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An unusual presentation of forearm intramuscular cysticercosis

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Abstract

Cysticercosis is a parasitic infestation of the body caused by cystodes, the pork tapeworm called Taenia solium.[1] Cysticercosis is an endemic disease in the developing countries. It has a propensity to increase in Western countries and becomes a globalized disease due to the immigrant influx from endemic areas. Cysticercosis can affect various organs like brain, spinal cord, soft tissue, heart, but the clinical manifestations of patients vary upon the encysted larval sites. Isolated muscular involvement is a rare entity and because of non-specific symptomatology, it becomes a diagnostic challenge. Till date there are only handful of cases of isolated intramuscular cysticercosis reported in the literature. Herein, we report a case of isolated myocysticercosis as a forearm swelling in a 23year old female, which produced a diagnostic dilemma on the clinical presentation accurately diagnosed by Ultrasonography and MRI examination and successfully treated with surgical excision and a course of antihelminthic agent.

Keywords: Cysticercosis, tape worm, intramuscular, albendazole, MRI

Introduction

Cysticercosis is a parasitic infestation of the body caused by cystodes, the pork tapeworm called Taenia solium.[1] Cysticercosis is an endemic disease in the developing countries. It has a propensity to increase in Western countries and becomes a globalized disease due to the immigrant influx from endemic areas.[2] Cysticercosis is endemic to countries like India, Africa, Mexico, Indonesia, Eastern Europe and Central America, but has a worldwide spread due to increased travel and immigration.[3] Humans are the definitive hosts for Taenia solium and pigs are the usual intermediate hosts. Humans

may become the intermediate hosts when they either consume eggs of Taenia solium or directly ingest cysticercosis cellulose, which is the larval form of the parasite. Tapeworm infections are common in developing countries where there is poor availability of sanitation facilities and close contact between humans and animals and where pigs are a major source of food. Cysticercosis can affect various organs like brain, spinal cord, soft tissue, heart, but the clinical manifestations of patients vary upon the encysted larval sites.[4] Most muscular disease is associated with involvement of the central nervous system, presence of multiple muscular cysts or a combination of both.[5] Isolated muscular involvement is a rare entity and because of non-specific symptomatology, it becomes a diagnostic challenge.[6] It has been designated as a biological marker of the socio-economic development of the community.[7] Till date there are only handful of cases of isolated intramuscular cysticercosis reported in the literature. [8-12] Herein, we report a case of isolated myocysticercosis as a forearm swelling in a 23year old female, which produced a diagnostic dilemma on the clinical presentation accurately diagnosed by Ultra sonography and MRI examination and successfully treated with surgical excision and a course of antihelminthic agent.

Case capsule

A 23year old female patient presented in General Surgery Out-patient department with complains of initially neglected about painless swelling over the antero-dorsolateral aspect of the proximal forearm since last 2 months. Initially was small in size, gradually progressed to its current size. She approached the hospital once she noticed a painful finger extension movement. She never suffered any constitutional symptoms, such as fever, night sweats, fatigue or recent weight loss during this period. There was no history of any trauma to the limb. The patient's physical status was good and his medical and family history was non-contributory. The pain is of dull aching in nature and gets aggravated with extension movement of the finger. Patient consumed mixed diet habits, but did not consume pork any time. She denied any animal contact. Upon arrival to the OPD, her vital signs were as follows: temperature 97.6 degrees Fahrenheit, heart rate 76 beats per minute, saturation-100% on room air and Blood pressure 120/78 mm Hg. She was conscious and oriented. On systemic examination: CVS- S1S2 heard, RS- Air entry bilaterally equal with no added sounds. Rest per abdomen examination was normal. Blood investigations were under normal limits. Human immunodeficiency virus (HIV) serology, Hepatitis-B and Hepatitis-C viral markers were negative. Chest X-ray and ECG were normal. Examination revealed a mildly tender, non-pulsatile, non-mobile, soft to firm ovoid swelling of 5x3cm in dimension with indistinct margins. The swelling was not adherent to skin but probably laying plane deep in the extensor muscular compartment and was confirmed by finger movements. Plain radiograph showed an oval haziness in the proximal forearm.

