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Pseudo-Romaña Sign Due to Botfly Infestation

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Abstract

We report a patient who presented with unilateral periorbital edema after a short-term travel to an endemic area for Chagas disease and exposure to triatomine (reduviid) bugs. The diagnosis, however, was furuncular myiasis, not Chagas disease. This case emphasizes that there are multiple parasitic agents besides *Trypanosoma cruzi* that may be associated with clinical findings consistent with Romaña sign.

Keywords

Romaña sign; Chagas disease; *Dermatobia hominis* ; botfly

Romaña sign is a painless nonpitting edema of the palpebrae and periocular tissues that develops approximately 10 days after contamination of the conjunctivae with triatomine feces that contain trypomastigotes of *Trypanosoma cruzi*.[1,2] The periophthalmic edema is unilateral and typically associated with conjunctivitis and regional lymphadenopathy of the preauricular or submaxillary nodes, and often appears coincident with the onset of fever and fatigue. Romaña sign is an uncommon manifestation of acute Chagas disease, but its presence is highly suggestive of acute infection when it occurs in the appropriate epidemiologic setting.

Myiasis is the invasion of living vertebrate tissue by fly larvae.[3] Although cutaneous involvement is the most frequent manifestation, myiasis may also involve ocular, aural, nasal, oral, gastrointestinal, and genitourinary sites.[3] *Dermatobia hominis* is the species of fly most often associated with cutaneous myiasis in humans; this species may also cause external ophthalmomyiasis with eyelid or conjunctival involvement and preauricular

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lymphadenopathy.[4] We describe a case of furuncular myiasis due to *D hominis* that presented as unilateral periorbital edema mimicking Romaña sign.

CASE REPORT

A 28-year-old previously healthy surgical resident vacationed in Belize and Guatemala from March 23 to March 30, 2009. Seven days after returning on April 6, she developed pain on the left side of her face and left ear with painful pre-auricular and cervical lymphadenopathy on the same side. On April 8, she noted a palpable nodule on the left side of her scalp proximal to the hairline. An otolaryngologist prescribed amoxicillin-clavulanate for possible suppurative parotitis. A day after starting the antibiotic, she noted nontender facial edema involving her forehead and left periorbital region, including the eyelid, causing difficulty in opening the left eye (Fig. 1). She did not have fever. Because of swelling of the left periorbital area, she was evaluated at an emergency department on April 11 and received a dose of intravenous (IV) clindamycin. A computed tomographic scan of the neck with contrast revealed left periorbital soft tissue swelling extending into the left facial region with enhancing lymph nodes or nodules along the left parotid, periparotid, and cervical regions (Figs. 2A, B).

The patient's recent travel history was colorful. On March 23, she visited the Belize Zoo where her forehead was licked by a jaguar. She also went ziplining over jungle canopy in Tikal Ruins of Guatemala on March 24, waded through an underground river, and participated in rock climbing within a cave in Caves Branch, Belize on March 25. From March 23 to 25, she slept in thatch-roofed mud-walled huts in a jungle lodge in San Ignacio and in Caves Branch, both in Belize (Fig. 3), where she was exposed to triatomine bugs (order Hemiptera, family Reduviidae, subfamily Triatominae). She went hiking on March 26, scuba diving on March 28, and snorkeling in the ocean on March 29, also in Belize.

On physical examination, she was afebrile. Pertinent findings were swelling of the left periorbital area and the left side of her face with mild warmth and erythema. She had tender left submandibular and left cervical lymph nodes. Results of complete blood count and comprehensive metabolic panel were normal. Routine blood cultures were negative, and *T cruzi* was not observed on examination of a buffy coat preparation.

She completed a 1-week course of amoxicillin-clavulanate with resolution of symptoms. Serial testing for evidence of *T cruzi* infection was negative (Table 1). Less than 1 month later, on May 8, the patient noted swelling, sharp pain, fluctuance, and increase in the size of the scalp nodule. She did her own incision and drainage with local anesthesia using lidocaine, and a whitish substance was extracted. The specimen, placed in 70% ethanol, was identified as an early third-instar larva of the human botfly, *D hominis* (Linnaeus Jr; order Diptera, family Oestridae, subfamily Cuterebrinae; Fig. 4). The patient has remained asymptomatic for 28 months since the incision and drainage was done. The botfly larvawas deposited as avoucher specimen in the Cornell University Insect Collection, Ithaca, NY, under Lot Number 1267.

