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Isolated Forearm Swelling - A Rare Presentation of Soft Tissue Cysticercosis

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Abstract Cysticercosis is a common human infestation in the developing world. Neurocysticercosis is the most common clinical presentation of cysticercal infestation in the human body, but it may be rarely encountered in other body parts like skeletal muscle, subcutaneous tissue and eye. We report this case of an uncommon clinical presentation of soft tissue cysticercosis as an isolated forearm swelling. Only a few cases of cysticercosis presenting as isolated swelling in any body part have been reported in literature. A 30 year old lady presented with a painless swelling in the left forearm for about one year. It was diagnosed as cysticercosis located in the intermuscular plane between the brachioradialis and pronator teres and above the supinator muscles. Magnetic resonance imaging (MRI) and ultrasonography (USG) of the forearm confirmed a cystic lesion and revealed a scolex. She was treated with albendazole and steroids for three weeks but did not respond to medical treatment. The swelling was surgically excised. Hisptopathological examination of the cyst confirmed cysticercosis. Soft tissue cysticercosis presenting as an isolated forearm swelling is a rare presentation of cysticercosis. Treating physicians and surgeons need to have a high index of clinical suspicion for cysticercosis, especially in endemic areas in the developing nations, to facilitate an early diagnosis. Through this case report we also highlight that cysticercosis can be easily diagnosed by non invasive investigations like MRI and USG. It is usually medically treatable but may occasionally require surgical intervention as was needed in our case. This document gives formatting instructions for authors preparing papers for publication in the journal. Authors are encouraged to prepare manuscripts directly using this template. This template demonstrates format requirements for the Journal.

Keywords: *isolated, forearm, swelling, soft tissue, intermuscular septum, cysticercosis*

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1. Introduction

Cysticercosis is a common human infestation in the developing world.

It is a parasitic infestation of the body caused by cystodes, the pork tapeworm, Taenia solium. Humans may be either definitive hosts(adult tapeworm residing in the gastrointestinal tract) or intermediate hosts(larval stage residing in the tissues) for Taenia solium. Humans are the only definitive hosts for Taenia solium and pigs are the usual intermediate hosts [1].

Humans become the intermediate hosts when they either consume eggs of Taenia solium or directly ingest cysticerci, the larval form of the parasite. Cysticerci are ingested by eating undercooked pork. Eggs are usually ingested by eating contaminated foods, uncooked or inadequately washed raw vegetables or salads or by autoinfection through own feces, if a person is harbouring an egg releasing tapeworm in his intestine. Eggs can survive in the environment for many months [1,2].

When eggs are ingested by the intermediate host,the larva (oncosphere) is released which penetrates the intestinal wall, reaches the blood stream and gets lodged in various tissues as the encysted larva (cysticercus) which develops in 60-90 days. These cysticerci can survive for months to years in the tissues. These are responsible for the clinical presentations of cysticercosis [1,2].

Man can become the definitive host when he ingests a tissue containing a cyst and the scolex develops into an adult tapeworm. It is usually single and resides in the upper jejunum for many years. It keeps on releasing proglottids filled with eggs which are excreted in the feces and are infective to man and pigs, but does not lead to clinical cysticercosis in this particular individual [1,2].

Cysticerci can be found in any part of the body but are commonly detected in the brain and uncommonly in the skeletal muscle, subcutaneous tissue or eyes. Cysticerci have a predilection to involve the brain. As a result, neurocysticercosis is a common neurological problem in the developing world. The clinical presentation depends on the location, size, number of lesions and the

inflammatory response evoked in the body by the parasite [1,2,3].

Usually muscle or soft tissue infestation is encountered in association with brain involvement. Isolated soft tissue or muscle involvement with cysticercosis however is not common and only a handful of cases have been reported in literature in which cysticercosis has presented as an isolated swelling in any body part.

We report this case of an uncommon clinical presentation of soft tissue cysticercosis as an isolated painless forearm swelling located in the intermuscular septum.

2. Case Report

A 30 year old female presented to the outpatient clinic of the Department of Orthopaedics with the complaint of a swelling in the left forearm (Figure 1) just below the cubital fossa. She had first noticed this swelling about a year back and gradually it increased to the present size of about 7 X 5 cm. It was situated about 3 cm distal to the elbow on the volar aspect of forearm (Figure 1). On examination it was a painless, soft to firm, mobile mass, not fixed to the underlying structures or to the overlying skin and there was no induration or redness. The swelling was not causing any other symptoms. She did not have any swelling in any other body part. There was no history of any headache, seizures or visual disturbances. Her physical examination was normal apart from the forearm swelling as described.

