Cysticercosis At Rare Sites: Our Experience At A Rural Medical College In Andhra Pradesh, India

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Abstract
Introduction: Cysticercosis is one of the most common parasitic infestations in humans. Neurocysticercosis is the commonest parasitosis of the central nervous system. This article highlights the infestation of this common parasite at eighteen unusual sites in eighteen different cases with review of literature. The sites of infestation are the subcutaneous tissue of chest wall, breast, left lumbar region, umbilicus, abdominal wall, back, extremities, neck region, medial canthus of the eye, and oral cavity. Materials and methods: Eighteen cases of Cysticercosis at rare sites are being discussed. In all the eighteen cases, formalin fixed tissue sections were stained with hematoxylin and eosin. Case 1: An 8-year-old girl presented with swelling over left side of umbilicus. Case 2: A 13-year-old girl presented with a swelling in the right paraumbilical region since 6 months. Case 3: A 40-year-old female presented with swelling in the left lumbar region. Case 4: An 18-year-old female presented with cystic swelling in the oral cavity. Case 5: A 12-year-old girl presented with a swelling in the submental region. Case 6: A 12-year-old girl presented with a swelling in the right paraumbilical region. Case 7: A 12-year-old girl presented with a swelling in the submental region. Case 8: A 20-year-old female presented with a swelling over the right side of chest wall. Case 9: A 25-year-old male presented with a swelling over left side of chest wall. Case 10: A 25-year-old male presented with a swelling over left side of chest wall. Case 11: A 25-year-old male presented with a swelling over the left side of chest wall since 6 months. Case 12: A 10-year-old boy presented with a firm swelling over the right shoulder. Case 13: A 35-year-old female presented with a swelling over the right supraclavicular region since 2 years. Case 14: A 25-year-old male presented with a swelling over the left side of chest wall. Case 15: A 16-year-old female presented with a lump in the right breast since 2 years. Case 16: A 10-year-old boy presented with a swelling over the anterior abdominal wall. Case 17: A 57-year-old male presented with a swelling over the back at the level of lumbar vertebrae. Case 18: A 6-year-old girl presented with a swelling over right side of neck since 8 months. Histopathologically all the eighteen cases were diagnosed as Cysticercus cellulosae. Results: Eighteen cases of Cysticercus cellulosae at rare sites are presented in this study. The mean age of patients was 19 years. Out of total 18 cases 12 (66.66 %) cases were females and 6 (33.33%) cases were males. Fifteen cases were of subcutaneous tissue of chest wall, breast, left lumbar region, umbilicus, abdominal wall, back, extremities, and neck region, 2 cases were of median canthus of eye, and 1 case was of oral cavity. Conclusion: Diagnosis of cysticercosis though common in central nervous system, it should also be kept as differential diagnosis even for the swellings at unusual sites such as the oral cavity, ocular and subcutaneous tissue in the endemic areas of Taeniasis infection.

INTRODUCTION
Cysticercosis is a serious public health problem in several developing countries. Taenia solium has a complex two-host life cycle. Humans are the only definitive hosts and harbour the adult tapeworm (taeniasis) and pigs are the intermediate host. Both humans and pigs can act as intermediate hosts and harbour the larvae or cysticerci. In cysticercosis, humans become the intermediate host by ingesting the infective eggs of Taenia solium from contaminated food and water and replace the pig as intermediate host. Humans are the only host for the adult tapeworm and thus the only source of cysticercosis for pigs or other humans. After entering the intestinal wall, the
embryo invade the blood stream and can lodge in various organs such as the central nervous system, eye, skeletal muscles, and subcutaneous tissue.\(^5\)

**MATERIALS AND METHODS**

Eighteen cases of Cysticercosis at rare sites are being discussed. In all the eighteen cases formalin fixed tissue sections were stained with hematoxylin and eosin.

Case 1: An 8-year-old girl presented with tender swelling over left side of umbilicus since one year. On gross examination, it was a greyish white, thin-walled cystic lesion measuring 1.5 x 1.0 x 0.4 cm.

