

Human Coenurosis in North America: Case Reports and Review

Michael B. Ing, Peter M. Schantz, and Jerrold A. Turner

From the Infectious Diseases Section, Medical Service, Department of Veterans Affairs Medical Center, and the Loma Linda University School of Medicine, Loma Linda, California; Epidemiology Branch, Division of Parasitic Diseases, Center for Infectious Diseases, Centers for Disease Control and Prevention, Atlanta, Georgia; and Division of Infectious Diseases, Department of Medicine, Harbor-UCLA Medical Center, Torrance, California

Coenurosis is a zoonotic disease of humans caused by the larval stage of *Taenia (Multiceps)* species. In North America, the adult tapeworm of *Taenia (Multiceps) serialis* is found in canids. The cystic larval forms (coenuri) are found in hares, rabbits, squirrels, and, rarely, in humans. We review in clinical detail the fifth case reported from North America, involving a child with extensive central nervous system involvement. We also report a sixth case, involving an adult with an intramuscular coenurus. The latter case was diagnosed by needle aspiration of the cyst. Although praziquantel administration may have been effective in killing the parasite in both patients, we are concerned about the production of marked inflammation as a result of treatment. The four other North American cases are reviewed, and the epidemiology of the infection in animals is discussed.

Human coenurosis is a zoonotic disease caused by the larval stages of *Taenia (Multiceps)* species. The definitive hosts are canids such as dogs, coyotes, and foxes, and intermediate hosts include hares, rabbits, and rodents. Humans may be infected as incidental intermediate hosts, with the cystic larvae usually developing in the CNS, eye, or subcutaneous or intramuscular tissues. Five cases of human coenurosis acquired in North America have previously been reported [1–5]. This review of human coenurosis in North America includes more clinical details regarding the fifth case [5] and a report of a sixth case.

Case Reports

Case 1. A 3½-year-old Native American girl was admitted to a hospital in Duluth, Minnesota, on 15 February 1983 with a 6-month history of progressive generalized muscle weakness, upper-extremity contractions, and inability to walk. She had been hospitalized 4 months earlier for delayed development, dehydration, vomiting, and a macular rash over her abdomen and chest. Her neurological status deteriorated. She was subsequently evaluated at another hospital in Duluth, where a head CT revealed massive hydrocephalus. The patient was treated with dexamethasone and transferred the following day back to the original hospital.

On admission her temperature was 38°C. Other vital signs were normal. Examination showed spastic quadriparesis and papilledema. Laboratory studies revealed a WBC count of 10,200/ μ L with a normal differential. The hemoglobin level was 10.7 g/dL, with a mean corpuscular volume (MCV) of 53 fL. Serum chemistry values and chest radiographic findings were normal. On the following day a right ventriculoperitoneal shunt was placed. Five days after shunt placement, the patient became febrile (temperature to 39.5°C). A shunt infection was suspected. Despite therapy with chloramphenicol and moxalactam, the patient remained febrile. The shunt was revised to drain externally. One week later, CSF studies showed the following values: WBCs, 360/ μ L, with 60% polymorphonuclear forms; RBCs, 2,220/ μ L; glucose, 28 mg/dL; and protein, 216 mg/dL. A left ventriculoperitoneal shunt was placed, and multiple carious teeth were removed.

On 18 March 1983, a CT scan of the head with intrathecal metrizamide showed multiple “bizarre lobulated serpiginous filling defects” extending from the posterior fossa, through the foramen magnum, and into the upper cervical subarachnoid space. A contrast myelogram with metrizamide also confirmed the presence of a mass with a “frond-like, lobular, cluster-of-grapes appearance” in this area. Differential diagnosis included ependymoma and arteriovenous malformation. Three days later the patient had a suboccipital craniotomy with subtotal removal of numerous “gelatinous, semicystic, multiloculated, mucinous masses.” These masses enveloped the brain stem and extended into the fourth ventricle. Histopathologic examination identified these structures as coenuri, an identification later confirmed by the Centers for Disease Control and Prevention (CDC).

