

Review Article

Oral cysticercosis due to taenia solium infection

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ABSTRACT:

Taenia solium is parasite which is endemic in many developing countries. Humans can be act as definitive host by eating undercooked pork and this condition is called taeniasis. Human can also act as intermediate host when consuming contaminated vegetables, water or through poor hand hygiene. *T. solium* adults that live as parasites in the human intestine are generally asymptomatic and if they cause symptoms, they are generally only mild. Worms are considered as parasites in humans which are quite important because the symptoms, they cause are quite fatal when humans are infected with their larvae. Larval infection is an extraintestinal infection, the disease is called cysticercosis. Cysticercosis occurs due to localization of *T. solium* larvae in various organs including brain tissue, muscles, skin, liver, lungs and heart. Oral cysticercosis are generally asymptomatic and are usually detected early because of their superficial location. The tissues of the oral cavity most frequently affected are the tongue (42.15%), lips (26.15%) - with the lower lips 64.7%, and buccal mucosa (18.9%). Treatment of oral cysticercosis can be accomplished by simple surgical excision and regular follow-up. Oral cysticercosis has a good prognosis and the prevention can be carried out through education about the life cycle of parasites and increasing awareness of the importance of personal and environmental health.

Keys words: Oral cysticercosis, *Taenia solium*

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INTRODUCTION

Taenia solium parasitic infection is a parasitic infection that is transmitted through food. This parasitic infection is endemic in many developing countries and humans will become infected if they eat undercooked pork, a condition known as taeniasis. Humans can also be infected by worm eggs if they eat vegetables or drink water contaminated with worm eggs, or through poor hand hygiene.¹ Worm cysts in ingested pork will develop into adult worms which will then attach themselves firmly to the mucosa of the upper small intestine because the worms have suckers and hooks on their heads (scolex). *T. solium* adults that live as parasites in the human intestine are generally asymptomatic and if they cause symptoms, they are generally only mild.²

T. solium belongs to the class Cestoidea, which is a parasitic worm whose adult stage is hermaphrodite and is covered with non-ciliated integuments. Scolex has a sucker vanity equipped with a hook that serves as a tool for attaching itself to its host tissue. Worms do not have a digestive tract system but have a

complete reproductive system inside the proglottids. These worms can be found in almost all countries in the world except in Muslim countries because humans can only be infected with worms if they eat undercooked pork. Worms are considered as parasites in humans which are quite important because the symptoms, they cause are quite fatal when humans are infected with their larvae. Larval infection is an extraintestinal infection, the disease is called cysticercosis with mild symptoms or even more severe symptoms than those caused by adult worms.³

TAENIA SOLIUM MORPHOLOGY AND LIFE CYCLE

The body of the worm consists of a scolex and a series of proglottids called strobila and contains one male and female sex units. The scolex is equipped with a rostellum, a rod-shaped uterus that extends medially and has lateral branches. Porus genitalis is located laterally and does not have a certain pattern. Adult worms can reach 1-5 meters in length and can

survive in the human body for 25 years or more.² Humans can be infected by adult worms through the mouth by eating pork that contains *T. solium* cysts (cysticercus cellulose). The larvae will come out of the muscle fibers of the meat with the help of gastric acid and then move to the duodenum and jejunum where then they evaginate and attach to the mucosa with a double row of 22 to 32 hooks and 4 suckers on the scolex, finally in 3-4 months the larvae will turn into adult tapeworms which is fully developed.⁴ Generally, humans are infected by one worm although it can be infected by many worms. The gravid proglottid which is located at the very end of the strobila will detach itself from the strobila strand, then leave the host's body with feces or migrate actively through the anus. In some cases, 3-4 proglottids at once move out with feces. The proglottid wall will break outside the host's body and release the egg. The eggs will remain alive in the soil for several weeks. If eggs are ingested by pigs or humans (via food contaminated with eggs) they hatch in the duodenum or jejunum after exposure to stomach acid. Furthermore, oncosphere larvae that come out will penetrate the intestinal mucosa and enter the mesenteric vein so that they are spread to various body tissues including subcutaneous tissue, intramuscular tissue, eyes, brain tissue and other tissues. *Cysticercus cellulose* is oval in shape, milky white or yellowish. The larvae are 5 mm long and 810 um wide. In brain tissue, the volume of the cyst can be high enough to reach 60 ml. The host will localize the larvae in one place by forming a fibrous tissue in the form of a capsule that covers the larvae except in brain tissue, especially in the ventricles.⁵