Case 2: A 13-year-old girl presented with a swelling in the right paraumbilical region since 6 months. On gross examination, there was an irregular greyish brown bit of tissue measuring 2 x 1.5 x 1 cm and a small translucent cyst measuring 0.5 cm in diameter. Histopathologically sections showed the body of cysticercus cellulosae along with inflammatory infiltrate composed of neutrophils, eosinophils, lymphocytes, plasma cells, and histiocytes.

Case 3: A 40-year-old female presented with swelling in the left lumbar region. On gross examination, it was an irregular soft, greyish brown tissue bit measuring 1.5 x 1 x 0.5 cm (Figure 1).

**Figure 1**
Figure 1: Photomicrograph [10X] showing trilaminated corrugated chitinous cell wall of the Cysticercus cellulosae with chronic inflammatory infiltrate.

Case 4: An 18-year-old female presented with cystic swelling in the oral cavity. On gross examination, it was an ovoid, soft cystic mass measuring 1 x 0.5 x 0.5 cm. Cut section, showed a small cystic cavity (Figure 2, 3, 4).

**Figure 2**
Figure 2: Photomicrograph [10X] showing cyst wall with fibrocollagenous tissue and the cysticercus bladder.

**Figure 3**
Figure 3: Photomicrograph [40X] showing cyst wall with fibrocollagenous tissue and the cysticercus bladder.
Figure 4
Figure 4: Photomicrograph [40X] showing chronic lymphoplasmacytic inflammatory infiltrate admixed with occasional eosinophils.

Case 5:A 12-year-old girl presented with a swelling in the submental region. On gross examination, there were two irregular firm swellings, the larger measured 2 x 1.5 x 1 cm and the smaller measured 1.5 x 1 x 0.5 cm. On cut section, the larger mass showed a cystic cavity rimmed by solid grey white area and the smaller was homogeneous grey white.

Case 6: A 12-year-old girl presented with cyst near medial canthus of the right eye. On gross examination, it was an ovoid, cystic, well circumscribed mass measuring 1 x 0.5 cm. Cut section, showed a white nodule in the cavity of cyst. Cyst contained clear fluid (Figure 5, 6, 7).

Figure 5
Figure 5: Photomicrograph [4X] showing cyst wall with cysticercus bladder and the adjoining suckers.

Case 7: An 8-year-old boy presented with cyst near medial canthus of the right eye. On gross examination, it was an oval, grey white, thin walled cyst measuring 1 x 0.5 cm. Cut section, showed a cystic cavity with a white dot and scanty watery clear fluid. Histopathologically sections showed the body of cysticercus cellulosae alongwith inflammatory infiltrate composed of neutrophils, eosinophils, lymphocytes, plasma cells, and histiocytes.

Case 8: An 8-year-old girl presented with swelling over the right side of chest wall. On gross examination, it was an oval greyish white tissue bit measuring 1.3 x 0.8 x 0.4 cm. Cut section revealed a cyst wall with two white nodules and it contained watery cystic fluid. Histopatholgically sections

Figure 6
Figure 6: Photomicrograph [10X] showing cyst wall with cysticercus bladder and the suckers

Figure 7
Figure 7: Photomicrograph [40X] showing cyst wall with cysticercus bladder.
showed a cyst wall lined by flattened epithelium. At one pole the larval form of cysticercus cellulosae seen. The larva showed bladder with multiple papillary invaginations lined by chitinous wall. Scolex with four hooklets observed. The cyst wall was made up of fibrocollagenous tissue infiltrated by the chronic inflammatory cells.

Case 9: A 20-year-old female presented with a swelling over the right side of chest wall. Swelling was soft cystic, pearly white measuring 1 x 0.5 cm.

Case 10: A 25-year-old male presented with a swelling over left side of the chest wall. On gross examination, it was an ovoid, cystic measuring 1.5 cm in diameter. On cutting open it contained watery clear fluid. Histopathologically sections showed trilaminated corrugated chitinous cell wall of the Cysticercus cellulosae with chronic inflammatory infiltrate (Figure 8).