On 1 April 1983, chemotherapy with praziquantel at a dosage of 150 mg/d (16.6 mg/kg per dose) was initiated. Three days later, an L3-L5 lumbar laminectomy was performed and multiple necrotic, degenerating, nonviable cysts were removed. The patient completed a 2-week course of praziquantel, resulting in

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Reprints or correspondence: Dr. Michael B. Ing, Medical Service, Infectious Diseases Section (111M), Jerry L. Pettis Memorial Veterans Affairs Medical Center, 11201 Benton Street, Loma Linda, California 92357.

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slight improvement of her neurological status. However, she continued to suffer from persistent hydrocephalus and shunt malfunction. Despite multiple shunt revisions and another 2-week course of praziquantel, her condition deteriorated, and she died on 18 August 1983. An autopsy request was not granted.

Case 2. A 39-year-old Hispanic man from Los Angeles with a long history of ethanol abuse and with liver cirrhosis and recently diagnosed hepatitis C infection was seen at a hospital in Downey, California, on 26 July 1994 for an enlarging mass on his left upper back. An excisional biopsy under local anesthetic agents was attempted; however, when a large intramuscular fibrous capsule was found, the procedure was terminated until a CT scan could be obtained. The patient was lost to follow-up and subsequently was admitted to a hospital in Monterey Park, California, with complaints of fever, chest, and back pain; dyspnea; nausea; vomiting; and the mass on the upper left side of his back, which had been progressively enlarging for the past 2 months. Fine-needle aspiration of the cyst showed scolices and hooklets consistent with a helminthic organism. A single dose of 900 mg of praziquantel was administered, and the patient was transferred to the Harbor-UCLA Medical Center (Torrance, CA) on 22 August 1994.

On admission, his temperature was 38.6°C and other vital signs were normal. Examination revealed a distended abdomen with a positive fluid wave and a 3-cm umbilical hernia. Neurological examination findings were normal. A cystic ovoid mass (8 ×

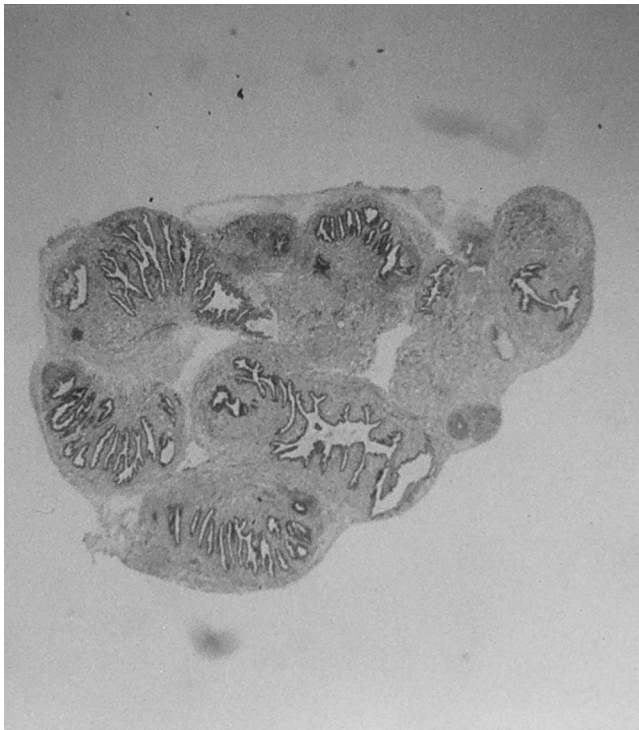


Figure 1. Photomicrograph of aspirate from the intramuscular cyst of case 2 (stain, hematoxylin-eosin; original magnification, $\times 40$). Multiple scolices are present.

