International Journal of Surgery Case Reports 103 (2023) 107867

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

Muscular hydatid cyst in Iran: A case report

Mahmoud Agholi [a], Hamid Reza Heidarian [a], Zahra Montaseri [b,*], Fatemeh Khajeh [c]

[a] Department of Parasitology and Mycology, Fasa University of Medical Sciences, Fasa, Iran
[b] Department of Infectious Diseases, Fasa University of Medical Sciences, Fasa, Iran
[c] Department of Pathology, Fasa University of Medical Sciences, Fasa, Iran

ARTICLE INFO

Keywords:
Echinococcosis
Skeletal muscle
Iran
Case report

ABSTRACT

Introduction and importance: Hydatid disease, caused by the larval stage of *Echinococcus granulosus*, is a common parasitic infection of humans and herbivores. Although livers and lungs are the most commonly affected organ, hydatid cysts may develop in any body part. Primary muscular hydatid cyst is extremely rare.

Case presentation: We reported the case of a 40-year-old-woman with the presentation of a soft, mobile, and non-tender lump in the dorsal part of her left upper arm (triceps brachii), which emerged one year ago. Her past medical history was unremarkable. The arm sonography revealed a single uniloculated cystic mass (6.5 cm × 5.5 cm) with a thick wall containing cystic lesions. It suggested the diagnosis of echinococcosis. The patient underwent surgery, and the hydatid cyst was excised. Histopathological examination confirmed hydatidosis.

Clinical discussion: Hydatid cysts occur rarely (about 4 %) in muscles even in endemic regions. The study is the first case of hydatidosis found in triceps brachii in Fars province, Iran. In endemic regions, considering the hydatid cyst possibility is very important because it presents with many diversities. As it clinically presents a painless slow-growing mass, may be misdiagnosed with benign soft tissue tumors.

Conclusion: Although muscular hydatidosis is extremely rare, it should be considered a differential diagnosis of any growing subcutaneous or muscular masses or tumors. Imaging modalities and blood tests are highly relevant for diagnosis. Surgical excision, a choice of treatment, should be done with cautions and is combined with anthelmintic therapy to reduce the risk of recurrence.

1. Introduction

Human echinococcosis (hydatidosis, or hydatid disease) causes by larval stages of cestodes (tapeworms) parasite from the genus *Echinococcus* (E). *E. granulosus* and less commonly *E. multilocularis* are the primary species responsible for human hydatid cysts [1,2]. In the life cycle, the adult tapeworm lives in definitive hosts (canines, mainly dogs) and releases eggs, passed in the feces. The infected eggs were then ingested by intermediate hosts (livestock, especially sheep). Humans, known as aberrant intermediate hosts, can be infected by ingesting the eggs. Humans are known as aberrant intermediate hosts [1]. The hexacanth embryo or oncosphere of *Echinococcus* penetrates the intestinal wall and is commonly transferred to the liver via portal circulation [1,2]. The two main types of the disease include cystic echinococcosis (CE) and alveolar echinococcosis. CE is asymptomatic in the early phases of the infection; however, it becomes symptomatic when the cysts become larger or complicated [1]. Clinical manifestations of CE vary and depend on the size, location, and condition of the cystic structure [1]. The commonest clinical presentation is an asymptomatic slow-growing mass. Hydatid cysts involve various organs predominantly the liver (60–70 %) and the lungs (20 %) [3]; however, the primary muscular occurrence is very rare (0.7–0.9 %), and it represents a benign soft tissue tumor in the muscle, even in endemic countries [4].

Hydatid cyst is endemic in many parts of the world, especially in the Middle East, including Iran, where about 1 % of all hospital surgeries are accounted for this disease [1,2,5]. The disease is one of the most important parasitic diseases in the Fars province, Southwest Iran [5].

The study aimed to report a rare case of a hydatid cyst in the triceps brachii muscle without identifiable hepatic or pulmonary hydatid involvement. Furthermore, the research reviewed previous hydatid cyst cases of the muscular system from Iran to delineate the most important demographic findings and locations of the disease in the country.

