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Primary Hydatid Cyst of the Submental Neck Region in a Child: A Rare Case Report with Surgical Management

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ABSTRACT

Hydatid cyst of the neck considered rare condition. In literature there are few published case reports, but none involved the submental region. This report is about a 11 years old boy who had a swelling at the submental region of few months duration. Examination revealed a swelling that extended to anterior part submandibular region, red overlying skin, no discharge, sound teeth. Radiology revealed multilobulated cystic mass. Surgery done under general anesthesia, the cyst completely removed and sent to histopathology examination which concluded as hydatid cyst. Albendazole was prescribed with subsequent follow-up for 6 months with no recurrence.

Keywords: Hydatid cyst, Submental region, Neck mass, Pediatric patient, Echinococcosis, Surgical management, Albendazole therapy

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INTRODUCTION

Hydatid cyst (HC) is one of the known parasite infections of human body. It is caused by Echinococcus species. Among others, E. granulosus possibly is the most common responsible of cystic formations. Other species which may cause the disease include E. multiocularis, E. oligarthrus and E. vogeli.[1] The most common sites to be infected are liver (50-77%), Lungs (15-47%), Spleen (0.5-8%) and Kidneys (2 -4%). Occurrence of HC in other parts of the body is not common.[1] Primary HC of subcutaneous sites sometimes can be seen and a solitary HC accounts for 1.6%.[2]

Through the search of literature, there were few published case reports of HC in the neck. In the neck, possibly the favorite site is the thyroid gland reported by many published materials.[1, 3-12] The following common site in the neck are salivary glands[13] and subcutaneous.[14,15]

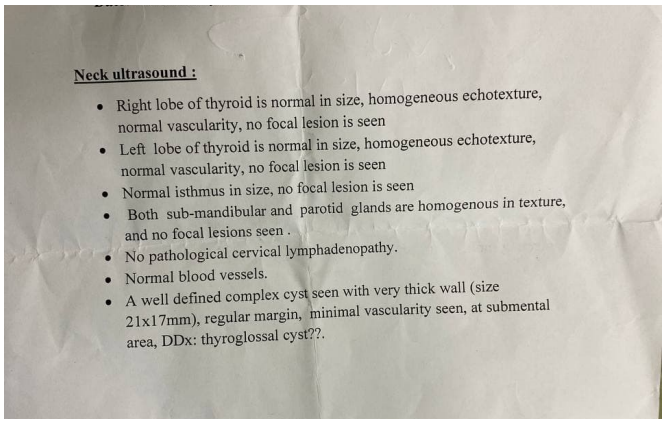
Here we are reporting a case of HC in the submental region of the neck

Case Report

An 11 year old boy presented with large swelling of submental region of few weeks duration that was painful and gradually increasing. Physical examination revealed large mass at submental region covering the area between the chin, hyoid bone and anterior part of submandibular space. Red discoloration of skin in some parts, soft to palpation and tender, fixed to surrounding tissues (figure 1). Intra oral examination revealed no carious and/or destructed teeth and normal mucous memebran.

Figure 1: Clinical picture before surgery

The ultrasound of neck reported a large complex cystic lesion with very thick wall, size 21*17 mm, minimum vascularity with regular margins. The neck otherwise was with no pathological features (figure 2).



Neck ultrasound :

- Right lobe of thyroid is normal in size, homogeneous echotexture, normal vascularity, no focal lesion is seen
- Left lobe of thyroid is normal in size, homogeneous echotexture, normal vascularity, no focal lesion is seen
- Normal isthmus in size, no focal lesion is seen
- Both sub-mandibular and parotid glands are homogenous in texture, and no focal lesions seen .
- No pathological cervical lymphadenopathy.
- Normal blood vessels.
- A well defined complex cyst seen with very thick wall (size 21x17mm), regular margin, minimal vascularity seen, at submental area, DDx: thyroglossal cyst??.

Figure 2: Ultrasonography report

The contrast CT of neck revealed a multilocular cystic lesion with multi-lobular outline, showing post contrast peripheral rim enhancement, size 2*2.5 cm, with multiple enlarged lymph nodes of submandibular region, suggesting an abscess (figures 3 and 4).

