



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Solitary hydatid cyst in the forearm: A case report

Raheef Alatassi^{a,*}, Saeed Koaban^a, Meshari Alshayie^a, Ismail Almogbil^b^a Security Forces Hospital, Department of Orthopedic Surgery, Riyadh, Saudi Arabia^b King Saud Medical City, Department of Orthopaedic Surgery, Riyadh, Saudi Arabia

ARTICLE INFO

Article history:

Received 12 August 2018

Received in revised form

14 September 2018

Accepted 17 September 2018

Available online 27 September 2018

Keywords:

Echinococcosis

Forearm

Flexors

Hydatid cyst

Muscle hydatidosis

Case report

ABSTRACT

INTRODUCTION: Hydatid cyst is a parasitic disease caused by *Echinococcus* that is mostly found in the Mediterranean and Gulf States regions. Hydatid cysts usually arise in the liver and lungs but rarely occur in the forearm.

PRESENTATION OF CASE: In this report, we present a rare case of a solitary intramuscular forearm hydatid cyst in a 32-year-old woman. The cyst exhibited positive radiological findings and was treated surgically, leading to a complete resolution of the patient's symptoms with no history of recurrence.

DISCUSSION: This case was unique because although the patient presented in her fourth decade of life and lived in a Gulf country, the hydatid cyst was in a solitary and unusual location and had been present for one year. It had all positive findings in a radiological investigation without any signs of systemic illness. Additionally, the cyst was attached to the median nerve and a complete excision was performed without affecting the nerve.

CONCLUSION: By reporting this rare case of a solitary intramuscular forearm hydatid cyst, we aim to increase the awareness of unusual sites for the appearance of hydatid cysts.

© 2018 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Hydatid cyst is a parasitic disease caused by *Echinococcus*. It is considered one of the five most diagnosed zoonotic diseases in the Mediterranean and Gulf States regions. The most widespread species is *Echinococcus granulosus*, which usually causes a unilocular hydatid cyst. Several animals are considered definite hosts, such as dogs and wolves, or as intermediate hosts, such as sheep and cattle [1–4].

A hydatid cyst can develop in almost any part or organ of the body; however, the most commonly affected organs are the liver and lungs. Concurrent involvement of both organs is reported at 5%–13% [4–6]. Primary muscular hydatid cysts in the absence of liver, lung, or bone involvement encompass <0.7%–3% of these cases. These types are particularly rare and are most commonly found in the trunk and lower extremity musculature. Hydatid cysts in the upper extremities are even rarer and only a few cases have been reported in the literature [3–10].

Herein we report a rare case of an unusual solitary intramuscular forearm hydatid cyst in a 32-year-old woman. In this case, surgical treatment led to a complete resolution of the patient's symptoms. The current case report was written according to the recently pub-

lished SCARE criteria [11], which are used to support transparency and accuracy in the publication of case reports.

2. Presentation of case

A 32-year-old female patient, a Gulf States national, presented to our hospital with a history of a right forearm painless mass that had appeared 1 year prior. The mass had slowly grown over the past year without affecting the range of motion in her elbow. She resides in an urban area without any history of contact with sheep or cattle. Moreover, she had no history of any chronic medical illnesses.

Upon examination, she had an obvious firm mass on the volar aspect of her mid-forearm that was approximately 8 cm distal to the cubital fossa and measuring approximately 6 × 3 × 3 cm. There was no tenderness over the mass. The mass was mobile only in the horizontal plane and decreased in size with the contraction of the flexor muscles of the forearm. Laboratory tests including antibodies against *Echinococcus* and a computed tomography (CT) scan of the thorax and abdomen were all unremarkable.

A lateral plain radiograph revealed a well-defined benign lesion with very thin calcified wall containing a multilocular mass mixed with coarse trabeculae that was not in contact with the bone in the soft tissue of the right forearm (Fig. 1). An ultrasound showed multilocular soft tissue cystic lesions (multiple daughter cysts) (Fig. 2). A CT scan without contrast and an MRI of the right forearm revealed a well-defined hyperdense capsule with a multilocular cystic lesion

* Corresponding author at: P.O. Box: 3643, Riyadh, 11481, Saudi Arabia.
E-mail addresses: raheef332@hotmail.com, ralatassi@med.sfh.sa (R. Alatassi).



Fig. 1. Lateral radiographs of the right forearm showing a well-defined benign lesion in the soft tissue.