The criteria for determining that the larva was an early-stage third-instar were as follows. The specimen's cuticular spines possessed features that are diagnostic for a third-instar larva: color, chestnut reddish-brown; bases, smoothly oval; and size, relatively small. In comparison, and counterintuitive to expectations for a less mature larva, the cuticular spines of a second-instar larva are heavier and more robust in form, black, and possess irregularly angulate bases. Three cuticular fragments possessing the heavy black spines diagnostic for a second-instar larva were loose in the specimen container, further indicating that the larva had recently molted to the third instar. The body length of the uncontracted specimen was approximately 10 mm, indicating that it was relatively immature compared to the body length of approximately 18 mm for a relatively mature and uncontracted third-instar larva in the arthropod reference collection of the New York State Department of Health Western Regional Office.

DISCUSSION

The initial presentation of the patient was consistent with the description of Romaña sign, and the patient had some risk of exposure to *T cruzi*, as the parasite is endemic to both Guatemala and Belize. Two important clinical findings merit emphasis. The patient presented with facial pain, but notably the periorbital edema was nontender. Although the classic description of Romaña sign is that of painless edema, pain has been reported in cases of confirmed acute Chagas,[5] so nontender edema may not be a key finding. More importantly, the patient did not have a fever or other systemic symptoms, which was a clue that alternative diagnoses should be considered. Additionally, transmission of *T cruzi* to short-term visitors to endemic areas is thought to be rare; the first reported case in a US traveler was recently published.[5] Data on transmission suggest that long, intense exposure to the vector are needed for transmission to occur.[6]

There are numerous parasitic infections or infestations besides Chagas disease that may cause unilateral periorbital swelling in a returning traveler (Table 2).[1–4,6–3] *D hominis*, also known as human botfly, American warble fly, or tórsalo in Central America, is endemic to the region extending from southern Mexico to northern Chile and Argentina.[18] The female botfly captures a zoophilic or anthropophilic arthropod (usually flies) and glues her eggs in clusters to its abdomen.[16,17,19–23] The egg carrier or "porter" subsequently conveys the embryonated eggs to a vertebrate host, where they hatch while the arthropod feeds.[22,23] Porters include day-flying mosquitoes (particularly *Psorophora* species; order Diptera, family Culicidae) and muscid flies (eg, *Sarcopromusca, Stomoxys*, and *Synthesiomyia* species; order Diptera, family Muscidae).[22,23] The enclosed larva abandons the egg on the porter and penetrates the skin of the host via the bite puncture (when the porter is a hematophagous fly), a hair follicle, a skin fold, or areas of moist skin in contact with clothing or bedding.[22,23] Development occurs at the point of entry in a boil-like pocket called a furuncular lesion where the botfly larva causes local inflammation.[4,18,22,23] Development through 3 (possibly 4) larval instars usually takes 5 to 10 weeks after which the mature larva enlarges the opening in the skin and drops to the ground to pupate.[22,23]

Furuncular myiasis is the condition in which furuncular lesions develop as a result of skin invasion by parasitic fly larvae.[19] Diagnosis of furuncular myiasis may be made by

knowledge of the exposure and travel history in a patient with one or more nonhealing furuncles on exposed skin with evidence of serous, serosanguineous, or seropurulent drainage from a central pore, with local symptoms of inflammation; perception of movement inside the lesion may be a helpful diagnostic clue. On examination, a small, white, wormlike organism may be seen protruding from the lesion when pressed laterally.[20]

Treatment of furuncular myiasis is mainly directed at causing paralysis or asphyxiation of the larva, facilitating removal. In the tropics, remedies include application of paraffin oil, petroleum jelly, occlusive ointment, beeswax, pork fat, or bacon, which interrupts the larva's respiration, thus forcing the larva to migrate to the surface of the lesion where it can be removed easily. If the larva is still in the early stage of development or small, then mechanical pressure on either side of the furuncle may be all that is required for removal. Injection of lidocaine will cause paralysis of the larva, making removal much easier.[3] Another option is surgical removal by doing a small incision to open the cavity with local anesthetic; the disadvantage of this approach is that the larvae can break, inciting an inflammatory reaction and incomplete extraction of the larva.[17] Systemic ivermectin may be used when there are multiple lesions, or when local occlusion treatment cannot be done easily, such as in the case of ophthalmomyiasis.[21]

This case emphasizes that causes other than *T cruzi* infection may be associated with clinical findings consistent with Romaña sign even in a patient with triatomine bug exposure.

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Consent has been obtained from the patient for the use of the image in Figure 1.

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FIGURE 1.
Left facial edema involving forehead and periorbital area, mimicking Romaña sign.