2. Case presentation

A 40-years old female from the Qashqai nomad, the name of a tribal

* Corresponding author.
E-mail address: montaserizahra90@gmail.com (Z. Montaseri).

https://doi.org/10.1016/j.ijscr.2022.107867
Received 16 September 2022; Received in revised form 26 December 2022; Accepted 30 December 2022
Available online 3 January 2023

people, who lived in a rural area of Fars province, Southwest Iran, was admitted to Fasa Valiasr hospital. On physical examination, there was a soft, mobile, and painless mass in the dorsal part of her left upper arm. The mass was first noticed a year ago. She had no history of trauma, surgery, drug and medication, or other diseases such as infections, genetic diseases, or malignancies. All her family members were healthy. While she had a history of caring for animals such as sheep, goats, and dogs. The preoperative diagnoses included benign tumors such as myxoma, cystic lymphangioma, and giant epidermoid cysts.

Arm sonography revealed a single uniloculated cystic mass with a thick wall with a dimension of 6.5 cm × 5.5 cm containing cystic lesions. Furthermore, to find hydatid cysts in other organs, such as the liver and the lungs, a pre-surgical computed tomography (CT) scan of the chest and ultrasonography of the abdomen was performed. The abdominal ultrasonography was normal, and the CT scan showed no lung involvement. Her laboratory, hematology, biochemistry, and serologic findings were normal, but the erythrocyte sedimentation rate (ESR) was 35 mm/h, higher than the upper limit of normal (25 mm/h for this case). The patient's weight was 70 kg and she was administered albendazole Table 400 mg PO BID twice a day for one month preoperative.

The patient underwent surgery to resect the mass from her triceps brachii in the surgical ward of Fasa Valiasr hospital. The procedure was performed by a senior orthopedic surgent. The cyst with the size of 6.5 cm × 5.5 cm and its surrounding tissue was completely excised without any gross spillage of its containing fluid inside the surgical field. The surgical field was lavaged with hypertonic saline. The procedure was tolerated appropriately without any unusual complications related to surgery or anesthesia. The patient was discharged in a good clinical condition after a day of post-operation and prescribed albendazole Table 400 mg PO twice a day for two 28-day cycles, separated by 14 drug-free days. At the 6-month follow-up, the patient did not complain of any symptoms, and no signs of hydatid recurrence were found by sonography in the hospital.

In gross pathology, a soft cystic mass contained gelatinous material and multiple daughter cysts. A histopathological cross-section was provided from the resected hydatid cyst, subsequently stained with haemotoxylin and eosin (H&E). The section with laminated and germinal layers, and brood capsules containing multiple protoscolices were considered as diagnostic keys (Fig. 1). The work has been reported in line with the SCARE 2020 criteria [6].

3. Discussion

Hepatic (60–70 %) and pulmonary (20 %) hydatid cysts are the most frequently affected organs in humans, while primary muscular hydatid cyst without liver or lung involvement is a rare entity in hydatid disease [1,7]. In endemic areas, muscular hydatid cyst is uncommon and accounts for 0.5–2.5 % of all hydatidosis cases [1]. A few cases of muscle involvement have been reported in Iran [4,8–18]. In a review of Iranian muscular hydatid cyst case reports, 15 cases, including 8 males and 7 females, with age range of 12–80 years (mean age of 43.73 years) [4,8–18], had the cysts located in the latissimus dorsi muscle, cervical muscle of the paraspinal area, gluteal, inferior rectus, biceps femoris, and thigh muscles [4,8–18]. To the best of our knowledge, this is the first study reporting a hydatid cyst in the triceps brachii in Iran.

Usually, hydatid cysts in muscles present as benign soft tissue tumors [9,13,16]. The commonest clinical presentation is a painless slow-growing mass [9,13,16]. The rarity of muscular hydatid cysts is hypothesized due to the high level of lactic acid in the muscles and contractility that confine the larva growth [13].

Total surgical excision is the main treatment when a single parasitic cyst is located in the subcutaneous or muscular tissue that should be performed to prevent severe complications caused by the advanced and ruptured cysts [2]. Anthelmintic drugs (such as albendazole) are routinely administered before and after the surgery [19]. Risk related to leakage, such as anaphylactic reactions and recurrence after surgery needs to be addressed [1,2]. Washing the surgical field with hypertonic saline during the operation, postoperative examination, pre- and post-operative treatments with albendazole are considered to reduce echinococcosis recurrence risk [20] that was implemented for the present case.