Figure 8
Figure 8: Photomicrograph [10X] showing trilaminated corrugated chitinous cell wall of the Cysticercus cellulosae.

Case 11: A 25-year-old male presented with a swelling over the right lower side of chest wall since 6 months. Ultrasound revealed a swelling in the intermuscular plane. On gross examination, the mass measured 2 x 1.5 x 0.5 cm. Cut section, showed grey white solid area and a cystic area.

Case 12: A 10-year-old girl presented with a firm swelling over the right shoulder. On gross examination, it was an irregular tissue bit measuring 5 x 1 cm. Histopathologically sections showed an undulating degenerative cyst wall lined with underlying inflammatory infiltrate composed of lymphocytes, plasma cells, and foamy histiocytes. Larval form of cysticercus cellulosae was seen.

Case 13: A 35-year-old female presented with a swelling over the right suprascapular region since 2 years. On gross examination, it was an ovoid soft to firm nodule measuring 3 x 2 x 1 cm. Cut section showed cystic area of 2 x 1 cm and solid grey white area of 1 x 1 cm.

Case 14: A 25-year-old male presented with a swelling over the medial aspect of left thigh. On gross examination, it was an oval, greyish cyst with adherent soft tissue to the surface measuring 9 x 7 x 5 cm. On cut section, the cyst contained watery clear fluid and brownish thin folded membrane with a whitish spot. Histopathologically sections showed the bladder of cysticercus cellulosae along with inflammatory infiltrate composed of neutrophils, eosinophils, lymphocytes, plasma cells, and histiocytes.

Figure 9
Figure 9: Photomicrograph [10X] showing cyst wall with fibrocollagenous tissue and the cysticercus bladder.

Case 15: A 16-year-old female presented with a lump in the right breast since 2 years. On gross examination, it was a grey brown, soft tissue mass measuring 2 x 1.5 cm. Cut section, showed solid grey white areas with interspersed yellowish and hemorrhagic areas. Histopathologically sections showed trilaminated corrugated chitinous cell wall of the Cysticercus cellulosae with chronic inflammatory infiltrate.

Case 16: A 10-year-old boy presented with a swelling over the anterior abdominal wall. On gross examination, it was a grey white soft tissue mass measuring 2.5 x 2 cm. Cut section showed a cystic area of 1.5 x 1 cm and homogeneous grey white area of 1 x 1 cm. Histopathologically it was diagnosed as Cysticercus cellulosae.

Case 17: A 57-year-old male presented with a swelling over the back at the level of lumbar vertebrae. On gross
examination, it was a grey white cystic mass measuring 2 x 1 cm. Cut section showed a white nodule measuring 0.5 cm in diameter. Histopathologically it was diagnosed as Cysticercus cellulosae

Case 18: A 6-year-old girl presented with a swelling over right side of neck since 8 months. On gross examination, it was a grey brown soft tissue mass measuring 1.5 x 1 cm. Cut section showed mucoid material. Histopathologically sections showed trilaminated corrugated chitinous cell wall of the Cysticercus cellulosae with chronic inflammatory infiltrate.

All the 18 cases were histopathologically diagnosed as cysticercosis of the respective sites.

RESULTS

Eighteen cases of Cysticercus cellulosae at rare sites are presented in this study. The mean age of patients was 19 years. Out of total 18 cases 12 (66.66 %) cases were females and 6 (33.33%) cases were males. Fifteen cases were of subcutaneous tissue of chest wall, breast, left lumbar region, umbilicus, abdominal wall, back, extremities, and neck region, 2 cases of eye, and 1 case of oral cavity.

Table 1 summarizes the different modes of presentation with the age and sex of our cases.

