Isolated primary hydatid disease of thyroid presenting as a solitary nodule: A rare case report

Dileep Ramesh Hoysal[1], Srikanth Kulkarni[2]

From [1]Associate Professor, [2]Professor, Department of Surgery, BGS Global Institute of Medical Sciences, Bengaluru, Karnataka, India

Correspondence to: Dr. Dileep Ramesh Hoysal, Flat No. 305, Chirag Residency, 7/2, Mysore Road "A" Cross, Opp. Police Station, Byatarayanapura, Bengaluru, Karnataka, India. E-mail: dileephoysal@gmail.com

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ABSTRACT

Echinococcosis is a significant health problem in endemic areas with a prevalence rate of 2–6%. Hydatidosis is reported in the liver in 70% of cases and lungs 15%, and rarely <1% are reported in breast, bones, gallbladder, pancreas, thyroid, and ovaries. Primary hydatid cyst of the thyroid is a rare clinical entity. Review of literature reveals <60 cases of isolated hydatid cyst of the thyroid. Here, we present the case of primary hydatid cyst in thyroid presenting as a solitary nodule. Uncommon presentation of thyroid in endemic areas during initial radiological workup should raise suspicion and warrant careful further evaluation. Immunologic tests have low sensitivity and specificity and, hence, have a limited role. Neck ultrasonography demonstrating daughter cysts is characteristic of hydatid cyst. Although enucleation and intracystic injection of albendazole are suggested, excision of cyst avoiding spillage of contents is currently the approved treatment strategy. Surgical excision with perioperative albendazole gives excellent results with low recurrence.

Key words: *Echinococcosis, Hydatid cyst, Hydatidosis, Thyroid nodule*

Hydatid disease is a significant health problem with a prevalence rate of 2–6% in endemic areas. Hydatid cyst disease is commonly seen in the liver in 70% of cases and lungs in 15%, and rarely in <1% of cases, it is reported in breast, bones, gallbladder, pancreas, thyroid, and ovaries [1,2].

Primary hydatid cyst of the thyroid gland is a rare clinical entity, even in the endemic areas. Review of literature reveals <60 cases of isolated hydatid cyst of thyroid [3-7]. Hydatid cyst in the thyroid is a rare clinical entity and can pose diagnostic and treatment challenges if a careful evaluation is not done and the same has been reflected in this case report.

CASE REPORT

A 14-year-old female presented with swelling in front of her neck of 4 years duration which was painless and progressive with no history of hyper- or hypo-thyroidism or pressure symptoms.

General examination and vitals were unremarkable. On local examination, solitary smooth, non-tender, firm, oval swelling of size 5 × 3 cm in the thyroid region was noted. Clinically, the patient was diagnosed to have solitary nodule in the left lobe of thyroid gland.

Ultrasonography (USG) of the neck revealed a well-marginated, hyperechoic, multivesicular cystic lesion (suggestive of daughter cysts) with positive split wall sign (Fig. 1). Serum *Echinococcus* immunoglobulin G (IgG) antibodies were positive. Diagnosis of hydatid cyst was made based on USG neck findings and positive IgG titer on enzyme-linked immunosorbent assay (ELISA).

Radionuclide thyroid scintigraphy revealed a large cold nodule involving almost entire left lobe. Systemic workup with USG abdomen and magnetic resonance imaging of the brain showed no evidence of any other cysts. Fine-needle aspiration cytology (FNAC) was not done due to the risk of anaphylaxis and spillage.

The patient was posted for the left hemithyroidectomy with all the necessary precautions to avoid the spillage of contents (Fig. 2). Cetrimide was used intraoperatively as the scolicidal solution to prevent the dissemination of the disease. Well-encapsulated cyst occupying almost entire left lobe of the thyroid was noted. The right lobe on intraoperative examination was normal. Hence, the left hemithyroidectomy was done. Cut surface shows a collapsed cyst wall (Fig. 3).

Post-operative period was uneventful and the patient was started on the post-operative course of albendazole 400 mg BD (3 cycles). Normal thyroid tissue with an encapsulated cystic lesion was seen. Histopathological examination with hematoxylin and eosin stain under ×40 showed cyst wall with laminated membranes (Fig. 4). Outer acellular laminated layer and a thin inner germinal layer along with scolices and hooklets along the capsule. All the above-mentioned features are suggestive of hydatid cyst. The patient is on close regular follow-up once in 3 months. No recurrence is noted till date.

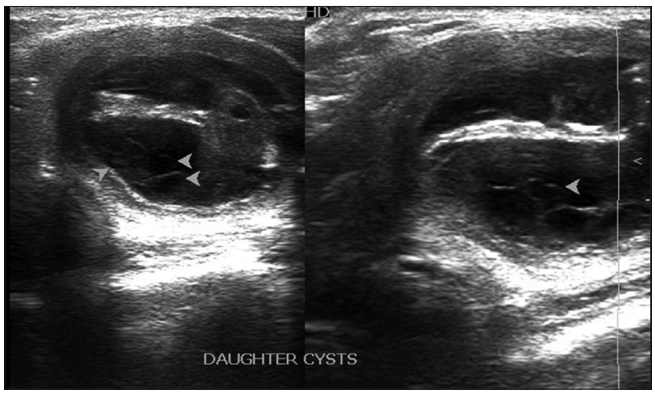

Figure 1: Ultrasound neck showing multivesicular cystic lesion with daughter cysts

