ORIGINAL RESEARCH

SUBCUTANEOUS CYSTICERCOSIS: ROLE OF HIGH RESOLUTION ULTRASOUND IN DIAGNOSIS

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ABSTRACT

Background: Though the commonest site of extraintestinal infestation with Taenia solium is brain, Subcutaneous cysticercosis is fairly common in asia. The advent of high resolution ultrasound, FNAC, and a heightened clinician awareness of the existence of isolated soft tissue cysticerci has probably supplanted the need for surgical intervention and excision biopsy in asymptomatic subcutaneous cysts, as cysts have high rate of spontaneous resolution.

Objectives: - To observe role of high resolution ultrasound in diagnosis and need of surgical intervention in treatment of subcutaneous cysticercosis.

Materials & Methods: retrospective study of seven cases of extraneural cysticercosis, all involving the subcutaneous tissues or muscles over the arms and torso. Either high resolution ultrasound, FNAC, or excision biopsy, or a combination of these were used to arrive at a diagnosis. All patients were followed up with serial ultrasounds. All patients received oral nitazoxanide for autoinfection. Surgical excision was resorted to in two patients, in whom it was possible to obtain a histopathologic diagnosis.

Results: of the seven cases of subcutaneous cysticercosis all have rural background, most of the patients (6) were vegetarian and one was non vegetarian. Age & gender of patient, size and duration of lesion were insignificant in establishing the diagnosis. High resolution ultrasound was highly significant in establishing the diagnosis over FNAC and histopathology. Five of the cases resolved spontaneously and surgical intervention was required only in two cases.

Conclusions: With heightened clinician awareness of the existence of isolated subcutaneous cysticercosis in patients with close animal contact, and the widespread availability of high resolution ultrasound and FNAC, subcutaneous cysticercosis can be diagnosed readily. Surgery can be avoided in the great majority of these patients, as the cysts mostly resolve on their own, and do not require any specific treatment. Identification and treatment of autoinfection, and of another source of infection in the household is equally important.

Keywords: Subcutaneous cysticercosis, high resolution ultrasound, vegetarian

INTRODUCTION

Cysticercosis is a common parasitic infection of the human soft tissues, caused by infestation of humans by ingestion of the eggs of *Taenia solium*. It is endemic in Asia, Latin America, Sub-Saharan Africa and parts of Oceania region [1]. Unlike infestation with adult *Taenia*, cysticercosis does not require the patient to eat infected pork as the route of transmission is feco-oral [2]. This also raises the possibility of auto infection [1, 2], and the prevalence of the disease is expected to be high in areas with poor sanitation facilities and open defecation practice. The most common area affected is the brain, followed by the eye, subcutaneous tissue, and muscles. Extraneural cysticercosis is more common in Asia and Africa than in the rest of the world [1]. In India, the prevalence is higher in North than in South India [3]. Though the extraneural tissues are usually involved synchronously or metachronously with the brain, isolated infestation of extraneural tissues in the absence of neural involvement may be more common than previously thought in endemic areas. Excision biopsy [1,2], FNAC [4, 5, 6, 7, 8], and high resolution ultrasound [9, 10, 11, 12, 13] provide means of definitive diagnosis in most cases, and in the recent years high resolution ultrasound has emerged as the initial investigation of choice [6]. The advent of high resolution ultrasound, FNAC, and a heightened clinician awareness of the existence of isolated soft tissue cysticerci has probably supplanted the need for surgical intervention and excision biopsy in asymptomatic subcutaneous cysts.

Here we report a series of seven consecutive patients of isolated subcutaneous or intramuscular cysticercosis, most of whom were managed with expectant treatment with good results. The paucity of non cysticercal subcutaneous or intramuscular cystic lump differentials combined with high resolution ultrasound and astute follow-

up sufficed in our experience as treatment modalities, as the cysts have a very high rate of spontaneous resolution [1].

