

Recurrent Cystic Hygroma In Adults: a rare case report

Dr.Dinesh Dutt Sharma¹,Dr Manish Kumar Mahala²,Dr Rishabh Bhargava³ Dr Ajay Malviya⁴

¹Assistant Professor Department of surgery,S.N Medical college Jodhpur

²IIIrd year Resident Department of surgery,S.N Medical college Jodhpur

³IIIrd year Resident Department of surgery,S.N Medical college Jodhpur

⁴Senior Professor and Unit head Department of surgery,S.N Medical college Jodhpur

DOI: 10.29322/IJSRP.9.12.2019.p9685

<http://dx.doi.org/10.29322/IJSRP.9.12.2019.p9685>

Abstract- Cystic hygroma is an aberrant proliferation of lymphatic vessels resulting from abnormal development of the lymphatic system^{1,2}.

Recurrent cystic hygroma can result from inadequate excision or specific tumour characteristics.³

We report a case of recurrent cystic hygroma in right axilla in a 18 year old male the surgical management of which was challenging due to large size, proximity to vital structure, unusual location.

Index Terms- Recurrent,Cystic hygroma,Adults,

I. INTRODUCTION

Cystic hygroma is a benign lymphatic malformation most commonly occurring in the cervicofacial region and age less than 2 years. It arises from sequestration of lymphatics or obstruction of lymphatic vessels⁴.Cystic hygroma occurs as a result of sequestration or obstruction of developing lymph vessels in approximately 1 in 12,000 births.⁴Although the lesion can occur anywhere, most common site is posterior triangle of neck.The mass may be apparent at birth or may appear and enlarge rapidly in early weeks or months of life as lymph accumulate; mostly present by age of 2 years.⁴

Diagnosis can be made clinically as swelling is soft, partially compressible and brilliantly transillumination on examination compare to other neck masses. Diagnosis is often aided by the use of fine needle aspiration for cytology, tissue histology, and ultrasound, MRI, or CT for definition of the mass. Loculations or cysts can often be appreciated and affected structures can be identified⁵.The modern management of lymphatic malformation includes combination of complete surgical excision and image guided sclerotherapy.⁴

II. CASE PRESENTATION

A 19 year old male, presented in surgery outdoor with complain of swelling over right upper chest since 5 days. Swelling was sudden in onset with no history of any preceding respiratory complaint or trauma. Patient had history of surgery for large mass at his age of 1.5yr, as told by patient(no document available).Patient is admitted for further management.

On examination swelling is visible over right upper chest, normal skin over swelling, linear horizontal scar visible just below right breast extending toward right axilla. On palpation swelling is soft cystic, non-tender, no local rise of temperature, swelling does not move in any direction, swelling is transilluminant and fluctuation present.

After admission of patient routine investigations performed. Chest x-ray is performed which suggest a soft tissue shadow near right shoulder joint which does not show any connection with pleural cavity. Patient sent for ultrasonography which suggest volume collection of around 100 ml, surrounded by irregular wall in intramuscular plane. This collection is inferiorly displacing subclavian vessels. On CT scan of chest, collection of size 6*8*7 cm seen in anterolateral chest wall and extending into right axillary region. The collection located in between pectoralis major and pectoralis minor muscles, collection showing focal loss of fat plane with right subclavian artery



Fig 1: CECT Thorax showing

The patient underwent surgical exploration of lesion, incision made over swelling along with skin crease extending towards axilla. The mass was encapsulated within a thin wall and adherent to pectoralis major and pectoralis minor muscles. Adhesions were divided by blunt dissection by artery forceps. Complete removal of wall of cyst was achieved. All the important nerves and arteries encountered during dissection are preserved. Macroscopically lesion measured 8*7 cm in horizontal and vertical dimensions. Histopathological examination revealed cystic space lined with endothelium like cells consistent with diagnosis of cystic hygroma.



Fig 2: Intra operative picture of Cystic

Hygroma anterior chest wall

Post-operatively patient recovered well without any movement restriction of shoulder joint. Patient discharged on postoperative day three after removal of suction drain and histopathological examination report received.