Musculoskeletal Ultrasonography examination revealed a well-defined oval hetero genous solid-cystic intramuscular lesion in the proximal forearm measuring around 5.3 x 2.4 x 3.2cm having irregular wall with mobile internal debris and internal septations with mildly increased peripheral vascularity. She was then subjected to MRI, showed a solitary multi-cystic swelling lesion showing T1 hypo, T2 hyper signal intensity in the intramuscular plane in the extensor compartment muscle involving the extensor digitorum communis and brachioradialis. On contrast it showed peripheral and intralesional moderate enhancement with irregular non involving intralesional areas. Titre of ELISA for Taenia

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solium was positive and raised- 21.6 (Negative Titre: <9, Positive titre: >11). She was then given a course of Albendazole 400mg twice daily but the swelling never subsided. Surgical excision of the swelling was done under Supraclavicular block with Albendazole 400mg twice daily for 6 weeks given. On exploration of the affected extremity, swelling was present in the extensor digitorum communis tendon of the left forearm. The swelling was attempted to separate out from the muscle fibres in order to preserve the muscular function, but the cyst was involving the middle and ring finger musculotendinous area in an inseparable way and hence it was excised in toto by including the middle and ring finger tendinous area at the musculotendinous junction. To preserve the extensor function of middle and ring finger, tenodesis of cut end to index and little finger was done. Plaster maintained in extension for 6 weeks and then active mobilization of all finger joints was done. Patient got a satisfactory hand function with no difficulty in daily movement. Clinically it was noticed 5-degree ring finer lag during bulk extension of the Metacarpophalangeal joint of the left hand. Histopathological examination of the swelling revealed circumscribed lesion, showing skeletal muscle with extensive areas of sparse to focally dense infiltration by mononuclear inflammatory cells interspersed by poorly formed epithelial granuloma with few giant cells of foreign body. At places, a coiled and highly convoluted structure with degenerated wall with multiple in folding formation seen, the coiled structure showed degenerated curved refractile hooklet like structures. Patient was followed every alternate day in first week and later on, every week for 6 weeks. Up to 6th week, there was no any recurrence of disease and MRI done at this follow-up shown complete resolution of lesions without any residue.

Discussion

Cysticercosis is caused by encysted larvae of the tapeworm Taenia solium due to ingestion of food or water contaminated with viable eggs of Taenia solium or proglottids regurgitation of an adult worm from the intestine into the stomach during the episodes of violent emesis.[13] Cysticercosis is endemic in Southeast Asia, Mexico, India, Africa, Central America where poor hygienic practices are still prevalent, where pigs are raised as a food source.[14] The increase in number of cases in western countries may be related to immigration and the increase in travel to tropical regions.[15] Cysticercosis is thus defined as an encystment of larval form of this tapeworm in various body tissues.[15] Humans are the only definitive hosts for Taenia solium.[15] When people ingest undercooked infected meat, gastric enzymes lyses the outer shell of the cyst, leaving behind the scolex which is called the head of the worm. The scolex has suckers and hooklet that helps in intestinal wall attachment.[16] Once attached, the scolex proliferates and become an adult tapeworm over 8 weeks. These tapeworms have a 4-year intestinal survival period. They measure 2 to 7 meters in length. Adult worms produce eggs called the proglottids, which mature, become gravid and migrate to the anorectal region or are passed in stools. When pigs ingest the eggs from contaminated soil, the life cycle begins again.[16] Pigs are intermediate hosts in the life cycle of this cystode. It is transmitted to humans through feco-oral contamination by ingestion of contaminated water or vegetables by the proglottids. The eggs are broken down in the stomach by enzymes, releasing oncospheres in the small bowel that penetrates the enteric mucosa and reach various tissues like brain, muscles, eyes and others via hematogenous route. Once they reach any particular organ, they develop into cysticercus cellulose, which is

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the encysted larval form of the tapeworm. Taenia solium has a viability period of around 9-10 years in humans.[17]

Amongst cysticercosis, neurocysticercosis is the most prevalent form affecting the central nervous system and the greatest cause of acquired seizures worldwide.[18] Neurocysticercosis is known to occur in endemic areas for cysticercosis. Cardiac muscle cysticercosis, Brown syndrome, Ocular cysticercosis, Cysticercosis involving the biceps brachii muscle, temporalis muscle, Iliopsoas muscle, tendoachilles tendon sheath, pronator teres muscle have been described.[7]

Our case presented as an isolated forearm swelling which is a rare presentation of myocysticercosis of Extensor digitorum communis and has been uncommonly reported in the literature.

Most muscular cysticercosis are asymptomatic and goes unnoticed for the life. Clinical suspicion of cysticercosis is difficult unless the differential diagnosis are kept in mind and importance is given to the history of the patient.