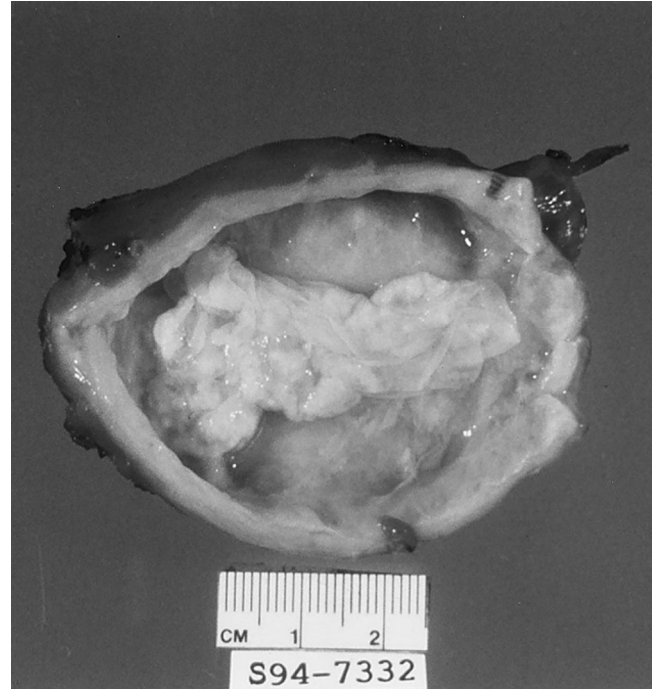


Figure 2. The gross surgical specimen from case 2, showing the cystic cavity within the muscle and the collapsed, membranous, parasitic cyst removed from the cavity.

13 cm) was noted on the left upper back. The mass was tender and warm, without erythema. Laboratory analysis revealed a WBC count of 4,900/ μL with 48% neutrophils, 25% lymphocytes, 23% monocytes, and 3% eosinophils. The hematocrit was 28.4%, with an MCV of 102.3 fL and a platelet count of 58,000/ μL . Other significant laboratory data included the following values: aspartate aminotransferase, 72 U/L; alanine aminotransferase, 37 U/L; lactate dehydrogenase, 236 U/L; total protein, 7.1 g/dL; and albumin, 2.5 g/dL. The creatine phosphokinase level determined 1 week before transfer was 645 U/L, with 2.2% MB isoenzymes. A CT scan of the head was negative. A chest CT revealed a 2 × 3 × 9-cm mass in the left paravertebral area, predominantly located within muscle.

A review of the fine-needle aspirate revealed a larval cestode cyst with multiple scolices most consistent with coenurosis (figure 1). At surgery, a 5 × 7-cm fibrous mass was removed. Incision of the mass revealed a 2 × 5-cm coenurus containing multiple scolices (figures 2 and 3). The patient did well and was soon discharged to home. He has had no recurrence of the mass after >1 year of outpatient evaluation.

Discussion

Human coenurosis was first reported by Brumpt in 1913 [6] and continues to remain a rare disease, with most cases being reported from Africa [1, 7]. Johnstone and Jones reported the first North American case in 1950 [1]. Since that time only

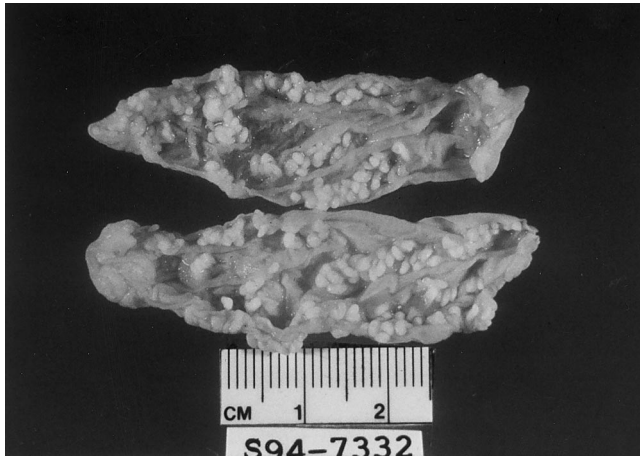


Figure 3. The parasitic cyst from case 2 has been divided into two parts, demonstrating the multiple nodular scolices on the inner surface of the cyst membrane.

four additional North American cases have been described in the literature [2–5].

Adult tapeworms of *Taenia (Multiceps)* species are found in the small intestines of canid definitive hosts. Gravid proglottids, passed in the feces, disintegrate and release eggs that are ingested by intermediate hosts. Herbivores such as sheep, cattle, goats, and horses are intermediate hosts for the species *Taenia (Multiceps) multiceps*. The larvae of *T. multiceps* develop in the CNS, causing a disease in sheep called gid or staggers. *Taenia multiceps* infections are still enzootic in Europe and Asia but have not been reported in the United States for the past 60 years [8].

