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CASE REPORT

Isolated Muscular Cysticercosis: A Rare Pseudotumor and Diagnostic Challenge, can It be treated Nonoperatively? A Report of Two Cases and Review of Literature

Sushil S Rangdal, Sharad Prabhakar, Sarvdeep S Dhatt, Mahesh Prakash, Mandeep S Dhillon

ABSTRACT

Cysticercosis, an infection with the larval form of pork tape worm, *Taenia solium*, commonly presents with multiple muscular cysts or CNS involvement. Due to vague clinical presentation and unfamiliarity of clinicians with this entity, it is difficult to diagnosis when seen as an isolated cyst. Traditional treatment is surgical excision and a course of deworming agents. We present two cases of isolated muscular cysticercosis diagnosed by USG, MRI and blood tests, who responded successfully to oral medication without need of operative intervention.

Case reports: A 45-year-old male patient had a tender 2 week swelling of 4 x 5 cm on medial right proximal calf. A 26-year male presented with discomfort and diffuse 6 cm swelling at inner left forearm. In both ultrasound showed characteristic hypoechoic cyst with hyperechoic scolex, and MRI revealed isolated cyst surrounded by inflamed muscles. FNAC confirmed cysticercosis, and the patient responded to oral albendazole (3 weeks) with oral steroids (2 weeks).

Conclusion: Although rare, possibility of isolated muscular cysticercosis should always be considered in any small musculoskeletal soft tissue swelling presenting with nonspecific clinical findings. Both our patients responded to medical therapy and were disease free, as confirmed at 3 months by follow-up MRI. These cysts can be confidently diagnosed on the basis of ultrasound and MRI, and can be very well-treated nonsurgically with an oral drug regimen consisting of albendazole and steroid.

Keywords: Cysticercosis, Isolated muscular, Pseudotumor, Drug therapy.

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INTRODUCTION

Cysticercosis in humans is infection with the larval form (cysticercus cellulosae) of the pork tapeworm *T. solium*.[1] The occurence of cysts in humans in order of frequency is the central nervous system, vitreous humor of the eye, striated muscle, subcutaneous tissue and rarely, other tissues.[2] Most muscular disease is associated with central nervous system involvement, presence of multiple muscular cysts or both.[3] Isolated muscular involvement is a rare finding[4] and because of the nonspecific symptoms, isolated soft tissue cysticercosis is very difficult to diagnose.[5]

Till date there are only handfull cases[6-13] of isolated muscular cysticercosis are reported. We are reporting here two cases of isolated myocysticercosis which produced diagnostic dilemma on clinical presentation, accurately diagnosed by ultrasound and MRI examination and successfully treated noninvasively with oral antihelmenthic medication and steroid combination.

CASE REPORTS

Case 1

An otherwise healthy 45 years male presented to us with pain and swelling in right proximal calf region since two weeks. Patient is office attendant by occupation and got his leg hit against a wooden sofa a week before starting of symptoms. He never suffered any constitutional symptoms, such as fever, night sweats, fatigue or recent weight loss during this period. The patient's physical status was good and his medical and family history was noncontributory. The pain is of dull aching in nature and gets exacerbated with movements of limb, particularly with climbing stairs. The pain and swelling gradually enhanced since then and presently needs diclofenac sodium 50 mg preparation for at least twice a day. Patient has mixed diet habits, but did not consume pork any time. He denied for any contact with animals raised for food but is a habitant of known endemic region for taeniasis.

On physical examination, there was no any obvious swelling was observed in the area pointed out by patient as region of discomfort. Skin over the region was normal. There were no signs of inflammation. Tenderness was localized over posteromedial aspect of proximal right calf approximately 5 cm distal to popliteal crease. Deep palpation revealed a nonpulsatile, soft to firm globular swelling of 3 × 5 cm in dimension with indistinct margins. The swelling was not adherent to skin and probably laying plane deep in the muscles. No other swelling was encountered elsewhere in body. No change in size of swelling was observed in association to knee movements or gravity. Movements of knee joint were within normal limits.