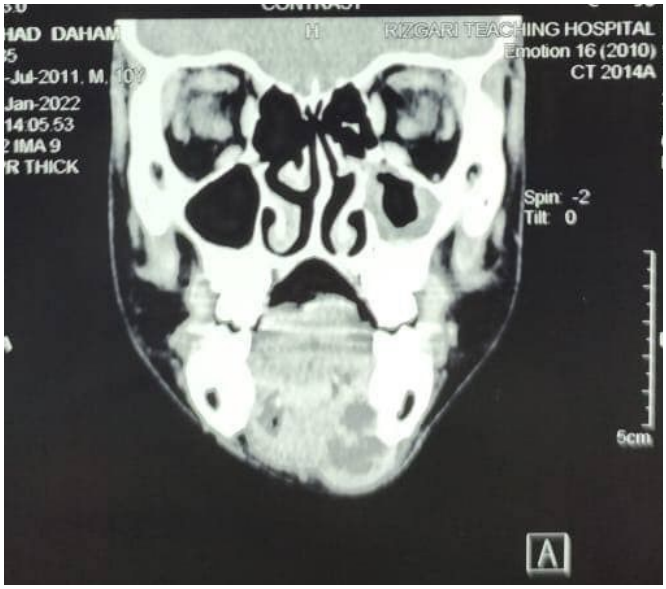
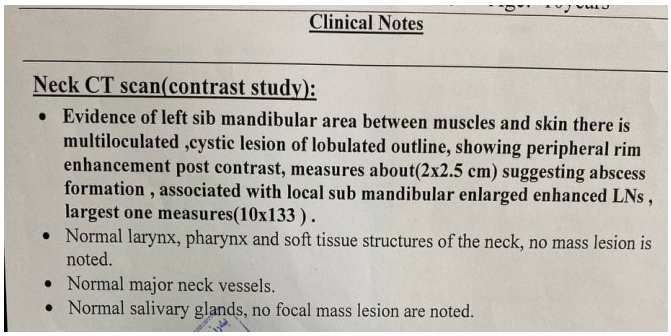


Figure 3: Coronal view of CT of the head and neck with report

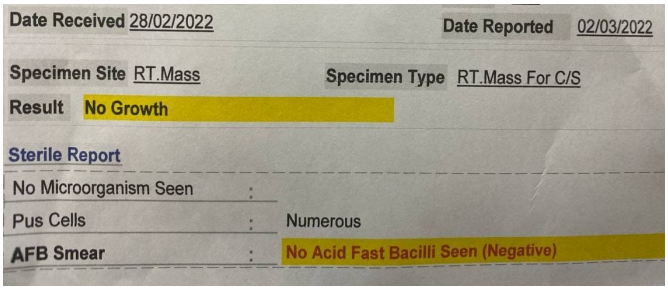


Clinical Notes

Neck CT scan(contrast study):

- Evidence of left sib mandibular area between muscles and skin there is multiloculated ,cystic lesion of lobulated outline, showing peripheral rim enhancement post contrast, measures about(2x2.5 cm) suggesting abscess formation , associated with local sub mandibular enlarged enhanced LNs , largest one measures(10x133) .
- Normal larynx, pharynx and soft tissue structures of the neck, no mass lesion is noted.
- Normal major neck vessels.
- Normal salivary glands, no focal mass lesion are noted.

Figure 4: Coronal view of CT of the head and neck with report



Date Received 28/02/2022 Date Reported 02/03/2022

Specimen Site RT.Mass Specimen Type RT.Mass For C/S

Result No Growth

Sterile Report

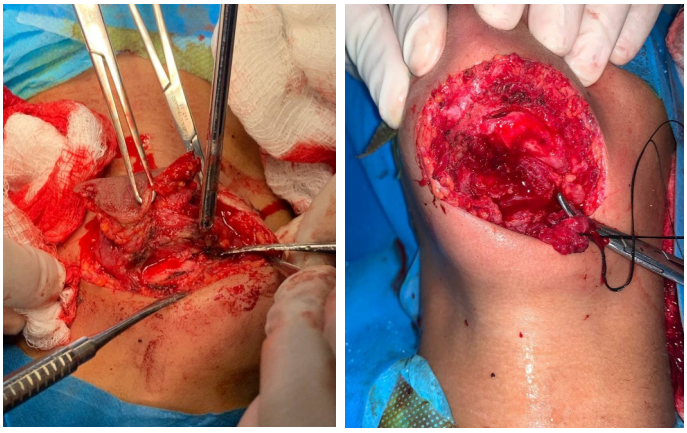
No Microorganism Seen	:	
Pus Cells	:	Numerous
AFB Smear	:	No Acid Fast Bacilli Seen (Negative)

Figure 5: Bacteriology report of antimicrobial susceptibility

The aspiration culture and sensitivity test showed no growth, multiple puss cells and negative for acid fast bacilli (figure 5).

