

Presentation and management of hydatid cyst of the thigh: A systematic review of literature

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ABSTRACT

Hydatid cyst of the thigh is an extremely rare variant of the condition. The aim of this study is to systematically review the reported cases of hydatid cyst of the thigh. Science Direct, Web of Science, Medline on OVID, PubMed and Google scholar were scrutinized for articles which at least contain one new case of hydatid cyst of the thigh. Sixty-four patients were collected and discussed. In conclusion, hydatid cyst of the thigh is a very rare parasitic manifestation presenting as a painless swelling, diagnosed typically by magnetic resonance imaging and managed with en bloc resection.

Keywords: Hydatid cyst, Parasite, Thigh

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INTRODUCTION

Echinococcus, a cestode of the Taeniidae family although lives outside the human body, causes a cystic parasitic infestation in mankind [1, 2]. The organism possesses both definitive and intermediate hosts. Wolves, dogs, and foxes (the definitive hosts) pass the worm's eggs in their excretes into the surroundings, subsequently, the intermediate hosts (sheep, cattle, humans, horses, goats, and camel) develop hydatidosis [3]. The organs which are most incessantly affected by the disease are liver and lung, the latter is more common in the pediatric age group [4]. Other seldom affected organs are skeletal and smooth muscles, bone, viscera, and mediastinum [5]. Hydatid cysts (HCs) of the thigh is a very rare variant of the parasite manifestation, still, it occurs in endemic areas of the Middle East [6]. Presentation of the thigh HC, epidemiology, its clinical courses, diagnosis, and management are not well described. As there is no summary paper about thigh HC, this study was attempted to systematically review the reported cases of HC of the thigh worldwide.

METHODS

Search and information sources

Science direct, Web of Science, Medline on OVID, PubMed, Elsevier, Scopus, Wiley online and Google scholar were scrutinized for English-language articles distributed before 2018. The keywords were thigh hydatid cyst, thigh hydatidosis, hydatid cyst of the thigh, Echinococcosis of thigh. The data assemblage was enhanced by references from the included articles (Figure 1).

Eligibility criteria

For an article to be considered for this review, it had to include at least one new case of HC of the thigh. Exclusion criteria was articles without atleast one new case of HC of the thigh.

Data assemblage and review process

Data were excerpted from the included studies by two authors independently (second and fourth authors). Authors of the included articles have not been contacted to get and confirm the data. Several data were quoted and some of them were pooled including socio-demographic characteristics of the patients, sample size, presentations, duration of presentation, the method of diagnosis, options for management, recurrence rate and complications.

Summary measures and synthesis of results

Some data were calculated and re-analyzed. According to the variables, they were demonstrated as the percentage, mean values, and ranges of variation

and percentages. Some other papers were illustrated and summarized in brief.

RESULTS

The search through literature found 70 papers. From which 20 papers were excluded by titles. Other three papers were ostracized either because of scanty information or incomplete report. Two papers were omitted as a result of failure to be retrieved. The remaining 45 papers were analyzed and 64 patients were collected (Table 1). Thirty-seven of them (57.8%) were females, 27 (43.2%) were males. The age of the patients ranged from eight to 83 years with a mean age of 32.6 years. Almost all the patient presented with slow-growing mass. One of the patients presented with signs and symptoms of abscess (erythema, tender and swelling) [7]. The duration of the presentation was variable ranging from three days to 30 years with a mean period of the presentation was two years. The left side was affected more commonly than the right side (41/58, 70.6%). All patients received antihelminthics from the time of diagnosis (pre or postoperatively) until a variable length of time. All of the cases were managed by total resection. Three of the patients (4%) had HC other than thigh HC [8–10]. Two of them had retroperitoneal HCs another case had liver HC.

