# Case study

## A rare case of acquired axillary cystic hygroma in an adult patient.

3

1

2

4

### 5 **ABSTRACT:**

A case of cystic hygroma was found in an adult male patient of 43 -year- age in the left axilla, which was **non-tender** cystic mass ( $57 \times 40 \times 49 \text{ mm}$ ) filled with haemorrhagic blood. It was successfully removed surgically without any complications. Being very rare presentation in the axilla of an adult, this case is reported.

### 6 Keywords: Cystic hygroma, Adult, Axilla, Haemorrhage.

7 8

#### 1. INTRODUCTION

9

10 Cystic hygroma, a benign and congenital malformation of the lymphatic system, also known as lymphangioma, usually occurs in children, especially in the neck. [1-3]. Cystic hygroma in the adult 11 is very rare and less than 150 cases of cervicofacial cystic hygroma are reported in English 12 literature up to 2011 [2]. Hygroma means a water containing tumour. This lesion does not have a 13 14 connection with the lymphatic or venous system. It may have unilocular or multilocular cystic lesions and may be of variable size. In microcystic varieties, the cyst may be less than 2 cm 15 diameter but in macrocystic size is more than 2 cm (also known as cystic hygroma) and in mixed 16 variety the size of cysts is variable [4,5]. Most often it is found in the cervical region of children 17 and less frequently found in the axilla or another site. Other uncommon sites are soft tissue, orbital 18 cavity, mediastinum, pancreas, liver, ovary, and fallopian tubes [6]. There are only five reported 19 cases of cystic hygroma in axilla of adult patient in English literature. [7-10]. 20

A literature searches (PUBMED, google and science direct) did not show any published report from Malaysia. In view of rare occurrence in adult especially in the axilla, this case is reported. To the

best of our knowledge, this is the first reported case of cystic hygroma in the axillary region in an
 adult patient from Malaysia.

25

### 26 **2. CASE HISTORY**

A 43 -year- old man, was referred to the surgical clinic of this hospital on 7<sup>th</sup> July 2015 with a

complaint of a mass in left axilla since 10 years which was very small, but recently in last few

29 months, it was increasing in size and patient had problem in moving the left upper arm. The

30 surgeon examined the case and found a cystic mass in left axilla about 60 mm x 50 mm in size. It

31 was cystic and fixed. There was no history of trauma, and there was no loss of weight or appetite by

the patient. The patient gave the history of chronic use of the deodorant spray.

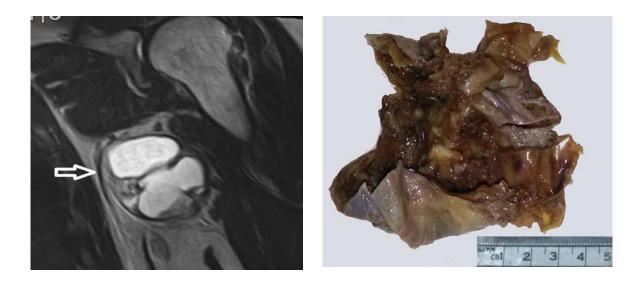
33 The patient was referred to the radiology department for MRI of left axillary mass which revealed a

34 well-defined oval cystic mass in left axilla measuring 57 x 40 x 49 mm. it was not attached to

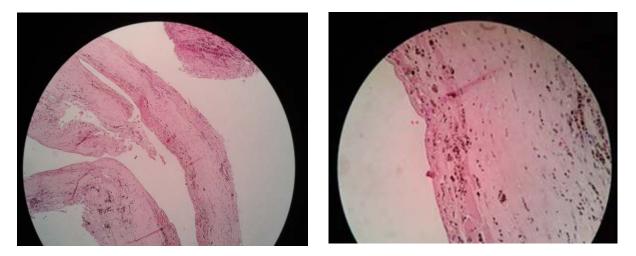
35 surrounding structure like blood vessels and muscles etc. The mass was cystic and no solid

