.

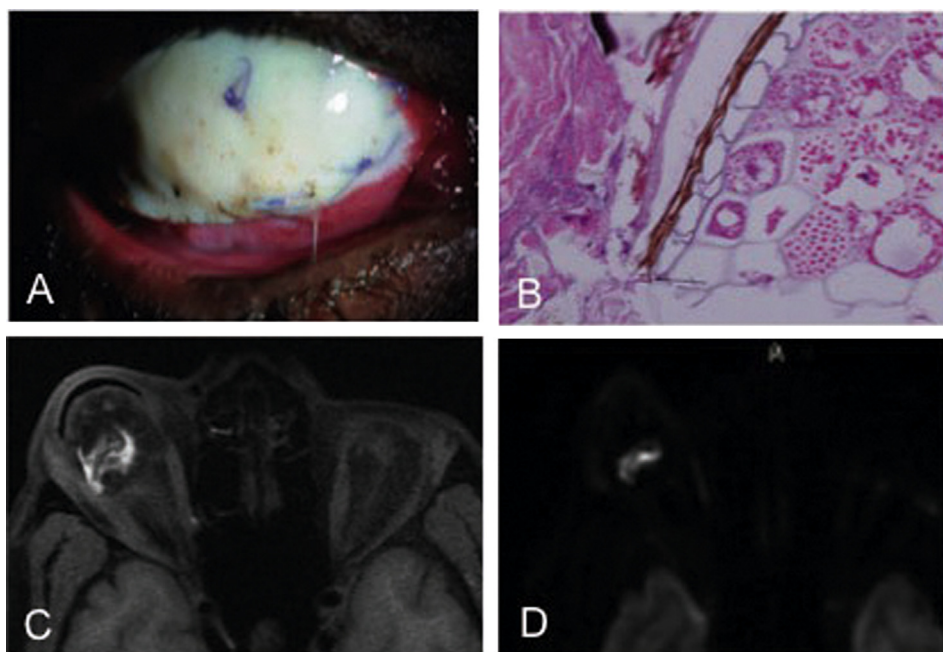


Fig. 1. (A) Clinical appearance at presentation (two weeks after DMFG implantation), showing vicryl sutures still in place, melted white dermis with brownish small dots overall in the inferior part of the dermis and severe conjunctival hyperemia; (B) portions of chitinous exoskeleton, insect leg (arrow) and ova embedded (*) in the collagenous dermis (hematoxylin and eosin; original magnification X 20) (C, D) Axial fast imaging employing steady-state acquisition (FIESTA) MRI, (C) Axial T1-weighted fat-suppressed postcontrast image showed dermal fat graft (long white arrow) with heterogeneous enhancement with a restricted pattern; (D) axial diffusion weighted image (thick arrow) with high signal intensity throughout the area of abnormal enhancement.

The final diagnosis of Tungiasis was based on a history of exposure and the histologic findings that suggested morphological characteristics previously reported⁷ in Tungiasis infected skin lesions including, the chitinous exoskeleton, ova and components of the leg.

Tungiasis is caused by the sand fly *Tunga penetrans* that causes an inflammatory skin disease.⁸ The sand fly is commonly found in the tropical regions in the Middle East, Africa, India, Caribbean, Central and South America. The flea is small and measures about 1 mm in size⁹ and the structure and size of parts identified on pathological examination corresponded to previously reported microscopic features.

The gravid female of *Tunga penetrans* penetrates the skin of the host. In our patient the sand flea, most likely, directly entered the dermis, easily reaching the graft. The fleas defecate and expel eggs and stay in contact with the air via an abdominal cone, leaving an opening which can serve as a portal of entry for microbes and other parasites. The gravid female begins laying eggs within 8–10 days and can lay thousands of eggs.⁷ This flea has the predilection to infect skin, hence, It was not surprising that the dermis fat graft was infected. The infection resulted in severe inflammation and necrosis. We believe that the small black dots over the DFG were likely the parasite.

Infestation of ocular and orbital tissues by fly larvae or ophthalmomyiasis or parasitic infections of the eyes,¹⁰ is a more common than flea infestation¹¹ and should be included in the differential diagnosis. Flea infestation is easily recognized by the typical granuloma with a central ulcer containing white larvae.^{12,13}

To our knowledge, Tungiasis has not previously been described in an anophthalmic socket or related to a DFG.⁵ Previous reports have published *Musca domestica* infestations of the conjunctiva, lingual tonsil, thigh, elbow, gluteal, ischial protuberances and knees.¹³

A Tungiasis lesion can be quite invasive, warranting surgical removal. In the current case, the graft infestation prompted us to remove the DFG. However, the patient complained of continued pain and odour and we performed a second procedure to excise the tissue surrounding the explanted DFG to ensure all the orbital tissues were free of fleas. Complete local recovery usually occurs after removal of the intact flea.

Other postoperative complications of DFG include, graft necrosis, hair growth on the graft, wound dehiscence in the donor area, keratinization of the socket, graft wound dehiscence,

donor wound hematomas and graft overgrowth requiring re-operation or debulking.⁵

In this case, the DWI sequence was very helpful when cross-referenced with conventional sequences in multiple planes for lesion localization and characterization. DWI improved diagnostic confidence when used in conjunction with contrast-enhanced imaging for localization of the infested regions.

In summary, Tungiasis should be considered in the differential diagnosis of a necrotic and chronically inflamed DFG in an anophthalmic socket, especially when patients are living in an environment conducive to fleas. Definitive treatment involves a surgical approach to eliminate the fleas, surrounding inflammation and necrosis.

Conflicts of interest and source of funding

The authors declared that there is no conflict of interest.

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