The other common swellings causing similar presentations and acting as a pseudotumors are lipomas, neuro fibromas, epidermoid cysts, pyomyositis, tuber culous lymphadenitis, intramuscular hematomas, soft tissue myxoma, rhabdomyosarcoma etc.[19]

Radiographs reveal soft tissue enhancement with soft tissue calcification of the inactive cysts which may appear as cigar or millet lesions which are oblong shaped.[10] High resolution Ultrasonography provides all the information available with MRI, and more regards to my cysticercosis. The cyst is better visualised by Ultrasonography than MRI, in muscular lesions.[10] Vijaya Raghavan et al. revealed four different types of sonographic appearances of cysticercosis.[20] The first type of appearance is a cyst cercus cyst with an inflammatory mass around it. Second appearance is an irregular cyst with minimal fluid on one side indicating leakage. The third appearance revealed irregular exudative collection within the muscle. Fourth appearance is of calcified cysts appearing as multiple elliptical calcifications in soft tissue.[20]

Stool test is insensitive for the diagnosis of cysticercosis and many samples may be needed over a period of several days. Indirect hemagglutination (IHA), Enzyme linked immune sorbent assay (ELISA), Indirect fluorescent antibodies (IFA), Enzyme linked immuno electro transfer blot (EITB) tests can be done for demonstration of specific serum antibodies.[21] EITB assay has nearly 100% specificity and 98% sensitivity. ELISA can be done if EITB test is not available, having around 63% specificity and 65% sensitivity with serum.[21]

Three different clinical types of muscular cysticercosis are described- the myalgic, myopathic and the nodular type and the pseudohypertrophy type in which multilocular cyst formation occurs in various group of muscles.[22] Myalgic type as a result of acute inflammation due the death of larvae and as a result there is leakage of cystic fluid.

A chronic inflammatory response due to slow intermittent leakage of fluid with collection of fluid around the cyst resulting in mass or abscess like type. Our case shows feature of nodular or mass like type.

Management of cysticercosis depends on the site of infestation, number of cysts and symptomatology. Isolated my cysticercosis requires no treatment unless is painful or affecting the quality of life and then simple excision may be required. Some studies support that antiparasitic therapy with Albendazole or Praziquantel, in conjunction with Corticoteroids, is very effective in the treatment of extraocular cysticercosis.[23]

Amongst the two mentioned antihelminthics, Albendazole is the preferred drug. It acts by inhibiting

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formation of microtubules, thus blocking glucose uptake in the larval and adult stages thereby depleting their glycogen reserve and ATP production causing immobilization and death of the worm.

Developed countries although, eradicated cysticercosis by improving sanitation and domestic pig raising control, but to date, no measure has achieved to interrupt the transmission process.

Humans with intramuscular cysticercosis are a health concern. Rates of human cysticercosis can be reduced either by detection and treatment of Taenia solium carriers, or by treatment of the whole population.[15]

Conclusion

This case demonstrates a rare incidence. The case is reported for the rarity of the clinical presentation of soft tissue intramuscular cysticercosis as an isolated forearm swelling lodged in the extensor compartment of forearm. Isolated myocysticercosis should always be kept as a differential diagnosis in a patient from endemic zone, presenting with small pseudotumor of uncertain origin. The cyst can be diagnosed completely by non-invasive means such as Ultrasonography and MRI, and can be treated medically with combination of oral antihelminthics and steroids or surgical excision in medically unresponsive swellings.

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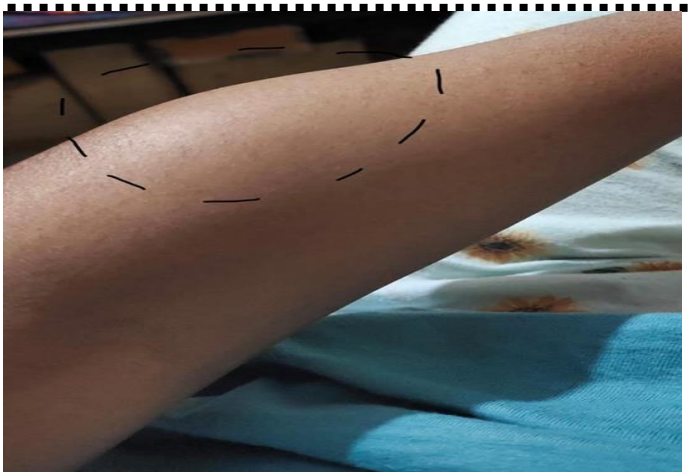

Figure 1: Pre-operative image: 5x3cm swelling over the antero-dorsolateral aspect of the proximal left forearm