MATERIALS AND METHODS

From September 2012 to December 2013, seven cases of extraneural cysticercosis, all involving the subcutaneous tissues or muscles over the arms and torso presented to our institute. All the cases were studied retrospectively. Either high resolution ultrasound, FNAC, or excision biopsy, or a combination of these were used to arrive at a diagnosis. Taenia serology was not done due to economic considerations. Stool microscopy was not done considering the poor sensitivity of microscopy in identifying autoinfection [1]. Stool coproantigen assays were not done due to non availability. All patients were followed up with serial ultrasounds. USG was performed with a 6.2 MHz probe using a Seimens Accuson 300 scanner. All patients received oral nitazoxanide for autoinfection. Surgical excision was resorted to in two patients, in whom it was possible to obtain a histopathologic diagnosis.

RESULTS

The tests of statistical significance used were the Z test, two tailed Chi square test, and a single tailed Chi square distribution. The relationship of the diagnosis of cysticercosis with age, sex, size, or duration of the lesion was not found to be significant. A vegetarian diet was found to be a significant factor with a P value of 0.04, as is suggested by the natural history of the parasite. Amongst the diagnostic procedures, FNAC was not significantly associated with either a positive or negative predictive value. A high resolution ultrasound of the affected soft tissues and histopathological examination of the surgical specimen had a highly significant positive predictive value. The details of these observations are discussed in the following tables.

Table 1: Test for significance of parameters

Parameter	N=7	Mean	S.D.	P-value
Age	7	37.14	19.28	0.5
Sex	7	-	-	
Male	3			0.8
Female	4			0.6
Diet	7			
Veg	6			0.04 HS
Non veg	1			0.6
Size of lesion	7	28.28	35.25	0.5
Duration of lesion	7	4.43	2.07	0.5
Type of diagnostic procedure				
a. FNAC	6			NS
+ diagnosis	2			
b. USG	6			VHS
+ diagnosis	6			
c. H/p	2			VHS
+ diagnosis	2			

Table2: Demographic profile of patients

Case No.	Gender	Age (yrs)	Residence	Diet
1.	Female	35	Rural	Non Veg
2.	Male	38	Rural	Vegetarian
3.	Female	53	Rural	Vegetarian
4.	Female	24	Rural	Vegetarian
5.	Female	36	Rural	Vegetarian
6.	Male	7	Rural	Vegetarian
7.	Male	67	Rural	Vegetarian

There were four females and three male patients. The mean age was 37.14 (±19.28) years with an age range of 7-67 years. The diagnosis was most commonly made in the fourth decade of life. All patients were rural with prolonged history of close contact with animals and history of pig rearing in the community. Six out of the seven patients studied were strict vegetarians. In this study, vegetarianism was significantly associated with the risk of cysticercosis with a P value of 0.04. A non vegetarian diet, on the other hand, was not associated significantly with the risk of cysticercosis with a P value of 0.6.

Table 3: Clinical characteristics of the lumps

Case No.	Site	Size*	Consistency	Surface	Dagnosis#
1	Right lateral chest wall, at the level of the fifth intercostals space in the mid axillary line, subcutaneous plane	0.7	Firm to hard	Smooth	Neurofibroma
2	Right lumbar region of anterior abdominal wall, 2 cm lateral to the midclavicular line, subcutaneous plane	1.2	Firm to hard	Smooth	Neurofibroma
3	Left anterior chest wall, one cm below and medial to the mid clavicular line, subcutaneous plane	1.4	Firm to hard	Smooth	Neurofibroma
4	Left lateral chest wall, at the level of the seventh intercostals space, one cm posterior to the anterior axillary line	0.8	Firm to hard	Smooth	Neurofibroma
5	Left flank, at the tranverse level of the first lumbar vertebra, just posterior to the posterior axillary line, subcutaneous plane	2.0	Firm to hard	Smooth	Neurofibroma
6	Left iliac fossa, deep to the deep fascia (the swelling became less prominent on muscular contraction)	1.6	Firm to hard	Smooth	Neurofibroma
7	On the upper back, just medial to the left scapula	10	Soft	Lobulated	Lipoma

