



Case report

Hydatid cyst in the neck, an unusual localization of the disease: A case report

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ABSTRACT

Introduction and importance: Hydatid cyst disease or hydatidosis is an old parasitic disease. Humans represent an accidental intermediate host. Neck is considered a rare location of hydatid cysts. It is usually overlooked as a deferential diagnosis in any cystic lesion in the neck, even in the regions where the disease is considered endemic.

Case presentation: A 26 year old female was admitted to Al-Mouwasat Hospital with the complaint of an anterior cervical mass causing compressive symptoms. Examinations determined a big cystic lesion at expense of the thyroid gland and extended to the level of aortic arch, in addition to smaller lesions in the two thyroid lobes. Multinodular goiter was diagnosed. However, Hydatid cyst disease was suspected during surgery. Cystectomy and total thyroidectomy were performed. Histopathology confirmed the diagnosis of hydatid cyst disease.

Clinical discussion: Hydatid cyst disease in the neck is usually asymptomatic unless it compresses near structures. Histopathological study makes the definitive diagnoses. The best treatment of thyroid hydatid cysts is total surgical excision of all cysts with intensive care not to spread the disease.

Conclusion: Hydatid cyst disease in the neck is extremely rare. Hydatid cyst should be included in differential diagnosis of cystic lesions in the neck especially in patients from endemic countries. Post-operative surveillance is important to diagnose the complete healing or recurrence of hydatid disease in operated or other locations but also for follow up of replacement therapy after total thyroidectomy as the patient require lifelong thyroid hormone replacement.

1. Introduction

Hydatid cysts disease or hydatidosis is an anthropozoonosis disease caused by *Echinococcus* species [1].

Canid are the definitive hosts, whereas herbivores are the intermediate hosts. Humans represent an accidental intermediate host in the parasite life cycle [2].

This disease is endemic in the Mediterranean countries, Middle East, South America, South Africa, New Zealand, Iceland, Australia, and India [3,4].

Hydatid cysts may reside in almost all organs of human body.

Neck is rarely involved by the disease, even in areas where it is endemic [5,6]. The incidence rate of hydatid cysts in the thyroid gland is 0.5-1% in Turkey which is an endemic country [7].

We present here a rare case of a 26 year old female having a very

large hydatid cyst in the neck extending into the level of aortic arch in the mediastinum. This work has been reported in line with the SCARE 2020 criteria [8].

2. Case report

A 26 year old female farmer presented at our clinic complaining from a clear anterior cervical mass growing in size during a one year span. She had compressive symptoms such as dysphagia and shortness of breath. The symptoms worsened as the mass got bigger.

The patient came from a rural region. She was admitted to the hospital.

Physical examination clearly showed an anterior cervical mass located at the midline inclined to the right side and extended downward to the base of the neck. It was measuring about 5*4 cm. It wasn't mobile

Abbreviations: CT, Computed tomography; MRI, Magnetic resonance imaging; FNA, Fine needle aspiration; E., *Echinococcus*.

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with deglutition. We made an echography in which a big septated cystic lesion was seen. It had a well-distinguished wall and located at the midline extending into the retrosternal region measuring about 5 cm. It was suspected to have a relation with the right thyroid lobe and might present a thyroid cyst. Small hypoechoic lesion in the right lobe of the thyroid gland measured 8 mm and another in the left lobe measured 10 mm were also noticed. Multinodular goiter with big thyroid cyst was diagnosed.

Computed tomography "CT" was obtained to determine the relation between the cyst and its surroundings. It showed a big walled-cystic lesion in the neck [Fig. 1]. It was pushing the surrounding structures [Fig. 2]. The cyst was extending into the mediastinum to reach the level of the aortic arch measuring 7.5*5*5 cm [Fig. 3]. Otherwise, chest and abdomen were normal.

Laboratories including complete blood count, thyroid function tests, renal function tests and blood glucose were all normal.

Differential diagnoses included multinodular goiter, malignant formations of the thyroid gland, big thyroglossal cyst, branchial cleft cyst, cold abscess and hydatid cyst disease.

After discussing the case in our general surgery department, it was decided to perform fine needle aspiration "FNA".

FNA was made and revealed scattered benign follicle cells which declared to be Bethesda II. Malignancy was ruled out.

The patient then was prepared for surgery to have total thyroidectomy.

