Review article

Isolated intramuscular cysticercosis: Clinicopathological features, diagnosis and management – A review

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ABSTRACT

Cysticercosis is an emerging parasitic disease affecting worldwide population. There is large volume of data present in the literature for neurocysticercosis. The isolated intramuscular involvement of cysticercosis is uncommon and there are only sporadic case reports available. There is no review available in the literature which gives in detail clinicoradiological features and management of isolated intramuscular cysticercosis. Knowledge about such a presentation is important especially in developing countries and to differentiate the condition from a variety of other similar conditions.

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1. Introduction

Cysticercosis is a parasitic infection caused by encysted larvae of *Taenia solium*, the pork tapeworm.¹ It occurs worldwide and is the most common human parasitic infection of the nervous system in immunocompetent individuals.² Cysticercosis is an ancient disease and has been found even in Egyptian mummies.³ It is endemic to countries like Mexico, Central and South America, Africa, India, China, Eastern Europe, and Indonesia, but due to increased travel and immigration of people it has now spread over worldwide.^{4,5}

Tapeworm infections are common in developing countries where there is poor access to sanitation facilities and close interaction between humans and animals and where pigs are a major source of food. Isolated muscle involvement by cysticercosis is rare^{6,7} and is difficult to diagnose due to nonspecific clinical presentation mimicking a variety of other diseases.⁸ We performed a thorough search on PubMed and Google search engines using the combination of keywords 'cysticercosis', 'muscle', 'isolated', 'intramuscular', 'diagnosis', 'radiology', 'complication' and 'treatment'. Articles related to neurocysticercosis, ocular cysticercosis, disseminated muscular involvement were excluded. We have found no review available in the literature which gives in detail clinicoradiological features and management of isolated intramuscular cysticercosis.

2. Life cycle

For *T. solium* man is the definite and pig is the intermediate host. *T. solium* infection in human occurs when its cysts are ingested from undercooked pork. The outer shell of the cyst is broken by the gastric secretions, leaving behind the head of the parasite known as scolex. The scolex has four cup-shaped suckers and double row of prominent hooks (rostellum) that help in anchorage to the small intestinal mucosa. After attachment, the scolex proliferates and becomes a mature tapeworm by 5–12 weeks. The adult tapeworm then sheds eggs and proglottids into human feces that can contaminate the pig food supply. Eggs are thick shelled and hence are not destroyed in the soil for long time. When pigs or humans ingest the eggs, the gastric secretions break the outer wall of eggs. The oncospheres are released which attach and penetrate the intestinal wall. On the surface of oncospheres hooks and suckers are present which helps in attachment and penetration of intestinal wall. Oncospheres enter into the mesenteric venules and via bloodstream lodge in various tissues, and develop into cysts.⁹ Cysts may lodge anywhere in the body but brain, eyes, subcutaneous tissue and muscles, are more commonly affected.¹⁰ Other rare sites which have been reported are tongue, oral cavity, breast, heart and lungs.^{11–15} The cysts are infectious within 10 weeks. Humans can act as intermediate hosts when fecal-oral transmission occurs via infected food handlers who do not appropriately wash their hands before working, or by fruits and vegetables fertilized with contaminated human waste. Autoinfection involves the retrograde transmission of proglottids from the intestines into the stomach

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due to regurgitation with subsequent release of *T. solium* eggs into the human gut.¹⁶ Ingestion of encysted pork does not directly cause cysticercosis; rather, it produces an intestinal infection of the adult tapeworm and a carrier state for the *T. solium* eggs that, when ingested, produces the clinical syndrome of cysticercosis.

3. Pathogenesis

The encysted larval form of *T. solium* called cysticercus cellulosae, can remain viable in this stage for long time in humans, even up to 10 years.^{10,17} The total number of cysts can range from a solitary lesion to several hundred. Living larvae evade immune recognition and do not elicit inflammation.¹⁰ This phase may last for years and is often clinically silent except when cyst location or size causes signs or symptoms. When the larva dies, it induces a vigorous acute inflammatory response that may produce symptoms, depending on the anatomic location.^{10,15} The resulting acute inflammation may result in local pain and myalgia.¹⁰ Alternatively, degeneration of the cyst may result in intermittent leakage of fluid, which elicits a chronic inflammatory response, with collection of fluid around the cyst, resulting in a mass-like, pseudotumor, or abscess-like lesion.¹⁰ Alternatively, the cyst retracts, its capsule thickens, and the scolex calcifies.¹⁰ The cyst may be completely calcified later on. When the muscle burden of the cyst is large, pseudohypertrophy of the muscle results, characterized by multiple nodules. Such patterns are seen in hyperendemic areas.¹⁸