*in cm, maximum dimension; # Clinical diagnosis before investigation

Five of seven lumps were situated on the left side of the body. Two of seven lumps were on the back, three on the anterior surface and two on the lateral surfaces of the trunk. All lumps were situated on the trunk, away from common sites of lymph node distribution. One of the lumps was clinically identified to be in the muscular plane, as it diminished on active contraction of abdominal muscles, all the rest were in the subcutaneous plane. The average size of the lumps was 2.83 cm (± 3.53 cm), with a range of 0.7 to 10 cm in the maximum dimension. Out of the seven patients, the diagnosis of subcutaneous cysticercosis was made without the need for surgical intervention in six patients. One of these six patients was taken up for surgical excision due to the development of pain and local inflammation, which was due to secondary infection of the cyst in the third month of follow-up. In the rest five patients, the lumps disappeared spontaneously without the need of any specific therapy directed towards local control of the disease. In the seventh patient, the diagnosis of cysticercosis was obtained only after excision and histopathology.

Table 4: Findings of diagnostic studies performed

Case No	FNAC	USG	Histopathology
1.	Non diagnostic	Subcutaneous cyst with edema.	Not performed.
2.	Non diagnostic	Subcutaneous cyst.	Not performed
3.	Non diagnostic	Subcutaneous cyst.	Not performed
4.	Diagnostic	Subcutaneous cyst.	Not performed
5.	Non diagnostic	Subcutaneous cyst.	Not performed
6.	Diagnostic	Intramuscular cyst with surrounding edema.	S/o cysticercosis.
7.	Not Performed.	Not performed.	S/o cysticercosis.

Table 5: Natural history of the lumps under clinical follow-up

Case No	Surgical intervention	Duration of disappearance of lump (in months)
1.	Not done	8, spontaneous.
2.	Not done	5, spontaneous.
3.	Not done	4, spontaneous.
4.	Not done	8, spontaneous.
5.	Not done	4, spontaneous.
6.	Done due to non resolution and local symptoms	3, taken up for surgery due to super-infection of the cyst.
7.	Done as a diagnosis of cysticercosis was not made before the surgery	Not applicable, history of 4 months before surgical excision.

The first investigation done in these cases with subcutaneous or intramuscular lumps not in the territory of recognized lymph nodes was an ultrasound. In all the cases except case 7, the ultrasound showed a subcutaneous cyst without a scolex with or without surrounding edema. The patients were next investigated with an FNAC. In cases 1, 2, 3, and 5, The FNAC drew clear fluid with a mixed inflammatory infiltrate and could not identify the parasitic nature of the lesion. The patients were kept on close follow-up and disappearance of lump, a positive USG, and non specific FNAC were considered diagnostic. In case 4, the FNAC drew clear fluid with an inflammatory infiltrate and histologic features of a scolex were identified. The patient was placed on follow-up and the lump disappeared. In case 6, an FNAC was done as the lump did not resolve with follow-up and there was associated pain and inflammation. The FNAC yielded pus with a neutrophilic infiltrate and the parasite was correctly identified. This patient was taken up for a surgical excision, and postoperative histology confirmed cysticercosis. In case 7, a preoperative diagnosis of a lipoma on the back was made and a direct surgical excision planned under general anesthesia. It was appreciated intra-operatively that the lesion was cystic and was sent for histopathology which showed cysticercosis. In the present study, the FNAC was not expected to be strongly diagnostic as measured by a Chi square test for a univariate sample. USG was found to be strongly diagnostic by the same test, as was histopathological examination.

Fig 1: Image of excised surgical specimen of an intramuscular cyst

Five of the seven patients had spontaneous resolution of the lumps without the need for surgical intervention. In one patient, surgical excision was taken up due to

local signs of inflammation. In another patient, a preoperative diagnosis of a large lipoma on the back was made, and the cystic nature of the lump was appreciated only during the surgery. In this patient, the lump had been present for 4 months prior to presentation for surgery. The diagnosis was established after histopathological examination of the surgical specimen. The average duration of disappearance of lumps in our study was 4.43 (±2.07) months, with a range of 3 to 8 months.

