

Case Report

Hydatid cyst of the submandibular region

Hossain MF¹, Rahman M², Amin R³, Ahmed T⁴, Karmaker A⁵

Abstract

Hydatid cyst is caused by larval stage of *Echinococcus* parasite, mainly/largely by *E. granulosus*. It usually involves one organ. It may affect many organs also. Hydatid cyst of the head and neck region is uncommon and involvement of submandibular region is very rare. Here we present a case of left submandibular region hydatid cyst. This patient is an elderly gentleman of age 61 years from Soydabad, Sirajgonj. He noticed a painless peanut size swelling in his left submandibular region 5 years back. It had started increasing in size for last 4 months and become cricket ball size. Examination revealed a soft, nontender lump in that region. Skin and subcutaneous tissues are free of lump adherence. Chest X-ray was unremarkable. CT Scan showed a benign cystic swelling. Excision was performed and pathological examination revealed a hydatid cyst.

Keywords- *Echinococcus*, Hydatid cyst, Submandibular region, Scolicidal agent.

Introduction

Hydatid disease, a zoonotic infection, caused by larval form of tapeworms of the genus *Echinococcus*, found in the small intestine of CARNIVORES. It is still an important health problem in Endemic regions^{1,2}. This disease is more frequent in the middle east, central Europe, Australia and south America³. Two of the four recognized species of *Echinococcus*: *E. granulosus* and *multilocularis*, cause cystic and alveolar Echinococcosis in human body respectively. Hydatid cyst which generally involve the liver and the lungs are uncommonly found in submandibular region even in endemic zones². Involvement of this region is extremely uncommon, because implantation at this site would require passage through the filters of the liver and lung⁴. We present a patient with lump in the left submandibular region for 5 years and later was found to have Hydatid disease.

Case Presentation

A 61 years aged gentleman MR. Abdur Rashid A-140501030282, R-140401089326, from Soydabad,

Sirajgonj, Bangladesh was referred to KYAMCH by local physician for a rapidly progressive swelling in his left submandibular region for 4 months, although patient had noticed this swelling 5 years back when it was peanut in size. 4 months back it had started growing rapidly and reached to a current cricket ball size. It is associated with mild pain, difficulty in chewing and swallowing for the same duration. No H/O trauma, excessive salivation, halitosis, discharge inside the oral cavity, chronic cough, fever, jaundice or any other systemic manifestation.

On examination he was afebrile. The skin overlying the soft, nontender, mildly mobile mass showed no sign of inflammation. The size of the mass was $\approx 8 \times 6$ cm. Intraoral examination revealed no stones in the left submandibular gland duct. Milking of the submandibular gland showed normal clear fluid. Examination of chest and abdomen were unremarkable. Biochemical investigation revealed UTI. CT scan of the neck revealed a large margined cystic lesion in the left lateral neck region.

1. Mohd. Farid Hossain, Assistant Professor, Department of General Surgery, KYAMCH, Enayetpur, Sirajgonj.

2. Masudar Rahman, Registrar, Department of General Surgery, KYAMCH, Enayetpur, Sirajgonj.

3. Ruhul Amin, Registrar, Department of General Surgery, KYAMCH, Enayetpur, Sirajgonj.

4. Tanvir Ahmed, Medical Officer, Department of General Surgery, KYAMCH, Enayetpur, Sirajgonj.

5. Apu Karmaker, Intern, Department of General Surgery, KYAMCH, Enayetpur, Sirajgonj.

Correspondence : Dr. Mohammad Farid Hossain, Asst. Prof. Dept. of General Surgery, KYAMCH, Enayetpur, Sirajgonj.