In Fars province, Iran, EC occurs more frequently among nomadic communities, settled in rural areas, because they have close contact with dogs (the definitive host of *E. granulosus*) [5]. Fars province is one of the centers of animal husbandry and agriculture in Iran, and one of the most important and populated tribal nomads (e.g., Qashqai) resides in this area. This case was a rural housewife living in Fars Province, which is endemic for hydatid cyst, and she had a history of contact with dogs, sheep, or other livestock.

4. Conclusion

The occurrence of muscular hydatidosis is extremely rare. However, in endemic areas, it should be considered as a differential diagnosis of

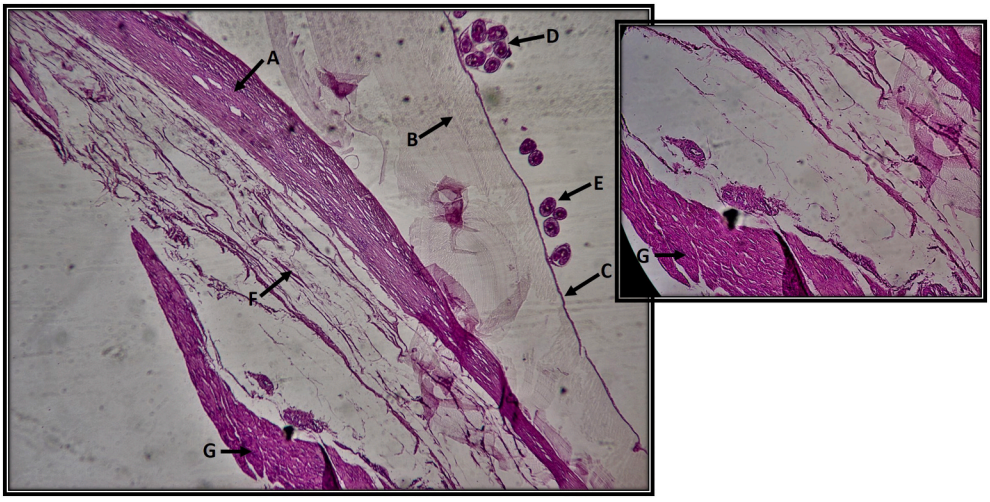

Fig. 1. Cross-section of a muscular hydatid cyst, stained with hematoxylin and eosin stain (H & E stain). Host tissue (A) encapsulates the hydatid cyst wall, which is composed of an acellular laminated layer (B) and a nucleate germinal layer (C) from which the brood capsule (D) arises. Inside the brood capsule, there are six protoscolices and liberated protoscolices from the brood capsule (E). At higher magnification, it is shown a striated muscle (G) and fibrous connective tissue (F).

other subcutaneous or muscle masses to avoid the risk of fine-needle biopsy and leakage of cyst contents. Imaging modalities (such as sonography and CT scan) and blood tests are applicable for EC diagnosis. Surgery as a choice of treatment should be performed with caution.

Consent

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

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards Fasa University Medical Science (IR.FUMS.REC.1400.103) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Sources of funding

N/A.

Author contribution

MA: Study design, data collections, and writing.
HRH: data collection, editing.
ZM: Study design, manuscript editing.
FK: Data analysis, writing.

Guarantor

Mahmoud Agholi and Zahra Montaseri.

Registration of research studies

N/A.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declaration of competing interest

N/A.

Acknowledgments

This study was supported by Clinical Research Development Unit, Valiasr Hospital, Fasa University of Medical Sciences, Fasa, Iran.

