



Case Report

Cystic Hygroma of the Neck in Adult Patient

Burkan Nasr^{1*}, Abdulhakim Al Tamimi², Mohmmed Issa³, Ali Amri³, Saeed Al Bahlooli⁴, Anwar Al Jounaeed¹, Ahmed Ramadan⁵, Abdulkarim Saleh⁶, Abdulfattah Al Domini⁶

Received: 03 December 2021; Accepted: 10 December 2021; Published: 17 December 2021

Citation: Burkan Nasr, Abdulhakim Al Tamimi, Mohmmed Issa, Ali Amri, Saeed Al Bahlooli, Anwar Al_Jounaeed, Ahmed Ramadan, Abdulkarim Saleh, Abdulfattah Al Domini. Cystic Hygroma of the Neck in Adult Patient. Journal of Surgery and Research 4 (2021): 778-783.

Abstract

Cystic Hygroma is hamartomatous tumor of lymphatic channels, with Most commonly these malformations occur in the head and neck. neonatal and infant more effected and Presentation in adulthood is very rare, we report 25 years old female with diagnosis cystic Hygroma and discuss the presentation, diagnosis and management options for such a presentation.

Keywords: Cystic Hygroma; Adult; Neck cyst; Lymphangioma

1. Introduction

Cystic Hygroma is benign hamartomatous soft tissue tumor due to malformation of lymphatic channels, It can occur in cervico-facial regions mainly at neck, but can be occure in the head, axilla, groin [1]. In 1834 Wernker give name Cystic Hygroma for this pathology [2]. Cystic Hygroma or Lymphangioma is generally known as a disease of childhood, present as a congenital blockage of lymphatic system, because in this age there is active lymphatic growth. But quite

¹Department Surgeon, Saudi Hospital at Hajjah, Yemen

²Department of Surgery, Aden University, Yemen

³Department of Surgery, Sanaa University, Yemen

⁴Department of Surgery, Thamar University, Yemen

⁵Department of Anesthesia, Al Azhar University, Egypt

⁶Department of Radiology, Saudi Hospital at Hajjah, Yemen

^{*}Corresponding author: Burkan Nasr Rashed Shaif, MD, FEBS, MRCS, FRCS, FACS. Consultant General and Laparoscopic surgery Al Thawra Modern General/Teaching Hospital Sana`a and Suadi Hospital at Hajjah, Yemen.

uncommon present in adult age [3]. Presentation in adult age attributes to delayed proliferation of the congenital or acquired lymphoid rests following trauma or preceding respiratory infections [4]. They have no gender predilection, adult cystic hygroma asymptomatic presentation, present as a painless mass that progressively enlarges. Typically, the mass is soft, nontender and fluctuated. Symptoms may develop where it compresses surrounding tissue. There may be obstructive symptoms such as dysphagia, dysphonia, and airway obstruction, however, this is rare in adults [5]. Nearly 150 have been reported in the literature for adult cystic Hygroma, That uncommon adult cystic Hygroma make challenged in its diagnosis and management [6].

2. Case Presentation

A 25-year-old female came to Surgery Department with complaint of painless swelling on left side of neck since 2 years. Patient was asymptomatic 2 years back after which he noticed a swelling in the neck; which was initially single and small and had gradually increased. Patient not complain of dysphagia or dyspnea. Patient did not give any history of trauma or upper respiratory tract infection. On examination, there was a large swelling in the left side of neck measuring 12x 10 cm. The swelling was soft lobulated surface, non-tender, fluctuant, brilliantly translucent extending into both anterior and posterior triangle of the neck (Figure 1). The computed tomography with contrast showed a well-defined non enhancement,

lobulated, hypodense, multiloculated thin walled cystic lesion (Figure 2). From history, examination and computed tomography scan radiology findings provisional diagnosis as adult cystic Hygroma.