Surgery

Under general anesthesia the cyst was approached through submental horizontal elliptical incision extended to the anterior part of submandibular space. The red fixed part of skin was included within the elliptical incision. Carful dissection revealed heavy granulation tissue attached to the capsule like tissue.

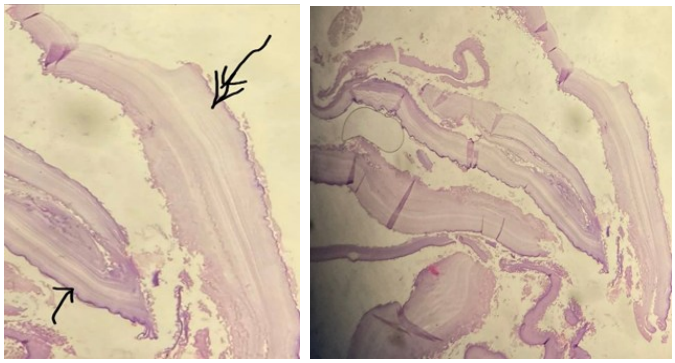


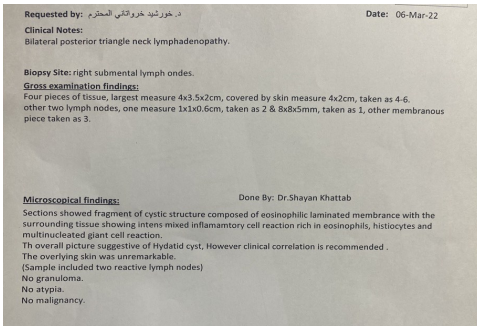
Figures 6 and 7: The operation site during surgery

The pathology resected down to the muscular base.

The site was treated with hydrogen peroxide 3% for few minutes then irrigation with normal saline. Suturing layer by layer (figures 6 and 7). The antibiotic therapy was Ceftriaxone vial 250 mg by two and metronidazole 250 mg by three for ten days.

The histopatholgical examination showed multi laminated tissue with conclusion of hydatid cyst (figures 8 and 10).





Requested by: د. خورشید خروانئی المحترم Date: 06-Mar-22

Clinical Notes:
Bilateral posterior triangle neck lymphadenopathy.

Biopsy Site: right submental lymph ondes.

Gross examination findings:
Four pieces of tissue, largest measure 4x3.5x2cm, covered by skin measure 4x2cm, taken as 4-6. other two lymph nodes, one measure 1x1x0.6cm, taken as 2 & 8x8x5mm, taken as 1, other membranous piece taken as 3.

Done By: Dr.Shayan Khattab

Microscopical findings:
Sections showed fragment of cystic structure composed of eosinophilic laminated membrance with the surrounding tissue showing intens mixed inflamamtory cell reaction rich in eosinophils, histiocytes and multinucleated giant cell reaction.
Th overall picture suggestive of Hydatid cyst, However clinical correlation is recommended .
The overlying skin was unremarkable.
(Sample included two reactive lymph nodes)
No granuloma.
No atypia.
No malignancy.

Figures 8–10: Histopathological examination and report

After above mentioned diagnosis by HPE, screening for chest and liver done, both were clean. Three cycles of Albendazole therapy was applied with following regimen: 15 mg/kg in two divided doses/day for 28 days, followed by 14 days drug free period.

Six months post operatively the patient was rechecked for recurrence and/ or occurrence of HC in other organs. Both chest x-ray and ultrasonography revealed no pathology (figures 11 and 12). The site of operation was silent with no features of recurrence.

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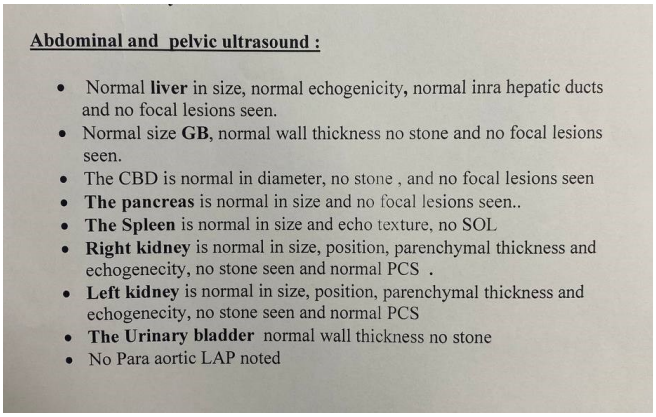
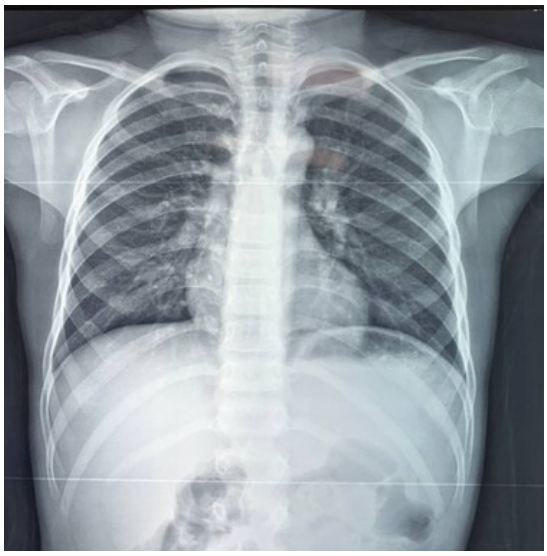