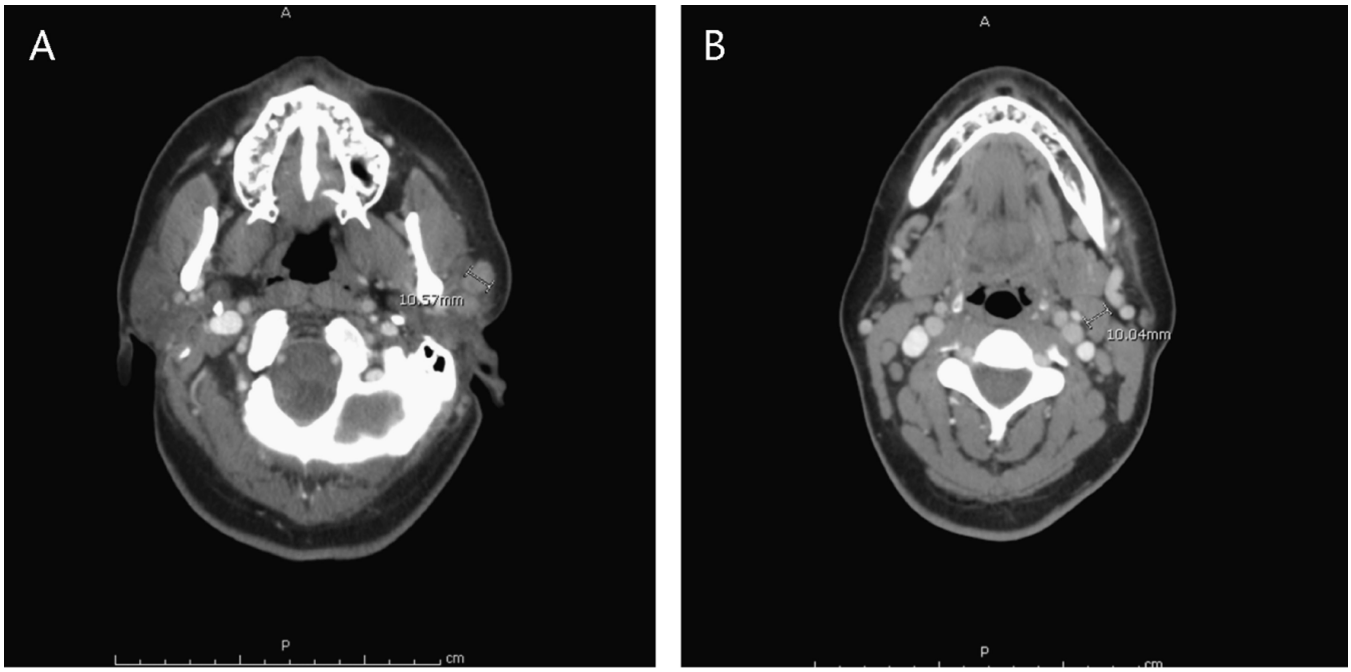

FIGURE 2.

A, B, Computed tomographic image of neck showing reactive lymph nodes along left side of the face, and parotid and cervical chains.

FIGURE 3.
Thatch-roofed, mud-walled huts in Belize.

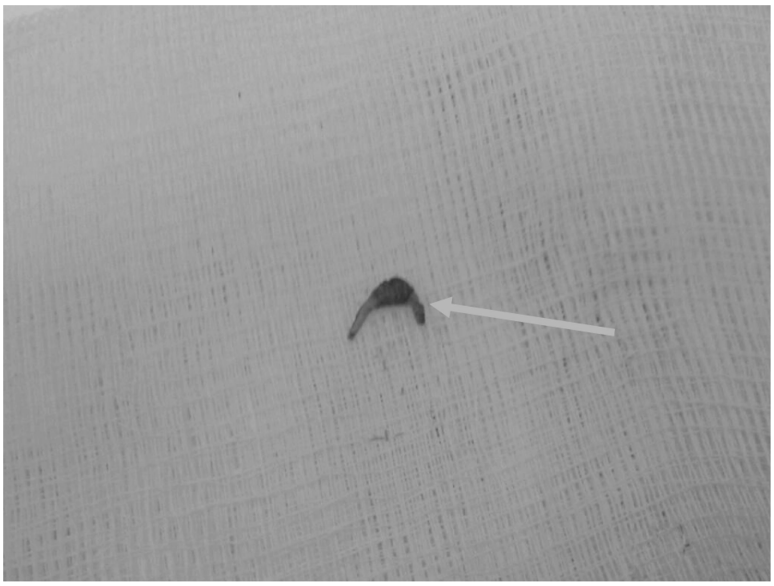

FIGURE 4.
Early third-instar larva of the human botfly, *Dermatobia hominis*, which was removed from the patient.

TABLE 1.

Evaluation for *T cruzi* Infection in our Patient (All Tests Done at the Centers for Disease Control and Prevention)

	April 27, 2009	June 10, 2009	Sep 16, 2009	Sep 21, 2009
T cruzi IFA [*]	Negative	Negative		Negative
T cruzi EIA [†]	Negative	Negative		Negative
T cruzi PCR [‡] on blood	Negative	Negative	Negative	Negative
T cruzi culture of blood	Negative	Negative		
T cruzi wet mount of blood	Negative	Negative		Negative

[*] CDC in-house IFA.

