CASE REPORT

Disseminated cysticercosis incidentally diagnosed in a patient of fracture shaft of femur

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SUMMARY

Cysticercosis is caused by the larval form of *Taenia solium* tape worm, cysticercus cellulose. The life cycle of *T. solium* consists of 2 host, the definite host is a man who harbours the adult forms of the worm in the intestine, while the intermediate host is the pig, where the larval form (cysticercus cellulose) is found in the skeletal muscle. Cysticercosis develops when human beings incidentally become the intermediate host and the eggs mature within their small intestine. The route of entry of the eggs into the human intestine may occur through autoinfection or by ingestion of contaminated food or water. Further, these eggs develop into the larval form which spread throughout the intestinal wall and are disseminated by the blood stream to brain, muscles, subcutaneous tissues or any other organ. Until now, only 50 cases of disseminated forms have been reported in the literature, with the majority of the cases being from Indian subcontinent. Regarding the clinical presentations, symptoms usually depend on the location, size and number of cysts in the involved lesion. However, it may present occasionally with dementia, muscular hypertrophy or subcutaneous nodules with relative absence of focal neurological signs or raised intracranial pressure. We, here, report a case of disseminated cysticercosis, detected incidentally in a man aged 52 years who presented with an open fracture of the right femur.

BACKGROUND

Cysticercosis is a parasitic infection caused due to the larval form of pork tapeworm, *Taenia solium*.^{1 2} The symptoms of disseminated cysticercosis depend on the site of cyst, cyst burden and the host reaction. The most common site of involvement is the central nervous system usually presenting with epilepsy, muscle enlargement, subcutaneous nodules etc. Dissemination into other organs is very rare. Asymptomatic cases are infrequent and have been rarely reported, thus emphasising on documentation of such cases.

CASE PRESENTATION

A man aged 52 years presented to orthopaedics outpatient department of Jawaharlal Nehru Medical College, Aligarh, with a trauma to the right thigh due to road traffic accident 20 days back followed by an open wound along with intense pain. For this, the patient went to a peripheral health centre but due to super-aided infection, the definitive management could not be performed and the patient was referred to our centre. On detailed work-up, there was no history of fever, pain or swelling of any

other site and no history of any weight/appetite loss or loss. Among the routine investigations, complete blood count showed haemoglobin of 12 g/dL, total leucocyte count of 7700 cells/cm, differential leucocyte count showed polymorphs 66%, lymphocyte 30% and eosinophils 4%. Renal and liver function tests were normal. X-ray showed fracture shaft of the femur along with multiple round to oval radio-dense lesions in the muscular plane (figure 1A). Following this, X-rays of the chest and pelvis were advised which also revealed similar multiple radio-dense shadows suggesting calcified cysticerci cysts (figure 1C, D). The patient was then enquired about history related to neurocysticercosis but was only found to have mild weakness in both the lower limbs without any headache, vomiting, sleep disturbance, seizures/fits, pain in joint/ muscle/ abdomen/chest or any eye symptoms in the past. Based on these findings, a provisional clinical diagnosis of infected compound grade III B fracture shaft of the right femur along with multiple calcified lesions suggestive of cysticercosis was made. Being an open, infected fracture with soft tissue involvement, the patient underwent provisional fixation using external tubular fixators and screws (figure 1B). During the peroperative phase, muscle biopsy from the degenerated calcified area was also taken and the biopsy specimen was sent for imprint cytology and histopathological examination. Imprint cytology showed degenerated epithelioid cell granulomas along with degenerated fibrillar eosinophilic material (figure 2A, B), while histopathology revealed amorphous eosinophilic hyalinised material with scattered foreign body type multinucleated giant cells (figure 2C, D). Microscopic examination of stool samples for three consecutive days did not show eggs or larvae of *T. solium*. CT/MRI of the brain/chest or pelvis could not be performed, as the patient was immediately undertaken for operation of the fracture femur. However, serum cysticercal IgG antibody (qualitative ELISA) was found to be positive. On the basis of a high incidence and variable presentations of cysticercosis in our set up, supported by the investigatory findings, the diagnosis was confirmed and the patient was advised anticysticercal treatment in the post-operative period, which comprised of prednisolone 1 mg/kg body weight for 1 week followed by albendazole in a dose of 15 mg/kg/day for 1 month. After 6 week of therapy, fixator was removed and definitive fixation was performed by open reduction and internal fixation with femoral interlocking nail and ipsilateral iliac crest graft. Postop, the patient was ensured to have a healthy healing process and was

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Figure 1 (A) X-ray showing fracture of the shaft of the right femur with multiple white radiodense lesions. (B) Postoperative X-ray showing reduction followed by internal fixation of the shaft of the right femur. (C, D) X-ray of the chest and pelvis showing similar multiple white radiodense lesions.