(multiple daughter cysts) and there were no signs of bone destruction (Fig. 3). T1- and T2-weighted images revealed a hypointense and hyperintense multilocular cyst with a low-intensity rim, suggestive of an intramuscular hydatid cyst (Fig. 4a) (Fig. 4b). The patient was advised to take albendazole for 8 weeks to be followed by possible surgery after 2 months. At the 2-month follow-up visit, the size of the mass had been reduced clinically and the patient was admitted to the hospital for its elective excision.

During the operation, the skin was incised longitudinally over the mass along the volar aspect of the right forearm. A blunt dissection was performed; the mass was identified below the flexor digitorum superficialis muscle and adherent to the belly of the flexor digitorum profundus muscle (Fig. 5). Moreover, the inferior surface of the cyst was adherent to the median nerve. The whole mass was excised in an en bloc manner without rupturing the cyst or damaging the median nerve (Fig. 6). The mass was sent for histopathology, which revealed scoleces, daughter cysts, and degeneration of the adjacent muscle fibers.

The patient's postoperative course was unremarkable. A complete resolution of the swelling was confirmed clinically. In addition, no nerve injury was documented. At the 2-year postoperative follow-up, she remained well, with full elbow mobility, no documented recurrence of symptoms, and no median nerve injury.

3. Discussion

Echinococcosis is a parasitic disease of tapeworms of the *Echinococcus* type. The two main types of the disease are cystic echinococcosis and alveolar echinococcosis. Humans are intermediate hosts in the parasite life cycle when they ingest eggs via contaminated food or water [12,13]. The ingested parasite ova enter the portal system, penetrating the intestinal wall and becoming entrapped in the hepatic sinusoids. Some reach the lungs and sys-

temic circulation, passing through the liver, thus causing hydatid disease in other organs accounting for solitary cysts at uncommon sites. Haque et al. [14] and Bal et al. [4] suggested that the involvement of unusual primary sites of hydatid cysts can be explained by oncospheres that bypass the liver and lungs through precapillary anastomosis between pre- and post-parenchymal circulation. Moreover, primary striated muscle involvement is rare with a 3% rate due to high lactic acid levels and may occur primarily due to direct seeding of the oncospheres or secondarily via blood circulation.

This disease can present at any age, but the peak is between the third and fifth decades. The world's highest prevalence of the disease is found in the Mediterranean and Gulf countries [10]. Our patient is in her third decade and lives in one of the Gulf countries, which is a geographic region where the infestation is known to occur.

Primary muscular hydatid cysts are commonly asymptomatic and most patients may present only with a painless mass or vague symptoms. This explains the delay of diagnosis and the need for a high amount of suspicion with such presentation. A diagnosis cannot be obtained only by clinical presentation and further tools are needed to confirm the diagnosis. These include plain radiographs, taking into consideration that only 38% of patients will show soft tissue masses and calcification via plain radiography [15,16]. Ultrasonography is considered the gold standard and magnetic resonance imaging is more valuable in terms of confirming the diagnosis. Magnetic resonance imaging helps to identify the anatomic relation of the cyst and is useful to differentiate a hydatid cyst from other soft tissue malignancies. These lesions usually have high signal intensity on T2-weighted images and low intensity on T1-weighted images with a low-intensity rim on both T1- and T2-weighted images [4,10–18,19].

