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Case Report

A rare phenotype of breast hydatid cyst causing misdiagnosis and unnecessary intervention: a case report *,**,*

Amal A. Alareqi, MD^{a,b}, Sultan Abdulwadoud Alshoabi, MD^{c,*}, Fahad H. Alhazmi, PhD^c, Abdullgabbar M. Hamid, MD^d, Walaa M. Alsharif, PhD^c, Moawia B. Gameraddin, PhD^c

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ABSTRACT

Hydatid cyst (cystic echinococcosis) is a chronic parasitic infection by the larval stage of the cestode that is called Echinococcus granulosus (E. granulosus) resulting in the development of cystic lesions in animals and humans. In this report, we describe a rare phenotype of hydatid cyst in the breast of a 23-year-old female presented with breast mass in the left upper outer quadrant for 3 months with palpable left axillary lymph nodes. Both US and mammography provided a picture of complex suspicious cystosolid lesion with amorphous micro-calcification (BIRAD-4A). Surgical consultation was performed and Tru-Cut biopsy was recommended. Histopathology results revealed multiple viable protoscolices of E. granulosus and suggested the final diagnosis of breast hydatid cyst. The patient returned with ruptured and infected hydatid cyst of the breast and started treatment with Albendazole 400 mg twice daily in addition to antibiotics. Following-up after two months of Albendazole treatment showed a dramatic shrinkage in the size of the cystic lesion. In conclusion, hydatid cyst should be considered as a differential diagnosis of any cystic or cystosolid lesion in the breast and any organ in the body from head to toe apart from the hair and nails. Radiologists should be aware to the benign lesions that may mimic breast cancer to avoid misdiagnosis and unnecessary invasive procedures and consequent complications.

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E-mail address: alshoabisultan@yahoo.com (S.A. Alshoabi).

^a Department of Radiology, University of Science and Technology Hospital (USTH), Sana'a, Republic of Yemen

^bRadiology department, 21 September university of medical and applied sciences, Sana'a, Republic of Yemen

^c Department of Diagnostic Radiology Technology, College of Applied Medical Sciences, Taibah University, Almadinah Almunawwarah, Kingdom of Saudi Arabia

^d Department of Radiology, Rush University Medical Center, Chicago, IL, USA

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^{*} Corresponding author.

Introduction

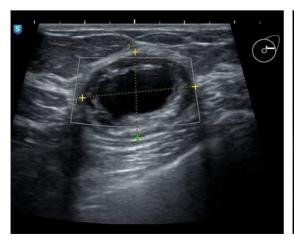
Hydatid cyst (cystic echinococcosis) is a chronic parasitic infection by the larval stage of the cestode (tapeworm) that is called Echinococcus granulosus (E. granulosus) resulting in the development of cystic lesions in animals and humans. It has a wide geographic distribution and it is an endemic in the middle east and other parts of the world. According to the World Health Organization (WHO), more than 1000,000 people are affected annually [1]. Infection occurs after oral ingestion of the infective eggs (an oncosphere containing hexacanths) that is coming from the stool of the definitive host such as dogs into the intermediate host such as human. The ingested eggs hatch the small intestine, penetrate the mucosa, enter the blood circulation, and migrate to various organs. It commonly affects the liver and lungs where the hydatid cysts are developed, and filled with protoscoleces and fluid [2]. Hydatid cysts affect usually adult female and male. It remains asymptomatic even into advanced age and the clinical manifestations depend on the site, size, and the load of cyst [3]. Ultrasound (US) imaging is considered as the primary imaging modality to diagnose hydatid cyst in the liver. Computed tomography (CT) is the imaging modality of choice to diagnose and characterize lung hydatid cysts. CT and magnetic resonance imaging (MRI) are important for further evaluation, differential diagnosis, and classification of liver hydatid cysts and in the diagnosing of central nervous system and musculoskeletal hydatid cysts. Hydatid cyst has typical imaging patterns on US ultrasound imaging, CT, and MRI which are commonly seen in extrahepatic lesions. Medical imaging is the cornerstone in diagnosing and differentiating of hydatid cyst from other cystic lesions and neoplasms in different parts of the body [4,5]. In the present case, we report a case study of patient with hydatid cyst in an unusual site in the breast that had led to misdiagnosis and mismanagement with unnecessary intervention.