A plain radiograph showed an oval haziness in the proximal forearm. MRI scan of the left forearm revealed a well defined ring enhancing lesion in the intermuscular plane, lying superficial to the supinator and between the brachioradialis and pronator teres muscles. The lesion was oval and well defined measuring approximately 3.7 X 2.2 cm, iso to hypointense on T1W and hyperintense on T2W images. Within this lesion a rounded lesion was present which was about 1.3 cm in diameter, located eccentrically towards the lateral aspect, hypointense on T1W and hyperintense on T2W images suggestive of a cystic nature. On T2 STIR sequences, a hyperintensity was noted in the surrounding muscle and fascia, suggesting oedema. The lesion showed a distinct enhancement on contrast administration. The underlying bones and the forearm vessels were normal [Figure 2, Figure 3, Figure 4, Figure 5].

An ultrasonographic examination of the left forearm was also performed, which revealed a well defined oval lesion with dense internal echoes within the intermuscular plane, showing an eccentrically located clear anechoic cystic lesion with a scolex within. On the basis of MRI and USG findings a diagnosis of soft tissue cysticercosis was made.

She was investigated further to look for evidence of asymptomatic cysticercosis elsewhere in the body. Her ophthalmological evaluation including fundus examination was normal. The blood investigations showed a hemoglobin of 13.5 gm%, total leucocyte count of 9900/mm^3 (Neutrophils 73, Lymphocytes 17, Monocytes 10, Eosinophils 0). Erythrocyte sedimentation rate was 8 mm first hour rate. Renal and liver function tests were also normal. A plain and contrast CT scan of the brain and USG abdomen were normal. Serological tests for anticysticercal antibodies were negative.

The patient was a non vegetarian but there was no history of pork ingestion and none of her family members had any history of symptomatic cysticercal infestation in any form.

She was treated with albendazole 15 mg/kg/day (400 mg twice daily) for three weeks with prednisolone in tapering doses for three weeks. This led to a decrease in the size of the forearm swelling initially, but at the end of three weeks the size of the swelling appeared almost the same. A repeat ultrasonographic examination was done which had similar findings as detailed earlier. Although the swelling was painless and was not increasing in size further the patient opted for a surgical removal of the cyst. A brownish white cyst of about 4.5X3X2.5 cm size filled with yellowish necrotic material was removed. Histopathological examination revealed a parasitic granuloma with scolex.

3. Discussion

Isolated cysticercal swellings have been reported in literature in muscles of mastication [4,5,6], neck [7,8] tongue [9,10,11], trunk [12], internal oblique [13] and biceps brachii muscles [14].

Our case presented as an isolated forearm swelling which also is a rare presentation of soft tissue cysticercosis and has been uncommonly reported in literature [15,16,17]. Moreover, our patient had a swelling located in the intermuscular plane rather than the muscle itself which is also an uncommon presentation.

The usual differential diagnostic considerations for a forearm swelling are lipoma, sarcoma, neurofibroma, soft tissue myxoma, rhabdomyosarcoma etc. Cysticercosis is not often considered as a diagnostic possibility when a patient with forearm swelling presents in the outpatient clinic. Though uncommon, it is important for the treating physician or surgeon to suspect cysticercosis in cases of isolated soft tissue or muscle swellings, especially so in the developing world where Taeniasis is endemic, since it is usually a medically treatable condition and can be reliably diagnosed by non invasive investigations like MRI and USG.

A definitive diagnosis of cysticercosis can only be made with definite demonstration of parasite in the body either by tissue biopsy)or by fundoscopy (showing the parasite in the anterior chamber, vitreous or subretinal spaces) or by neuroimaging showing cystic lesions with a scolex. Often diagnostic certainty is not possible and a clinical diagnosis is made on the basis of clinical presentation, imaging studies, serologic tests and exposure history [18].

High resolution ultrasound is an inexpensive and readily available modality to diagnose cysticercosis which has the typical sonographic apprearance of a cystic lesion with an echogenic mural nodule representing the scolex, which is a characterstic radiological feature of cysticercosis [19].

Live cysticerci can be diagnosed on MRI as cystic lesions which appear hypointense on T1 and hyperintense on T2 with an eccentric nodule, but scolex is better appreciated on ultrasonography. Also MRI gives a better

idea of the anatomical location of the lesion and the stage of the cysticercal cyst. Viable cysticerci do not show enhancement on MRI as they do not generate an inflammatory response. When the cyst degenerates fluid leaks out and creates inflammation which is seen as peripheral enhancement on MRI and CT. Also variable degree of oedema may be seen in the surrounding tissues [20,21].