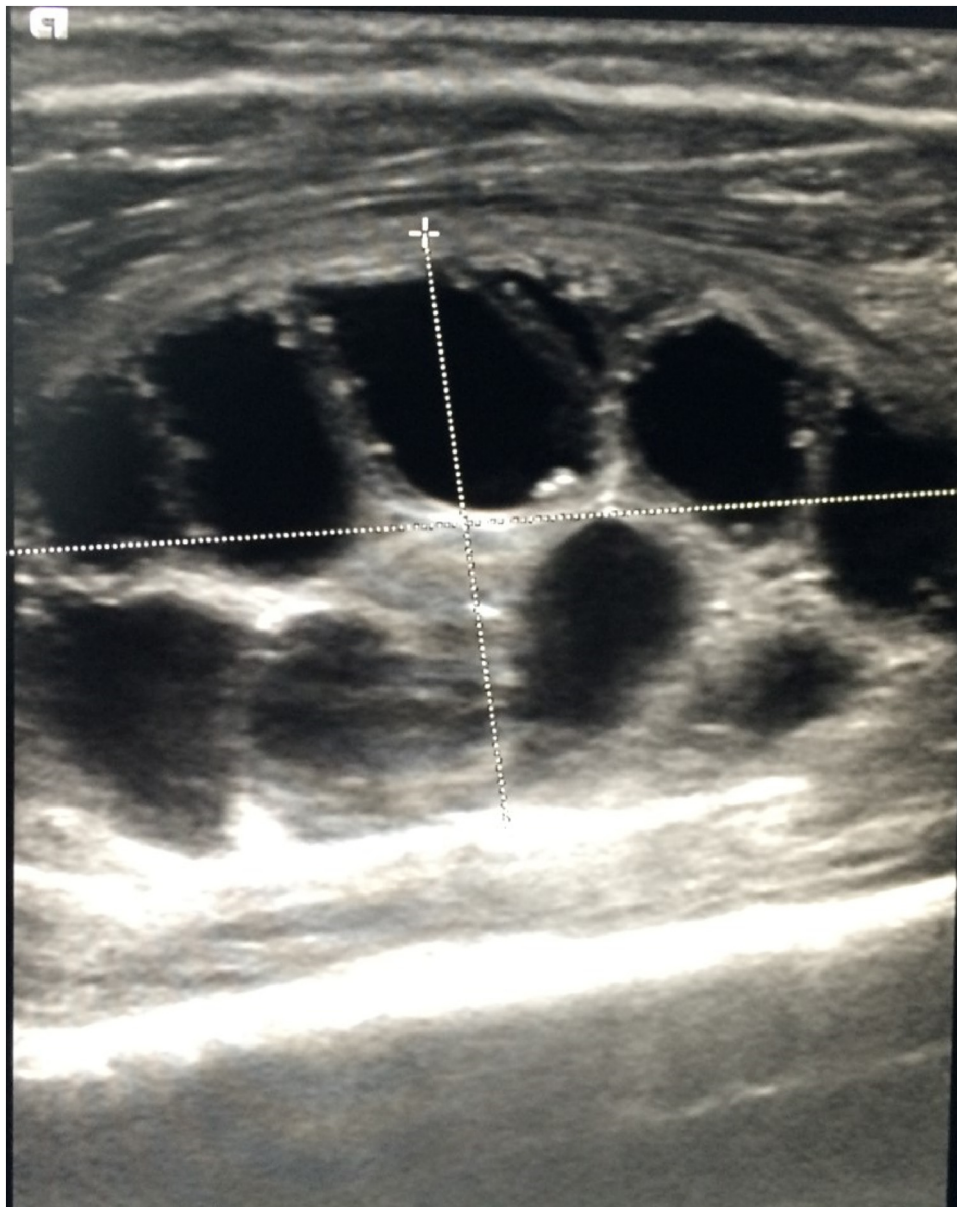


Fig. 2. Ultrasonography image revealing large multilocular soft tissue cystic lesions (multiple daughter cysts) of the right forearm lesion.

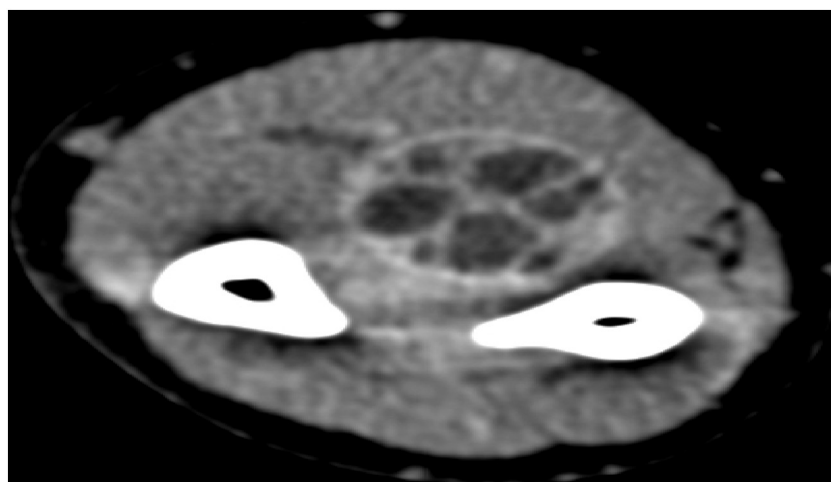


Fig. 3. Axial computed tomography scans of the right mid-forearm revealing a well-defined hyperdense capsule with multilocular cystic lesions (multiple daughter cysts) without any bony destruction.