DISCUSSION

Human cysticercosis is ubiquitous worldwide where consumption of semi- or improperly cooked pork, contaminated salad, and inadequate hygiene are all possible causes of taeniasis and thereby can cause autoinfection. Food habits, poor hygiene, autoinfection, or travelling patterns may be responsible for their distribution. The clinical features of cysticercosis vary with the number, size, location and stage of cysticercosis as well as the intensity of the host’s immune response. Ocular involvements include cysts in eyelids, extraocular muscles, orbit, conjunctiva, anterior chamber, uvea, retina–vitreous, and optic nerve. Brain and eye cysts cause the most morbidity, with the brain being most common location for cysts (60–90% of all cases) and eye being the least common (1–3 %). In our case 2, Cysticercosis was found near the medial canthus of right eye. Total number of cysts can range from a solitary lesion to several hundred. On review of all documented cases of ocular cysticercosis, it was found that 35% of the cysts were reported in subretinal space, and 22% in vitreous. Cysticerci can lodge themselves in any part of the ocular and extraocular tissues and associated brain parenchyma involvement is also quite rare. Pushker et al
studied 20 patients with ocular and extraocular cysticercosis, of whom only 2 (10%) had associated cysts in the brain parenchyma. Our cases 6 and 7 had medial canthus eye swelling, where in CNS involvement was ruled out. Skeletal muscle or subcutaneous cysticercosis can cause localized pain and nodules. Cysticercosis may cause cystic swellings or nodules in mouth, and these may be the only evidence of disease as was in our case 4. Subcutaneous lesions can help in diagnosis of neurocysticercosis.

Human beings acquire cysticercosis through faecal-oral contamination with Taenia solium eggs from tapeworm carriers. Thus, vegetarians and other people who do not eat pork can also acquire cysticercosis. Although cysticercosis has been known for ages, its relation to the adult tapeworm was not clear until it was shown by Kuchenmaister in 1855; he fed condemned prisoners with cysticercosis-infected pork and recovered young tapeworms at autopsy. The larva evaginates in the small intestine; the head (scolex) attaches to the mucosa and begins forming segments (proglottids).

Taenia solium has a scolex with four suckers and a double crown of hooks, a narrow neck, and a large strobila measuring 2–4 mm and consisting of several hundred proglottids. About 2 months after infection, gravid proglottids begin to detach from the distal end and are excreted in the faeces; each segment contains 50–60×10^3 fertile eggs. The worm attaches strongly to the mucosa of the upper small intestine by its suckers and hooks. The adult tapeworm causes only mild inflammation at the implantation site. Abdominal pain, distension, diarrhoea, and nausea are documented, but most patients seem to be free of symptoms. Viable Taenia solium cysts often do not produce symptoms and can evade host immune defences by producing taeniaestatin and paramyosin, which seem to inhibit complement activation.

Carriers of Taenia solium will neither look for medical care nor notice the tapeworm segments in their stools. Conversely, patients infected with Taenia saginata notice passage of numerous, motile proglottids, larger than those of Taenia solium. Identification of Taenia solium is important because of the risk of cysticercosis in the carrier or the immediate environment.

The tapeworm carrier can be found in the patient’s household. Stool examination has poor sensitivity but screening of the patient and household members is recommended so that sources of infection can be detected and eliminated. The lifespan of the adult Taenia solium is unknown. Parasitology textbooks and reviews cite the lifespan as 20–25 years. Some studies suggest it to be probably less than 5 years.

The invasive oncospheres (embryos) in the eggs are liberated by the action of gastric acid and intestinal fluids and cross the bowel wall, enter the bloodstream, and are carried to the muscles and other tissues. At small terminal vessels, they establish and encyst as cysticerci, reaching their definitive size of about 1 cm in 2–3 months.

Extraneural cysticercosis causes no major symptoms. Subcutaneous cysticercosis presents as small, movable, painless nodules commonly noticed over the arms or chest. After a few months or even years, the nodules become swollen, tender, and inflamed, and gradually disappear. In our cases 1, 2, 3, 5, 12, 13, 14, 16, 17, 18 the subcutaneous nodule was over the umbilicus, paraumbilical region, lumbar region, submental region, shoulder, suprascapular region, thigh, breast, anterior abdominal wall, lower back, neck respectively. In cases 8, 9, 10, 11 the subcutaneous nodule was over the chest. Subcutaneous cysticercosis is common in Asia and Africa. Biopsy or fine-needle cytology of subcutaneous nodule helps to confirm the diagnosis of cysticercosis.