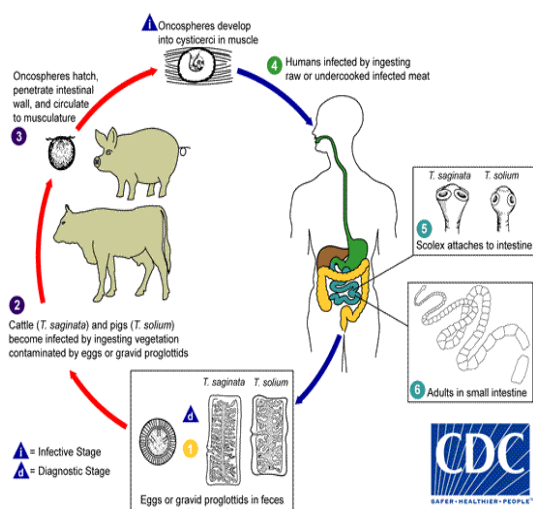


Fig. 1 Life cycle of *Taenia solium*

TAENIASIS

Humans can act as definitive and intermediate hosts for *T. solium*. If humans consume uncooked pork or contaminated pork, in this condition humans act as definitive hosts where the larvae will reach the small intestine and develop into adult worms. This condition is a normal cycle of *T. solium* and the

disease is called taeniasis.⁵ Humans can suffer from taeniasis for a long time and contaminate the environment continuously. Automatic infestation in humans can occur through contaminated fingers or through intestinal peristalsis where the gravid proglottids that are released from the strobila chain will re-enter the intestine, this is equivalent to swallowing thousands of eggs. Some of the eggs that come out of the proglottids will be digested in the stomach, and some of them develop in to oncospheres which will not penetrate the small intestinal mucosa to then spread throughout the body through arterio-venous and lymphatic channels. In general, the dispersed oncospheres will form cysts in the subcutaneous tissue; striated muscles, brain and eye tissue. This condition is also found in vegetarian individuals.^{6,7}

Adult tapeworms cause only mild inflammation at the implantation site, without significant intestinal damage, this causes taeniasis which is generally asymptomatic or characterized by mild symptoms such as abdominal pain, distension, diarrhea with constipation, nausea, loose motions, sensation of hunger, bodyweight loss. Common symptoms include headache, fever and myalgia.^{2,7,8}

Several obstacles can hinder the diagnosis of *T. solium* infection, namely microscopic sensitivity to poor stool samples, and the morphological similarity between *T. solium* and *T. saginata* eggs. If a worm scolex is found and the presence of a double hook (only in *T. solium*) is seen, it can provide definite identification of the species. Genital morphology in mature proglottids or gravid uterus in distal proglottids may also allow species identification. Hematoxylin-eosin staining of the histologic sections of the proglottids can also aid identification. Adult gravid proglottids are rare, so a morphological diagnosis of species based on proglottid material is technically difficult.²

CYSTRICERCOSIS

Cysticercosis is an infection of *cysticercus cellulose*, the larval stage of *T. solium*. *T. solium* goes through its life cycle in two hosts. This disease affects individuals who act as intermediate hosts for *T. solium* and is more common in endemic areas such as Latin America, Asia, Africa and Eastern Europe. Cysticercosis occurs due to localization of *T. solium* larvae in various organs of the human body. Worm larvae will penetrate the intestinal mucosa, enter the blood and lymphatic vessels and then are distributed throughout the body, especially to brain tissue, muscles, skin, liver, lungs and heart. Worm larvae can also be found in the oral and perioral tissues, especially the muscles of mastication. In tissues other than the intestinal mucosa, the larvae eventually die and act as foreign bodies for the body, causing the formation of granulomas and calcified scar tissue as the body's defense against larvae. This process of formation of granulomas and scar tissue occurs about

3 months later after larval infestation. These areas in the tissue are referred to as cysticerci. In this situation, humans act as intermediate hosts who acquire infection through food contaminated with eggs. Symptoms that arise depend on the part of the body that is affected and can spread to various body tissues.^{5,6}

Larvae that develop within cysticercosis can trigger a series of inflammatory reactions including infiltration of neutrophils and eosinophils, lymphocytes, plasma cells and sometimes giant cells, followed by fibrosis and necrosis of the capsule with caseation or calcification of the larvae. It is estimated that after 10 weeks, the egg becomes a cysticercus and the cysticerci can survive for years. The first stage of cysticercal involution is the colloid stage, in which a thick, cloudy fluid replaces the transparent vesicular fluid. The next stage is the granular stage where the scolex shows signs of hyaline degeneration which is followed by thickening of the cyst wall and the scolex turns into coarse mineralized granules. Eventually, the formation of granulomatous tissue will occur which is characterized by the development of histiocytes, epithelioid cells, and foreign body giant cells, which causes fibrosis of the supporting stroma and calcification of parasitic debris. These reactions occur in a similar pattern in the various organs involved, and this evolution indicates the age of the infestation. The exact duration of each of these stages varies widely, mainly because of the considerable differences in the host immune response.⁷