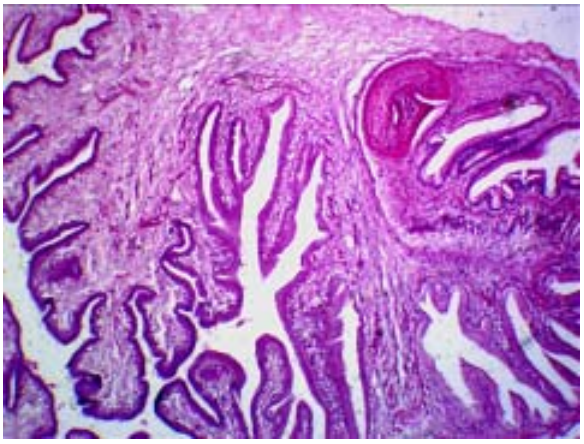

Fig 2: Photomicrograph of the excised surgical specimen

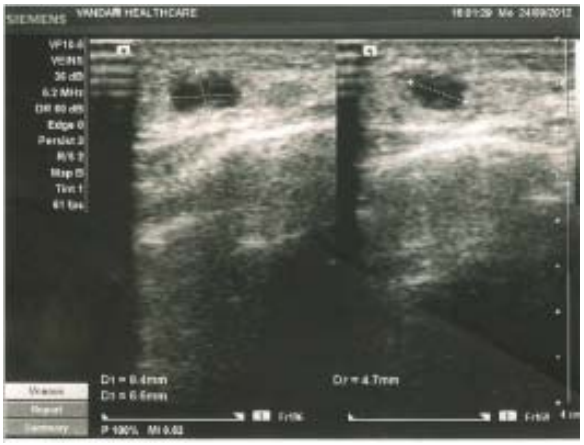

Fig 3: USG Image

DISCUSSION

Cysticercosis is the form of Taeniasis that occurs due to infestation of soft tissues with larval forms of the parasite *Taenia solium* when man acts as the intermediate host rather than the definitive host [1]. Man can become the intermediate host through eating vegetables or water contaminated with *Taenia* eggs [8], though this represents a dead end for the parasite. The ingested eggs and larvae distribute throughout the human body in the same fashion as they do in the pig. The commonest form of extraintestinal taeniasis is neurocysticercosis [1]. Extraneural cysticercosis is also fairly common in Asia and Africa [1] and presents with or without neural involvement. It is much less common than neurocysticercosis [14]. It mostly presents as small, isolated, movable, painless nodules on the trunk and arms. The nodules may become tender and inflamed and eventually disappear over months to years [1]. The parasitic load in muscular cysticercosis may be so large as to induce limb hypertrophy, a condition labeled muscular pseudo hypertrophy [1, 15, 16]. Ocular [1, 2] and intracardiac [1] cysts have also been described. Rarer forms of cysticercosis include thyroid gland [17], mesenteric lymph nodal cysticercosis [18], and pharyngeal involvement [19]. Rarely very large cysts mimicking tumors have been reported [20].

The frequency of autoinfection with intestinal *Taenia solium* is estimated to be 15-25% [1, 21]. The carrier in many other cases resides in the household members [1]. The diagnosis of co-infection with intestinal *Taenia solium* or *T. saginata* is difficult as stool microscopy has a poor sensitivity missing up to 60-70% of cases, and coproantigen detection ELISA is not readily available. The ELISA has a much better specificity (99%) and sensitivity (95%) [1]. Autoinfection is important as the adult worm residing in the intestine can be the source of the cysticerci. In our series, autoinfection was treated with empiric oral nitazoxanide[22, 23]. Niclosamide and praziquantel are preferred agents [1], but could not be used as they were not available locally.