DISCUSSION

The prevalence of thigh HCs is not notorious. Apart from two case series studies, all cases of thigh HCs accessible in the literature are case reports [11, 12]. Ammari and his colleagues published the registry of 13 years with nine cases of thigh HCs, seven patients were female, their mean age was 37 years [12]. Madhar et al presented seven cases of thigh HCs in a solitary paper from which six patients were female and their ages ranged from 19 to 56 years with a mean age of 30 years [11]. Thigh HC is either primary or secondary. In the latter, HCs should disturb concomitantly at least one of the common sites like liver, lung or spleen. Acu and his associates disclosed a case of thigh HC with concomitant HC of the left lobe of the liver occurring in a 20-year-female patient [8]. Sarda and his companions reported a 60-year-female with secondary HC of the thigh, the patient presented with swelling of the thigh with a palpable mass in the suprapubic region, during operation, they realized that there were two HCs. One of the thigh and other in the retroperitoneum communicating with each other via sub-facial plane [9]. In endemic countries, the disease might be prevented by several mechanisms including regular supplementation of praziquantel to the hosts, preventing dog's access to intermediate hosts, vaccinating sheep, boiling or safe disposal of offal [13]. Up to date, 63 patients of thigh HCs have been reported in the medical literature [6–53] The age of the cases ranged from 8 to 83 years [4, 23].

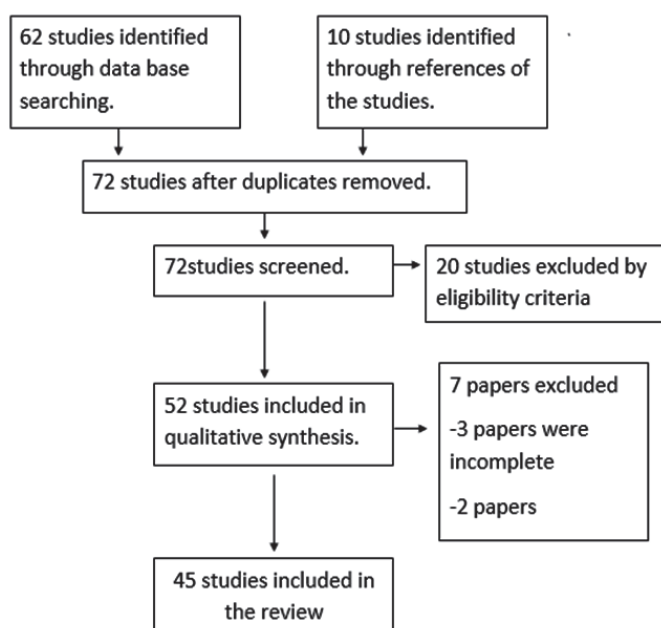


Figure 1: Flow chart of the included articles.