Surgery was performed in Al Mouwasat University Hospital by the three authors. Under general anesthesia, we performed Kocher incision then midline dissection through the strap muscles to reach the thyroid gland. A very big cyst was noticed taking up most of the thyroid gland and extending into the mediastinum. With much care we exposed the cyst by dissecting it from its surroundings, but we couldn't dissect it from the thyroid. We highly suspected the hydatid cyst disease. Moistened gauze with hypertonic saline solution were put around the cyst in which we made a very small incision in its anterior wall very carefully with immediate suction to its inner contents. Many cysts looked like daughter cysts were suctioned. We extended the incision in the cystic wall and resected the endothelium membrane that could be the germinal layer. After suctioning the cystic components the inferior borders of the cystic

wall were clearly exposed. The resection of the whole cystic wall was then proceeded in addition to performing total thyroidectomy since bilateral lesions in the right and left lobes of the thyroid were known.

Two corrugated drains were put, one in each side.

Three days after the procedure, the drains were pulled out. Symptoms of dysphagia and shortness of breath were resolved. She was discharged without putting her on anti-parasitic medications since no contamination happened during surgery as there was no evidence of other cysts in other organs such as liver and lungs.

Three and six months after surgery, echography was repeated. It was normal.

3. Discussion

Hydatid cysts disease is one of the oldest known disease [9]. It is anthrozoosis caused by *Echinococcus* species. *E. granulosus* is the most common responsible of cystic formations. Other species which may cause the disease include *E. multilocularis*, *E. oligarthrus* and *E. vogeli* [1].

It is endemic in the Mediterranean countries, Middle East, South America, South Africa, New Zealand, Iceland, Australia, and India [3,4].

Canids are the definitive hosts whereas sheep, goats, cattle, camels and pigs are the intermediate hosts in the parasite life cycle [2].

Humans represent an accidental intermediate host after ingesting food contaminated with parasite eggs [2].

Eggs penetrates the wall of small bowel's host and migrate through the circulatory system into other organs especially the liver and lungs. They develop there to become cysts [10].

The common organs to be infected are liver (50-77%), Lungs (15-47%), Spleen (0.5-8%) and Kidneys (2-4%) [6,11].

Less commonly brain, musculoskeletal system, heart and pancreas. Neck is rarely involved by the disease process even in areas where the disease is considered endemic [5,6].

The incidence rate of hydatid cysts in the thyroid gland is 0.5-1% in Turkey which is an endemic country [7].

Hydatid cyst disease in the thyroid is usually asymptomatic. Cysts then may increase in size and compress the surrounding structures causing symptoms of dyspnea, dysphagia and hoarseness [4,12,13].

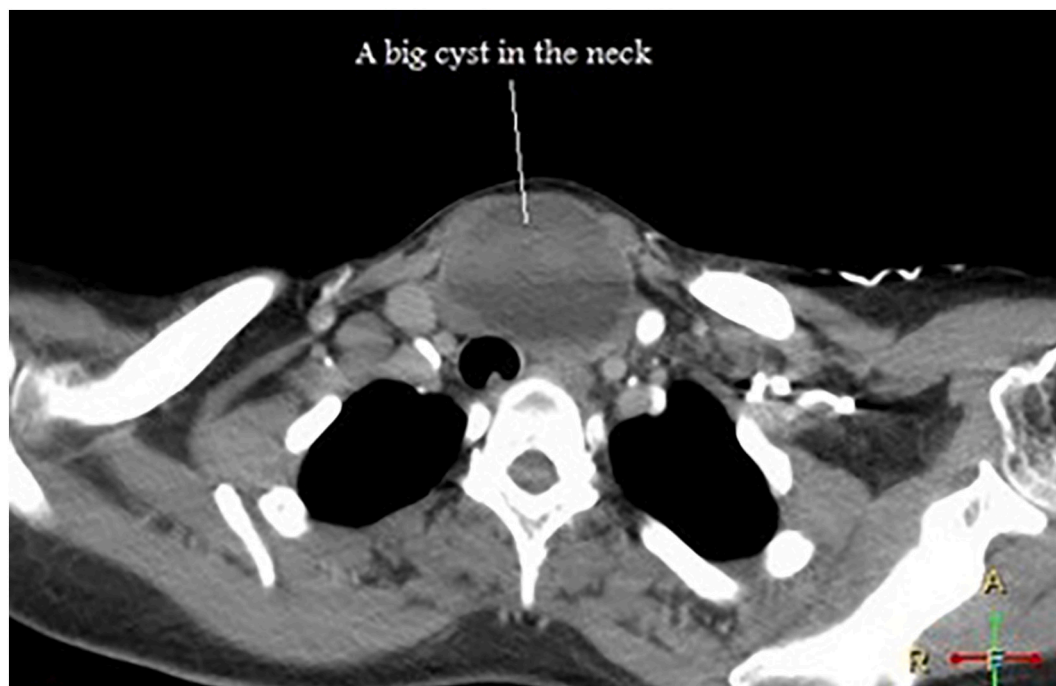


Fig. 1. A Big cyst in the neck.