[†] Wiener EIA or Chaga test ELISA recombinant version 3.0.

[‡] CDC in house PCR.

EIA indicates enzyme immunoassay; IFA, immunofluorescent antibody; PCR, polymerase chain reaction.

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TABLE 2.

Parasitic Infections and Infestations That may be Associated With Unilateral Periorbital Edema Consistent With Romaña's Sign in a Returning Traveler

Parasitic Infection	Endemic Region	Localized Manifestations	Other Manifestations
American trypanosomiasis/ Chagas disease	South America, Central America, Southern half of United States	Painless, unilateral nonpitting periophthalmic edema, possibly associated conjunctivitis and regional lymphadenopathy[7]	Fever commonly accompanies local symptoms; acute myocarditis or meningoencephalitis can rarely occur[8]
Loiasis/African eye worm	West and Central Africa	Localized, usually nonpitting, nontender edema (Calabar swelling); adult worm may visibly migrate under the conjunctiva with accompanying pain, conjunctivitis, and photophobia[7,9]	Calabar swelling usually occurs on limbs, near joints; may be associated with local or systemic pruritus[9]
Dirofilariasis/Dog heartworm	Worldwide	Pruritic painful edema[7]; can cause orbital infections and result in proptosis, ptosis, or diplopia[10]	Fever and lymphadenopathy occasionally seen; can penetrate tissue and migrate to internal organs[10]
Trichinellosis	Worldwide	Eyelid and/or facial edema, usually becomes bilateral and symmetric, may develop painful eye movements; subconjunctival hemorrhage can occur; vascular edema and/or vascular lesions of the conjunctiva, uvea, and retina can occur[11]	DiffuDiffuse myalgias common, fever common, may develop urticaria or maculopapular rash; may develop myocarditis, thromboembolic disease, and encephalitis[11]
Cysticercosis	Mexico, Central and South America, Africa, Southeast Asia (India, Philippines), Southern Europe	PainfPainful, periorbital edema, although a painless firm nodule may occur; invasion of the eye or surrounding structures can result in diplopia, blurred vision, or ptosis[7]	PrecePreceding gastrointestinal symptoms may occur[7]; cysts can be located in muscle, causing pain or edema or in the central nervous system, where the cysts may be asymptomatic, cause seizures, or other neurologic sequelae
Cutaneous leishmaniasis	Old World *Leishmania* species: Middle East, North and East Africa, Southeast and Central Asia, Europe New World *Leishmania* species: South America, Central America, Mexico	Localized edema followed by inflammatory and/or necrotic lesion with ulceration[7,12]; eyelid lesions, chronic blepharoconjunctivitis, scleromalacia, interstitial keratitis, and iridocyclitis; ptosis may occur[13]	LympLymphatic spread and lymph-gland involvement may precede lesion development[12]
Dracunculiasis/Guinea worm disease	Sudan, Ghana, Mali, Ethiopia, Chad	Localized area of painful edema, with eventual appearance of blister and accompanying cellulitis[14]	Worm emerges from lower extremities in 80%–90% of cases; fever and urticarial rash precedes the blister; GI symptoms may occur[14]
Gnathostomiasis	Japan, SE Asia particularly Thailand, Central and South America, Mexico	Migratory, nonpitting edema; there may be associated pain, pruritus, erythema, or subcutaneous hemorrhagic tracks; invasion of the eye can result in intraocular hemorrhage, glaucoma, uveitis, iritis, retinal scarring, and detachment[7,15]	Facial involvement increases the risk of CNS invasion, which results in pain along the nerve the parasite tracks along with subsequent paralysis of the extremities and/or cranial nerve palsies, and eosinophilic meningitis; preceding GI symptoms may occur[15]
External ophthalmomyiasis	Botflies: Tropical Latin America (Southern Mexico to northern Chile and Argentina), Trinidad Blow flies: Tropical Africa, Tropical Latin America, Tropical Asia, Worldwide	Bitin Biting and movements of larvae and chemical irritation from its feces creates an allergic reaction that may affect eyelids, nasolacrimal ducts, lacrimal sac and conjunctiva, causing acute redness, irritation, discharge[7,16] and preauricular lymphadenopathy on the same side as the ocular lesion[4]	Larvae can parasitize skin, vagina, urinary tract, gastrointestinal tract, nasopharynx, sinuses, and auditory canals[17]

Parasitic Infection	Endemic Region	Localized Manifestations	Other Manifestations
	Flesh flies: Mediterranean basin, Eastern Europe		