We prepared the patient for operation, complete all routine investigation, Complete blood count (CBC), Creatinin, Liver Function tests and chest X-ray was all within normal. Risk consent was obtained mainly explain incomplete cyst excision and neurovascular injury. The patient was taken under general anesthesia supine position head extended with tilt toward Left side, aspiration by syringe was clear yellow fluid (Figure 3), and result of aspirated fluid biochemistry was LDH 171U/L, Glucose 108mg/dl, RBC 188, WBC 620, PMN 60% and MONO 40%.

Transverse incision over the swelling meticulous homeostasis and dissection done (Figure 4) was complete lobulated cystic lesion excision with some adherence to internal jugular vein was released successful, send the specimen for histopathology, small tube draining was done inside residual cavity for one day, patient discharge next day with improve their general condition. The result of histopathology showed collagenous stroma with a dense lymphocytic infiltration, endothelium cell lined wide and large lymphatic channels, that histopathology and clinical examination and radiology findings consists with adult Cystic Hygroma.



Figure 1: Female 25 years adult with Cystic Hygroma

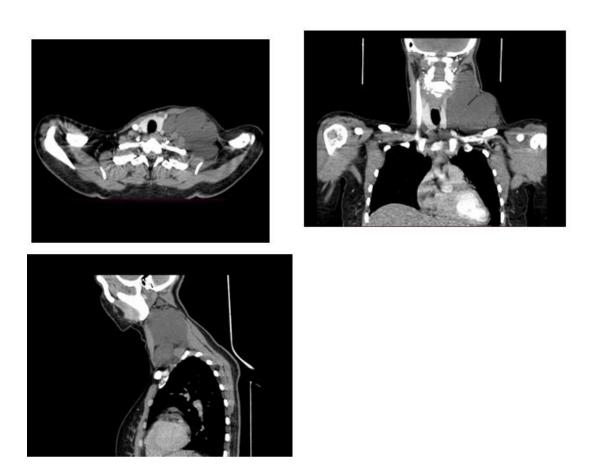


Figure 2: CT scan Lt Neck Cystic Hygroma



Figure 3: Fluid Aspiration straw clear fluid



Figure 4: Inoperative finding lobulated cystic lesion and clean field post cyst excision

3. Discussion

Cystic Hygroma, benign congenital or acquired malformation of lymphatic vessels that result of sequestration of lymphatic tissue that has retained its potential for growth or lymphatic system fails to communicate with the normal jugular vein [1,7]. More than 90% of cystic Hygroma present itself in pediatrics age period, present as a birth defect (congenital) neck swelling rarely became symptoms or present with complication, presentation in adulthood is

rare, and the cause controversial or uncertain, but mostly due to delayed proliferation of the congenital lymphatic malformation although trauma and upper respiratory tract infection have both been suggested as possible triggers for the onset [6,8]. In our case painless neck swelling, asymptomatic gradually increase in size and no compression symptoms, this presentation similar to cases report by Gow et al. [6] and Kalsotra et al. [9] they described the most common adult presentation as asymptomatic painless swelling

and very rarely compresses major structures of neck such as larynx, trachea, oesophagus, branchial plexus and great vessels. Bhatia, Varsha et al. [10]. In his case, it was described large swelling causing mass effect with patient complaining of difficulty in deglutition and pain in the right ear. Most commonly cystic Hygroma occurs in the head and neck, although they have been described in a variety of other axilla (20%), anatomical locations such as mediastinum (5%), groin, retroperitoneal space and groin [7]. Diagnosis in adults is considered to present a greater challenge than in children, and final diagnosis is usually based on postoperative histology [6]. Our patient underwent neck CT scan and ultrasonography (USG), show neck swelling in left posterior triangle with extended to left triangle. Morley SE [3] and Karakas O [11] report that neck CT scan and ultrasonography (USG) and Magnetic Resonant Imaging (MRI), it is essential to evaluate neck cystic hygroma and it's intrathoracic extension, which might be seen in 10% of cases. There are many differential diagnosis for neck cystic hygroma in adults like branchial cleft cyst, dermoid cyst, lipomas, hemangiomas, tumours and metastatic disease [6,12]. Although after clinical and radiological examination, the above-mentioned diagnosis may look similar to cystic hygroma but histopathology plays a critical role in differentiating it from others. Cystic hygroma could classified into septated (multiloculated) or nonseptated single cavity (nonloculated) types [13], our case was septated (multiloculated) cystic lesion. There are three histological subtypes of cystic hygroma- Cavernous lymphangioma is the most common subtype, capillary lymphangioma, and cystic lymphangioma [14]. Our case was of cystic lymphangioma. Total surgical excision was performed in this case. As surgical excision is the best management option for adult lymphangioma or cystic Hygroma. Cystic Hygroma consider difficult to manage due to locally aggressive and recurrence of the tumor following surgery, complete excision of a cystic hygroma has been shown to have an 81% cure rate with a recurrence rate of 18-21%. When only part of a lymphatic malformation is excised, there is an 88% recurrence rate [15]. Saxena et al. [12] described that cystic hygroma might infiltrate in to adjacent structures which can lead to recurrence if complete excision is not done. Different others management options alternatives to surgery for adult lymphangioma or cystic Hygroma, including laser surgery, cryotherapy, electrocautery, steroid administration, sclerotherapy, embolization and radiation therapy [7].