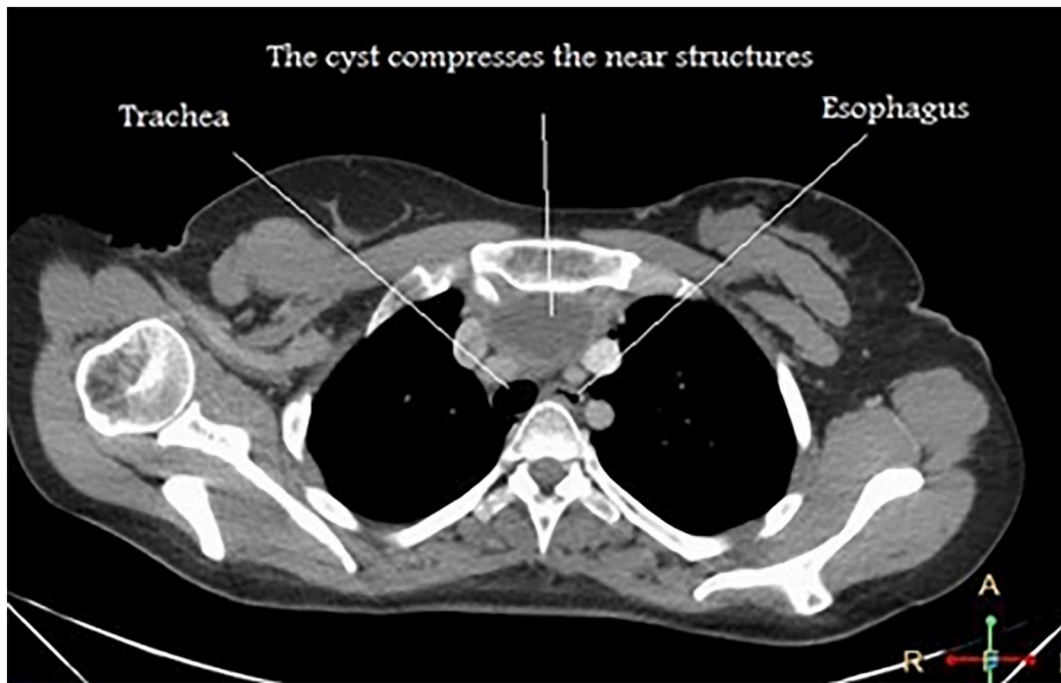


Fig. 2. The cyst compresses trachea and esophagus.

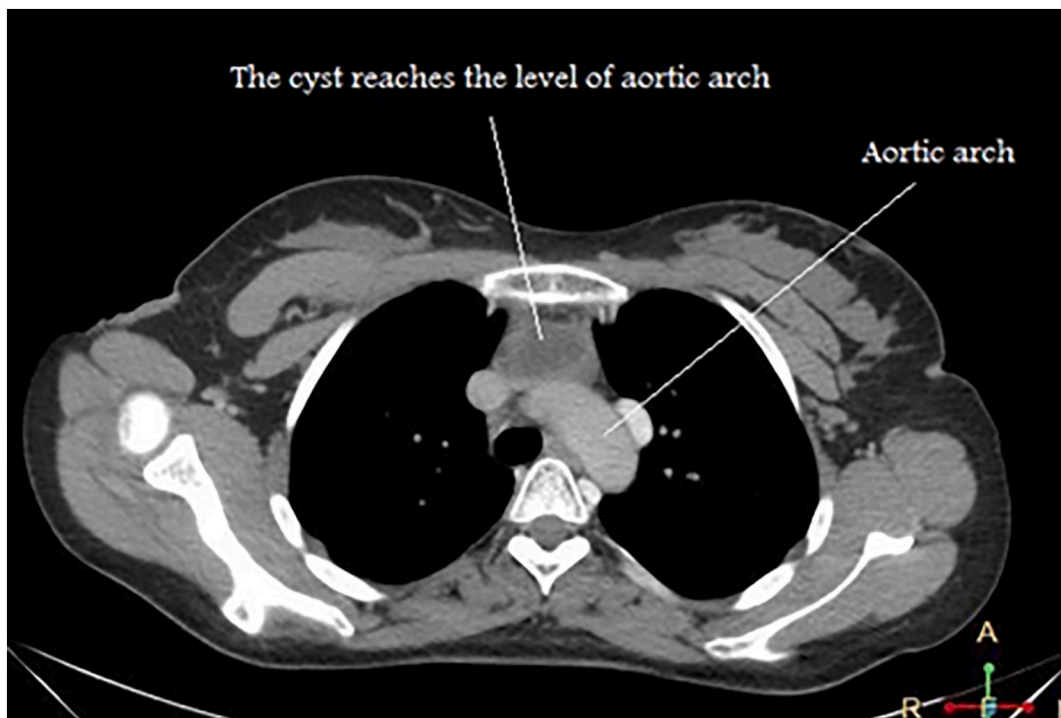


Fig. 3. The cyst extends to the level of aortic arch.

