



Case series

Hydatid disease as a rare cause of neck swelling: Two cases report

Makram Tbini^{*}, Rim Lahiani, Houda Guelmami, Habib Jaafoura, Ines Riahi, Mamia Bensalah

Head and Neck Surgery Department, Charles Nicolle Hospital, Tunis, Tunisia

ARTICLE INFO

Keywords:

Hydatid disease
Echinococcosis
Neck swelling
Surgery

ABSTRACT

Introduction: Despite the fact that hydatid disease is endemic in some countries, its cervical location remains very rare.

Presentation of case: We report two cases of primary cervical hydatid disease in two children from rural areas. The imaging was not specific, since the location is rare; the diagnosis was only confirmed after surgery.

Discussion: Hydatid cyst is a rare pathology. Thus clinical presentation, radiological examinations, fine needle aspiration and serology can be useful to suspect the diagnosis.

Conclusion: Hydatid disease should be considered when dealing with slow-growing recurrent neck swelling in endemic countries.

1. Introduction

Echinococcus granulosus the causative parasite of hydatid disease requires two mammalian hosts for completion of its life cycle; a canid (dog, wolf, dingo, or jackal) as the final host, domestic ruminant (sheep) as the intermediate host and human can be an accidental intermediate host [1]. Hydatid disease mainly affects liver and lung in endemic regions such as South America, Middle East, Africa, Australia, and the Mediterranean region [1].

Head and neck hydatid cyst is still an exceptional localization even in endemic countries, accounting for less than 1% of the localizations and diagnostic is quite challenging [2].

The aim of this work is to study the epidemiological, clinical, therapeutic features and to highlight the diagnostic difficulties posed by head and neck localization of hydatid disease.

This article has been reported in line with SCARE criteria [3].

2. Presentation of case

2.1. 1st case

A 17-year-old child from a rural origin, asthmatic, presented with an isolated swelling in the left supraclavicular fossa, growing over a period 2 years.

Physical examination revealed a left supraclavicular tumefaction of 6 cm long axis, well limited, homogeneous, depressible, soft, mobile, painless and covered with healthy skin.

Cervical ultrasound objectified a cystic mass of 66 mm multivesicular.

A cervical MRI was requested showing a cystic lesion with a low signal intensity on T1-weighted image and a high signal intensity on T2-weighted image associated with a floating membrane (Fig. 1).

We suspected a hydatid cyst and a requested hydatid serology came back positive.

The patient had a cystectomy (Fig. 2) with simple operative follow-up. Anatomopathology concluded a hydatid cyst. Abdominal ultrasound and chest X-ray were without abnormalities.

2.2. 2nd case

A 6 years old boy from a rural origin was admitted to our department for a 3 cm swelling in the right anterior triangle of the neck.

The mass was mobile, painless and non-tender with healthy skin.

Cervical ultrasonography showed a cystic formation with unilocular liquid content.

Cervical MRI was performed and showed a unilocular cystic lesion with a low signal intensity on T1-weighted image and a high signal intensity on T2-weighted image (Figs. 3 and 4).

Branchial cleft cyst or a cystic lymphangioma were suspected so the cyst was removed intact, and histological examination confirmed the diagnosis of hydatid disease.

The postoperative course was uneventful. Abdominal ultrasonography and chest X-Ray were normal. No recurrence was observed with a mean follow-up of 2 years.

^{*} Corresponding author at: Head and Neck Surgery Department, Charles Nicolle Hospital, Boulevard du 9-Avril-1938, 1006 Tunis, Tunisia.

E-mail address: makramtb@yahoo.com (M. Tbini).