MRI and USG in combination can thus be confidently used to diagnose non invasive cysticercosis in soft tissue, as was possible in our case also. Live cysticerci cannot be seen on plain radiographs and CT scan but the final calcified stage can be visualized as elongated foci of calcification along the muscle fibres and can be used as ancillary investigations in the diagnosis of cysticercosis.

Another major diagnostic criterion is detection of specific anticysticercal antibodies in serum by EITB (enzyme linked immunoelectrotransfer blot), which is > 99% specific and sensitive [22]. Positive serum or CSF ELISA for cysticercal antibodies or antigens is included as a minor criterion because it is not as specific as EITB and may be falsely positive. However, it may be negative in single cysticercal lesions as in our case.

Resolution of lesions spontaneously or after anticysticercal treatment is also considered one of the major diagnostic criterion [18]. Our patient though had an initial reduction in her forearm swelling after medical treatment, which could be attributed to the decrease in inflammation due to steroids, did not respond much to albendazole and had to be taken up for surgical excision of the swelling.

Treatment of cysticercosis is usually symptomatic. Especially in neurocysticercosis involving the brain parenchyma, it is highly debatable whether to treat with anticysticidal drugs or not. Many studies have not shown any clinical benefit of treating parencymal neurocysticercosis with anticysticidal drugs, however a faster radiological clearance has been observed. So albendazole (15 mg/kg/day for 8-28 days) or praziquantel (100 mg/kg in three doses over a single day) can be used in parenchymal neurocysticercosis with moderate lesion load (1-5 lesions). But these drugs may induce an inflammatory response around the dying parasite therefore, concomitant use of glucocorticoids is advocated. Steroids may interfere with praziquantel by inducing its first pass metabolism so albendazole remains the preferred drug in clinical practice. However, use of anticysticidal drugs is absolutely contraindicated in cases of multiple neurocysticercosis, ocular, spinal medullary and subependymal cysticercosis, owing to the risk of inflammation and irreversible tissue injury [18].

Cysticercosis is a preventable disease. Preventive measures comprise of good personal hygiene including proper handwashing and sanitization, adequate washing and cleaning of vegetables and salads which are consumed raw, proper disposal of feces, treatment and prevention of human intestinal infections and proper cooking of pork to kill cysticerci.

4. Conclusion

We report this case for the rarity of the clinical presentation of soft tissue cysticercosis as an isolated forearm swelling lodged in the intermuscular plane between brachioradialis and pronator teres and above the supinator muscle. We also wish to highlight the facts that cysticercosis should be considered as a differential diagnosis in painless isolated swellings, particularly in endemic areas. Soft tissue cysticercosis can be confidently diagnosed non invasively on MRI and USG. Medical treatment often cures it, but surgical excision may be required, as in our case, in medically unresponsive swellings.