4. Clinical features

Any age and gender may be affected but children commonly suffer because of increased chances of fomite infection.¹⁹ There may be a history of travel to an endemic zone.²⁰ Both vegetarians and non-vegetarians may be infected.⁶ The clinical features of cysticercosis depend on the location, cyst burden and the associated inflammation.^{10,19} Brain and eye cysts cause the most morbidity, with the brain being the most common location for cysts (60–90% of all cases) and the eye being the least common (1–3%).¹⁹ The probable cause of growth of cysticercal larva in human brain, eyes, and muscles is due to high blood supply in these tissues compared with other tissues. Solitary cysticercosis of muscle without involvement of central nervous system is rare. Most soft tissue and muscular cysticercal infection is associated with central nervous system involvement or multiple cysts.²¹ Therefore central nervous system or ocular involvement should always be ruled out by history, examination and investigations.¹⁹ In contrast to neurocysticercosis, isolated muscle involvement is generally not fatal.²² In the muscular form, three types of clinical manifestations have been described: the myalgic type; the mass-like, pseudotumor or abscess-like type; and the rare pseudohypertrophic type, depending upon the pathogenesis as described above.^{18,23,24}

The physical factors such as the firm nonexpansile nature of the host tissue may limit the growth of the parasite. Sometimes cyst grows near neural structure causing compressive neuropathy.^{25,26} Vascular compromise along with inflammatory response to infection leading to fibrosis of the muscle fibers and subsequent Volkmann's contracture has been reported.²³ These patients present with deformity depending upon the location of the cyst.

The course of the disease may be waxing and waning.²⁵

Fever is usually absent.²⁷ There may be history of trauma which triggers inflammation within asymptomatic cysts. The clinical presentation ranges from asymptomatic cases to those presenting with painful, tender swellings at various sites. Head, neck, trunk and upper extremity involvement has more commonly been seen

than lower extremity involvement but the cause of this cranial preponderance is not known.^{21,27}

5. Diagnosis

Diagnosis of intramuscular cysticercosis is often delayed and missed due to nonspecific clinical presentations.²⁸ Radiological evaluation is often needed to clinch the diagnosis early. The diagnosis of intramuscular cysticercosis can be difficult clinically with several clinical differentials such as lipomas, epidermoid cysts, neuroma, neurofibromas, pseudoganglia, sarcoma, myxoma, pyomyositis, cold abscess or tuberculous lymphadenitis.^{19,21}

5.1. Imaging modalities

The imaging appearance depends on the stage of the parasite.²⁵ In the active phase, plain radiography rarely shows cysticerci but calcified cysts are apparent on plain radiographs.²⁹ Multiple such cysts are seen as millet seed-shaped elliptical calcifications in the muscles on radiographs or computed tomography (CT) scan, so called starry-sky appearance.³⁰ Gradient refocused echo (GRE) imaging is the imaging method of choice for demonstration of the scolex in a CT calcified lesion.³¹ Calcified cysts are usually detected incidentally in an asymptomatic patient.

High-resolution ultrasound has advantages of being non-invasive, non-ionizing modality which is quick to perform, easily repeated with minimum patient discomfort and at a lower cost. On ultrasonography the intramuscular cyst has a characteristic appearance of an eccentric echogenic scolex.^{21,32,33} Four different sonographic patterns of muscular cysticercosis have been described.³⁴ The first type is cysticercus cyst with an inflammatory mass around it, as a result of death of the larva. The second type is an irregular cyst with very minimal fluid on one side, indicating leakage of fluid. The eccentric echogenic protrusion from the wall due to the scolex is not seen within this cyst. This type of appearance may be due to escape of scolex outside the cyst or due to partial collapse of the cyst. In the third type there is a large irregular collection of exudative fluid within the muscle with the typical cysticercus cyst containing the scolex, situated eccentrically within the collection. This may be due to chronic inflammatory reaction around the cyst. This appearance is similar to an intramuscular abscess. In all these three types of appearances, the diagnostic feature is that of the cysticercus itself, which appears as an oval or round well-defined cystic lesion with an eccentric echogenic scolex in it. Fourth is calcified cyst appearing as multiple elliptical calcifications in soft tissue.