In humans, the parasite commonly infects the central nervous system, causing neurocysticercosis, a pleiomorphic clinical disorder. After entering the central nervous system, the cysticerci can live and cause slight inflammatory changes in the surrounding tissue. Cysticerci can survive for a long time at this stage, are protected by the blood-brain barrier, and the body's defense system will be activated by cysticercuses.^{2,6}

The existence of larvae in various parts of the human body is highly dependent on state of the patient's immune system, type of leukocyte antigen, nutritional status of patients, number of infecting eggs, and *T. solium* strain. Neurocysticercosis disease can affect men and women of various ages with a peak incidence at the age of 30-50 years. Factors related to the pathogenesis of neurocysticercosis include immune response to parasites and their tolerance to strong inflammatory responses, location of the parasite in the brain, the number of infecting cysticercus cellulose, and the stage of the lesion, for example, a cyst has just been seen, the inflammation has formed an exudate, a granuloma has formed, calcification has occurred and fibrotic tissue residue has occurred. Symptoms of brain tissue infection due to worm larvae can be caused by the invasion of these larvae into brain tissue or caused by dead larvae that stimulate the surrounding tissue. It is widely reported that epilepsy suffered by individuals who do not have

genetic factors for the symptoms are strongly suspected to be caused by cysticercosis. Other symptoms of brain cysticercosis are abnormal behavior, transient paresis, intermittent obstructive hydrocephalus, loss of balance, meningoencephalitis, and visual disturbances. Cysticercus can be found in various parts of the eye such as the eyelids or conjunctiva. If the parasite is left unchecked, severe eye damage will occur, especially if the parasite is dead so that it can cause a severe inflammatory reaction.² Efforts to prevent cysticercosis can be carried out through various activities include inspection of pork, thoroughly washed vegetables, consumption of filtered or boiled water, and efficient hand washing before eating and food preparation.⁹

ORAL CYSTICERCOSIS

Oral cysticercosis is a rare occurrence. It develops in humans due to consumption of contaminated food or contaminated drinking water, eating with unclean hands, consumption of raw or improperly cooked pork and reflux of proglottids from the stomach.¹⁰ Oral cysticercosis is a serious condition as a result of ingested eggs developing into embryos (oncospheres) in the intestine which spread through the vascular or lymphatic circulation to develop into cystic larvae (cysticercuscellulosae) in the oral tissues.⁵ Cysticercosis can occur in various tissues but the most commonly affected area is the subcutaneous tissue. Subcutaneous oral cysticerci are generally asymptomatic and are usually detected early because of their superficial location.⁸ The tissues of the oral cavity most frequently affected are the tongue (42.15%), lips (26.15%) - with the lower lips 64.7%, and buccal mucosa (18.9%). A patient might suffer from multiple foci of cysticerci. Once a person is infected with cysticercuscellulosae, cysticercosis can develop in various organs and tissues.⁹



Fig.2 Oral Cysticercosis

A person with cysticercosis does not need to be a resident of an endemic area or have a history of consuming beef or pork. Clinical symptoms depend on the location and number of cysticerci in the body. During the invasion stage there are no symptoms or there is mild muscle pain which may be accompanied by a low-grade fever. Cysticerci are well tolerated by tissues when they are alive but if they are dead, they

will cause a toxic reaction to the surrounding tissue.¹⁰ The patient's most common chief complaint is swelling without pain unless secondary infection is present. The intraoral appearance mostly shows a soft, well-defined, painless swelling that may resemble a fluctuating lesion such as a mucocele. The clinical differential diagnosis depends on the site involved.¹¹ Clinical identification of oral cysticercosis is often confused with benign lesions because of its relatively infrequent occurrence. In the case of solitary nodules on the tongue, lined by normal mucosa, oral cysticercosis can be differentiated with benign neoplasms of neural origin such as neurofibromas and benign schwannomas, granular cell myoblastomas, vascular neoplasms, vascular leiomyomas, fibromas, lipomas, pyogenic granuloma, or rhabdomyoma. In the case of labial and buccal cysticercosis, the differential diagnosis is mucocele, benign tumor of mesenchymal origin, benign tumor of minor salivary glands, such as pleomorphic adenoma, fibroma, lipoma, mucocele, pyogenic granuloma or pleomorphic adenoma.^{7,9,12} Lesions on the tongue can interfere with tongue movement, causing discomfort when speaking and eating. Although, oral cysticercosis shows the spread of the infestation but the majority of patients with oral cysticercosis do not show any systemic complications.⁹