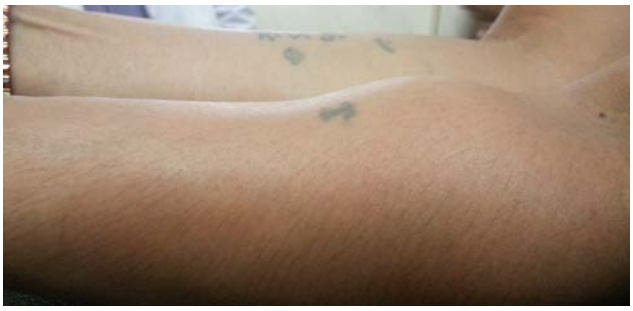

Figure 1. Left forearm swelling below the cubital fossa

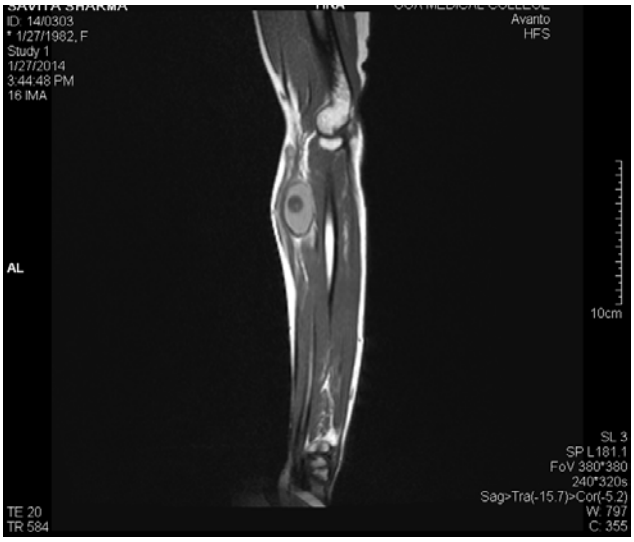

Figure 2. MRI Forearm TI W Spin Echo Sagittal image

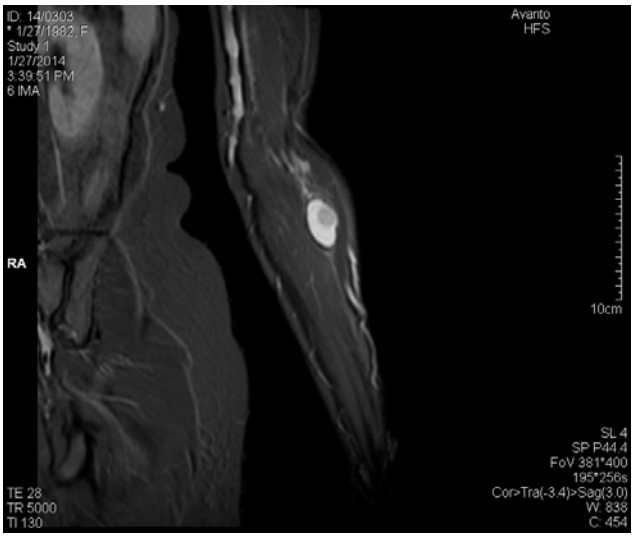

Figure 3. MRI Forearm T2 W STIR Coronal image

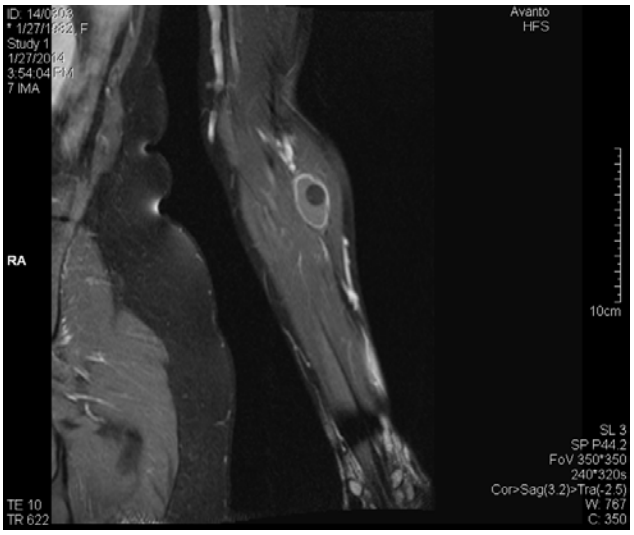

Figure 4. MRI Forearm Post contrast T1W FS Coronal image

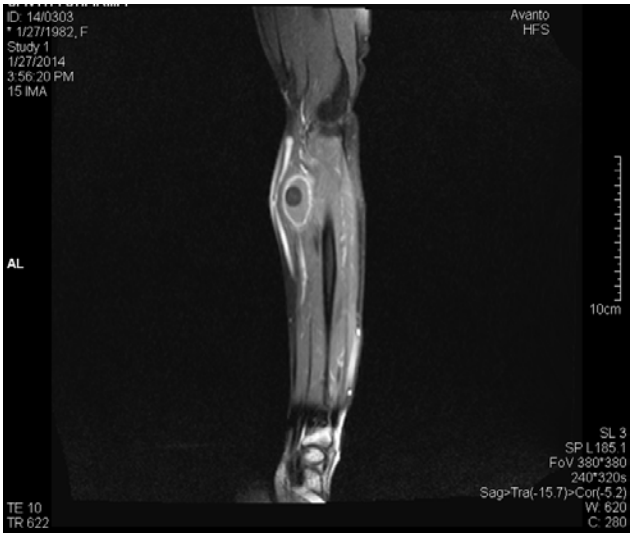

Figure 5. MRI Forearm Post contrast T1 FS sagittal image

Figure 1, Figure 2, Figure 3, Figure 4, Figure 5: An oval well defined lesion measuring approximately 3.7X 2.2 cm in the left forearm lying superficial to the supinator and in between the brachioradialis and pronator teres muscle, appearing iso to hypointense on T 1W sequence and hyperintense on T2W sequence. This lesion is seen to contain a well defined rounded lesion measuring measuring 1.3 cm in diameter located eccentrically towards the lateral aspect appearing hypointense on T1W and hyperintense on T2W sequence suggesting a cystic lesion. Hyperintensity seen surrounding muscle and fascia on T2 STIR sequence suggestive of oedema. The lesion shows contrast enhancement.

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