Radiographic examination did not revealed any gross abnormality (Fig. 1). To know about the nature of swelling and extent in soft tissue, the patient undergone magnetic resonance imaging. MRI showed oval well-defined lesion

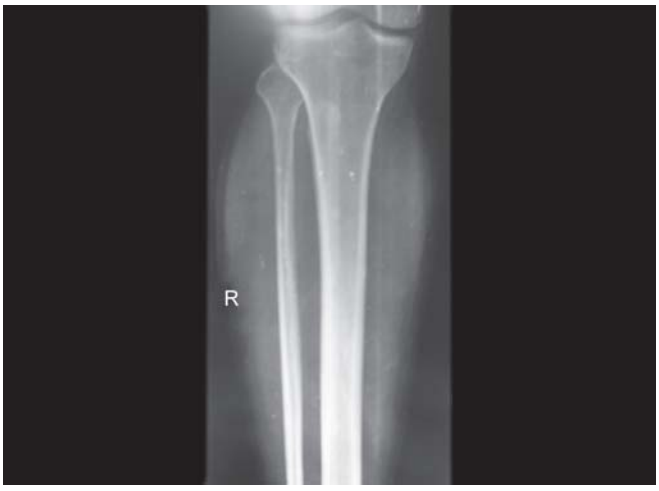


Fig. 1: Radiograph of right leg

in right gastrocnemius muscle. Characteristically it contained rounded cyst of 0.8 × 0.73 cm diameter in center, which was surrounded by inflammatory phlegmon of approximately 3 × 5 cm in dimension (Figs 2A and B). The cyst had low signal on T1-weighted images and high signal on T2-weighted images indicative of fluid content. The phlegmon had constant high intensity in STIR images indicative of edema. Radiologist was in two opinions, either of parasitic cyst or infective abscess and further advised for ultrasound examination. In absence of constitutional symptoms possibility of abscess was very unlikely.

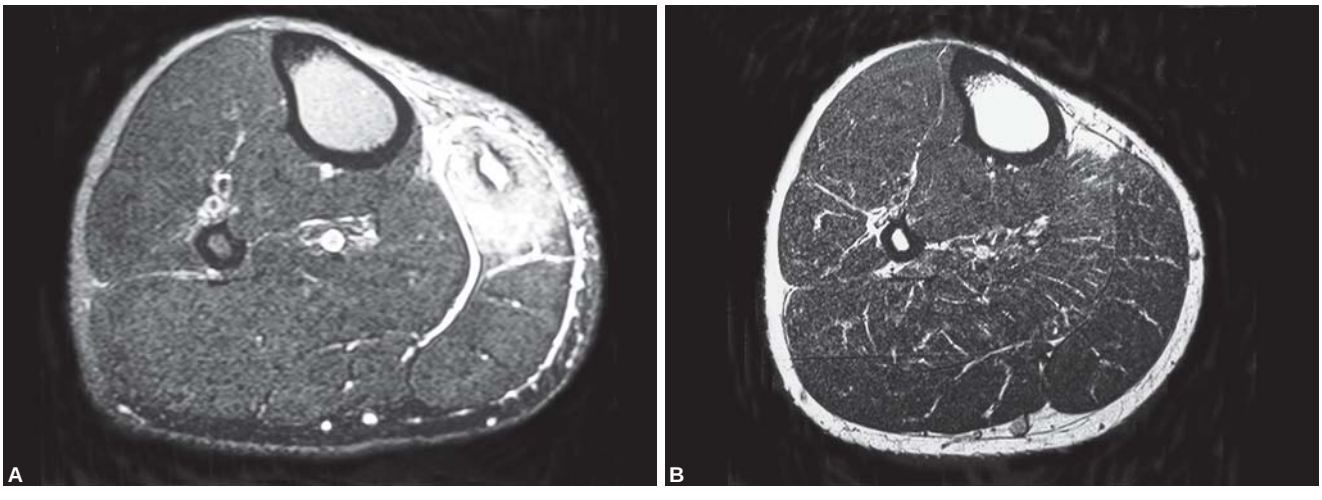
Ultrasound examination reconfirmed the MRI findings and further supported the cyst being a paracytic one. Presence of hypoechoic cyst with eccentrically placed hyperdense signal (scolices) within it and surrounded by inflamed hypoechoic muscle of 3 × 5 cm dimension; these were the features which cleared the dilemma excluded the infective abscess. At this stage radiologist was sure of the diagnosis and denied any other possibility than muscular cysticercosis.