In a case of an isolated neck mass, it is not common to think of a hydatid cyst but shouldn't be forgotten especially in endemic countries.

Like in our case, diagnosis was not easy. It was actually accomplished during surgery and the confirmation was after examination the specimen under microscope.

Patient history, physical examination, serologic tests and imaging studies should all integrate to reach the diagnosis of such rare cases.

The patient's history which include working closely with livestock or in which other family members were diagnosed with Hydatidosis should

raise the probability of any cystic lesion to be a hydatid cyst.

Although physical examination doesn't give the specific diagnosis, it is useful to describe the characteristics of the mass and to compare with the typical masses of the thyroid like being mobile with swallowing or protrusion of the tongue.

In our case the big cyst was mobile with neither swallowing nor protrusion of the tongue so we didn't consider it at the expense of thyroid gland. However, we performed FNA to rule out malignancy.

Imaging studies are very helpful to accurately describe the cyst in

addition to discover whether there are another cysts in other organs.

Ultrasound is the first-line modality to evaluate any neck mass. It is useful to distinguish between solid and cystic lesions. It can suggest the hydatid cyst as a differential diagnosis of cystic lesions based on many features like detachment of the membrane, multilocular appearance and peripheral calcifications as in Gharbi's classification [14].

Ultrasound sensitivity and specificity are estimated to be 90–95% [15,16].

Computed tomography “CT” and magnetic resonance imaging “MRI” have also high sensitivity and specificity. They are indicated to determine the relations between the cyst and adjacent structures when they are not clear at ultrasound [17,18].

FNA is widely performed to evaluate thyroid lesions and other cystic lesions in the neck. It is not widely accepted in case hydatid cyst is a deferential diagnosis since it could cause anaphylactic reactions and may disseminates the disease [17,18].

However, no complications have been reported in patients whom hydatid cyst disease incidentally diagnosed after histopathology examination, like in our case [19].

More studies are needed to evaluate the safety of performing FNA of hydatid cysts in the neck.

Histopathological study makes the definitive diagnoses of hydatid cyst disease [13,20].

The best treatment of thyroid hydatid cysts is total surgical excision of all cysts with intensive care not to spread the disease in case of cyst rupture and dissemination of the scolices.

Treatment with anti-parasitic drugs such as Mebendazole and Albendazole is recommended before surgery to prevent contamination by cyst contents during surgery. Anti-parasitic drugs may be described as the only treatment in patients who can't have surgery, in this case they should be followed up for long time [21,22].

Post-operative surveillance is important to diagnose the complete healing or recurrence. It is recommended to have an ultrasound examination in 3 months, 6 months and one year after surgery. Serology can be performed 6 months and one year after surgery.

Computed tomography can be obtained in suspected recurrence [23].

4. Conclusion

Hydatid cyst disease in the neck is extremely rare especially in western population. Physicians should be aware of this disease and hydatid cyst should be included in differential diagnosis of cystic lesions in the neck especially in patients from endemic countries. Post-operative surveillance is important to diagnose the complete healing or recurrence of hydatid disease in operated or other locations but also for follow up of replacement therapy after total thyroidectomy as the patient require lifelong thyroid hormone replacement.

Ethical approval

This study was performed in accordance with the Declaration of Helsinki.

Consent for publication

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.

Guarantor

Dr. Mohamad Zuheir Al kazzaz.

Patient perspective

The patient was cooperative. She had the ability to breathe and to eat more comfortably after surgery.

Availability of data and materials

Non-digital data supporting this study are curated at Al-Mouwasat hospital, Damascus, Syria.

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CRediT authorship contribution statement

Mohamad Zuheir Al kazzaz was the supervisor of the patient's case at hospital and revised the paper.

Muhammad Ghanem performed the surgery, analyzed, interpreted the patient data and was a major contributor in writing the manuscript.

Alnour Soliman contributed to surgery and writing the manuscript. All authors read and approved the final manuscript.

Declaration of competing interest

The authors declare that they have no competing interests.

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Registration of research studies

N/A.

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