DIAGNOSIS

In general, the diagnosis of cysticercosis can be established through biopsy, radiography, serology, and laboratory tests. Histopathological examination can confirm the diagnosis of cysticercosis by detecting the presence of a cystic space containing cysticercuscellulosae which is lined by three layers, namely the inner cuticle, the middle granular layer, and the outer loose parenchyma layer. The diagnosis of oral cysticercosis can be made by direct histologic examination. In each case, a detailed examination should be carried out to detect the presence of parasites in other areas. MRI and computerized tomography (CT) examinations can be used to evaluate the presence of parasites in the CNS area. Confirmation of the diagnosis of cysticercosis in endemic areas can be done only by histopathological examination.¹¹ FNAC is also a good and fast procedure for diagnosing cysticercosis before surgery to assist doctors in planning treatment. Other diagnostic tools such as radiological imaging and serology may be used. Apart from normal radiographic examination, other imaging modalities are very effective in detecting the cysticercus such as computerized tomography, ultrasonography and magnetic resonance.⁹ Cysts can attack various locations, therefore all patients with cysticercosis should undergo ophthalmologic examination to detect ocular involvement. All patients with extra-neurological cysticercosis should undergo computed tomography or brain magnetic resonance imaging to detect neurocysticercosis. Laboratory blood tests

showed an increase in eosinophils and Immunoglobulin E. Another very important laboratory test is a positive enzyme-linked immunosorbent assay (ELISA) test for cysticercus cellulose. Immunodetection of cysticercosis can be performed by examining samples from serum, cerebrospinal fluid and saliva by ELISA or enzyme-linked immunoelectrotransfer blot. It is important to consider that individuals living in endemic areas may have antibodies due to exposure rather than established infestation.^{9,12}

TREATMENT

Treatment of cysticercosis depends on the symptoms and the accessibility of the lesion. Neurocysticercosis and some other cysticercosis can be treated with drugs such as Praziquantel and Albendazole. The treatment of choice for accessible solitary lesions is surgical excision.⁸ Recent studies have shown that albendazole and praziquantel are effective for the treatment of neurocysticercosis. Treatment of superficial cysticercosis and oral cysticercosis characterized by a solitary asymptomatic nodule can be accomplished by simple surgical excision and regular follow-up.⁹ Follow-up is important for a detailed medical evaluation in each case, to confirm the presence of parasites in other areas.⁷ Treatment of oral cysticercosis should be tailored to the specific needs of the patient regarding the use of anthelmintic drugs or surgery.¹¹ Early diagnosis helps to initiate appropriate therapy either surgical or medicinal. FNAC is the investigation of choice for cases of cysticercosis in endemic areas where a definitive clinical diagnosis cannot be made.¹⁰ Prevention and eradication of disease that can be done is through educational programs about the life cycle of parasites and the importance of personal hygiene.⁷ Alternative therapies need to be further considered considering the nature of the epidemic and the severity of the disease by reducing the rate of worm disease spread, for example by vaccination.¹³

PROGNOSIS

Oral cysticercosis is usually easy to excise and the prognosis is good. In almost all cases, simple surgical excision proved successful in removing the lesion completely without postoperative complications. Treatment of multiple cysts may not be necessary in asymptomatic individuals after confirmation of the diagnosis. Thorough clinical and epidemiological surveys should be carried out with the aim of identifying the possible sources and magnitude of problems in a certain community.¹³

CONCLUSION

Oral cysticercosis is a rare infection caused by cysts of the worm *T. solium*. There is no gender or age predilection. The risk factors for cysticercosis are the consumption of undercooked pork, poor personal and environmental hygiene and a history of contact with

feces containing tapeworm proglottids. The diagnosis of oral cysticercosis can be made by clinical examination, biopsy, laboratory and radiography. Oral cysticercosis often affects the mucous membranes of the lips, buccal and tongue. Oral cysticercosis can be differentiated with benign tumor, malignancy and mucocele. Oral cysticercosis can be treated with simple surgical excision without the use of anthelmintic drugs. Oral cysticercosis has a good prognosis and prevention can be done through educational programs and improving personal and environmental hygiene.

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