Mean while patient undergone laboratory tests. It showed hemoglobin of 14 gm%, total leukocyte count of 9500/mm^3. Differential count was as, neutrophils of 69%, lymphocytes 25%; monocytes 4% and eosinophils 2%. Erythrocyte sedimentation rate was 10 mm at the end of 1 hour. Titer of ELISA for *T. solium* was not significantly raised (<1: 800). It was necessary to rule out presence of cysts in more frequently involved and important sites. So patient undergone MRI of brain and eyes which revealed no cysts in these organs. On the basis of MRI and ultrasound picture we confirmed this cyst to be of muscular cysticercosis and which later was redocumented with FNAC report.

We managed this case of isolated muscular cysticercosis conservatively and started him on oral medication. Patient was prescribed tablet albendazole with dose of 15 mg/kg/day for 3 weeks and oral prednisolone with dose of 2 mg/kg/day for 2 weeks, tapering it in the next 1 week. Patient was followed every alternate day in first week and later on, every week for 12 weeks. Patient started responding from fifth day of this course and at the fourth week of follow-up, there was complete resolution of the swelling on palpation and patient fully recovered symptomatically. Up to 12th week, there was no any recurrence of disease and MRI done at this follow-up shown complete resolution of lesions without any residue. Patient was able to do normal stair climbing which was his main concern at presentation. When asked regarding submission of his data for publication, he agreed and given consent.

Case 2

Previously normal 26-year-old male presented to us with complaints of mild discomfort in left forearm, since 3 weeks



Figs 2A and B: (A) MRI of right calf showing cyst with hyperintense signals on T2-weighted images surrounded by inflammatory edema in medial head of gastrocnemius muscle, (B) which completely resolved after a course of medical therapy

which turned in to intense pain and palpable swelling since last 5 days after episode of trivial trauma. Strongly gripping any object and complete extension of fingers of same hand were the exaggerating factors. There are no any constitutional symptoms he suffered during this period. He did not suffered any major illnesses in past and his family history was not contributory. He did not noticed swelling at any other site. Patient is a pure vegetarian of normal build and health and did not had any contact with pork.

On local examination we observed diffuse bulge on ulnar aspect of middle of left forearm. Skin appeared slightly indurated. Palpation revealed firm to hard diffuse swelling in medial one-third of flexor compartment of left forearm extending in middle segment of length of forearm approximately for 6 cm (Fig. 3). It was nonpulsatile, fusiform in shape without distinct margins and moderately tender for deep palpation. The swelling was minimally mobile in horizontal plane, with no movement possible in longitudinal plane and probably arising from layer deep in muscle. Axillary lymph nodes were palpable and mildly tender. There was no other swelling found in body. Typically passive extension of fingers, particularly that of ring and little fingers produced intense pain.

X-ray of left forearm was normal. Further radiological evaluation was performed with MRI study. This shown 1.5 × 0.7 cm hyperintense lesion in flexor carpi ulnaris on T2-weighted images. It was isointense in T1 views and distinctly visualized with ring enhancement in gadolinium contrasted images (Fig. 4). There was inflammatory edema of subcutaneous fat and fascia. The impression was of cysticercal cyst. Ultrasound examination revealed centrally placed hyperechoic scolex within the hypoechoic cyst which was not visualized in MRI (Fig. 5).

The diagnosis of myocysticercosis was confirmed with FNAC which shown fragments of cysticercus admixed with sheets of polymorphs, histiocytes, lymphocytes and few plasma cells.

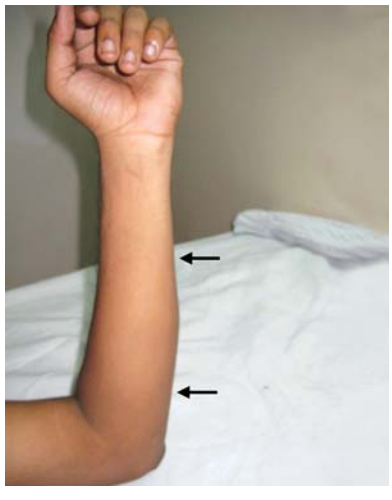


Fig. 3: Extent of swelling on medial third of flexor compartment in mid—6 cm of length of forearm