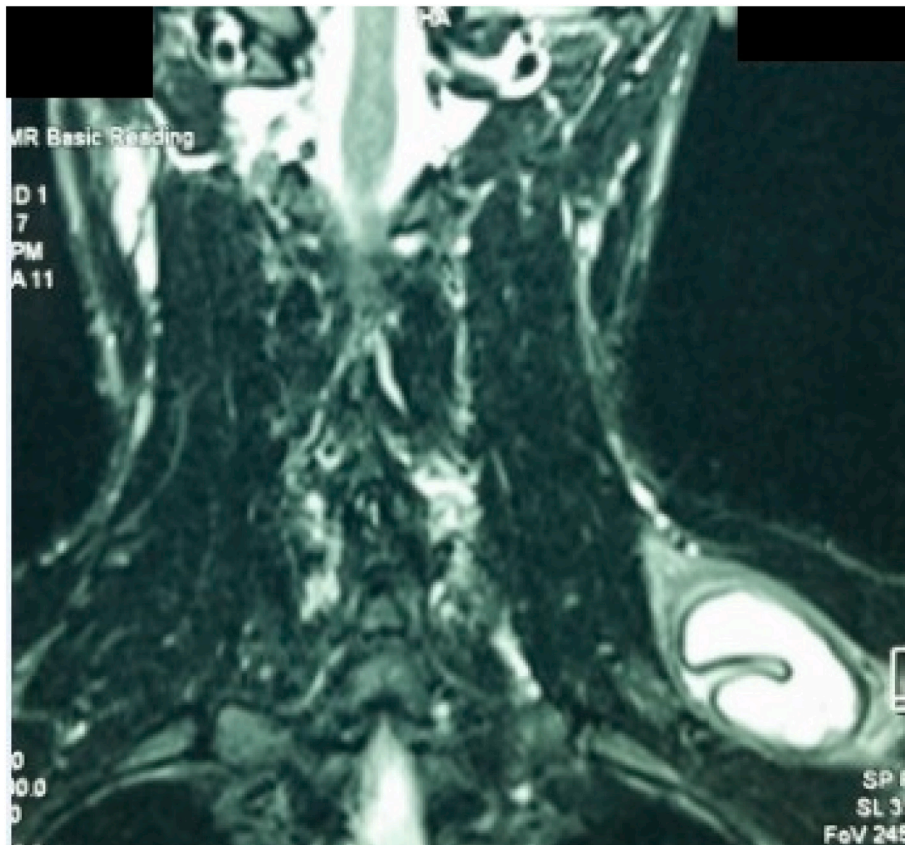


Fig. 1. MRI coronal cut: left side supraclavicular cystic lesion with a high signal intensity on T2-weighted image associated with a floating membrane.

3. Discussion

Hydatid disease is caused by *Echinococcus granulosus*. Dogs are the main hosts and sheep are the common intermediate hosts. Humans are accidental intermediate hosts. Food and drinks contaminated with infected dog feces containing *Echinococcus* eggs usually infect humans. Once in the digestive system, the oncosphere infiltrates the gut wall and circulates in the blood to the liver then possibly to the lungs, but rarely oncospheres escape pulmonary and hepatic filters into the general systematic circulation and develop in other organs. Then oncospheres develop to become hydatid cyst [1].

Hydatid disease is endemic in regions where sheep rearing is common like the Mediterranean regions, southern and central parts of Russia, central Asia, China, Australia, South America, north Africa and east Africa [4]. However even in those regions, head and neck hydatid disease have been rarely reported and thus the disease is rarely mentioned as a differential diagnosis of head and neck swellings [5].

The diagnosis is evoked based on epidemiological data such as personal history of liver or lung hydatid disease, ingestion of infested raw or undercooked meat in rural areas, contact with dogs. Clinical presentations are non-specific. Patient may be asymptomatic and symptoms are caused by the compression of adjacent organs based on the location, size, host immune response, and complications of the cysts [6].

Imaging methods such as ultrasound, CT scan, and magnetic resonance imaging can help make the diagnosis. Ultrasound can reveal hydatid sand-fine internal debris, floating membranes and daughter cysts. Computed tomography detects calcifications, bone involvement, while magnetic resonance imaging is useful for detecting neural involvement,

provides a better contrast resolution, and thus allows precise definition of the internal contents of the cyst. Hydatid cysts are classified into five types based to their characteristics (type I: unilocular cyst, type II: Multivesicular cyst, type III: Floating membranes, type IV: Heterogeneous cyst, type V: calcified) [7,8]. Since it is a rare location, there are no clear recommendations on imaging modality. Nevertheless, like other cystic cervical masses, ultrasound is the best initial investigation tool given its cost and availability, followed by MRI which is preferred over CT, especially in T2 sequence for better tissue resolution allowing to anatomically locate the cyst and appreciate its extensions [9,10].

Fine needle aspiration cytology (FNAC) is widely used in head and neck swelling. However its use in case of hydatid disease is controversial due to the risk of hydatid spread and anaphylactic reaction but this risk is rare and FNAC can help detect hydatid cysts [11,12].

A variety of serological methods for hydatid disease exist including indirect hemagglutination (IHA), immunoblotting, enzyme-linked immunosorbent assay (ELISA), indirect fluorescent-antibody (IFA), latex agglutination test, and immunochromatography test. Hydatid serology can guide the diagnosis when positive but negative serology does not exclude the diagnosis [13,14].

Clinical presentation, imaging, FNAC and serology are non-specific even in endemic regions making hydatid disease rarely considered as a differential diagnosis of neck cyst. However, hydatid disease should always be kept in mind to avoid perforating the capsule during surgery [15].

