Neglected rare human parasitic infections: Part II: Coenuriasis

Review Article Wael M Lotfy

Department of Community Health Nursing, Faculty of Nursing, Matrouh University, Egypt

ABSTRACT

Coenuriasis (also called coenurosis, gid or sturdy) is another neglected parasitic infection that is implicated in human clinical complications. It is a rare zoonotic infection with either *Taenia multiceps* or *T. serialis*. The information available in the literature about human infection is scattered and sometimes argumentative.

Keywords: Cestoda, coenuriasis, coenurus, flatworms, gid, metacestode, staggers, sturdy, T. multiceps, T. serialis

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Corresponding Author: Wael M Lotfy, Tel.: +20 1008154959, E-mail: waelotfy@mau.edu.eg

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Human coenuriasis is a zoonotic parasitic infection with a metacestode of either Taenia multiceps or T. serialis (Cestoda: Cyclophyllidea) which is called coenurus. Coenuriasis occurs mainly in rodents, rabbits, sheep, goats, horses, cattle, and occasionally human (Figure 1). On the other hand, adult worms develop in the small intestine of some canids causing taeniasis. However, human cannot be a definitive host for these species. The intermediate host becomes infected by ingesting vegetation contaminated with eggs and/or gravid proglottids of the parasite. In the small intestine, the oncospheres hatch, invade the intestinal wall and migrate mainly to the striated muscles where they develop into metacestodes called coenuri (singular coenurus). This metacestode is in the form of a vesicle that contains multiple inverted protoscolices, attached to the internal membrane of the cyst. Daughter cysts may be seen inside some coenuri. The definitive host becomes infected by ingesting raw or undercooked infected meat. In the host intestine, the metacestode develops into an adult tapeworm, with a prepatent period that varies according to the parasite species. Adult worms reside in the small intestine by attachment to the intestinal wall. After maturity and fertilization, eggs or gravid proglottids are passed in the host's faeces to complete the life cycle^[1]. Coenuriasis occurs very rarely in humans, and many cases probably existed years before it was recognized or discovered^[2]. This review summarizes current knowledge on the biology and public health significance of human coenurosis, and the recommended methods of prevention and control.

1- Taenia multiceps (Leske, 1780) Versteer, 1967

Synonyms: Multiceps multiceps (Leske, 1780) Hall, 1919; M. gaigeri Hall, 1916; M. skrjabini Popov, 1937; *Coenurus cerebralis* (Batsch, 1786) Rudolphi, 1808.

This cestode has a wide distribution especially in Europe and Asia^[3,4]. It has a well-established pastoral life cycle in dogs and domestic livestock and a sylvatic cycle in wild canids and wild herbivores. It colonizes

domestic and wild canids mainly *Canis familiaris, C. lupus, C. latrans, C. mesomelas, Alopex lagopus* (the arctic fox), *Vulpes vulpes* (fox), and *Nyctereutes procyonoides* (raccoon dog). The parasite rarely infects felids like *Felis concolor* (cougar, puma)^[4]. Experimental infections have been established in *Vulpes corsac* (fox), but this species was not as susceptible as dogs^[5]. Coenuri are found in domestic cattle, sheep, goats, pigs, and many wild hosts. There are occasional reports of the larvae in primates like *Theropithecus gelada*^[6].

Infection usually causes morbidity and mortality in livestock. After infection of the intermediate host, oncosphere larvae penetrate the intestinal wall and circulate with the blood. In the central nervous system, moving larvae leave migratory tracts. Heavy infections may result in acute meningoencephalitis, where migratory tracts and encephalitis are visible at necropsy. A disease variously called gid, sturdy, or staggers usually results from the presence of larvae in the central nervous system^[7]. Poor motor coordination is quite common. Vision, posture, and gait may be affected. The presence of one coenurus in the cerebral hemisphere may result in moving of the infected sheep in circles in a direction opposite to the side of the brain in which the larva is located. The presence of the larvae in the spinal cord may cause paralysis of the hindquarters. In infected sheep, depression, anorexia, emaciation, and death are not uncommon^[8,9].

Although, the parasite cannot be transmitted to man from the intermediate hosts, the meat infected with coenuri of *T. multiceps* is considered suitable for human consumption if only a few cysts are present and if they are removed; but, if many coenuri are present in the meat the entire carcass should be judged unsuitable for human consumption^[10]. The first identified human infection was in Paris in 1913. A man was suffering from loss of ability to speak and understand speech, convulsions, and aphasia. His autopsy revealed two coenuri in the brain, most probably *T. multiceps*; one was degenerated and the other contained 75 scolices^[11]. Subsequently, more human cases of

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cerebral coenuriasis were reported from Africa^[12,13], Europe^[14-17], Asia^[2,18-20] and the Americas^{<math>[21-26]}</sup>. Coenuri</sup></sup> were reported mainly in the submeningeal cortex. They were in the form of cysts that grow and compress the surrounding tissue. However, coenuri have been less frequently found within the cerebral parenchyma and the spinal cord^[2,25]. The most common symptoms of human cerebral coenuriasis include headaches, vomiting, and papilloedema that are related to the increased intracranial pressure caused by the mass effect of the cystic lesion. Focal neurological deficit, such as cranial nerve palsy and motor disturbances are also frequent^[20]. In more than 50% of the human cases, cysts are sterile (without protoscolices) which makes diagnosis difficult. Apart from imaging techniques, there is no clinical way of differentiating cerebral coenuriasis from cases of cysticercosis or echinococcosis^[27].