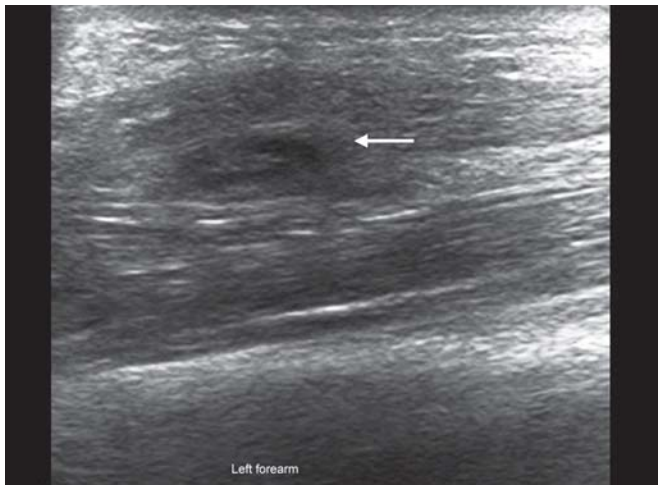


Fig. 4: Ultrasound of forearm showing hypoechoic cysticercal cyst typically containing hyperechoic scolex within it

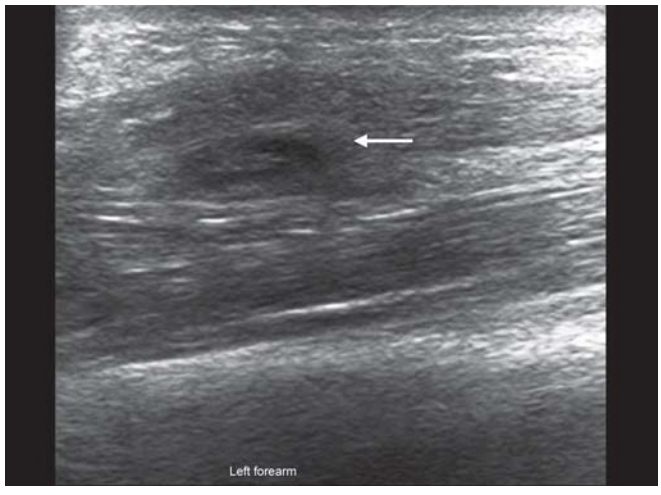


Fig. 5: MRI image of same patient showing hyperintense signals on T2-weighted images surrounded by inflammatory edema in medial aspect of left forearm

Patient was prescribed albendazole with dose of 15 mg/kg/day for 3 weeks and oral prednisolone with dose of 2 mg/kg/day for 2 weeks, tapering it in the next 1 week, the patient responded this combination excellently and was asymptomatic at 3 weeks of follow-up. Patient given consent for publication of any of data for academic purpose. There was no any relapse of disease up to tenth week of follow-up. We ruled out the possibility of any residual lesion at twelfth week by doing MRI which was absolutely normal.

DISCUSSION

Taenia solium (Pork tapeworm) infection and the resulting symptoms are endemic in Southeast Asia, Mexico, Central and South America and Africa,[5] where poor hygienic practices are still common and where pigs are raised as a

food source.[1] The cases of this infestation in the western nations may be related to immigration and the increase in travel to tropical countries.[14] Cysticercosis is encystment of larval form of this tapeworm in various tissues of body.

Humans are the only definitive host for this parasite. When people ingest the raw or undercooked infected meat, stomach enzymes lyses the outer shell of the parasite cyst, leaving the scolex (head) behind. The scolex has suckers and hooks (rostellum) that aid in attachment to the intestinal wall.[15] Once the parasite has attached itself to the intestinal wall, the scolex proliferates and becomes an adult tapeworm over 2 months; these tapeworms can survive 4 years within the human intestines. They may reach 2 to 7 meters in length. Adult tapeworms produce eggs (proglottids), which mature, become gravid, detach from the tapeworm, and migrate to the anus or are passed in the stool. When pigs ingests the eggs from infected soil, the cycle begins again.[15,16]