Magnetic resonance imaging (MRI) is a gold standard technique for diagnosis of intramuscular cysticercosis. MRI is superior to CT scan in evaluating and detecting the stage of the disease and showing perilesional edema.^{35,36} Recently Tripathi et al. demonstrated the role of MRI in the diagnosis of solitary intramuscular cysticercosis in six patients. MRI can show live scolex and cysts and degenerating cysts as well.²¹ Typically, the cyst is hypointense on T1 weighted images (WI), hyperintense on T2WI, oval or elliptical directed along the course of muscle fibers.²⁸ There is presence of post-contrast perilesional enhancement.²⁸ Scolices can range from 1.5 to 2.5 mm in thickness with intermediate to low signal intensity on T2WI and of intermediate signal within the low-signal fluid on T1WI.²¹ Edema of varying degrees can be seen in all the cases. In the initial stage, when parasite is alive, a fluid-filled lesion without peripheral enhancement is seen.³⁵ In this stage patient may or may not be symptomatic. In the later stage, due to leakage of fluid and resulting host response, peripheral rim enhancement and perilesional edema is seen.³⁵ The patient is symptomatic.²¹

Fig. 1. (A) MRI findings of a 32-year-old female with pain and swelling in the left thigh, revealed a well defined cystic lesion in the left semimembranosus muscle with surrounding soft tissue edema. (B) T2W hyperintense cystic lesion with typical orientation along the direction of muscle fibers. (C) CT scan showed cystic lesion with eccentric intramural calcified focus suggestive of calcified scolex.

CT scan is more sensitive than MRI in showing small calcifications (Figs. 1–5).^{20,36}

The clinicroadiological picture is correlated with stage of the parasite in Table 1.

5.2. Laboratory studies

Hematological investigations are generally non-conclusive and nonspecific with normal blood count. There may be raised ESR and eosinophil counts that may indicate a helminthic infection.²⁸ Eosinophilia is transient and is seen in the early course of the disease associated with migration of the larvae through blood or tissue.²²

5.3. Immunodiagnostic studies

Serological tests available are Enzyme Linked Immunosorbant Assay (ELISA), Complement Fixation Test and Enzyme Linked

Immunotransfer Blot (EITB). Currently, the most effective method for the detection of specific anticysticercal antibodies in serum is EITB.^{37,38} According to the Centre for Disease Control and Prevention, EITB has a specificity of 100%, a sensitivity of 98% for patients with multiple cerebral lesions, and a sensitivity of 60–85% for patients with a single cystic lesion.³⁹

5.4. Histological studies

The definitive diagnosis requires histological demonstration of the parasite.^{40,41} However modern imaging techniques have obviated the need for a formal histologic confirmation.^{28,42} Fine needle aspiration cytology (FNAC) is rapid, safe, cheap, and reliable diagnostic tool for such lesions and it may obviate the need for future biopsy.^{43,44} On cytology there are eosinophils, neutrophils, palisading histocytes and epithelioid cell granulomas with giant cells. Fragments of larval cuticle and parenchyma are strongly

Fig. 2. (A) MRI and (B) USG findings in a 21-year-old journalist with difficulty in writing showed well circumscribed intramuscular circumscribed cystic lesion in the thenar eminence just posterior to the 1st metacarpal.

Fig. 3. (A, B) MRI and (C) USG findings of a 35-year-old female with acute swelling in her right elbow region revealed a circumscribed cystic lesion measuring about 1.5 cm within the substance of brachialis muscle with eccentric nodular focus with marked circumferential myofascial and subcutaneous edema.

suggestive of cysticercosis. Large dagger shaped hooklets are diagnostic which are present within the acute inflammatory exudate (Fig. 6). However, the sample obtained may not be representative and many of the times the scolex and hooklets are missed.

Histopathological examinations of excised material confirm the diagnosis but are usually not required in the advent of modern imaging modalities (Fig. 7). The diagnostic appearance is that of a scolex with four suckers and a double row of hooklets.^{40,41} Mitochondrial DNA analysis of extremely calcified cysts in a histological specimen has also been used to diagnose intramuscular cysticercosis.⁴⁵