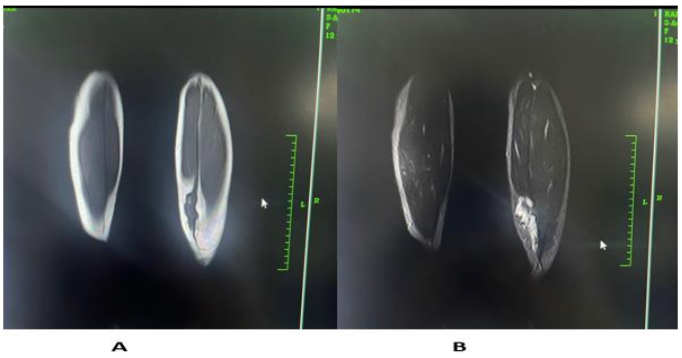

Figure 2: A) Sagittal T1- weighed

B) Sagittal T2- weighted, preoperative MRI revealing a well- defined solitary multi cystic lesion with internal content located in the periphery of left extensor digitorium communis muscle with peripheral and intralesional moderate enhancement

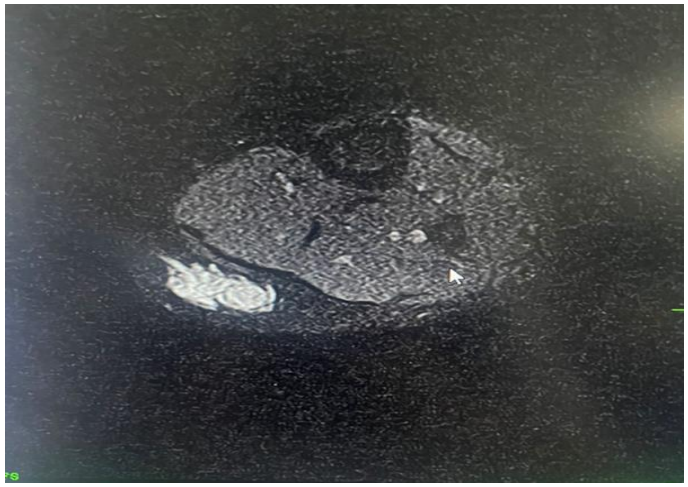

Figure 3: Axial T2- weighted, preoperative MRI

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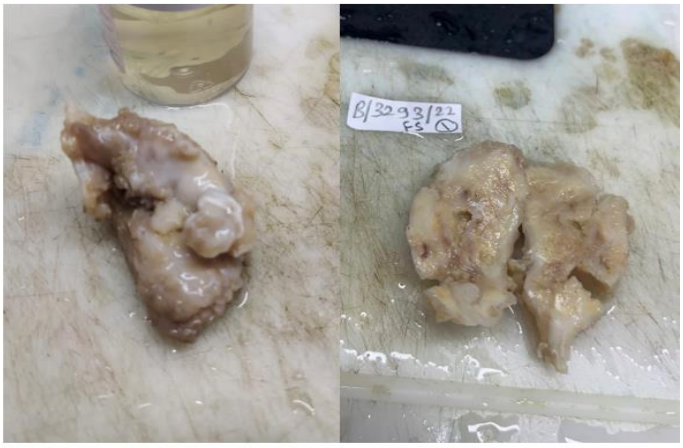
revealing a cyst located in the periphery of extensor digitorium communis muscle. Surrounding muscle tissue appears normal, without oedema



A B

Figure 4: A) Gross image: Greyish white to greyish brown, soft to firm tissue measuring 5x3x2.5cm containing a characteristic greyish white cheesy material B) Cut section: greyish brown, soft to firm tissue

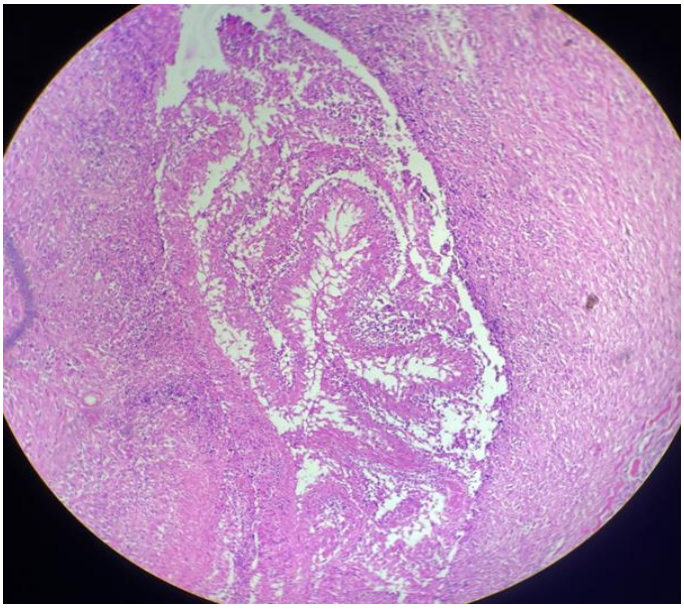


Figure 5A: Histopathological examination showing a convoluted structure (cysticercosis, worm) (stain, Haematoxylin and eosin; original magnification, ×10)