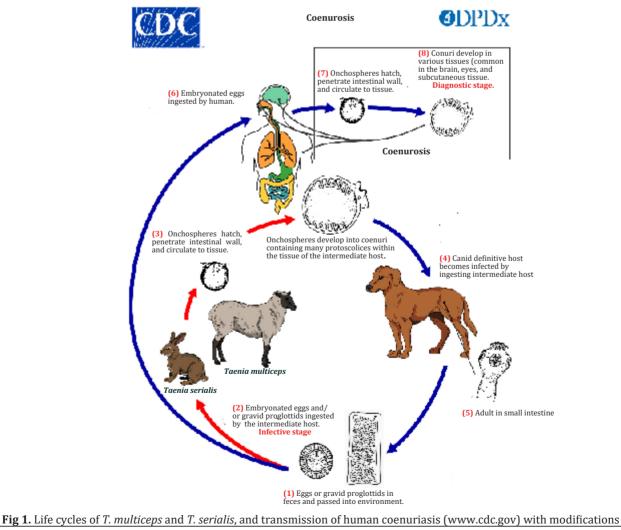
2- Taenia serialis (Gervais, 1847) Baillet, 1867 sensu lato

Synonyms: Coenurus serialis Gervais, 1847; Multiceps serialis (Gervais, 1847) Stiles et Stevenson 1905.

In 1969, Verster divided this species into two subspecies: *T. s. serialis* and *T. s. brauni*. Identification of each was based on biological rather than morphological characters. The former subspecies has a cosmopolitan

distribution while the latter is present exclusively in Africa. Adult worms of both subspecies colonize canids. Coenuri of *T. s. serialis* were reported from lagomorphs and, less frequently, rodents, while coenuri of *T. s. brauni* were reported from rodents and primates^[28].

The first authenticated case of human coenuriasis due to T. s. serialis was reported in a 59-year-old French woman by Bonnal and colleagues in 1933. The coenurus that had been growing under her skin was extracted from her subcutaneous tissue and fed to a dog. Seven characteristic scolices were collected later from the dog^[29]. Thereafter, more human cases were reported in Europe^[30,31]. It is believed that *T. multiceps* was imported into North America, and currently it is no longer present or at least uncommon there^[3]. Cases of human cerebral coenuriasis that were reported from North America may be caused by *T. s. serialis* that is enzootic there^[25]. The cystic larvae of *T. s. serialis* are usually found in the subcutaneous and intramuscular tissues. Experimental infection with *T. s. serialis* produced CNS involvement^[32], and at least one case of fatal infection caused by the T. s. serialis larva in the brain of a cat has been described^[33]. Also, cases of infection without a CNS involvement were reported in USA^[24,34,35].



Taenia s. brauni is endemic in animals exclusively in Africa^[36-38]. Turner and Leiper in 1919, reported a human infection in Nigeria with a coenurus which they identified as *Coenurus glomerulatus*^[39], a parasite which was described previously in gerbils by Railliet and Henry in 1915^[40]. However, this coenurus is identical with T. s. brauni and is therefore considered a synonym^[41]. Many human cases infected with *T. s.* brauni were reported in Tropical Africa. Mainly from Burundi, Kenya, Rwanda, Nigeria, the Democratic Republic of the Congo, and Uganda. Larvae usually develop in the subcutaneous tissue and intramuscular connective tissue, and eve. In rare cases, a few larvae were also found in the brain^[30,41-47]. Non-ocular coenuriasis caused by T. s. brauni is the most benign form of the human infection. The subcutaneous cysts resemble lipomas or sebaceous cysts^[48]. Cases of human intraocular infections with T. multiceps were reported in tropical and South Africa^[49-51]. However, all the coenurus infections of the eye in Africa are said to have conformed to the species *T. s. braun*i^[44,52,53].

Prevention and control: Human coenuriasis has no vaccination. However, experimental vaccination of sheep was found successful and could lead to a halt in the life cycle of *T. multiceps*^[54]. In enzootic areas, the communities should make sure their water supply remains safe and uncontaminated with faeces of the definitive hosts. Also, it is recommended to control transmission of infection through control of the wild definitive host populations. People in enzootic areas should wash fruits and vegetables thoroughly before eating. Also, they should make sure that their dogs are not infected with either *T. multiceps* or *T. serialis*.

For treatment of human coenuriasis, it is recommended to use a combination of surgical excision of the cysts and suitable anthelmintic medication^[2,25]. Surgical intervention is the most common treatment. However, surgical excision is not always possible. The infection becomes more complicated and severe when the oncospheres settle in the CNS tissues. Praziquantel and albendazole are the recommended drugs. Glucocorticoids can be used to help subdue the inflammatory symptoms of the disease. The best treatment for an intravitreal infection of the eye is the removal of the cyst through a closed vitrectomy^[44].

Conclusion: Two species of Taenia: *T. multiceps* and *T. serialis* are incriminated in causing human coenuriasis. Other species reported to infect humans in the literature are synonymous. Coenuriasis is among the neglected human parasitic infections because it occurs very rarely in humans, and many cases of coenuriasis probably existed years before it was recognized or discovered. However, the disease may have CNS involvement and fatal sequelae. Thus, more studies are needed to gain a better understanding regarding the incriminated species and their biology.

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