5. Conclusions

Cystic hygroma are rare presentation in adult age, usually asymptomatic, rarely complicated, has many differential diagnosis as cystic neck mass and complete surgical excision are best management option.

Declaration of patient consent

I obtained verbal consent from patient, that was understand like this publishing help to improve medical scientific activities in our country also I explained for the patient that name and identity will not be published.

Financial support and sponsorship

Nil.

Conflicts of interests

All procedures were conducted in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

References

- Potdar PD, Deshpande S, Chaugule S. Development and molecular characterization of cystic hygroma cell line to be used as an in vitro model system to study the progression of hygroma in young children. Pediatr Res Int J 13 (2013): 1-13.
- Rashid A. Cystic hygroma. Indep Rev 16 (2014): 141-144.
- Morley SE, Ramesar KC, Macleod DA. Cystic Hygroma in an adult: a case report. J R Coll Surg Edinb 44 (1999): 57-58.
- 4. Kumar VB, Martis S, Joseph J, et al. Rare case of neck swelling in an adult. Int J Med Health Sci 2 (2013): 373-375.
- Ali G, Akif A, Faik C. Cystic hygromas in adults: Reports of two cases. Bakirkoy Tip Derg 2 (2006): 101-103.
- Gow L, Gulati R, Khan A, et al. Adult onset cystic hygroma: A case report and review of management.
- Fidan V, Sütbeyaz Y. Lingual cystic lymphangioma in an elderly patient. Kulak Burun Bogaz Ihtis Derg 18 (2008): 260-262.

- 8. Kumar VB, Martis S, Joseph J, et al. Rare case of neck swelling in an adult. Int J Med Health Sci 2 (2013): 373-375.
- Kalsotra P, Prusty A, Gupta A, et al. Cervical cystic hygroma in an Adult. JK Science 3 (2001): 191-193.
- Bhatia, Varsha. Cystic hygroma in an adult male: A Diagnostic Challenge. Iranian journal of pathology 4 (2015): 310-313.
- Karakas O, Karakas E, Boyaci FN, et al. Cervicomediastinal giant cystic hygroma: A Case Report. J Clin Med Res 5 (2011): 61-63.
- Saxena P, Chandra D. Cystic Hygroma of neck in an adult female: Case Report & literature review. Int J Surg 22 (2010): 10-12.
- Mallick KC, Khatua RK, Routray S, et al. Cystic hygroma in adults - A rare case report. J Evol Med Dent Sci 3 (2014): 2561-2564.
- Guner Ali, Aydin A, Celik F. Cystic Hygromas in Adults: Reports of two cases. Medical Journal of Bakirkoy 2 (2006): 101-103
- 15. Rolekar NG, Shah DK. Recurrent cystic lymphangioma of the neck in an adult: A case report of rare entity. Int J Med Sci Public Health 3 (2014): 243-245.



This article is an open access article distributed under the terms and conditions of the Creative Commons Attribution (CC-BY) license 4.0