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Slides:(1): H/E Broken

TITLE: ORBIT WITH BROWN SPOTS

CASE REPORT

A 44-year-old Saudi male farmer with live stock (camels and sheep) underwent bilateral evisceration after trauma. The left socket did not receive an implant and the right socket received a Dermis fat graft. The fat was placed inside the orbit and the perimeter of the dermis was sutured to the rectus muscles and conjunctiva with 6-0 polyglactin. There were no intraoperative complications. Postoperatively, the patient was prescribed a combination antibiotic-steroid drop. One month later, the patient presented with pain and pruritus in the right socket, with discharge and a foul odor. 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 brown dots superficially. Magnetic resonance imaging (MRI) indicated inflammation restricted to the graft showing heterogeneous infiltration with necrosis, infected process within the fat graft with restricted pattern on diffusion-weighted images and peri-graft infiltration suggestive of graft infection with fat necrosis. The patient was admitted and a graft swab cultures were positive for Methicillin-resistant *Staphylococcus aureus*. The patient underwent intravenous administration of Gentamicin 100 mg 8 hours, Cefazoline 1gr every 8 hours and Vancomycin hydrochloride (preservative free)10 g every 8 hours. Also, the patient was prescribed a topical combination drop of Neomycin, Polymyxin B and Dexamethasone ointment qid. However, the infection was non-responsive to the antibiotic regimen and after two days the DFG was surgically removed.

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. 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 infestation by **Tunga penetrans.**

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

MRI (T2) showed residual heterogeneous signal intensity along the central aspect of the orbital fat, extending posteriorly (Figure 1F). A second surgical procedure was performed to remove the remaining orbital tissues surrounding the previous DFG. Histopathology indicated chronic non-granulomatous inflammation with no fleas' parts 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 on a farm in contact with flea infested animals in southwest Saudi Arabia. He 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[6] resulted in extensive DFG necrosis, with secretion and a foul odor.
The final diagnosis of Tungiasis was based on history of exposure and the histologic findings, that suggested morphological characteristics previously reported in skin lesions of Tungiasis that included the chitinous exoskeleton, ova and leg structures.
Tungiasis is caused by the sand fly *Tunga Penetrans* that causes an inflammatory skin disease.[7] The sand fly is commonly found in the tropical parts of Middle East , Africa, India, Caribbean, Central and South America. The flea is small and measures about 1 mm in size[8] and the structure and size of parts identified on pathological examination corresponded to reported microscopic features.

The gravid female of *Tunga penetrans* penetrates the skin of the host. In our patient the sand flea directly likely entered the dermis, easily reaching the graft. The fleas defecate and expels eggs and stays in contact with the air via its abdominal cone, leaving an opening which can be a portal of entry for microbes and other parasites. The gravid female begins laying eggs within 8 to 1[illegible] days and can lay thousands of eggs.[6] It is therefore not surprising that this flea that has the predilection to affect the skin wa[illegible] found in the dermis fat graft of this patients causing severe inflammation and necrosis. We suggest 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, is a more common condition than fleas[9] and should be in the differential diagnosis, easily recognized by the typical granuloma with central ulcer containing white larvae.[10,11]

To our knowledge, Tungiasis has not previously been described in an anophthalmic socket or related to a DFG. Previous reports have published infestations of the conjunctiva, lingual tonsil, thigh, elbow, gluteal, ischial protuberances and knees.[12]

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 odor and we performed a second procedure to excise the tissue surrounding the removed DFG ensuring all the orbital tissues were free of fleas. Complete local recovery usually occurs after removal of the intact flea.

In summary, Tungiasis should be considered in the differential diagnosis of a necrotic and chronic 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.

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