Ophthalmic cysticercosis is less common than neurocysticercosis (1–3 % of all infections), Taenia solium is the most common intraorbital parasite. The ocular manifestations can be destructive because the cysticercus gradually increases in size, leading to blindness in 3 to 5 years. The death of the parasite causes release of toxins, leading to intense inflammatory reactions and eye damage. In our ocular cases there was no history of affection of vision.

**DIAGNOSIS**

Two problems hamper the diagnosis of infection with Taenia solium: the poor sensitivity of stool microscopy, and morphological similarity between the eggs of Taenia solium and Taenia saginata. Haematoxylin-eosin staining of histological sections of proglottids can help. Visualisation of taenia eggs by microscopy was the only diagnostic method available until early 1990s. Perianal scraping with adhesive tape (Graham’s test) is highly sensitive for Taenia saginata but not for Taenia solium. Coproantigen detection ELISA is a good tool for taenia-specific molecules in faecal samples with sensitivity of 95% and specificity of 99%. It confirmed that microscopy was poorly sensitive, missing
60–70% of cases. DNA-based assays to discriminate Taenia solium from Taenia saginata infections, \[17,18\] and a serological assay for specific identification of tapeworm carriers have been described.

The most commonly used ELISA cross-reacts with Hymenolepis nana and Echinococcus granulosus, which are common cestode infections. The enzyme-linked immunoblot assay has sensitivity of 98% and specificity of 100%, and is better than ELISA. \[18\] Biopsy of brain, skin, or muscle provides a definitive diagnosis.

**SEROLOGIC DIAGNOSIS**

Advances in serologic diagnosis include the identification and synthesis of specific antigens to obtain consistent and highly sensitive assays.

**ANTIBODY ASSAYS FOR CYSTICERCOSIS**

The Western blot for cysticercosis or the enzyme-linked immunoelectrodiffusion transfer blot (EITB), which uses lentil lectin purified glycoprotein (LLGP) antigens extracted from the metacestode of Taenia solium, has been the “gold standard” serodiagnostic assay since it was first described in 1989. GP50, a Taenia solium protein diagnostic for cysticercosis has been cloned, sequenced, and characterized. It is another diagnostic component of the LLGP antigens that has been used for antibody-based diagnosis of cysticercosis with the EITB assay. GP50 is a glycosylated and glycosyl-phosphatidylinositol–anchored membrane protein. GP50 purified from cysticerci has two homologs expressed in the adult worm: Taenia solium excretory/secretory (TSES) and TSES. Both are diagnostic for taeniasis.

A preliminary evaluation of recombinant GP50 (rGP50) in the EITB assay showed a specificity of 100% for cysticercosis and a sensitivity of 90% for cysticercosis-positive serum samples reactive with the GP50 component of LLGP. \[6\]

**ANTIGEN-DETECTION ASSAYS FOR CYSTICERCOSIS**

Detection of circulating parasite antigen reflects the presence of live parasites, establishes the presence of ongoing viable infection in the absence of definitive radiologic features. \[6\]

**ANTIBODY ASSAYS FOR TAENIASIS**

Humans can be infected with either the tapeworm and/or the larval form of Taenia solium, resulting in taeniasis or cysticercosis, respectively. The diagnosis and treatment of taeniasis is particularly important because this stage of the parasite produces large numbers of infective ova that after ingestion result in cysticercosis in humans and pigs. Treatment and elimination of tapeworms in carriers would eventually result in eradication of cysticercosis. \[6\]

**CONCLUSION**

Cysticercosis though common in central nervous system, it can also be seen in the rare sites like the eye, skeletal muscles, and subcutaneous tissue, as is in our cases. We had seen cysticercosis in the oral cavity, eye and subcutaneous tissue. Hence it should also be kept as differential diagnosis even for the swellings at unusual sites such as the oral cavity, ocular and subcutaneous tissue in the endemic areas of Taeniasis infection.

**References**

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