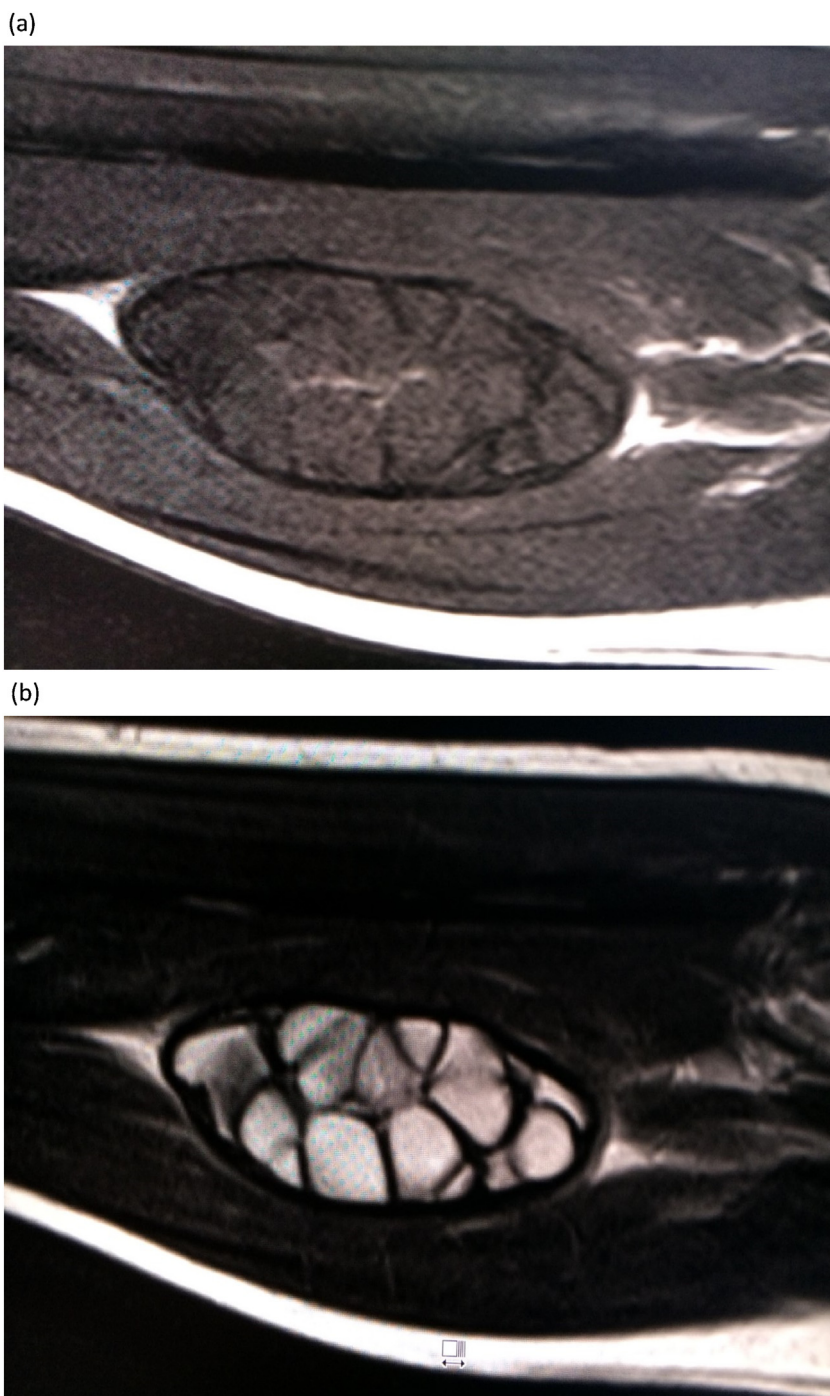


Fig. 4. (A) T1 and (B) T2 magnetic resonance imaging of the right mid-forearm revealing a hypointense and hyperintense multilocular cyst with a low-intensity rim suggestive of an intramuscular hydatid cyst in the middle third between the flexor digitorum profundus and the flexor digitorum superficialis.

Our patient noticed a painless swelling without any signs of systemic infection one year prior to presentation at our clinic. She had undergone all of the standard radiological investigations to diagnose the hydatid cyst including multiple daughter cysts, which are considered pathognomonic signs and are the most characteristic features of hydatid disease and imply viability.