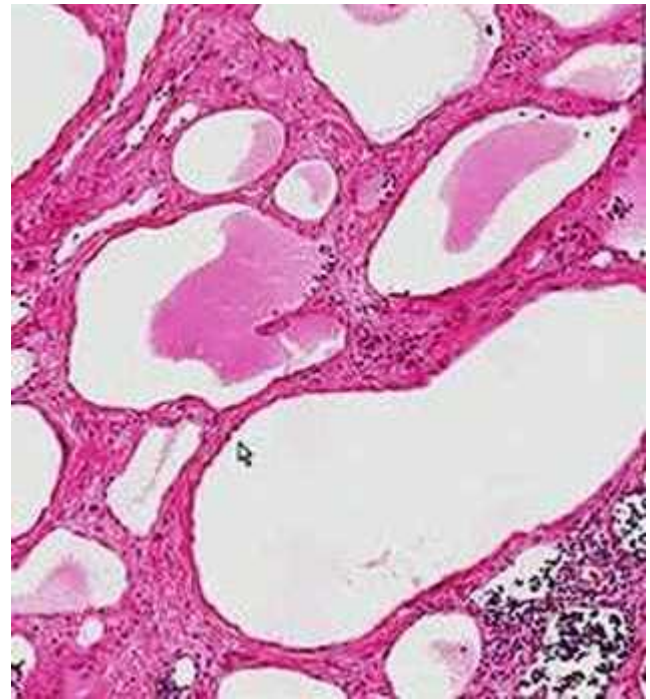


Fig 3: Represents Histopathological image of cystic hygroma

III. DISCUSSION

Most cystic hygromas present in-utero or in infancy and therefore most of the literature on management focuses on paediatric cases. Cystic hygroma considered to arise from congenital malformation of the lymphatic system in which a failure of communication between the lymphatic and venous pathways leads to lymph accumulation. The effect of these lesions depends on their position and their relationship to surrounding structures. Most common adult presentation is of a painless lump in an otherwise asymptomatic patient.⁶

Differential diagnosis for a cystic hygroma includes soft tissue sarcoma, abscess synovial cyst and haematoma.⁷ Although it was strongly suspected in initial examination as in this case patient had history of surgery in his childhood and swelling was transilluminant.

Pre-operative investigations were done to find out the location of the lesion and its relation to surrounding vital structures and also because of the extreme rarity of this condition in adults.

Treatment of cystic hygroma historically required surgical excision⁸. In this case it was considered that the ideal treatment would be complete surgical excision. Successful surgical excision has been found to correlate histology, encapsulation completeness.

Complications of surgery like scar, injury to blood vessels and nerves wound infection and recurrence were informed to patients before consent was obtained for surgery.

Although sclerotherapy is now well established in treatment of neonatal and paediatric cystic hygroma, there have been very few cases reported of its use in adults. Some success has been observed in small number of adults with sclerotherapy agents such as OK-432.⁹

IV. CONCLUSION

Cystic hygroma is a rare entity in adults. Management of cystic hygroma particularly for more advanced stage lesions, remains a complicated issue and decision should be made on individual case basis.

We report this case of recurrent cystic hygroma in an adult and it could be only source to inform the surgeons about the management outcomes and should be encouraged.

This case contributes to the evidence supporting the role of cystic hygroma in differential diagnosis for cystic masses in adults. Further the future management of cystic hygroma in axilla with proximity to vital structures is better informed with our addition to examples of uncomplicated resection.

REFERENCES

- [1] Bloom DC, Perkins JA, Manning SC. Management of lymphatic malformations. *Curr Opin Otolaryngol Head Neck Surg* 2004; 12: 500–4.
- [2] Naidu SI. Lymphatic malformations of the head and neck in adults: a case report and review of the literature. *Ann Otol Rhinol Laryngol* 2004; 113: 218–22.
- [3] Kamath BS, Chatterjee AS. GIANT RECURRENT CYSTIC HYGROMA: A CASE REPORT. *Journal of the west african college of surgeons*. 2014;4(2):100–11
- [4] Brunicaudi F, Charles. *Schwartz's PRINCIPLES OF SURGERY*. 10th ed. United states of America. McGraw-Hill EDUCATION; 2010;1602-1603.
- [5] GowL, GulatiR, KhanA, MihaimeedF. Adult-onsetcystichygroma:acasereport and review of management.2011;11(1):5–11.

- [6] Schefter RP, Olsen KD. Cervical lymphangioma in the adult. *Otolaryngol Head Neck Surg* 1985; 93: 65–69. PMID:3920626.
- [7] Mulliken JB, Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. *Plast Reconstr Surg* 1982;69(3):412–22.
- [8] Perkins JA, Manning SC, Tempero RM, Cunningham MJ, Edmonds JL et al. Lymphatic malformations: review of current treatment. *Otolaryngol Head Neck Surg* 2010;142:795–803.
- [9] Smith MC, Zimmerman MB, et al. Efficacy and safety of OK-432 immunotherapy of lymphatic malformations. *Laryngoscope* 2009; 119: 107–15. doi:10.1002/lary.20041. PMID:19117316.

AUTHORS

First Author – Dr.Dinesh Dutt Sharma, Assistant Professor
Department of surgery,S.N Medical college Jodhpur

Second Author – Dr Manish Kumar Mahala, IIIrd year Resident
Department of surgery,S.N Medical college Jodhpur

Third Author – Dr Rishabh Bhargava, IIIrd year Resident
Department of surgery,S.N Medical college Jodhpur

Fourth Author – Dr Ajay Malviya, Senior Professor and Unit
head Department of surgery,S.N Medical college Jodhpur

Corresponding Author: Dr. Rishabh Bhargava, Mob No.:
9462143739, Email id: dr.rishabhbhargava@gmail.com
Address: C-37, Bapu Nagar, Jaipur, Rajasthan-302015