Email: drfarid85@yahoo.co.in

FNAC of the swelling revealed clear fluid with no malignant cell in it suggestive of benign cystic lesion. With the impression of a benign cystic mass in the left submandibular region, Surgical Exploration was initiated with Horizontal incision about 4 cm below the lower edge of the left mandible. After dividing platysma muscle, a cystic swelling was found. Its lower part was pearly white in color suggestive of classical appearance of hydatid endo cyst. But superior medial wall could not be delineated properly due to dense, inseparable muscle adherence with it. Left submandibular salivary gland appeared normal, although its inferior surface was densely adhere with the cystic wall and was inseparable. During mobilization, Cyst got ruptured, cystic fluid spilled, daughter cysts were noted inside and about to exit. Immediately 10% povidon iodine solution soaked sponges were placed all around the cyst. Ruptured area was repaired to stop further leakage. Entire cyst was removed along with few muscle fibers and submandibular nerve fibers which were inseparable from cyst wall. Cut section of the cyst showed numerous daughter cysts and clear fluid. Patient was discharged on POD2 with features of neuropraxia at the left mandibular region and tongue. Later on pathological report revealed Hydatid cyst. Patient was given Tab. Albendazole 400 mg 8 hourly for 28 days to prevention of Echinococcosis recurrence.



Figure: 1



Figure 2

Discussion

Hydatid cyst of submandibular region is very rare^{5,6}. In a large series, the distribution of hydatid cysts outside the liver and lungs were reported as 9% only⁷. Reports of primary submandibular region hydatid cyst is very rare. we could find a single case after reviewing of literature. In our case, pt had not undergone any previous surgery for hydatid cyst and no hydatid cysts were found in other organ. Therefore pt was diagnosed as having primary hydatid cyst. The mechanism of primary localization of hydatid cyst in submandibular region is unclear.

After being ingested orally, under the act of gastric and intestinal enzyme, the oncosphere is released; it penetrates the intestinal wall, join the portal system and reaches liver. If the egg attached to the liver, an hepatic hydatid cyst develops. These eggs can pass to the systemic circulation and cause disease in other end organs. Larvae must pass through two filters (Liver and Lung) to form a solitary hydatid cyst, but this is very difficult. It is very much possible that systemic dissemination via lymphatic route may accounts for cases with solitary cysts in uncommon sites⁸. Direct spread from adjacent sites may be another mechanism of infection, provided a micro puncture has occurred⁹.

It is important to establish the diagnosis preoperatively in order to limit the risk of anaphylactic shock or dissemination of viable protoscolices in the event of puncture or accidental opening of the cyst during resection. Chest radiography, ultrasound, computed tomography (CT Scan) and magnetic resonance (MR) imaging are all can be used to depict hydatid cyst. However the imaging method to be used depends on the organ involved and the growth stage of the cyst. Submandibular region hydatid cyst can cause a variety of diagnostic problems specially in absence of typical radiological findings. That had happened in our case where CT Scan suggested a simple (benign) cyst.

Nevertheless, USG is the diagnostic test of choice for the initial work-up and is particularly useful for detection of the floating membranes, daughter cysts and hydatid sand in purely cystic lesion¹⁰. It is also used for staging and classification of *E. granulosus* infection¹⁵. CT Scan is best for detecting cyst wall calcification and modality of choice in peritoneal seedling¹¹. Both CT and MR imaging can detect cyst wall defects as well as the complication such as rupture and infection of cysts¹².

Immunodiagnosis can also play an important complementary role for primary diagnosis and also for follow up of pt's after surgical or pharmacological treatment. Detection of antibody against E.granulosus in sera remains the method of choice¹³.

Surgical treatment remains the standard therapy since there is no response to drug administration¹⁴. Complete total excision offers best hope for permanent cure¹ without opening the cyst. If the cyst cannot be excised without opening the fluid contents should be removed first. The laminated membrane should be totally excised and the cyst pouch should be irrigated with protoscolicidal solution¹⁵.

Subcutaneously located cysts are more prone to rupture since they have not been diagnosed preoperatively. In our case cyst got ruptured while mobilizing, we have mopped the operative field with scolical agent soaked sponges. Hypertonic saline (15-20%), cetrimide (0.5%), chlorhexidine, hydrogenperoxide, ethyl alcohol (70-95%), povidine iodine (10%) are some of the compounds used as scolical¹. Used povidine iodine (10%) because of its ready availability in the operation theater. Spillage of daughter cysts result in recurrence. Concomitant drug treatment with antihelminthics such as Albendazole or praziquantel reduces the risk of secondary echinococcosis and recurrence^{1,16}.