Table 1: Sociodemographic and clinical summary of the included articles

Authors	Country	Age/sex	Presentation	Sites	Other HCs	Durations	Medication/ duration	Surgery
A.A. Abebe	Ethiopia	25 Yr/F	Cystic swelling	Left thigh	Not mentioned	2 years	Not mentioned	Not mentioned
Leyla Acu	Turkey	20 Yr/F	mass	Right thigh	Liver	Not mentioned	Anthelmintic therapy for 3 months	Surgery
Nicolas Argy MD	Turkey lived in France for 20 years	60 Yr/F	swelling	Right thigh	Not present	six months	400 mg of albendazole per day for 10 days before and three weeks after surgery	Surgery nine months after the first operation, a second pericystectomy done
Muhammet Arslan	Turkey	32 Yr/M	Mass	Thigh??	Not mentioned	18 months	Albendazole (10 mg/kg/day) duration not obvious	All remaining components of the mass were evacuated
Rajesh Kumar Bansawal	India	60 Yr/F	Large soft tissue swelling	Medial aspect of left thigh	Not present	30 years	Not mentioned	Surgery
SaphaBarkati	Canada	57 Yr/ worker	Painful erythematous 7 × 6 cm mass	Lateral aspect of the right thigh	Not present	Not mentioned	Prolonged albendazole treatment	Complete surgical resection
KA Bothale	India	70 Yr/F	Slowly growing mass	Posterior aspect of left thigh	Not present	2 years	Not mentioned	Surgical excision
S. Bouomrani	Tunisia	34 Yr/ patient	Smooth Tumefaction	Left thigh	Not present	6 months	Anthelmintic drug	Radical surgery
Xavier Chevallier	Portugal lived in France for 20 years	40 Yr/M	Slow-growing, slightly tender mass	Posterior aspect of the left thigh	Not mentioned	Not mentioned	The patient was then given 400 mg of albendazole daily for 2 (after Surgery)	Surgical excision
AbuzerDirican 1st case	Turkey	64 Yr/M farmer	Swelling	Right medial thigh	Not mentioned	1 year	Not mentioned	Surgical excision
AbuzerDirican 2nd case	Turkey	67 Yr/M farmer	Swelling	Left palm	Not present	1 year	Not mentioned	Surgical excision
FuatDuygulu	Turkey	8 Yr/F	Slow-growing, painless mass	Left thigh	Not present	Not mentioned	Albendazole therapy, 200 mg twice daily, was given for six weeks (after Surgery)	Surgical excision
Lorenzo Garagnani	Italy	59 Yr/M	Chronic localized pain and swelling thigh	Over the root of his left thigh	Not present	8 years	Three courses of Albendazole at a dose of 800 mg/day (400 mg twice daily) were administered (after Surgery)	Surgical excision
Arjun Goel	India	23 Yr/M farmer	Lump	Medial aspect of left thigh	Not present	1 year	Albendazole (400 mg bid) was started two weeks preoperatively The course of albendazole was continued postoperatively	Surgical excision
Gupta A	India	38 Yr/M	Swelling	Medial aspect of the right thigh	Not present	10 months	Albendazole at a dose of 400 mg (after Surgery)	Surgical excision

Table 1: (Continued)

Authors	Country	Age/sex	Presentation	Sites	Other HCs	Durations	Medication/ duration	Surgery
SaadY.Ibrahim	Iraq	35 Yr/M	Slowgrowing, painful lump	Medial aspect of the mid third of the left thigh	Not mentioned	6 months	Patient was put on medical treatment for one month (after Surgery)	Local Journal
ZehraKazmi	Pakistan	23 Yr/M farmer	Swelling	Medial aspect of the left thigh	Not present	5 months	Not mentioned	Surgical excision
ZaferKoc	Turkey	23 Yr/M	Swelling and pain	Left thigh	Not present	6 months	Oral anthelmintic therapy consisting of albendazole (400 mg, twice daily) for 3 months	Surgical excision
A. Kocakusak	Turkey	37 Yr/M	Cystic mass	Left thigh	Not present	4 years	Postoperatively he underwent Albendazole treatment	Surgical excision
ManellLandolisi	Tunisia	27 Yr/M	Swelling	Right thigh	Not present	6 months	Not mentioned	Surgical excision
Mohamed Madhar	7 cases from Marrakech, Maroc	average age is 30 six women and one man	Soft-tissue swelling	Adductor compartment for 4 cases and in the anterior compartment for 3 cases. All localizations were to the left.	Not present	1 year on average	Not clear	Not clear
Abdelhalim Mahmoudi	Morocco	14 Yr/F farmer	Swelling	Distal medial left thigh	Not present	2 years	Not mentioned	Surgical excision
Sanjay Marwah	India	11 Yr/F	Progressively increasing swelling	Right thigh	Not present	1 year	(Albendazole). three weeks pre Op. and six weeks post Op.	Surgical excision
Omar Mourafiq	Morocco	Middle age/F	Gradually growing mass	Left thigh	Not present	8 months	Albendazole (400 mg/day) for two months postoperatively	Surgical excision
Tarun Kumar Pathak	India	30 Yr/F	Smooth, large swelling	Anteromedial side of her right thigh	Not present	4 year	Oral Albendazole (10-15 mgm/kg/day) for 4 weeks pre Op. and 1 month post Op.	Surgical excision
InderPawar	India	26 Yr/F	Large firm to hard painless lump	Anteromedial part of left thigh	Not mentioned	14 months	Albendazole 10 mg/kg per day for three months post Op.	Surgical excision
HARRY PERELMAN	U.S.	61 Yr/M	Large, hard, painless lump	Right upper thigh	Not present	4-6 months	Not mentioned	Surgical excision
Rehana Shaikh	Pakistan	40 Yr/F	Very slowly growing mass	Medial aspect of left thigh	Not present	2 years	Pre Op. anthelmintic therapy given, post-surgical period was uneventful	Surgical excision
Gabriel Rodrigues	India	62 Yr/F	Swelling	Anterior aspect of the right thigh	Not mentioned	7 years	Preoperative albendazole 400 mg twice daily for 4 weeks, Postoperatively, for 3 months	Surgical excision
George S. Abi Saad	Lebanon	53 Yr/F	Painless, progressively enlarging mass	Medial aspect of right thigh	Not mentioned	4 month	The patient was maintained on albendazole at a dose of 400 mg twice a day for a period of 6 weeks post operatively	Surgical excision