It is generally accepted that the cases of coenurosis involving humans that are reported from North America are caused by *Taenia (Multiceps) serialis* that is enzootic in this region. The larval stages of *T. serialis* develop in rabbits, hares, and rodents. The cystic larvae are usually found in the subcutaneous and intramuscular tissues; however, experimental infections of rodents have produced CNS involvement [9].

Human coenurosis usually involves CNS, intramuscular, or subcutaneous tissues. The CSF pathways are often involved, causing arachnoiditis and ependymitis. Intraparenchymal brain and spinal cord lesions also may occur [10, 11]. Basilar leptomeningitis, leading to arteritis and transient hemiparesis, has also been reported [12]. The spectrum of CNS involvement resembles that of the more common infection, neurocysticercosis, caused by larvae of *Taenia solium*. Seizures, hydrocephalus, and meningitis may occur in both infections.

Intramuscular and subcutaneous cysts, which have been reported most frequently from Africa, often involve the intercostal regions and the anterior abdominal wall [7]. Soft-tissue infections in Africa are usually considered to be caused by the related species, *Taenia brauni*. Intraperitoneal disease, resulting in extrinsic biliary compression and obstructive jaundice, has also been described [13].

Although cause and effect could not be established, pectoral muscle involvement in a 38-year-old Canadian woman was associated with severe systemic symptoms that resembled lymphoma [4]. Lymph nodes and the spleen showed no evidence of lymphoma, and symptoms did not recur after removal of the coenurus. It is also impossible to determine the causal relationship between the presence of the coenurus and the onset of systemic symptoms that occurred in case 2. Ocular coenurosis may involve the vitreous, anterior chamber, or subconjunctival tissues [7, 14, 15].

There is little recent information on the prevalence of taeniid cestodes in wildlife. In 1974, Lieby and Dyer summarized surveys of the prevalence and geographic distribution of *T. multiceps* and *T. serialis* in wild mammals in North America [16]. Adult parasites and coenuri were widely recovered from definitive and intermediate hosts throughout North America. The prevalence of adult tapeworms in definitive hosts ranged from 3% to 29%. Coenuri were found in 4%–19% of intermediate hosts. In later investigations, *T. serialis* was recovered from 12% of 429 dogs surveyed on Navajo reservations in Arizona and New Mexico [17]. A survey reported in 1988 showed that North American intermediate hosts such as black-tailed jack rabbits (*Lepus californicus*) continue to be infected with *T. serialis* [18].

Epidemiological observations on the Bad River Indian Reservation, where the patient in case 1 was born and raised, revealed that there were numerous wild animals in the area including deer, black bear, coyotes, foxes, snowshoe hares, and cottontail rabbits. Numerous dogs also lived on the reservation. There were three dogs and a pig living outside the patient's house. The child played frequently in the yard area, but the child's parents denied that she had displayed pica behavior. Several dogs owned by local volunteers were later purged by the arecoline hydrobromide method, and their bowel contents were examined. A single tapeworm (*Taenia pisiformis*) was recovered from one dog.

The patient in case 2 was born and raised in Southern California but traveled annually to Juarez, Mexico, to visit relatives. While in Mexico, he purchased food from local street vendors. He owned a pet dog that continued to remain in good health. Although there are no recent surveys of the prevalence of cestode parasites in animals in Southern California, *T. serialis* has been recovered from coyotes in the Los Angeles County area (Patrick Ryan, D.V.M., Los Angeles County Veterinary Services, personal communication). There are also no recently published surveys of the prevalence of cestode parasites in Mexican wildlife. However, prevalence data would be expected to be similar to that reported from Arizona and New Mexico.

There is no distinct clinical syndrome for this disease; however the diagnosis of human coenurosis may be suspected by findings from various radiological studies. Calcified sterile cysts are easily visualized on CT scans. Viable cysts are often visualized as lucent lesions with rim enhancement. Cyst fluid seen on MRI scans is usually of similar signal intensity as CSF

Table 1. Summary of reported cases of human coenurosis in North America.