Surgery is the treatment of choice for hydatid cysts, however, anti-parasitic drugs (Albendazol or Meprobendazol) can be used in inoperable patients, in cases of multiple lesions and for a pre-operative or post-



Fig. 2. Intraoperative view: in-bloc resection of the cyst.



Fig. 3. MRI coronal cut: cervical right side unilocular cystic lesion with a low signal intensity on T1-weighted image.

operative treatment to prevent recurrence [16].

Although the traditional choice of treatment for hydatid disease is radical in-bloc resection to avoid anaphylactic reaction, dissemination of disease and recurrence, percutaneous treatment by PAIR technique (Puncture-Aspiration of cyst contents-Injection of hypertonic saline solution-Reaspiration) can be a safe alternative to surgical treatment [17].

4. Conclusion

Although the liver and lungs are the primary sites of infection, hydatid disease is also reported from various sites of the body such as the head and neck.

Clinical symptomatology is non-specific. Notwithstanding, a slow-growing recurrent swelling in the head and neck in a patient living in an endemic area should draw attention to the hydatid cyst. Thus, surgical precautions should be kept in mind when operating.

Prevention remains the best option.

Consent

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

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

This study is exempt from ethical approval at our institution.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Guarantor

Makram Tbini.

Research registration number

Not applicable.

Author contribution

Makram Tbini: Writing - Original draft
 Rim Lahiani: Writing - Review & editing
 Houda Guelmami: Writing - Original draft
 Habib Jaafoura: Data curation
 Ines Riahi: Supervision
 Mami Bensalah: Supervision.

Declaration of competing interest

The authors report no declarations of interest.