The total excision of the cyst to avoid its rupture and spillage is the gold standard treatment. Preoperative planning is important to achieve curative management without complications. Such lesions must be completely removed while protecting the neurovascular structures. In our case, the cyst was adherent to the median nerve, so careful dissection and excision were applied, especially to the

median nerve. The mass was completely removed and the patient had a functioning nerve postoperatively.

We have presented herein a rare case of an unusual location of a solitary and intramuscular hydatid cyst in the forearm of a healthy 32-year-old. The cyst was treated surgically with complete resolution without any recurrence or complications. Very limited similar cases have been reported in the literature.

4. Conclusion

This case confirmed that a solitary hydatid cyst can be found in the musculoskeletal system without a history of systemic infection

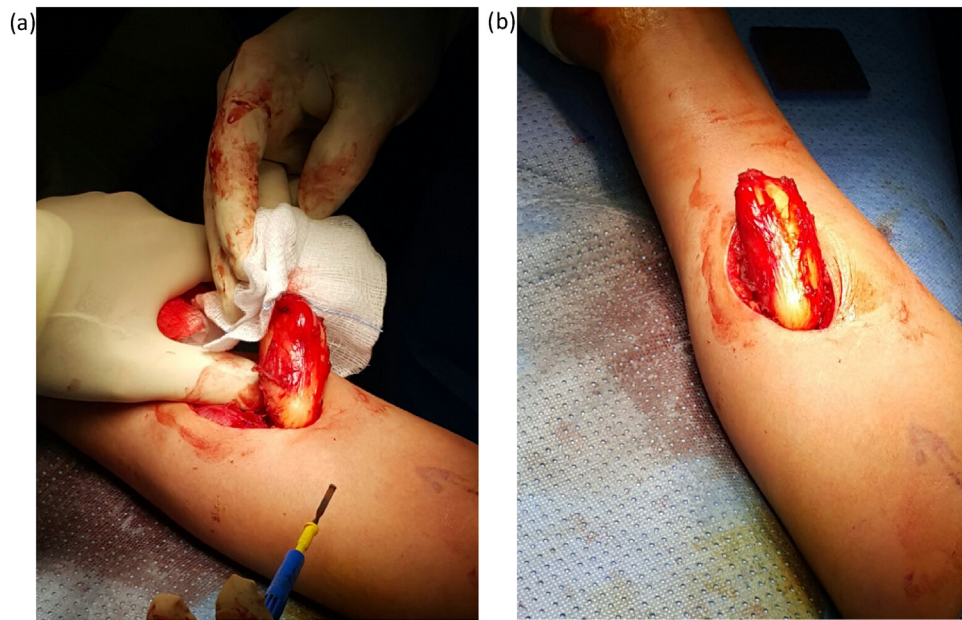


Fig. 5. (A) and (B) Intraoperative photographs of the exposed hydatid cyst.



Fig. 6. Intraoperative photograph of the hydatid cyst after excision.

or other similar cysts originating in the liver or lungs. This report aimed to increase the awareness of unusual sites of hydatid cysts and the importance of using radiological investigation to obtain a diagnosis and plan surgical treatment.

Conflicts of interest

The authors have no conflicts of interest to declare.

Funding

No specific grant from funding agencies in the public, commercial, or not-for-profit sectors was received for this work.

Ethical approval

We have reported a single case and ethical approval have been taken from our institution with valid reference number and without any conditions.

RN: 18 - 263- 28
 IRB: H-01-R-069

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

- Raheef Alatassi, surgeon: performed the literature review and data collection, designed the manuscript, wrote the manuscript and reviewed the final version of the manuscript.
- Saeed Koaban, surgeon: contributed to the manuscript writing.
- Meshari Alshayie, surgeon: contributed to the manuscript writing.
- Ismail Almogbil, surgeon, contributed to the manuscript writing.

Registration of research studies

We have reported a single case with no requirement for registry. This manuscript does not describe a clinical study.

Guarantor

Raheef Alatassi.

Provenance and peer review

Not commissioned, externally peer-reviewed.

