CASE REPORTS

Tracheopulmonary Myiasis Caused by a Mature Third-Instar Cuterebra Larva: Case Report and Review

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Myiasis is the infestation of vertebrate tissues with fly larvae (Diptera). Most human cases in North America are subcutaneous forms due to Dermatobia hominis imported from Central and South America. Human cases of myiasis acquired in North America are rare and are primarily subdermal or ophthalmologic forms of infestation caused by early stages of Cuterebra larvae. We report an unusual case of tracheopulmonary myiasis, resulting from the in situ development of a mature cuterebrine larva associated with high eosinophilia. Only two other cases of tracheopulmonary cuterebrid myiasis have been reported in humans, and they are reviewed herein. Cuterebra myiasis (cuterebrosis) remains a rare and aberrant cause of tracheopulmonary disease and is a newly described cause of eosinophilia in humans.

CASE REPORT

In early October 2002, a 60-year-old woman, a resident of Florida, presented in the emergency ward of Hôtel-Dieu Hospital in Paris, France. She had arrived in Europe for a vacation a week earlier, and there was no history of travel outside Florida in the previous 3 months. She reported a 3-week history of whooping cough with occasional bloody sputum, without fever. A few minutes before arriving in the emergency ward she coughed up bloody sputum which contained a large, brownish fly maggot. The larva was alive and mobile and measured 17 by 5 mm (Fig. 1). Chest auscultation and chest X-ray performed shortly after spontaneous expulsion of the maggot were normal. Additional larvae were not found after complete examination of the patient. A hemogram showed high eosinophilia (eosinophil count, 2.03 \( \times \) 109/liter) and a white cell count of 10.4 \( \times \) 109/liter. The level of C-reactive protein was 7 mg/liter. Detection of precipitating antibodies in the serum by a homemade method with Hypoderma lineatum and Gasterophilus antigens was negative. The day after maggot expulsion, the patient was referred to a chest specialist. Hemoptysis and cough had completely disappeared, and chest auscultation was clear. No further investigation or treatment was performed, and she reported no further complication. The larva was identified as a mature third instar (prepupal stage) in the genus Cuterebra based on size, characteristic cuticular plates and associated spines, and the posterior spiracular apparatus (Fig. 2 and 3). A more detailed examination of the external morphology revealed that the specimen was a member of the genus Cuterebra normally infesting lagomorphs (2). Because Cuterebra is restricted to the New World, and given the patient’s recent travel history, we are reasonably confident that the patient was infested in Florida.

Discussion. Myiasis is the condition caused by the invasion of human or animal tissues by fly larvae (called maggots) and can be classified as accidental, facultative (opportunistic), or obligate. Human cases are considered accidental and most often result in subcutaneous infestations (furuncular myiasis) caused by the tropical species Dermatobia hominis, the human botfly, native to Central and South America, and Cordylobia anthropophaga, the tumbu fly, in tropical sub-Saharan Africa (4, 20). As a result, human myiasis is rarely acquired outside tropical and subtropical areas, and very few cases in patients who had not recently (within several months) traveled to myiasis-prone regions have been described. Dipteran fly larvae in the genus Cuterebra Clark 1815 (Oestridae; Cuterebrinae), the rodent or rabbit botfly or larva fly, are obligate parasites on rodents and lagomorphs, rarely infesting humans. Most human cases of myiasis acquired in North America are caused by the genus Cuterebra, with cuterebrosis being primarily a subdermal or ophthalmologic form of infestation (4, 23). Nasal, oropharyngeal, or orotracheal myiasis cases have been occasionally reported but tracheopulmonary or intratracheal myiasis is a very unusual and aberrant form of the disease in humans (5, 7, 15, 25). To date, only four cases of tracheopulmonary myiasis have been reported in humans: two cases of pulmonary infections caused by Gasterophilus, the horse botfly (1), and two cases of tracheopulmonary cuterebrine myiasis (3, 24). No Cuterebra larvae in human hosts have been known (or allowed) to

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develop to maturity (6); however, the chance of human infestations with possible severe medical complications determines the medical importance of this genus.