6. Management

Therapeutic options include medications, surgery, or watchful waiting. The treatment decision is based on multiple factors,

including symptoms and the location, number, stage, and size of cysts. Isolated skeletal muscle or subcutaneous cysticercosis requires no specific treatment unless it is symptomatic.²⁸ According to some authors, for cysts outside the central nervous system, surgical removal is the optimum management.^{6,8,20,46} Surgery may be required in case of intramuscular cysticercosis if there is neurovascular compromise due to growth of the cyst.²³ Medical treatment should be considered if the site or number of the lesions makes surgical excision unfeasible.^{8,20} Concomitant intestinal taeniasis which is found in 25% of the cases should be investigated and treated as well.^{6,8} Antiparasitic therapy like praziquantel and albendazole can be used to hasten the involution of the cysts and corticosteroids can be added to reduce the inflammatory reaction evoked due to rupture of the cyst.²⁸ Albendazole is a vermicide that causes degenerative changes in the tegument and intestinal cells of the worm by binding to the colchicine-sensitive sites of tubulin, preventing its polymerization

Fig. 4. A 38-year-old female with acute onset swelling in her right side of posterior of chest wall. (A) USG revealed a cystic lesion in the intramuscular region of the upper chest wall (right latissimus dorsi muscle). An intraluminal rounded eccentric well defined soft tissue focus (2.3 mm × 2.2 mm) is noted within the lesion. A tiny echogenic calcified speck noted within the lesion. (B) On MRI well defined rounded lesion in the right latissimus dorsi muscle. The lesion was hypointense on T1 and on T2 hyperintense with well-defined hypointense rim and surrounding edema. Overall findings were suggestive of cysticercus cyst with perilesional inflammation.

Fig. 5. An 8-year-old male with four months history of painless gradually increasing swelling in the left leg, clinically diagnosed as cold abscess. PCR and culture for *Mycobacterium tuberculosis* was negative. ELISA for cysticercosis was positive. On MRI a well-defined oval shaped intramuscular lesion in the leg involving flexor digitorum longus. The wall of the lesion appears hypo tense on both T1 and T2 weight images. The interior of the lesion is mildly hyperintense on T1W1 and markedly hyperintense on T2W1. Significant fascial edema was noted in the intramuscular and intramuscular planes.

into microtubules. This blocks glucose uptake and decreases the glycogen stores of the parasite ultimately leading to immobilization and death of the worm.⁴² Some authors have found medical management alone to be curative.^{10,28} Surgical removal may be associated with rupture of the cyst wall causing leakage of antigens

Fig. 6. Ziehl-Neelsen stain shows acid fast hooklet of cysticercus in fine needle aspirate (original magnification ×1000).

and evoking an inflammatory response. Praziquantel (50 mg/kg/day for 3 weeks) is considered the preferred treatment.²⁰ However, some studies indicate that albendazole (10–15 mg/kg/day for 2 weeks) can be effective as well.^{47,48}

Fig. 7. Photomicrograph shows cysticercus with three layers including outer cuticular, middle cellular and inner parenchyma reticulum (H&E ×200).

Table 1
Clinicoradiological findings according to the stage of the parasite.

Stage	MRI findings	Clinical presentation	Pathology
Initial stage	Fluid-filled cyst without perimeter enhancement	±symptomatic	Parasite is alive
Later stages	Perimeter-enhancing lesion with perilesional edema	Usually symptomatic	Leakage of fluid or parasite dead
Final stage	Elliptical, calcified lesion in the muscle	Usually asymptomatic; incidentally detected	Calcified cyst

7. Prevention

Lifestyle changes, socioeconomic improvement, sanitary improvement, education to people about the disease and anthelmintic therapy are important in reducing the prevalence of the *T. solium* larval diseases. According to the Centre for Disease Control and Prevention Working Group on Parasitic Diseases, cysticercosis is a potentially eradicable disease.⁴ Efforts may include decreasing pork tapeworm carriers and reducing *T. solium* egg shedding through more intense meat inspection and preparation, eliminating exposure of pigs to human feces, and developing a vaccine against *T. solium*. Vaccination of pigs is one of the important measures to interrupt the life cycle and to prevent the transmission of *T. solium* in endemic areas thereby reducing the incidence of human cysticercosis. Recent studies have demonstrated the potential utility of various vaccines for use in pigs, but widespread use is not yet a reality.^{49–55}

8. Conclusion

Though solitary intramuscular cysticercosis is rare, the diagnosis should be kept in mind in patients presenting with an intramuscular or a subcutaneous mass, especially in endemic areas. USG and MRI are useful noninvasive diagnostic modalities to clinch the diagnosis. Only symptomatic cysts require treatment. Both surgery and medical management have been found to yield good results and thus the treatment may be individualized.

Conflicts of interest

The authors have none to declare.

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