Case report

A 23-year-old female with left breast mass in the left upper outer quadrant for 3 months with palpable left axillary lymph node that was referred for breast US imaging. US showed a well-defined cystic lesion (31 \times 27 \times 17 mm) with thick wall and internal thick components at 2 o'clock and 5 cm away from the nipple (Fig. 1), with axillary enlarged lymph node with thick cortex and compressed hilum suggesting reactive lymphadenopathy. The patient was referred for mammography which showed lobulated well-defined hyperdense lesion (35 \times 30 \times 23 mm) at the left upper outer quadrant with mild trabeculaion, mild tissue distortion, multiple internal amorphous micro-calcification giving a picture of suspicious lesion (Fig. 2 & 3). The US and mammography examination provided a picture of complex suspicious cystic lesion with amorphous micro-calcification (BIRAD-4A). Surgical consultation was performed and Tru-Cut biopsy was recommended. Tru-Cut biopsy was performed under US guidance and sample was sent to the histopathologist. Histopathology results revealed multiple viable protoscolices of E. granulosus and suggested the final diagnosis of breast hydatid cyst (Fig. 4). After 10 days, the patient returned with ruptured and infected hydatid cyst of the breast as a complications of the biopsy. The patient started treatment with Albendazole 400 mg twice daily in addition to antibiotics. Following-up after two months of Albendazole treatment showed a dramatic shrinkage in the size of the cystic lesion.

Discussion

This report stated a rare case of breast hydatid cyst that was initially misdiagnosed as suspicious breast mass. The Tru-Cut biopsy was performed and the cyst was complicated by rup-



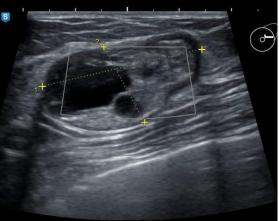


Fig. 1 – Ultrasound images of a 23-year-old female with breast lesion show a well-defined cystic lesion (31 \times 27 \times 17 mm) with thick wall and internal thick components in the left upper outer quadrant at 2 o'clock and 5 cm away from the nipple.

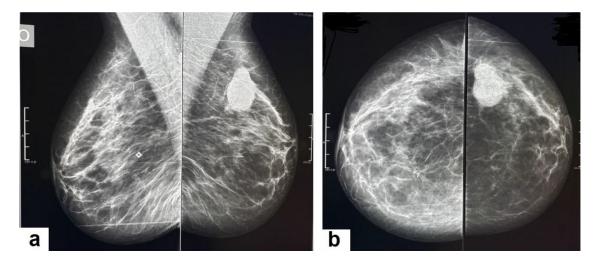


Fig. 2 – Mammography image of a 23-year-old female with breast lesion (A) Medio-lateral oblique view, and (B) Cranio-caudal view show a lobulated well-defined hyperdense lesion (35 \times 30 \times 23 mm) at the left upper outer quadrant with mild trabeculaion, mild tissue distortion giving a picture of suspicious lesion.

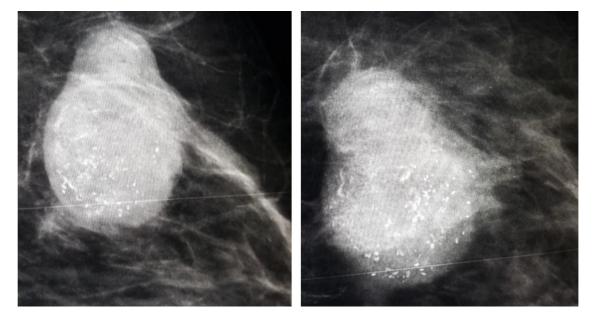
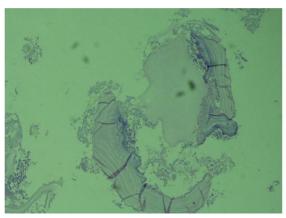


Fig. 3 – Mammography images of a 23-year-old female with breast lesion, medio-lateral oblique full field view show a well-defined lobulated hyperdense lesion with mild trabeculaion, mild tissue distortion, multiple internal amorphous micro-calcification giving a picture of suspicious lesion (BIRAD-4A).

ture and infection before reaching to the correct diagnosis. Zalaquett *et al.* reported that hydatid cyst commonly affects the liver, however, it can affect the lung, spleen, kidney, peritoneum, heart, muscles, and brain [4]. Sachar *et al.* reported that hydatid cyst can affect thyroid gland, breast, gallbladder, pancreas, uterus and adnexa, seminal vesicles, bones, muscles, skin and subcutaneous tissue [6]. In the literature, hydatid cyst was reported in the orbit [7], the breast of a male patient [8], the right ventricle of the heart [9], the thyroid gland [10], the subcutaneous region of face [11], over the plantar surface of the foot [12], and even in the posterior triangle of the neck of a 3-year-old child [13].