Table-1: Incidence of hydatid cyst affecting various

Organs/tissues	Incid
Liver	60%
Lung	30%
Kidney	2.5%
Heart	Less than 2%
Spleen	
Brain	
Bone	
Orbit	Only few cases reported
Urinary bladder	
Spinal extradural space	
Breast	Few cases
Submandibular gland	One case
Thyroid	
Muscle	Few cases

Even through hydatid cysts are abundantly found in the liver, lung and other areas, to our knowledge there are no reports to date concerning submandibular region involvement in Bangladesh, although few cases were reported in other countries. Therefore hydatid cyst should be included in the differential diagnosis of submandibular lesion lump.

Conclusion

For submandibular region lump, hydatid cyst is a differential diagnosis. Sometimes, Preoperative diagnosis is difficult. Total excision cure the disease. Therefore meticulous dissection of the cyst without rupturing the wall is the key of surgery.

References

1. Guidelines for treatment of cystic and alveolar echinococcosis in human. WHO Informal Working Group on Echinococcosis. Bull World Health Organ 1996;74:231-42.
2. Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. Clin Microbiol Rev 2004;17: 107-35
3. Kayaalp C: Hydatid cyst of the liver. In Surgery of the Liver, Biliary Tract, and Pancreas 4th edition. Edited by: Blumgart LH, Belghiti RJ, DeMatto RP, Chapman WC, Buchler MW, Hann LE, D'Angleca M. Philadelphia, PA: Saunders Elsevier; 2007:952-970.
4. Saswata Bharati, Mrityurjay Pal: Primary hydatid cyst in gastrocnemius Muscle. Nigerian Journal of Surgery Jan- Jun 2012, Vol. 18, Issue 1, 19-21
5. Gures N, Baysal K: Hydatid cyst in thyroid and submandibular salivary gland in a child. Z Kinderchir: 41(6):362-363, 1986.
6. Singh S: Submandibular hydatid cyst. J Laryngol Otol 86 (6): 647-650, 1972.
7. Prousalidis J, Tzardioglou K, Sgouradis L, Katsolis C, Alertan H: Uncommon sites of Hydatid disease. World J Surg 1998;22:17-22.
8. Engin O, Erdogan M: Solitary subcutaneous hydatid cyst. Am J Trop Med Hyg 2000, 62:583-584.
9. Safioleas M, Nikiteas N, Stamatakos M, Safioleas C, Manti CH, Revenas C, Safioleas P: Echinococcal cyst of the subcutaneous tissue, A rare case report. Parasitol Int. 2008, 57:236-238
10. Turgut AT, Akhn O, Bhatt S, Dogra VS. Sonographic spectrum of hydatid disease. Ultrasound Q 2008;24:17-29
11. WHO Informal Working Group. International classification of ultrasound images in cystic echinococcosis for application in clinical and field epidemiological settings. Acta Trop 2003;85:253-61

12. Pedrosa I, Saiz A, Arrazola J, Ferreiros J, Pedrosa CS. Hydatid disease: Radiologic and pathologic features and complications. *Radiographic* 2000;20:795-817
13. Zhang W, Li J, McManus DP. Concepts in immunology and diagnosis of hydatid disease. *Clin Microbiol Rev* 2003;16:18-36
14. Schwartz SL: Liver. In: Schwartz SI, Shires GT, Spencer FC, Huser W C (eds.), *Principles of Surgery*. New York:McGraw-Hill, pp. 1319-1366,1994.
15. Duncan GJ, Tooke SMT: Echinococcus infestation of the biceps brachii. *Clin Orthop* 1990, 261:247-250.
16. Shams-Ul-Bari, Arif SH, Malik AA, Khaja AR, Dass TA, Naiko ZA. Role of albendazole in the management of hydatid cyst liver. *Saudi J Gastroenterol* 2011;17:343-7
17. N A M Golam Mahbub, Unusual Presentation of Hydatid Cyst, *TAJ* 2005; 18(2): 137-139