References

- [1] A.K. Amir-Jahed, R. Fardin, A. Farzad, K. Bakshandeh, Clinical echinococcosis, *Ann. Surg.* 182 (November (5)) (1975) 541.
- [2] J. Prousalidis, K. Tzardinoglou, L. Sgouradis, C. Katsohis, H. Aletras, Uncommon sites of hydatid disease, *World J. Surg.* 22 (January (1)) (1998) 17–22.
- [3] N. Altintas, Past to present: echinococcosis in Turkey, *Acta Trop.* 85 (February (2)) (2003) 105–112.
- [4] N. Bal, N.E. Kocer, R. Arpacı, A. Ezer, F. Kayaselcuk, Uncommon locations of hydatid cyst, *Saudi Med. J.* 29 (7) (2008) 1004–1008.
- [5] K. Basarir, Y. Saglik, Y. Yildiz, T. Yetis, O. Cebesoy, Primary muscular hydatidosis mimicking soft tissue tumour: a report of five cases, *J. Orthop. Surg.* 16 (December (3)) (2008) 368–372.
- [6] F. Hekmatnia, M. Motiffard, H. Yazdi, A. Rizi, P. Hedayat, A.A. Hekmatnia, Large primary subcutaneous hydatid cyst in proximal thigh: an unusual localization, *Adv. Biomed. Res.* 7 (2018).
- [7] N. Argy, A. Abou Bacar, C. Boeri, C. Lohmann, A.W. Pfaff, Y. Hansmann, D. Christmann, E. Candolfi, N. Lefebvre, Primary musculoskeletal hydatid cyst of the thigh: diagnostic and curative challenge for an unusual localization, *Can. J. Infect. Dis. Med. Microbiol.* 24 (3) (2013) e99–101.
- [8] I.H. Bouraoui, O. Essid, H. Boughammoura, N. Arifa, R. Frikha, H. Jemni, K.T. Graiess, Forearm hydatid cyst: an unusual presentation, *East. Mediterr. Health J.* 17 (December (12)) (2011) 994.
- [9] M. Uzel, S. Aksu, E. Yorgancigil, M. Yildiz, A. Veral, I. Oztoprak, Skeletal muscle hydatidosis in the forearm: an unusual location, *Orthopedics* 29 (July (7)) (2006).
- [10] M. Bayram, A. Sirikci, Hydatid cyst located intermuscular area of the forearm: MR imaging findings, *Eur. J. Radiol.* 36 (December (3)) (2000) 130–132.
- [11] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, Orgill DP for the SCARE group, the SCARE statement: consensus-based surgical case report guidelines, *Int. J. Surg.* 34 (2016) 180–186.
- [12] J. Eckert, P. Deplazes, Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern, *Clin. Microbiol. Rev.* 17 (January (1)) (2004) 107–135.
- [13] S.E. Golzari, M. Sokouti, Pericyst: the outermost layer of hydatid cyst, *World J. Gastroenterol.* 20 (February (5)) (2014) 1377.
- [14] F. Haque, S.H. Harris, R. Khan, S.Z. Abbas, Primary hydatidosis of gluteus maximus, *J. Postgrad. Med.* 52 (October (4)) (2006) 300.
- [15] A. Combalia, S. Sastre-Solsona, Hydatid cyst of gluteus muscle. Two cases. Review of the literature, *Joint Bone Spine: Revue du Rhumatisme* 72 (October (5)) (2005) 430–432.
- [16] (a) M.Y. Booz, The value of plain film findings in hydatid disease of bone, *Clin. Radiol.* 47 (April (4)) (1993) 265–268;
(b) P. Taourel, B. Marty-Ane, S. Charasset, et al., Hydatid cyst of the liver: Comparison of CT and MRI, *J. Comput. Assist. Tomogr.* 17 (1993) 80–85.
- [17] A.I. Garcia-Diez, L.R. Mendoza, V.M. Villacampa, M. Cozar, M.I. Fuertes, MRI evaluation of soft tissue hydatid disease, *Eur. Radiol.* 10 (February (3)) (2000) 462–466.
- [18] M. Manisali, D. Ozaksoy, I. Kovanlikaya, The role of MR imaging in cardiac echinococcosis, *AJR Am. J. Roentgenol.* 168 (January (1)) (1997) 282–283.
- [19] G. Salamone, L. Licari, B. Randisi, N. Falco, R. Tutino, A. Vaglica, R. Gullo, C. Porrello, G. Cocorullo, G. Gulotta, Uncommon localizations of hydatid cyst. Review of the literature, *G. Chir.* 37 (July (4)) (2016) 180.

Open Access

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.