The diagnosis of subcutaneous or muscular cysticercosis can be definitively made with a combination of high frequency ultrasound [9, 10, 11, 12, 13] and clinical examination. The differential diagnoses include tubercular lymphadenopathy, lymphoma, neurofibroma, and lipoma [24, 25]. It may be noted that all differential diagnoses mentioned in literature are solid in nature, and a USG should suffice to differentiate the cystic nature of the lumps. USG is fairly specific in subcutaneous cysticercosis and in our experience is an important tool in the diagnosis of disseminated or isolated subcutaneous cysticercosis. The findings on USG include a cyst with or without a visible scolex, which may be accompanied by perilesional edema or abscesses [9, 10], all of which are readily identifiable on a high resolution ultrasound. In our series, FNAC was performed in six of seven cases but was diagnostic in two cases only. Other authors, however, have concluded that FNAC is diagnostic [1, 5, 6, 7, 8]. In a series from the KyungHee Medical Center in Korea, parasitic diseases were identified in about 0.07% of the surgical specimens submitted, of which a majority were cysticerca [26]. Biopsy, in our opinion, is rarely required, and mostly resorted to only when expectant management fails and surgical excision is required [1, 3]. In one of our cases, a differential diagnosis of subcutaneous cysticercosis was not considered preoperatively. The final diagnosis was reached only after surgical excision and histopathological examination of the cyst.

The treatment of subcutaneous or intramuscular cysticercosis is expectant [1]. Autoinfection with adult *Taenia* should be suspected in all patients. In the absence of readily available and sensitive tests to identify autoinfection, the treatment for autoinfection should be extended in all patients [1]. In addition screening of household members is also recommended for diagnosing the

source of infection [1]. The drugs recommended for the treatment of intestinal taeniasis are niclosamide (single 2 gm dose) or praziquantel (5-10 mg/kg single dose) [1], though both are very difficult to source. More readily available alternative agent is nitazoxanide [22, 23]. The treatment of cysticerca requires albendazole, though controversial, as is typified in the treatment of neural cysticercosis [1, 2, 27]. Albendazole is not required in extra-neural cysticercosis [1, 28]. Surgical excision should be reserved for patients in whom the diagnosis cannot be established, the patient demands an exact histological diagnosis or surgical excision, the cyst gets infected, or does not resolve with expectant management.

In conclusion, with heightened clinician awareness of the existence of isolated subcutaneous cysticercosis in patients with close animal contact, and the widespread availability of high resolution ultrasound and FNAC, subcutaneous cysticercosis can be diagnosed readily. Surgery, either with a therapeutic or diagnostic intent, can be avoided in the great majority of these patients, as the cysts mostly resolve on their own, and do not require any specific treatment. Identification and treatment of autoinfection, and of another source of infection in the household is equally important.