References

[1] A.T. Gessese, Review on epidemiology and public health significance of hydatidosis, Vet. Med. Int. 2020 (2020), 8859116.
[2] H. Wen, L. Vuitton, T. Tuxun, J. Li, D.A. Vuitton, W. Zhang, et al., Echinococcosis: advances in the 21st century, Clin. Microbiol. Rev. 32 (2019), e00075-18.
[3] M.A. Adelyar, S. Hesham, K. Walizada, E.A. Mushkani, R. Saadaat, Primary mesenteric hydatid cyst; a rare manifestation of hydatid disease through a case report and literature review, Int. J. Surg. Case Rep. 99 (2022), 107592.
[4] M.R. Motie, A. Rezapanah, M. Pezeshki Rad, H. Razavian, A. Azadmand, M. Khajeh, Primary localization of a hydatid cyst in the latissimus dorsi muscle: an unusual location, Surg. Infect. 12 (2011) 401–403.
[5] R. Shahriarirad, A. Erfani, M. Eskandarisani, M. Rastegarian, H. Taghizadeh, B. Sarkari, Human cystic echinococcosis in southwest Iran: a 15-year retrospective epidemiological study of hospitalized cases, Trop. Med. Health 48 (2020) 1–7.
[6] T.F. RA. Agha, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
[7] S. Mahmoudi, S. Mamishi, M. Banar, B. Pourakbari, H. Keshavarz, Epidemiology of echinococcosis in Iran: a systematic review and meta-analysis, BMC Infect. Dis. 19 (2019) 1–19.
[8] M.J. Akhavan, S. Mehrvarz, F. Panahi, Primary hydatid cyst in paravertebral muscle (a case report), Kowsar Med. J. 10 (2005) 223–226.
[9] Z.A. Aliabady, F. Berenji, M.R. Jamshidi, A case report of muscle hydatidosis from Iran, Iran. J. Parasitol. 10 (2015) 132.
[10] F. Hekmatnia, M. Motififard, H. Yazdi, A. Rizi, P. Hedayat, A. Hekmatnia, A large primary subcutaneous hydatid cyst in proximal thigh: an unusual localization, Adv. Biomed. Res. 7 (2018) 50.
[11] Z. Hosseini, R. Shahriarirad, B. Sarkari, Cystic echinococcosis: knowledge, attitude, and practices (KAP) among surgically operated cases in Fars Province,Southern Iran, J. Parasitol. Res. 2021 (2021), 976548.
[12] A. Karimi, K. Asadi, F. Mohseni, A.M. Hossein, Hydatid cyst of the biceps femoris muscle (a rare case in orthopedic surgery), Shiraz E Med. J. 12 (2011) 150–154.
[13] S.M. Nouriyan, M. Mokhtari, S.A. Fard, N. Nouriyan, Primary solitary hydatid cyst in paraspinal cervical muscles: a case report and review of the literature, Neurochir. Pol. 45 (2011) 387–390.
[14] M.T. Rajabi, S. Berijani, F.A. Amoli, S.S. Mohammadi, Hydatid cyst of inferior rectus, Ophthalmic Plast. Reconstr. Surg. 37 (2021) S168.
[15] H. Salehi, K.S. Boroujeni, S. Yaghoubi, Report of a hydatid cyst case with biceps brachii involvement, Adv. Biomed. Res. 8 (2019) 23.
[16] M. Samsami, S. Qaderi, J.Z. Bagherpour, D.E. Lucero-Prisno III, A case report of primary isolated extrahepatic hydatid cyst of the soft tissues of the breast and thigh, Int. J. Surg. Case Rep. 79 (2021) 475–478.
[17] S.S.S. Saravi, F. Sabouni, A. Arefidoust, R. Yaftian, S. Heydarzadeh, M.T. Rajabi, An orbital hydatid cyst involving inferior rectus muscle: a case report, Orbit 35 (2016) 109–112.
[18] M. Seyedsadeghi, A. Arabzadeh, A. Habibzadeh, A case of hydatid cyst in biceps femoris, Int. J. Surg. Case Rep. 66 (2020) 107–109.
[19] R. Tekin, A. Avci, R.C. Tekin, M. Gem, R. Cevik, Hydatid cysts in muscles: clinical manifestations, diagnosis, and management of this atypical presentation, Rev. Soc. Bras. Med. Trop. 48 (2015) 594–598.
[20] M.S.E. Akremi, M. Bellil, M. Souissi, M. Zaibi, W. Balti, M.B. Salah, Sciatica and femoral head osteonecrosis complicating a gluteal hydatid cyst: a case report, Int. J. Surg. Case Rep. 92 (2022), 106841.