Although, US can predict benign breast masses in nearly 97% of cases [14], our case was misdiagnosed as suspicious breast mass (BIRAD-4A) by both US and mammography. This was explained by the very rare site in the breast and the atypical morphology of the hydatid cyst in our case which is leading to low suspicion of the radiologist. Michaels *et al.* reported that fat necrosis, hematomas, and reactive lymph nodes are benign breast lesions which change the weight against benign morphology of breast lesions [15].

The diagnosis of hydatid cyst should be excluded before any surgical intervention or Tru-Cut biopsy to avoid leakage of cyst components and risk of anaphylactic shock and sec-



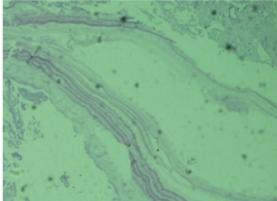


Fig. 4 – Histopathology microscopic images with hematoxylin and eosin (H&E) stain show multiple protoscolices of *Echinococcus granulosus* in a Tru-Cut biopsy of a 23-year-old female with breast lesion which confirmed the diagnosis of breast hydatid cyst.

ondary hydatidosis. In case you suspect hydatid cyst as a differential diagnosis, you can confirm the diagnosis by further radiological and serological tests. Magnetic resonance imaging (MRI) can help in diagnosis of hydatid cyst. The findings of cystic lesion with enhancing capsule are suggestive of hydatid cyst. Hydatid cyst appear, as any cystic lesion, hypointense on T1-weighted images (T1WIs) and hyperintense on T2-weighted images (T2WIs). However, a low signal-intensity rim "rim sign" is more evident on T2WIs. Daughter cysts may appear hypointense or isointense relative to the maternal matrix on T1WIs and T2WIs. The "serpent sign" or "snake sign" which represents collapsed membranes of cystic damage or degeneration of low signal intensity with all sequences is another specific imaging feature of a hydatid cyst [16].

The enzyme-linked immunosorbent assay (ELISA) is highly sensitive and specific for hydatid cyst of the liver but less sensitive and specific for it in other organs [17]. Other serological tests include casoni skin test, complement fixation weinberg (CF) test, indirect hemagglutination (IHA) test, and western blot (WB) [18]. Positive serological tests are valuable, however, negative serological tests do not exclude hydatid cyst [17,18].

In addition, US guided fine needle aspiration cytology (FNAC) is safe, fast, inexpensive and accurate preoperative diagnostic tool that can allow surgery to be avoided. Protoscolices, hooklets, and laminated membranes of E. granulosus can be identified in FNAC without complications [19,20]. Treatment of the breast hydatid cyst is cystectomy with pericystectomy to protect rupture and reinfestation in addition to Albandazole for further prevention [21]. Our case report suggests to add breast hydatid cyst to the differential diagnoses of breast lumps especially in endemic regions and to exclude hydatid cyst before any surgical intervention to avoid undesirable complications.

Conclusion

Radiologists should be aware to the benign breast lesions that may mimic breast cancer to avoid misdiagnosis and unnecessary invasive procedures and consequent complications. Hydatid cyst has different phenotypes on medical imaging modalities that can mimic malignant lesions. It should be considered as a differential diagnosis of any cystic or cystosolid lesion in the breast and any organ in the body from head to toe apart from the hair and nails.

Teaching points from this case

- 1 Hydatid cyst should be considered as a differential diagnosis of any cystic or cystosolid lesion in the breast to choose the appropriate diagnostic tests and avoid unnecessary intervention.
- 2 FNAC is a suggested safe and accurate preoperative diagnostic tool in beast lumps of questionable diagnosis for early accurate diagnosis and appropriate treatment.

Authors contribution

A.A.A. provided ultrasound imaging and data collection. S.A.A. wrote the final drafts of the manuscript. F.H.A. revised the manuscript and edited language. A.M.H. interpreted data. WMA revised the manuscript. MBG revised the manuscript.

REFERENCES

- [1] Mahmoudi S, Mamishi S, Banar M, Pourakbari B, Keshavarz H. Epidemiology of echinococcosis in Iran: a systematic review and meta-analysis. BMC Infect Dis 2019;19(1):929. doi:10.1186/s12879-019-4458-5.
- [2] Li X, Jiang S, Wang X, Hui W, Jia B. iTRAQ-based comparative proteomic analysis in different developmental stages of Echinococcus granulosus. Parasite 2021;28:15. doi:10.1051/parasite/2021012.
- [3] Joshi U, Subedi R, Jayswal A, Agrawal V. Clinical characteristics and management of the hydatid cyst of the liver: a study from a tertiary care center in Nepal. J Parasitol Res 2020;2020:8867744. doi:10.1155/2020/8867744.