Figure 2 (A) Imprint cytology smear showing degenerated epithelioid cell granulomas (Pap $\times 10$). (B) Imprint cytology smear showing degenerated fibrillar eosinophilic material (H&E $\times 40$). (C) Section of muscle biopsy showing amorphous eosinophilic collagenised material (H&E $\times 10$). (D) Section showing multiple foreign body-type giant cells (H&E $\times 40$).

further referred to plastic surgery for closure of the open wound using skin flap. After 16 weeks of follow-up, X-ray showed that the fracture united uneventfully and the general condition of the patient improved along with relief of locoregional symptoms.

DISCUSSION

Cysticercosis is an infection caused by larval stage (cysticercus cellulosae) of pork tapeworm *T. solium*. The life cycle of *T. solium* consists of two hosts. The definite host is the human who contains the adult tapeworm, while pig is the intermediate host harbouring the larval stage. The adult worm found in the human intestine sheds the eggs in stool, which infect pigs. On reaching the alimentary tract of pig (the intermediate host),

these eggs rupture, releasing the oncospheres. These oncospheres then penetrate into the gut wall, reach the systemic circulation and ultimately reside in different organs and muscles of the intermediate host where they mature into larval form, cysticercosis cellulosae.³ The organs commonly affected by cysticercosis are the subcutaneous tissues, skeletal muscles, lungs, brain, eyes, liver and rarely the heart. However, dissemination of the cysticerci can occur to any organ of the body. The signs and symptoms depend on the site of cysts, the number of cysts at a particular location and the associated inflammatory host reaction.⁴ Depending on the corresponding site of involvement, a few symptoms of disseminated form include epilepsy, dementia, muscle hypertrophy, subcutaneous or lingual nodules.² Among

these muscular enlargement is the most frequent manifestation of disseminated cysticercosis, followed by palpable nodules and then seizures.⁵ Banu and Veena have reported a case of cysticercosis with dissemination to skeletal muscles, lungs, subcutaneous tissues and brain, while the patient only presented with symptoms of arthritis.⁶ Similarly, our patient was also clinically asymptomatic for cysticercosis, presenting with open compound fracture of the right femur, for which X-ray was performed, which showed incidental multiple round to oval white radiopaque shadows, suggesting cysticercosis.

Besides X-ray, CT and MRI are considered beneficial for anatomical localisation of the cysts. MRI is much more sensitive than CT, as it can identify the scolex of the larvae, the live cysts as well as in knowing the response to treatment.² Non-enhanced CT of the muscles can reveal multiple white radiodense cysts against the black background of the muscle mass, giving the appearance of honeycomb or leopard spots.⁴ In our patient also, X-ray showed multiple white radiodense foci in muscles, which was later on confirmed on cytology and histopathology. CT/MRI could not be performed due to emergency operative condition of the patient.

The role of cytology in diagnosing cysticercosis was first described by Kung *et al*⁷ in 1989. Cytological smears depicting mixed inflammatory cell infiltrate, granulomas along with variable proportion of giant cells are highly suspicious of parasitic origin, even in the absence of a parasite.⁸ Quite similarly in our case also the imprint cytology smear showed many degenerated granuloma along with inflammatory infiltrate. However, giant cells were not seen.

On histopathology, cysticercosis can be vaguely classified into early, intermediate and late stages. Early stages usually show preserved morphology of the parasite with scolex and hooklets, intact subcuticular muscle layer, host tissue reaction in the form of diffuse epithelioid cell proliferation along with lymphocytes and eosinophils. The intermediate stage similarly shows diffuse histiocytic proliferation along with an outer collagen capsule, while the late stage shows well-developed collagenised capsular tissue with very scant lymphocytic infiltrate.⁹ Our case fell in the third category that is the late stage which showed only acellular eosinophilic collagenised/hyalinised material with occasional giant cells.

Regarding the management of disseminated cysticercosis, antihelminthic drugs such as praziquantel (10–15 mg/kg/day for 6–21 days) and albendazole (15 mg/kg/day for 30 days) are given, as they help in decreasing the parasite burden. Patients with symptomatic central nervous system involvement should additionally be given antiepileptics. The treatment of cysticercosis can be accompanied with severe reactions, resulting in

enlargement of the cysts, massive release of antigens leading to local tissue swelling or generalised anaphylactic reaction. Treatment with corticosteroids prior to antihelminthic therapy reduces the occurrence of such complications.^{2 6}

Learning points

- ▶ Cysticercosis is caused due to the larval form of tapeworm, *Taenia solium*.
- ▶ The central nervous system is the most common site of involvement, with the dissemination to other organs being very rare.
- ▶ Symptoms of disseminated cysticercosis are epilepsy, muscular pseudohypertrophy, subcutaneous nodules, etc.
- ▶ Asymptomatic disseminated cysticercosis is very rare, with very few cases reported in the literature.
- ▶ It is important to recognise this rare entity through radiological investigations, so that appropriate therapy can be initiated.

Contributors All the authors have contributed equally in reporting this case. NS and AE have thoroughly investigated and screened this case while SMV has reported this case.

Competing interests None declared.

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