Table 1: (Continued)

Authors	Country	Age/sex	Presentation	Sites	Other HCs	Durations	Medication/ duration	Surgery
Guiseppe Salamone	Italy	68 Yr/F	Mass	Upper medial side of right thigh	Not present	12 years	Preoperative albendazole 400 mg daily for 2 weeks, postoperative albendazole 400 mg daily for 2 weeks,	Surgical excision
Dinesh K. Sarda	India	60 Yr/F	Swelling	Left thigh since	Abdominally retroperitoneal HC, there was connection HC of thigh and retroperitoneal one	3 months	Albendazole for three months post operatively	Surgical excision
Aycan Kaylkcioğlu	Turkey	69 Yr/F	Painful mass	Right midthigh	Not present	6 months	Mebendazole for 6 months 40mg/kg/day post operatively	Surgical excision
Karin Thursky	Greek man lived in Australia	63 Yr/M farmer	Painful swelling	Left thigh	Not mentioned	3 days	1 week of albendazole therapy pre Op. and Treatment with albendazole at 400 mg b.i.d. was immediately commenced postoperatively But The appearance was thought to be consistent with disseminated hydatid cysts. Chemotherapy was changed to therapy with continuous albendazole and intermittent praziquantel at 40 mg/kg that alternated between 1 week on and 1 week off. After completing 3 cycles of praziquantel with 3 months of albendazole treatment, the patient made an uneventful recovery	Surgical excision
Gaetano La Greca	Italy	46 Yr/M	Mildly painful mass	Right groin and thigh, diffuse edema involving the right leg	The involvement of the psoas muscle and retroperitoneum from the Morrison's pouch up to the right iliac fossa and to the iliac region is evident	2 weeks	Not mentioned	muscle-sparing approach
Cissé AM	Not mentioned	59 Yr/F	Progressive diffuse painless swelling	Left thigh	Not mentioned	1 year	Not mentioned	Surgical management
Farzam	Zanjan	23 Yr/F	Mass	Right thigh	Not mentioned	3 years	Not mentioned	operation
S.M. Javad Mortazavi	Not mentioned	16 Yr/F	Swelling	Left thigh	Not mentioned	12 months	Albendazole (10 mg/kg) was administered for 2 weeks	Excision
Sarda	repeated							

Table 1: (Continued)