REFERENCES

1. Garcia HH, Gonzalez AA, Evans C AW, Gilman RH. Taenia Solium cysticercosis. Lancet 2003;361:547-56.
2. Bhalla A, Sood A, Sachdev A, Varma V. Disseminated cysticercosis: a case report and review of literature. Journal of Medical Case Reports 2008;2:137
3. Agrawal R. Soft tissue cysticercosis: Study of 21 cases. J of Clinical and Diagnostic Research. 2012;6:1669-1671.
4. Sahai K, Kapila K, Verma K. Parasites in fine needle breast aspirates –assessment of host tissue response. Postgrad Med J 2002;78:165-7.
5. Lakhey M, Hirachand S, Akhter J, Thapa B. Cysticerci in palpable nodules diagnosed on fine needle aspiration cytology. J Nepal Med Assoc. 2009;48(176):314-7.
6. Cytomorphological spectrum of subcutaneous and intramuscular cysticercosis: A study of 22 cases. Meenu Gill, Shivani Dua, PS Gill, Veena Gupta, Sumiti Gupta, Rajiv Sen. J Cytol. 2010 October; 27(4): 123–126.
7. Suchita S, Vani K, Sunila R, Manjunath GV. Fine needle aspiration cytology of cysticercosis – A case report. Case Reports in Infectious Disease Volume 2012 Article ID 854704, 2 pages, 2012. doi:10.1155/2012/854704.
8. Sawhney M, Agarwal S. Cysticercosis: Hooked by a Hooklet on Fine Needle Aspiration Cytology—A Case Report. Case Reports in Infectious Diseases, vol. 2013, Article ID 315834, 2 pages, 2013. doi:10.1155/2013/315834
9. Mittal A, Gupta S, Gupta S, Mehta V. Subcutaneous and intramuscular cysticercosis: High resolution sonography. Indian J Dematol Venereol Leprol 2009;75:515-6.
10. Vijayaraghavan SB. Sonographic appearances in cysticercosis. J Ultrasound Med 2004;23:423-7.
11. Sharma P, Neupane S, Shrestha M, Dwivedi R, Paudel K. An ultrasonologic evaluation of solitary muscular and subcutaneous cysticercosis. Kathmandu Univ Med J. 2010;8(30):257-60.
12. Asrani A, Morani A. Primary sonographic diagnosis of disseminated muscular cysticercosis. J Ultrasound Med 2004; 23: 1245-8.
13. Mittal A, Gupta S, Mehta V, Gupta R. Anterior abdominal wall cysticercosis—the role of high resolution USG. Indian J Radiol Imaging 2008; 18(3): 266-7.
14. Singrodia S, Joshi RG, Solanki RB, Rawal RC. Neurocysticercosis with subcutaneous nodules. Indian J Dermatol Venereol Leprol 2009;75:524-525.
15. Bandyopadhyay D, Sumit S. Disseminated cysticercosis with huge muscle hypertrophy. Indian J Dermatol. 2009;54(1):49-51.
16. Chopra JS, Nand N, Jain K, Mittal R, Abrol L. Generalised muscular pseudohyperptrophy in cysticercosis. Postgrad Med J. 1986;62:299-300.
17. Gupta S, Sodhani P. Clinically unsuspected thyroid involvement in cysticercosis: a case report. Acta cytol. 2010;54(5 suppl.):853-6.
18. Mohan H, Bal A, Aulakh R. Multiple cysticerca as an unusual cause of mesenteric lymph node enlargement: a case report. J of Med Case Reports. 2008;2:196.
19. Sobnach S, Khosa SA, Pather S, Longhurst S, Kahn D, Raubenheimer PJ. First case report of pharyngeal cysticercosis. Trans R Soc Trop Med Hyg. 2009;103 (2):206-8.
20. Bangal V, Tayade S, Kwatra A. Rare case of cysticercosis of rectus abdominis muscle presenting as pelvi-abdominal lump during puerperium. J Mahatma Gandhi Inst Med Sci. 2010,15(2),68-71 .
21. Gilman RH, Del Brutto OH, Garcia HH, Martinez M. Prevalence of taeniasis among patients with neurocysticercosis is related to severity of infection. Neurology 2000;55:1062.
22. Lateef M[1], Zargar SA, Khan AR, Nazir M, Shoukat A. Successful treatment of niclosamide- and praziquantel-resistant beef tapeworm infection with nitazoxanide. Int J Infect Dis. 2008 Jan;12(1):80-2.
23. Rossignol JF, Maisonneuve H. Nitazoxanide in the treatment of *Taenia saginata* and *Hymenolepis nana* infections.Am J Trop Med Hyg. 1984 May;33(3):511-2.
24. Rao RN, Krishnani N, Malhotra K, Suresh B, Mehrotra R. Dilemmas is cytodiagnosis of subcutaneous swellings: mimics and lookalikes of cysticercosis. J Clin Pathol 2010; 63(10):926-9.
25. Shah I. Subcutaneous cervical cysticercosis in a child. J Glob Infect Dis 2012;4(2):135.
26. Choi WH, Chu JP, Jiang M, Lee YS, Kim BS, Kim DG, Park YK. Analysis of parasitic diseases diagnosed by tissue biopsy at the KyungHee Medical Center 1984-2005 in Seoul, Korea. Korean J Parasitol; 48(1):85-88.
27. Rajbhandari KC. Therapeutic Efficacy of the cysticide albendazole in neurocysticercosis. http://www.healthnet.org.np/article/neurology/theraptic.html
28. Sinha S, TIwari A, Sarin Y K, Khurana N. Isolated soft tissue cysticercosis involving the trunk in children: report of 4 cases. ASPS J Case Rep 2013;4(3):35.