- [4] Zalaquett E, Menias C, Garrido F, Vargas M, Olivares JF, Campos D, et al. Imaging of hydatid disease with a focus on extrahepatic involvement. Radiographics 2017;37(3):901–23. doi:10.1148/rg.2017160172.
- [5] Bhutani N, Kajal P. Hepatic echinococcosis: A review. Ann Med Surg (Lond) 2018;36:99–105. doi:10.1016/j.amsu.2018.10.032.
- [6] Sachar S, Goyal S, Goyal S, Sangwan S. Uncommon locations and presentations of hydatid cyst. Ann Med Health Sci Res 2014;4(3):447–52. doi:10.4103/2141-9248.133476.
- [7] Assimakopoulos SF, Marangos M. Orbital hydatid cyst. N Engl J Med 2020;382(14):1352. doi:10.1056/NEJMicm1911903.
- [8] Limaiem F, Bouslama S, Haddad I, Bouraoui S, Lahmar A, Mzabi S. Hydatid cyst presenting as a breast lump in a male patient. Pathologica 2013;105(3):101–3.
- [9] Aljaber NN, Alshoabi SA, Qurashi AA, Daqqaq TS. Cardiac hydatid cyst in the right ventricle: an unusual case at a rare site. J Taibah Univ Med Sci 2020;15(3):249–52. doi:10.1016/j.jtumed.2020.02.004.
- [10] Eshraghi M, Shahmoradi L, Ghoddoosi M, Sadati SJA. Diagnosis of primary hydatid cyst of thyroid gland: a case report. Biomol Concepts 2019;10(1):106–10. doi:10.1515/bmc-2019-0013.
- [11] Nischal N, Verma A K, Bhangale D. Primary subcutaneous hydatid cyst of the face: a rare entity with emphasis on radiological evaluation, arch clin infect dis. 2017; 12(3):e63543. doi:10.5812/archcid.63543.
- [12] Ewnte B. Hydatid cyst of the foot: a case report. J Med Case Rep 2020;14(1):6. doi:10.1186/s13256-019-2337-8.
- [13] Giriyan SS, Navyashree N. An unusual presentation of hydatid cyst in the neck: a cytological diagnosis. Indian J Pathol Microbiol 2018;61(3):454–5. doi:10.4103/IJPM.IJPM_9_18.

- [14] Binnuhaid AA, Alshoabi SA, Alhazmi FH, Daqqaq TS, Salih SG, SA Al-Dubai. Predictive value of ultrasound imaging in differentiating benign from malignant breast lesions taking biopsy results as the standard. J Family Med Prim Care 2019;8(12):3971–6. doi:10.4103/jfmpc.jfmpc_827_19.
- [15] Michaels AY, Birdwell RL, Chung CS, Frost EP, Giess CS. Assessment and management of challenging BI-RADS Category 3 Mammographic Lesions. Radiographics 2016;36(5):1261–72. doi:10.1148/rg.2016150231.
- [16] Koc A, Sarici IS, Vurdem UE, Karabiyik O, Gumus UO. Unusual presentation of hydatid cyst in breast with magnetic resonance imaging findings. Case Rep Med 2017:6237435 2017. doi:10.1155/2017/6237435.
- [17] Samsami M, Qaderi S, J Zebarjadi Bagherpour, 3rd Lucero-Prisno DE. A case report of primary isolated extrahepatic hydatid cyst of the soft tissues of the breast and thigh. Int J Surg Case Rep 2021;79:475–8. doi:10.1016/j.ijscr.2021.01.087.
- [18] Meshram N, Kamal F, Gadkari R. Hydatid cyst of breast: unwelcome guest at unusual site; a case report and review of literature. Int J Contemporary Med Res 2021;8(1):A12–16.
- [19] Alamer A, Aldhilan A, Makanjuola D, Alkushi A. Preoperative diagnosis of hydatid cyst of the breast: a case report. Pan Afr Med J 2013;14:99. doi:10.11604/pamj.2013.14.99.2396.
- [20] Cancelo MJ, Martín M, Mendoza N. Preoperative diagnosis of a breast hydatid cyst using fine-needle aspiration cytology: a case report and review of the literature. J Med Case Rep 2012;6:293. doi:10.1186/1752-1947-6-293.
- [21] Moussaoui KE, Lakhdar A, Baidada A, Kherbach A. Hydatid cyst of the breast: case report. Int J Surg Case Rep 2020;77:325–8. doi:10.1016/j.ijscr.2020.10.109.