Authors	Country	Age/sex	Presentation	Sites	Other HCs	Durations	Medication/ duration	Surgery
Ammari T. 9 cases	Not mentioned	Mean 37 Y/ all were female	Isolated tumefaction	Thigh Lt. 7/9	Not mentioned	12 months average	Not mentioned	En bloc resection of the HC with peripheral muscle tissue in 6 cases and subtotal pericystectomy in 2
Bagatur AE	Not mentioned	83 Yr/M	Giant lesion	Posterior aspect of the right thigh	Not mentioned	Not mentioned	Not mentioned	Excision
Atmatzidis K	Not mentioned	73 Yr/F	Painless mass	Thigh	Not present	Not mentioned	Oral albendazole for six months postoperatively	Excision
Sipahioğlu S	Not mentioned	24 Yr/M	Not mentioned	Medial thigh	Not mentioned	Not mentioned	Anthelmintic drug	Excision
Hammami T 3 cases	Not mentioned	Not mentioned	Soft tissue mass	Thigh muscles	Not mentioned	Not mentioned	Not mentioned	Total pericystectomy was performed in all cases
Ekinci Y	Not mentioned	64 Yr/M	Not mentioned	Left anterolateral aspect of the thigh	Not mentioned	Not mentioned	Pre and post surgical medical Rx.	Surgical Rx.
Sakka SA	Not mentioned	Not mentioned	Not mentioned	Thigh	Not mentioned	Not mentioned	Not mentioned	Not mentioned
Ghoroobi	Not mentioned	child	Not mentioned	Left thigh	Not mentioned	Not mentioned	Not mentioned	Not mentioned
Sushma Yalavarthi	India	75 Yr/M	Painless swelling	Posterior aspect of the left thigh	Not mentioned	2 year	Not mentioned	Not mentioned
Lotfi Saeed Reza	Iran	69 Yr/f	Mass	Right inguina	Not mentioned	10 months	Albendazol was administered postoperatively	Excision

The particular characteristic of HCs in any organ of the body is that the cyst can remain silently for a long period of time without inducing a specific sign or symptom. Regarding thigh HC, it is either diagnosed incidentally or cause pressure symptoms [14]. Bansiwali and his comrades reported a 60-year-old female presented with slowly growing mass in the medial aspect of the thigh for 30-year duration, investigations confirmed thigh HC [15].

Preoperative diagnosis of thigh HCs is crucial. Specificity and sensitivity of serological investigations depend on the type of HCs, these are positive in 90% of hepatic HCs while negative in the majority of HCs in other parts of the body. The accuracy of ultrasound reaches 100% in typical cases of HCs. A better details regarding number, site, size, and architecture of the cysts also their relationship with the neighboring structures can be taken from computed tomography scan (CT scan). However, in HCs of atypical areas like subcutaneous or muscular hydatidosis, magnetic resonance imaging (MRI) is preferred because it gives more information about the mass and its surrounding soft tissue structure [6]. Almost all cases of thigh HCs were diagnosed by ultrasound and some confirmed by MRI. Apart from calcification, all features of HC can be visualized by MRI. The features include multivesicularity with or without rim sign (hypodense peripheral ring), cysts show high-signal intensity on T2 weighted images and low signal intensity on T1- weighted images [6–53]. Definitive management strategy is total surgical resection [15]. All reported cases were treated by complete surgical resection. Preoperative anthelmintics have given to those patients who were diagnosed before the intervention. Medication was continuous after surgery for the variable duration of times ranging from three days to six months [6–53]. Kayikcioglu and his teammates put their patient on 40 mg/kg/day of mebendazole for six months after complete surgical excision, while Salamone et al preferred 400 mg/day of albendazole after operation for two weeks [16, 17]. Specific criteria could not be found regarding choice of Antihelminthic drugs in the literature, while generally, albendazole preferred over others [14, 15, 23].

CONCLUSION

Hydatid cyst of the thigh is a very rare parasitic manifestation presenting as a painless swelling, diagnosed typically by MRI and managed with en bloc resection.

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Author Contributions

Abdulwahid M. Salih – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
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All relevant data are within the paper and its Supporting Information files.

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