Abdominal and pelvic ultrasound :

- Normal **liver** in size, normal echogenicity, normal inra hepatic ducts and no focal lesions seen.
- Normal size **GB**, normal wall thickness no stone and no focal lesions seen.
- The CBD is normal in diameter, no stone , and no focal lesions seen
- **The pancreas** is normal in size and no focal lesions seen..
- **The Spleen** is normal in size and echo texture, no SOL
- **Right kidney** is normal in size, position, parenchymal thickness and echogenecity, no stone seen and normal PCS .
- **Left kidney** is normal in size, position, parenchymal thickness and echogenecity, no stone seen and normal PCS
- **The Urinary bladder** normal wall thickness no stone
- No Para aortic LAP noted



Figures 11 and 12: Ultrasonography of the abdomen and pelvis with chest X-ray to detect any lesions in other organs

One year post operative follow up visit no revealed no pathology and no recurrence. So, the patient was dismissed.

DISCUSSION

As was mentioned above, it is not common to see HC in the neck region. The most common swelling in the upper neck are odontogenic infections, salivary gland pathosis and lymph nodes. Rare masses may include, thyroglossal cyst, dermoid, branchial clefts, carotid body tumor, tuberculosis and few others. In our case, we have excluded odontogenic infection both clinically and radiologicaly. The fact that the mass was below muscle layer under the skin of submental region, and ultrasonography reported normal glands, so salivary gland pathosis was excluded too. The texture, multilobulation was not going with dermoid cyst. Clinically and radiologically (ultrasound and CT) both thyroglossal cyst and vascular malformation were excluded too. The location, ultrasound and CT examination were not in favor of carotid body, branchial clefts. The culture and sensitivity test excluded abscess and tuberculosis.

Depending on above mentioned facts, the surgery was done for unknown mass as excisional biopsy. The antibiotic therapy was that of any possible infection in the region. The same challenge of diagnosis was reported by Ghanem et al (2021); Iranpour et al (2018); and majority of authors. The main tool definite diagnosis was histopathological examination. Since all other organs were clean from pathology, authors considered the case as primary HC of submental region.

It is common practice to do aspiration for cystic lesions and FNA for solid lesions. The same was done in this case mainly to differentiate vascular, abscess and usual cysts. However, such maneuver is not recommended in case of HC, since there is chances of anaphylactic reaction. The same procedure done by other authors who managed HC in the neck. The reason is the rarity of this disease in the neck.

Management of HC is challenging. However, there is guidelines for management. There are three options for management of HC: surgical, medical and combined. Recently an other option has been suggested with good prognosis for hepatic HC. The technique called PAIR that stands for: puncture, aspiration, injection, and re-aspiration.[16] This technique may be considered the best option unless we are talking about undiagnosed HC that obligate the surgical approach with or without chemotherapy. In current case the surgical approach yield good result in combination with Albendazole therapy. The reason for prescribing the last one was possibility of seeding during surgery.

In endemic countries like those in middle east, there should be HC in the list of differential diagnosis of any cyst in the neck region. Both ultrasound and CT can be helpful tools to expect such pathologies when ever they reported thick wall lobulated cyst. Authors recommending not to do aspiration and FNA for cystic swellings in the neck before ultrasound and CT reports, to prevent anaphylaxis if the case was HC.

CONCLUSION

Hydatid cyst is rare pathology of neck. However, care must be taken to rule out HC whenever there was a cystic swelling in the neck.

ETHICAL CONSIDERATION

The case was performed in accordance with the Declaration of Helsinki.

PATIENT APPROVAL

Patient's permission was granted for publication.

CONFLICT OF INTEREST

We declare no conflict of interest.

FUNDING

The study got no fund from any organization.

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