Human cases of myiasis caused by *Cuterebra* are rare (4, 19). In a published review, Baird et al. compiled 55 cases (4). Most of the cases were either subcutaneous abscesses or ophthalmomyiasis (4, 8, 9, 11, 16, 21). All patients with cuterebrid myiasis were infected with only a single larva. To our knowledge this report is the third described human case of tracheopulmonary myiasis due to a *Cuterebra* sp. and represents the first case showing apparent complete development of the maggot in a human. All three cases occurred in North America. The first was in northern Ontario, Canada, in September 1977. The larva was coughed up and identified as *Cuterebra fontinella* Clark, a rodent botfly. It is unclear whether the larva was a second or an early third instar (4, 24). The second case occurred in Oregon in August 1979, and the larva subsequently coughed up was an early second instar (3). In the case reported here, the larva was likely from Florida.

Only one genus of botfly, *Cuterebra*, is currently recognized as occurring in North America, of which at least 34 species are known to occur in the United States and Canada (22). The genus is also found in Mexico and Neotropical regions (17). Although more than 70 species of *Cuterebra* (6) have been described (13, 14, 22), much remains unknown about preimaginal stages and fly biology. The taxonomy remains poorly defined, and existing keys are considered inadequate for definitive separation of many of the species in the immature stages. Often, rearing the developed larvae to adults greatly augments species determination. Most recorded cases of human cuterebrane infestation, however, have been attributed to rabbit bots (6). Sabrosky subdivided the North American (Neartic) *Cuterebra* into four subgroups, two of which, the cuniculi group (two species) and the buccata group (seven species), naturally parasitize lagomorphs (22). Given the scarcity of the taxonomy keys for immature stages, the specimen identified in this case report can be identified as a rabbit bot but cannot be identified to species level.

Some knowledge of the cuterebrid life cycle helps us to understand how humans may become accidental hosts (6). The short-lived female botfly lays her eggs during the warmer spring months on vegetation and other objects, often near the ground burrows of potential rodent or rabbit hosts. Upon coming into contact with a host, eggs hatch and the infective first-instar larvae enter through the mucous membranes of the nose, eyes, mouth, or anus or may directly penetrate the skin. In natural host, the larvae commonly migrate through the trachea, pleural cavity, diaphragm, and abdominal cavity, after which they embed themselves into the subdermal layers and develop into second and third instars. After a period of time (often weeks, sometimes several months), the mature third instar (prepupal form) leaves the host to pupate in loose soil. The normal duration of the larval development cycle in most natural hosts is 3 to 5 weeks (4, 23). First-instar larvae can infest humans after contact with contaminated vegetation, infested wild rodents and rabbits, and pets that have been in the immediate environment harboring natural hosts. Because first-
instar larvae can invade the trachea of the natural host, it is not unreasonable to suspect that accidental tracheopulmonary infestation could result in humans. In such cases, the portals of entry are nasal and oral openings. As with the patient, the two previously described patients presented only mild respiratory symptoms, and their condition immediately improved after the larva was coughed up. This feature and the absence of skin or other organ involvement suggest that the larva remained within the respiratory tract (likely the trachea) during development. The spontaneous expulsion of the mature third-instar larva by our patient in early October was also consistent with the seasonal distribution of cuterebrid myiasis since nearly all cases yielding third-instar larvae occurred in late August to October in North America (4). The duration of the cycle in humans is unclear, but in our patient we suspect it was longer than 3 weeks. Most reported North American cuterebrid infestations have occurred in rural or semirural temperate areas of the northeast. Given her travel history, our patient is presumed to have been infested in a rural or semirural environment in central Florida (near Orlando), a distinctly humid and subtropical environment most times of the year. In their review, Baird et al. did not report any human cases from State of Florida, making this report a new distribution record for human infestation (4).

Eosinophilia has been associated with myiasis caused by Hypoderma species. Histopathologic examination of Cuterebra furuncular myiasis has shown eosinophil tissue infiltration around the infestation site (4). Reports of eosinophilia in cats with invasive cuterebrine myiasis are rare (12). This appears to be the first report of cuterebrine myiasis in humans associated with high eosinophilia (10, 18, 20).

This report is remarkable in three ways: (i) this is the first reported case of human tracheopulmonary cuterebrid myiasis with complete maturation of the larva in the respiratory tract, (ii) this is also the first reported case of human cuterebrid myiasis associated with significant eosinophilia, and (iii) the occurrence of cuterebridosis in a subtropical location in North America is unusual. Cuterebrid myiasis is a rare cause of tracheopulmonary disease and is a newly described cause of eosinophilia in humans.

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