Normaly pigs are intermediate host in life cycle of this cystode. However, rarely man acts as intermediate host and that manifest as cysticercosis. It is transmitted to humans by ingestion of eggs from contaminated water and vegetables,[17] or very rarely by internal regurgitation of eggs into the stomach due to reverse peristalsis, when the intestine harbors a gravid worm.[18] The eggs lysed in stomach, releasing oncospheres in small inestine that penetrate the bowel mucosa and enter the bloodstream to reach various tissues like brain, subcutaneous tissue, muscles, eyes and others. In these organs they develop to form cysticercus cellulosae, which is the encysted larval form of *T. solium*. These can remain viable in this stage for as long as 10 years in human.[19]

Neurocysticercosis is the most prevalent infection of the central nervous system and is the greatest cause of acquired epilepsy worldwide.[20] Muscles are involved most of the times along with involvement of central nervous system. Isolated muscular involvement by only one cyst is rare.

Most muscular cysticercosis is asymptomatic and goes unnoticed for life of the patient. Rarely, after death of worm in cyst or trauma to cyst there is release of antigens from cyst which initiates immune reaction and inflammation around cyst, making it symptomatic. Our first case in this report had history of trauma, so probably that caused leakage of antigens and subsequent inflammation.

Clinical suspicion of cysticercosis is difficult unless this differential diagnosis is kept in mind and due importance is given to history. The other common pseudotumors causing similar presentation includes lipomas, neurofibromas, epidermoid cysts, pyomyositis or tuberculous lymphadenitis.[10,19,21] In case one, lipoma or neurofibroma were our principle clinical differential diagnosis. In the absence of constitutional symptoms possibility of pyomyositis and tuberculous lymphadenitis was very unlikely. Other causes were excluded on the basis of characteric features on MRI and ultrasound. In case two, tenosynovitis and tuberculous myositis were initial impressions.

Three different clinical manifestations of muscular cysticercosis are described, that includes the myalgic, myopathic type; the nodular or mass like type and the pseudohypertrophy type in which multilocular cyst formation occurs in group of muscle.[17,22] During the death of the larva, there is leakage of fluid from the cyst and the consequent acute inflammation which result in myalgic type of cysticercosis. Alternatively, degeneration of the cyst may result in slow intermittent leakage of fluid, eliciting a chronic inflammatory response, with collection of fluid around the cyst, resulting in the masslike, pseudotumor or abscess like type.[23] Both cases in this report shows features of myalgic type. Blood counts are not helpful except for the elevation of eosinophils which is occasionaly seen in some patients of cysticercosis. But raised eosinophils are only a vague indicator of helmenthic infestation. Both our patients had normal hemogram. ELISA for *T. solium* proved nonspecific and nonsensetive than more precise enzyme linked immunoblot assay[24] which has shown to have greater than 98% sensitivity and specificity. However, in patients with single ring-enhancing lesions, sensitivity falls 60 to 80%.[25]

Plain radiographs rarely show cysticercie unless they are degenerated and get calcified. Multiple small elongated calcifications in soft tissues shadows of thigh and calf are very likely of cysticercosis.[26] Computed tomography is usefull in diagnosing CNS cysticercosis[27] and not beneficial in musculoskeletal lesions. Muscular cysticercie can be reliably diagnosed on ultrasound.[2,10,23,28,29] MRI assess the degree of infection and exact plane of lodgement of cyst in soft tissues. It visualises well the perilesional edema and the degenerative changes of the parasite. Sometimes it can also shows characteristic appearance of solitary cyst and a scolex within it. However, findings may differ according to the growth stage of parasite and host's immune response.[5] In reported case one it shown hypointense cyst on T1 images and hyperintense on T2-weighted images without obvious scolex, which was clearly seen in ultrasound, confirming diagnosis of cysticercosis. The typical features on MRI were fluid-equivalent signal and peripheral rim enhancement indicating parasitic cyst or small abscess. In absence of systemic features of pyogenic infection probability of abscess was very less. As there was no history of contact and constitutional symptoms of tuberculosis, possibility of this lesoin being tuberculous lymphadenitis also was unlikely. In case two, the MRI showed cyst without scolex

within it. On ultrasound this case showed large cyst with scolex lying in center surrounded by inflammatory phlegmon. In this patient tissue diagnosis was done by FNAC and this was confirmatory. Visualization of inflammatory edema around such a cyst is characteristic feature of cysticercal cyst and was present in MRI of both the reported cases.