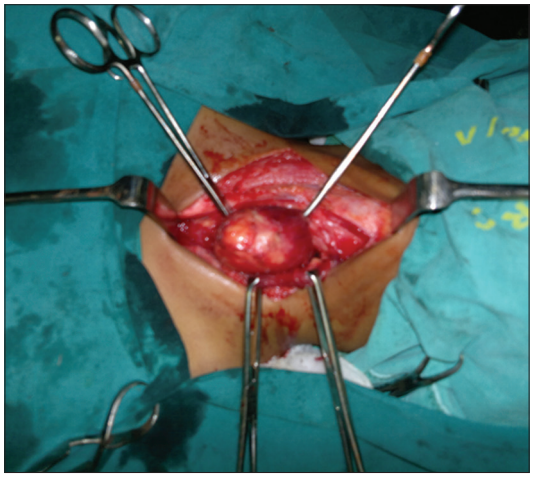

Figure 2: Intraoperative picture showing the left lobe of the thyroid

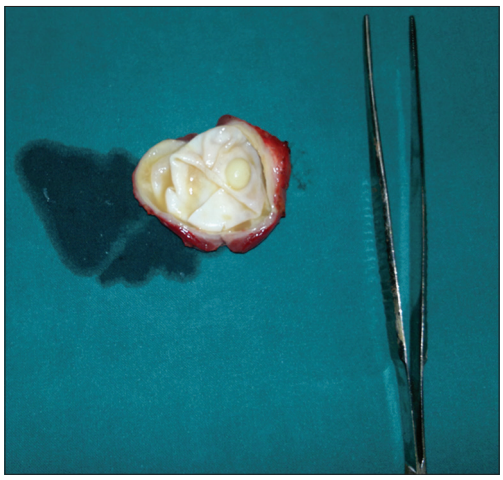

Figure 3: Cut surface of hydatid cyst wall

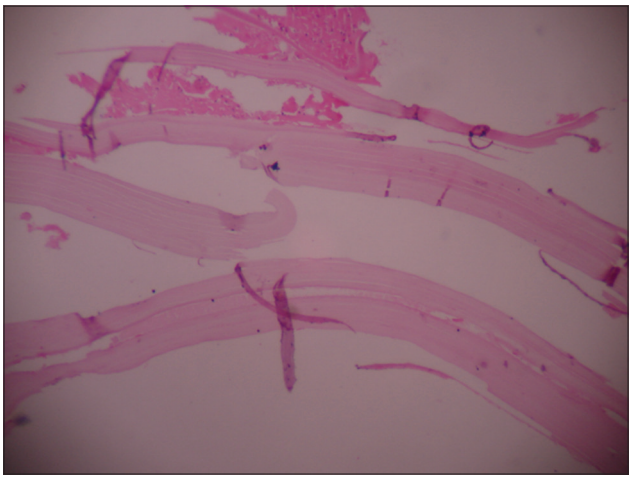

Figure 4: Histopathology on H and E stain under ×40 shows cyst wall with laminated membranes

DISCUSSION

Hydatidosis is a significant health problem, especially in the region where echinococcosis is endemic. Hydatid disease is endemic in the Mediterranean region, South America, Central and South Asia, and Southern Africa. In India, the high incidence is noted in Andhra Pradesh and Madras region. The prevalence rates of 2–6% or higher have been recorded [1,2]. Primary hydatid cyst of the thyroid gland is a very rare clinical entity with <1% prevalence, even in the endemic areas [8].

Diagnosis of thyroid hydatid cyst is made through a comprehensive multipronged approach using serological tests, imaging, and pathological confirmation. Indirect hemagglutination, ELISA, immunoelectrophoresis, and latex agglutination are the serological tests approved for hydatidosis. In a retrospective study of six cases of the primary hydatid cyst of the thyroid gland, hydatid origin was thought in 50% of patients and immunological tests showed 33% false-positive rates. The authors were of the opinion that the pre-operative diagnosis could be difficult based on serological tests [5]. However, in this case, a positive titer for IgG antibodies was noted. Serologic examinations have low sensitivity and specificity and are of limited use [6].

Evaluation of hydatid disease has been greatly facilitated by radiological imaging. USG is extremely sensitive in detecting daughter cysts and germinal vesicles which are the characteristic feature for pre-operative diagnosis of hydatid cyst. The multiple hypoechoic images noted within the cystic lesions are thought to represent germinal vesicles of a hydatid cyst [7,9].

Histological confirmation is diagnostic of hydatid disease. FNAC reveals characteristic findings of the fragments of hyaline, laminated cyst wall membrane, and confirmatory findings of scolices and hooklets in the smears [10]. However, a few cases of anaphylaxis and dissemination due to FNAC have been reported in literature [11].

The left hemithyroidectomy done in this case proved to be curative. The current recommendations in the treatment of thyroid hydatid cysts are complete removal of the cyst with avoidance of the spillage of contents, which may induce anaphylaxis. In difficult cases, enucleation can be tried, but it is associated with local relapse due to the presence of daughter cysts in the adjacent tissues. Perioperative albendazole has resulted in fewer recurrences. Intracystic injection of albendazole is a novel approach; however, its efficacy and benefit are yet to be determined [12].

CONCLUSION

Uncommon presentation of a thyroid finding in the endemic areas during the initial radiological workup should raise the suspicion and warrants careful further evaluation and management. USG of the neck is highly sensitive in diagnosing hydatid disease with a characteristic demonstration of daughter cysts. Complete surgical excision along with perioperative albendazole remains the gold standard treatment strategy and gives excellent results with low recurrence.

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