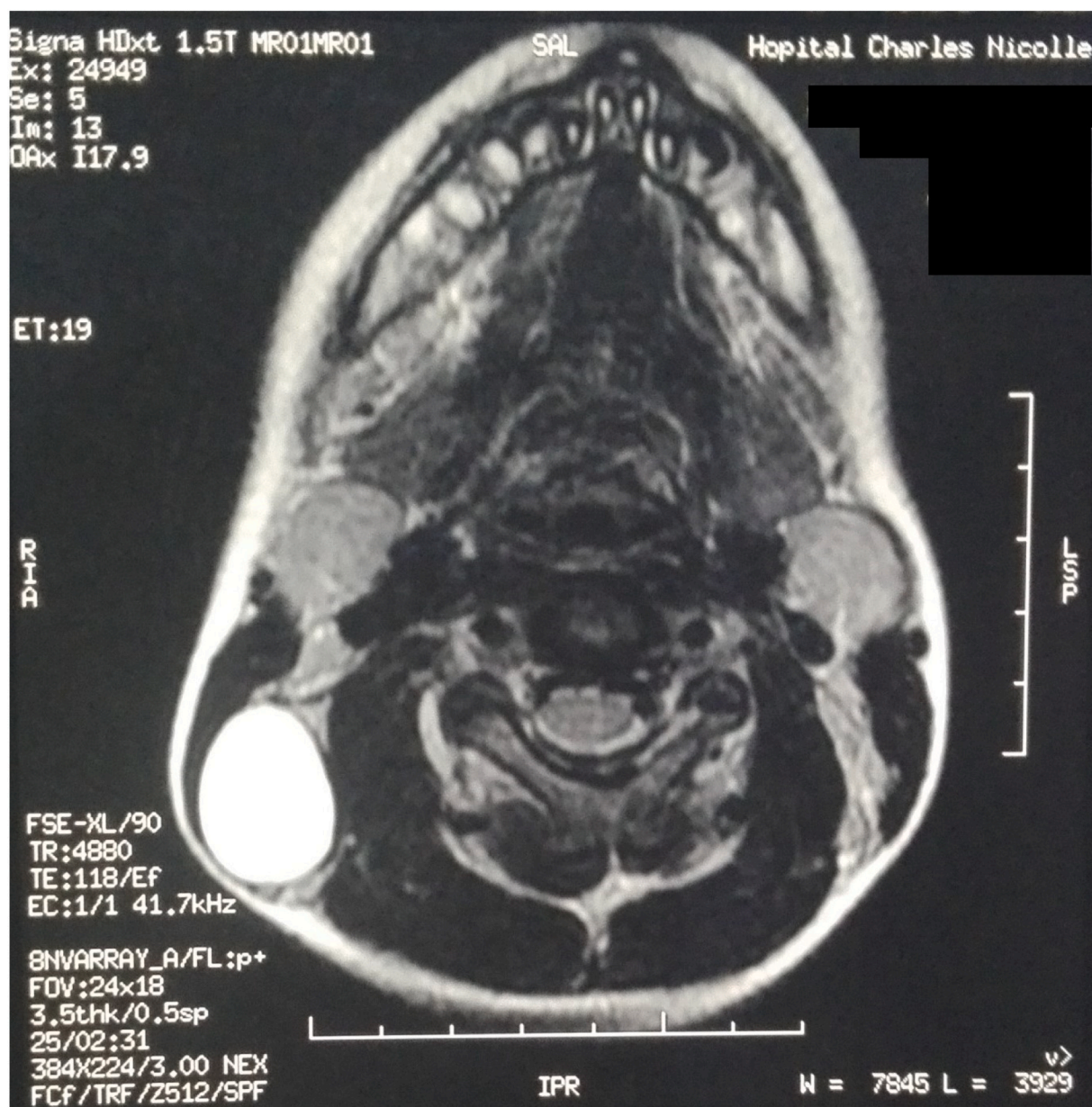


Fig. 4. MRI axial cut: cervical right side unilocular cystic lesion with a high signal intensity on T2-weighted image.

References

- [1] A.T. Gessese, Review on epidemiology and public health significance of hydatidosis, *Vet Med Int.* 2020 (2020) 8859116.
- [2] M.T. Rahimi, R. Jurat, M.A. Shirzad, H. Rahman, Primary hydatid cyst of the neck – EPSC 101593, *J. Pediatr. Surg. Case Rep.* 61 (2020), 101593.
- [3] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, Group S, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, *Int. J. Surg.* 84 (2020) 226–230.
- [4] G. Grosso, S. Gruttadauria, A. Biondi, S. Marventano, A. Mistretta, Worldwide epidemiology of liver hydatidosis including the Mediterranean area, *World J. Gastroenterol.* 18 (13) (2012) 1425–1437.
- [5] R. Khalifa, F. Nasser, A. Elsetouhy, I. Farag, Hydatid cyst of the neck. a case report and literature review, *Egypt. J. Ear Nose Throat Allied Sci.* 17 (2) (2016) 103–105.
- [6] M. Alam, S.-A. Hasan, S.-F. Hashmi, Unusual presentation of hydatidosis - neck lump causing costo-vertebral erosion, *Iran. J. Otorhinolaryngol.* 28 (88) (2016) 363–367.
- [7] M.R. Srinivas, B. Deepashri, M.T. Lakshmeesha, Imaging spectrum of hydatid disease: usual and unusual locations, *Pol. J. Radiol.* 81 (2016) 190–205.
- [8] E. Zalaquett, C. Menias, F. Garrido, M. Vargas, J.F. Olivares, D. Campos, et al., Imaging of hydatid disease with a focus on extrahepatic involvement, *RadioGraphics* 37 (3) (2017) 901–923.
- [9] K.T. Wong, Y.Y. Lee, A.D. King, A.T. Ahuja, Imaging of cystic or cyst-like neck masses, *Clin. Radiol.* 63 (6) (2008) 613–622.
- [10] S. Acharya, S. Panda, S. Biswal, Primary hydatid cyst of neck, a rare case report, *Indian J. Otolaryngol. Head Neck Surg.* (2021).
- [11] R. Singal, A. Mittal, M. Garg, M. Zaman, M. Chaudhry, S. Singal, et al., Unusual location of primary hydatid cyst diagnosed on aspiration cytology, *J. Cosmet. Dermatol.* 16 (4) (2017) e42–e44.
- [12] I. Chakrabarti, B.K. Goswami, Primary hydatid cyst of the neck diagnosed by aspiration cytology, *Trop. Parasitol.* 2 (2) (2012) 127–128.
- [13] S. Mahajan, S. Thapar, V. Khillan, P. Gupta, A. Rastogi, E. Gupta, Comparative evaluation of echinococcus serology with cytology for the diagnosis of hepatic hydatid disease, *J. Lab. Physicians* 12 (2) (2020) 98–102.
- [14] B. Sarkari, Z. Rezaei, Immunodiagnosis of human hydatid disease: where do we stand? *World J. Methodol.* 5 (4) (2015) 185–195.
- [15] K. Abasi, C.-x. Li, P. Pataer, Z.-c. Gong, Hydatid cyst of the oromaxillofacial region – a rare location: a case report and review of the literature, *J. Oral Maxillofac. Surg.* (2021).
- [16] J.Y. Chai, B.K. Jung, S.J. Hong, Albendazole and mebendazole as anti-parasitic and anti-cancer agents: an update, *Korean J. Parasitol.* 59 (3) (2021) 189–225.
- [17] O. Akhan, S. Ensari, M. Özmen, Percutaneous treatment of a parotid gland hydatid cyst: a possible alternative to surgery, *Eur. Radiol.* 12 (3) (2002) 597–599.