In recent years ultrasound emerged as a precise and reliable diagnostic tool for imaging intramuscular cysticercosis. There are many reports of cysticercosis being accurately diagnosed solely with the help of this noninvasive method and responded well to chemotherapy.[2,23,28,29]

Four different types of sonographic appearances of cysticercosis have been described by Vijayaraghavan.[23] First type of appearance is a cysticercus cyst with an inflammatory mass around it, as a result of the death of the larva. Similar picture was observed in both of our cases. Second appearance is an irregular cyst with very minimal fluid on one side, indicating the leakage of fluid. The eccentric echogenic protrusion from the wall due to the scolex is not seen within the cyst. This may be because the scolex escapes outside the cyst, or because of the partial collapse of the cyst. The third appearance 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 intermittent leakage of fluid from the cyst, leading to florid inflammatory exudates. Fourth appearance is of calcified cysts appearing as multiple elliptical calcifications in soft tissue similar to the pathognomonic millet seed-shaped elliptical calcifications described on plain radiograph.

In this report we combinely used both the proved imaging techniques in diagnosing myocysticercosis, i.e. MRI and ultrasound, and further reconfirmed it with tissue diagnosis by FNAC without relying solely on one modality.[2,23,28,29] Both the patients did not had any contributory history of seizures, neurological deficit or defective vision, although it is necessary to exclude the presence of asymptomatic cysticercal cysts in more commonly habited sites namely neural and ophthalmic tissues. Both reported patients undergone MRI of brain and eye, and did not reported cysts there. As per our knowledge only few cases of reported isolated muscular cysticercosis[5,28] undergone this screening for excluding neurocysticercosis and ocular cysticercosis. In absence of multiple muscular cysts and other systemic clinical features, we does not feel it is feasible to do whole body MRI to exclude cysts in rarely mentioned sites, though it is recommended by some authers.[5]

Treatment of cysticercosis depends on site of infestation, number of cysts and symptoms of patient. Isolated skeletal muscle or subcutaneous cysticercosis requires no specific treatment unless it is painful, and then simple excision may be required. But in recent past some case reports[2] emphasized on success full nonsurgical management of this disease. Few small case series also support that antiparasitic therapy with albendazole or praziquantel, in conjunction with steroids, is effective in the treatment of extraocular muscle involvement with cysticercosis.[30] In this context, we consider surgical excision of isolated muscular cysticercosis loosing popularity among practitioners when noninvasive promising treatment options are available. Among the two mentioned antihelmenthics, albendazole is the preffered drug. Praziquantel is known to cause abdominal discomfort, diarrhea, drowsiness, and pruritus along with myositis. In neurocysticercosis, inflammatory reactions to this drug may produce meningismus, seizures and mental changes. More so it is contraindicated in ocular cysticercosis because the host response can irreversibly damage the eye by inflammatory process.

Albendazole acts by inhibiting microtubule formation. The loss of the cytoplasmic microtubules blocks glucose uptake in the larval and adult stages of the parasites, thereby depletes their glycogen store and ATP production causing immobilization and death of the parasite. Steroids are used to controll the inflammatory phlegmon caused by these dying cysts. Accordingly our reported patients prescribed albendazole and steroid preparations. They responded well to this medical regimen without any side effects at the end of therapy. Both the patients improved symptomatically and shown complete resolution of their presenting swellings.

Although developed countries eradicated cysticercosis by improving sanitation and controlling domestic pig-raising, but to date, no measure has achieved sustained interruption of transmission. Individuals with muscular cysticercosis are a health concern; but, they do not pose a public-health risk unless they carry an intestinal tapeworm. Only the tapeworm carriers and the infected pigs are important in respect of transmission. Rates of human taeniasis can be decreased either by detection and treatment of the tapeworm carriers, or by treatment of the whole population. Porcine infection can be addressed either by mass antihelminthic treatment or by immunization of the pig population, if an effective vaccine becomes available in future.

CONCLUSION

Isolated myocysticercosis should always be kept in mind as differential diagnosis in a patient from endemic zone, presenting with small muscular pseudotumor of uncertain etiology. The blood picture may be misleading and serum markers have low sensitivity in small cyst. This lesion can

be certainly diagnosed noninvasively with ultrasound and MRI. This cyst can be completely treated nonsurgically with combination of oral antihelmenthic and steroid preparation.

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