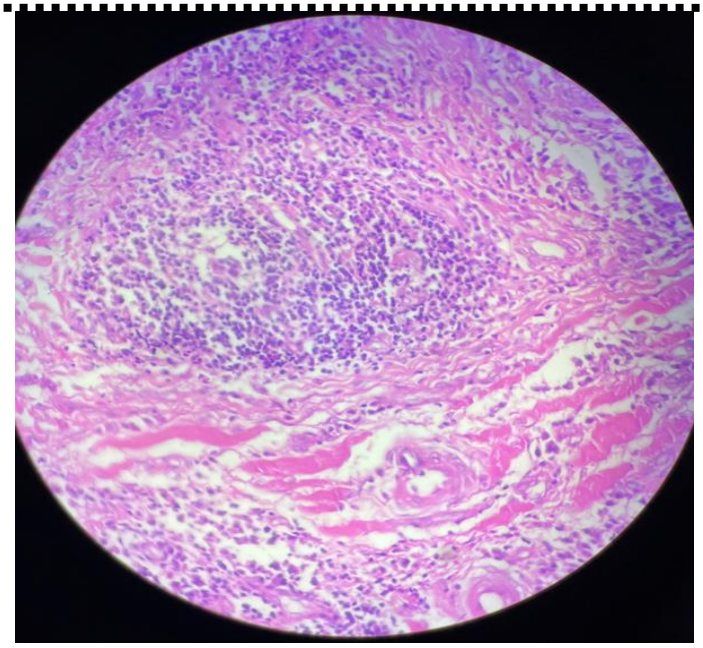


Figure 5B: Histo pathological examination showing aggregation of lymphocytes and plasma cells (stain, Haematoxylin and eosin; original magnification, ×40)

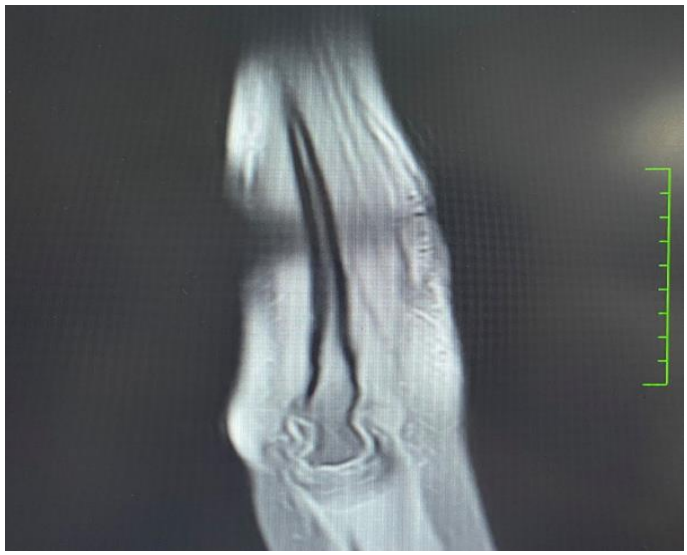


Figure 6: Post operative MRI after 6 weeks showing no cystic lesion without any residual tissue

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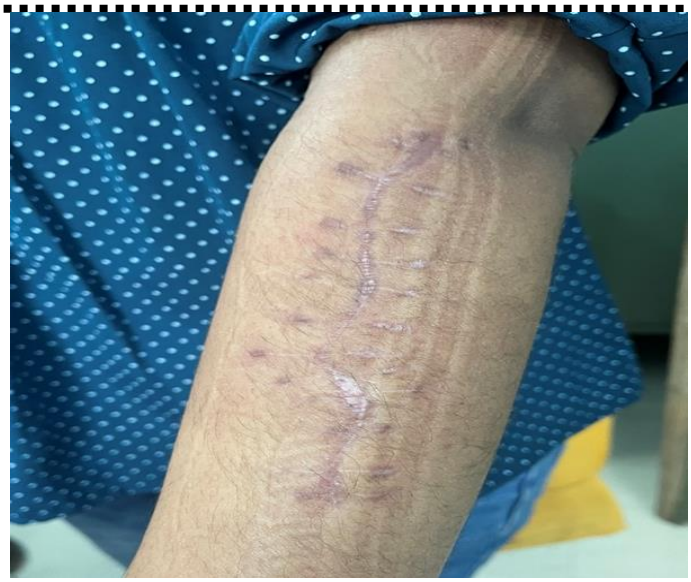

Figure 7: Post operative healed scar

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