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LEE, KUMAR AND OTHERS

CYSTICERCOSIS WITH AN ORBITAL TROPISM IN TWINS

Case Report: Cysticercosis with an Orbital Tropism in Twins

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Abstract.

Two fraternal twin sisters developed cysticercosis localizing to the right lateral orbit over the same period after a presumed common-source exposure in China. This case demonstrates that cysticercosis can be related to travel. Similar temporal and spatial occurrences of these infections suggest a genetic tropism of the infecting organism in these twins.

BACKGROUND

Cysticercosis is a parasitic infection that is caused by ingesting eggs of the pork cestode *Taenia solium* via fecal-oral transmission. Humans acquire the infection by consuming food contaminated with oncospheres from the parasite or via autoinfection. The most common form of systemic involvement is neurocysticercosis and may manifest with characteristic brain lesions leading to seizure. Ocular and ocular adnexal involvement may occur in the context of isolated disease or systemic dissemination and can present with mass effect with or without a change in vision.^{1,2}

Cysticerci can lodge within the orbit and ocular adnexa or intraocularly. Intravitreal, subretinal, and subconjunctival cysticercosis are most frequently reported.^{2,3} The extraocular muscles, particularly the medial rectus, are the most commonly involved tissue in the orbit. The next most commonly involved sites are the subconjunctival space, eyelid, optic nerve, retro-orbital space, and the lacrimal gland. However, most subconjunctival cysts are found to be adhering to the insertion site of an extraocular muscle, so these cases may be more accurately categorized as a type of myocysticercosis.^{1,2}

Although cysticercosis is endemic to rural areas of Latin America, Asia, and Africa, it can also occur in developed countries presumably as a result of food handlers and individuals migrating from endemic rural areas. In this way, individuals who do not eat pork or come into contact with pigs may also acquire cysticercosis. Here, we present the first known cases of twins

living in different states who, by history, apparently were infected during the same exposure to *T. solium* in China. Both developed orbital cysticercosis in the same anatomic location during the same timeframe.

CASE REPORT

Two fraternal twin sisters aged 55 years, who do not eat pork for religious reasons, toured China 2 years before presentation. They live widely apart, with Twin A living in San Diego, California, and Twin B living in Minnetonka, Minnesota. They were together for the last time during a 9-day trip to China in July 2012, during which they consumed all meals together in the same restaurants. They both denied other travel within the last few years.

Twin A first noticed a nodule overlying her right lateral orbital rim in August 2013. Over the course of 9 months, the lesion progressively enlarged, prompting her to seek ophthalmological consultation in May 2014. She denied any history of pain, itching, inflammation, or change in vision. She had no history of trauma in the region or animal contact. Review of systems, medical history, and surgical history were noncontributory. A computed tomography (CT) scan of the orbits was performed showing a $4 \times 6 \times 5$ mm clear, cystic lesion (Figure 1A) at the right lateral orbital rim interpreted by the radiologist as a lipoma. A lateral orbitotomy was performed and revealed an encapsulated cyst that was adherent to the periosteum and filled with milky fluid. Pathologic examination revealed a three-layered acellular cystic structure with an eosinophilic cuticular layer, a cellular layer with dark-staining nuclei, and a loose reticular layer suspicious of a *T. solium* cyst (Figure 1B). No definite scolex was identified, and review of the histology by the Center for Disease Control Parasites Division confirmed a cestode bladder. The final diagnosis was cysticercosis or coenurosis, the latter being possible due to the lack of a visualized scoleces, but considered unlikely given the clinical scenario. Testing for *T. solium* antibodies was negative, CT of the brain was negative, and at the patient's request, she was given five doses of albendazole without recurrence or complication.

Twin B presented for care in August 2014 with two 8–10 mm lesions again overlying the right lateral orbital rim. The lesions had been present for at least 6 months before May 2014, but the exact onset of the lesions was unclear since she was not concerned about the lesions until learning of her twin sister's diagnosis. Twin B also denied any history of pain, itching, inflammation, or change in vision. Magnetic resonance imaging of the brain and orbits was performed and showed the presence of two masses near the right lateral orbital rim (Figure 1C) but no intracranial lesions. One cyst measured 8×5 mm and was a tan cystic lesion filled with a creamy white-tan material. The other measured 7×8 mm and was described as a semi-firm pink-tan mass (Figure 1D). Histological sections revealed thin-walled cysts associated with chronic inflammation with a foreign body giant cell reaction. The cyst wall had structures suggestive of degenerating parasite cuticle. However, no larval parts were visualized, and at the patient's request, she was treated with a 1-week course of albendazole with no recurrence or complication.

DISCUSSION

We present a case of geographically separated twins who contracted cysticercosis while traveling in China. Both twins presented with gradually enlarging cysts overlying the right lateral orbital rim within 1–2 years of presumed exposure. Taken in isolation, these individual infections would not be unprecedented, as there have been previous reports of travel-related cysticercosis in tourists from non-endemic countries,⁴ as well as cysticercosis occurring in

individuals who do not eat pork for religious reasons.⁵ Most reports of travel-related cysticercosis have been of neurocysticercosis, and a recent review of neurocysticercosis among international travelers found that infections were most common among tourists visiting the Indian subcontinent, Latin America, and southeast Asia, but were rarely acquired during travel under 3 months duration.⁴ The nearly simultaneous occurrence of orbital cysticercosis in twin sisters deserves further consideration of the pathogenesis of this infection.

Once *T. solium* oncospheres penetrate the small intestine mucosa,⁶ the parasite spreads hematogenously and may end up in brain, eye, muscle, or skin. From an ocular standpoint, cysticercosis is known to present throughout the ocular and orbital system, including the vitreous, subretinal space, orbital region with a proclivity for the extraocular muscles, subconjunctival space, and even the optic nerve. In the case of these twins, the lateral canthal region has extensive arterial anastomoses between the lacrimal artery, the orbital branch of the superficial temporal artery, and the lateral palpebral arteries. As such, the blood supply of the lateral canthal region stems from the internal and external carotid circulations. However, tissue tropism in cysticercosis likely goes beyond hematogenous spread based on vascularization patterns and may involve molecular and genetic factors.

Infectious oncospheres contain myriad surface proteins, the best characterized being TSOL18,^{7,8} but which proteins might be involved in homing remains unknown. These cases of twins with periocular cysticercosis are particularly notable for localization to the lateral orbit (and by chance on the right side). Given their highly similar genotypes, the twins almost certainly share a genetic predisposition that may have resulted in identical clinical presentations. Future twin-pair studies may provide new insights into the mechanisms underlying clinical presentations of cysticercosis.

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FIGURE 1. Far left column, top and bottom: External photograph of cysticercosis lesions at the right lateral orbital rim in Twin A (top) and Twin B (bottom). Second from the left column, top and bottom: axial computed tomography (CT) scan of Twin A and T1 weighted magnetic resonance imaging (MRI T1) scan of Twin B showing radiolucent cystic lesions at the lateral orbital rim. Top row, second from right and far right: hematoxylin and eosin stains of surgical specimen from Twin A shows a cystic structure with mild chronic inflammation that on higher magnification reveals an epithelial-lined cyst with numerous calcifications. Bottom row, second from right: intraoperative photo of surgical specimen in Twin B. Bottom row, far right: hematoxylin and eosin stain of surgical specimen from Twin B shows a cystic structure associated with foreign body giant cell granulomas against potential degenerating hooklets. This figure appears in color at www.ajtmh.org.

Figure 1