[Reference]/case no.	Year of case	Patient's age (y) at onset/sex	Geographic area	Site of parasitic cysts	No. of cysts	Exposure to dogs?
[1]	1948	1/M	Idaho, Nevada	Intracerebral, brain stem	Multiple	Y
[2]	1966	62/F	Texas	Trapezius muscle	Single	Y
[3]	1967	2/M	South Dakota	Spinal cord, brain stem, cerebellum, intracerebral	Multiple	Y
[4]	1976	38/F	Ontario	Breast (pectoral muscle)	Multiple	Y
[5, PR]/1	1982	3/F	Wisconsin	Brain stem, spinal cord	Multiple	Y
[PR]/2	1994	39/M	California	Paravertebral muscles	Single	Y

NOTE. PR = present report; Y = yes.

[10]. CT with metrizamide contrast in the CSF may be valuable in demonstrating cysts within the basal cisterns [5].

The differential diagnosis of these cystic lesions would include the more common larval cestode infections, cysticercosis (*T. solium*) and hydatid disease (*Echinococcus granulosus*). Although the lesions produced by sparganosis (infection with larval stages of the tapeworm *Spirometra*) are not usually cystic, they may produce subcutaneous and intracerebral masses. Surgical resection remains the definitive method for diagnosis.

Case 2 appears to be the first example of human coenurosis diagnosed by fine-needle aspiration cytology, a technique used commonly in the diagnosis of subcutaneous cysticercosis [19–21]. There is no contraindication to the use of fine-needle aspiration for subcutaneous lesions. Use of needle aspiration of cystic lesions of viscera or the CNS would be subject to the concern that penetration of a hydatid cyst might cause serious allergic complications, including anaphylaxis.

Serological studies remain experimental, and rates of cross-reaction with other taeniid cestodes remain unclear. In case 2, serological tests performed at the CDC by the enzyme-linked immunoelectrotransfer blot assay method for cysticercosis were negative [22]. Among the other cases of North American coenurosis reviewed, there were two reports of serological testing for other taeniid cestodes. A pediatric case of CNS coenurosis had positive serology for both *E. granulosus* and *Taenia saginata* in serum and spinal fluid [3]. A case of intramuscular coenurosis was reported to have a negative serological test for cysticercosis [4].

PCR-based restriction fragment length polymorphism (PCR-RFLP) has been used experimentally to differentiate various species of taeniid cestodes, including *T. multiceps* and *T. serialis*. This technique may soon prove useful for systematic, epidemiological, and diagnostic purposes [23].

There is very limited information on the use of praziquantel for the treatment of coenurosis in sheep, nonhuman primates, and humans [14, 24, 25]. That information and the results of praziquantel treatment in the two cases reported above indicate that the drug kills the parasite rapidly. In case 1, there appeared to be a favorable response to the combination of surgical de-

compression and administration of praziquantel. However, it is impossible to determine the contribution of each treatment to the change in status. After only 3 days of praziquantel therapy, cysts recovered from the lumbar thecal sac were noted to be nonviable, necrotic, and degenerating.

In case 2, there was a marked local inflammatory response after only one dose of praziquantel. This intramuscular cyst was also nonviable. Although there is no available information on the use of albendazole in coenurosis, this drug would be expected to have an effect similar effect to that of praziquantel, on the basis of its activity in cysticercosis.

A case of successful praziquantel therapy for retroperitoneal, intramuscular, and subcutaneous coenurosis in a spectacled langur (*Presbytis obscura*) has been reported. The animal tolerated the therapy with clinical and radiographic improvement and no apparent adverse effects [24]. However, because of the severe inflammatory reaction seen in case 2, surgical resection would still appear to be the treatment of choice in accessible soft-tissue disease [4, 26]. Several patients with neurological disease have also undergone surgical resection, followed by long-term cure [11]. The development of toxic endophthalmitis and retinal detachment following praziquantel treatment of ocular coenurosis mandates extreme caution in the medical treatment of this type of infection [14].

In review of the six reported North American cases of human coenurosis (table 1), the following trends are noted. Disease prevalence does not appear to be localized to any one geographic area in North America. The three pediatric cases developed CNS disease, while the three adult cases had soft-tissue involvement. The two cases with single lesions were adults with soft-tissue cysts. All cases had significant prior exposure to dogs.

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