36 component was found in it [Figure 1]. The wall was smooth and thin and there was a fluid level

- 37 seen in the cyst. Axillary lymph nodes were not enlarged. The MRI diagnosis was a cystic hygroma
- 38 with haemorrhagic blood within it.
- 39



- 40
- 41 **Fig 1.** MRI of left axilla showing the wall of the cyst (large black arrow). The cyst is filled with
- 42 haemorrhagic blood (white in colour within the cyst wall -- large arrow mark).
- 43 Fig 2: Gross appearance of the inner surface of the wall showing some blood clots (dark brown).



- Fig3: Microscopic appearance of the cyst wall (H&E stain, X 100). The thin and smooth cyst wall
   composed fibrous connective tissue lined by flattened epithelium.
- **Fig 4:** High power microscopic (H&E stain, X 400) shows flattened epithelium and scattered
- 47 hemosiderin laden macrophage in the wall.
- 48 The patient was operated under general anaesthesia on 8<sup>th</sup> July 2015 and the cystic lesion was
- 49 completely removed. The removed specimen was sent in formalin for histopathology.

- 50 The pathologist noted that the outer wall was intact, smooth, and brownish black in colour and size
- of cut opened specimen was 93x83x10 mm as seen in the picture. The lumen of the cyst was
- 52 unilocular and bit irregular without any papillary structures. The wall was thin and smooth. There
- 53 were some blood clots within the cyst [Figure 2].
- 54 Few random areas were selected from the gross specimen and processed for microscopic
- examination. Microscopic features were characterised by a thin and smooth wall, which was
- 56 composed of fibrous connective tissue lined by very thin and flatten epithelium. Few hemosiderin-
- 57 laden macrophages were seen in the wall. There were no any papillary structures or any malignant
- changes in cyst wall [figure 3]. Histopathological diagnosis was cystic hygroma with some
- 59 haemorrhage in the cyst.
- 60 Post-operative stay in the hospital was uneventful. The patient was discharged from hospital on the
- 61 third postoperative day and was followed up at two monthly intervals. There was no problem
- 62 found during follow-up visits. The operated site showed healed scar without any recurrence in the
- 63 left axilla (disease free for four months)

### 64 **3. DISCUSSION**

- 65 Cystic hygroma (also known as cystic lymphangioma) is a congenital lesion usually seen in
- children under the age of 2 years. It is a malformation of lymphatics. It may occur at various places
- in the body but usually in the neck and uncommonly in another part of body of the children. Cystic
- hygroma is an uncommon lesion in adult and very few cases are reported in the literature. [2,4]. The
- 69 cystic hygroma is a benign and painless proliferation of lymphatics and occurs with no connection
- 70 between lymphatics and venous system.
- 71 The predisposing factors that are responsible for the development of cystic hygroma in the adult are
- an infection, trauma, being as a growth or iatrogenic stimuli etc. [1]. Surgical excision is the main
- treatment and other treatment includes injection of bleomycin, a sclerosing agent and laser surgery.
- 74 [1,3,5]
- A case of large, fluctuant and non-tender cystic hygroma (10x12x17 cm) was reported by Gelal et
- al in a 24-year-old, seven months' pregnant woman in left axilla, situated between anterior and
- 77 posterior axillary line [7].
- 78 Güner et al reported a case of axillary cystic hygroma in 83 -year- old male with the history of
- 79 discomfort during the inspiration phase of breathing for the past four months. It was a painless and
- 80 mobile right axillary mass. They did not find any recurrence during five months' follow-up period
- 81 [8].
- Michail et al reported a cystic hygroma in a female patient which developed rapidly in the axillaryregion in the absence of any predisposing factor [9].
- 84 McCaffreya at el reported an another case of cystic hygroma in a 58 -year- old male in right upper
- 85 flank extending up to axilla which was non-tender and large (20x12x7 cm). It was multiseptate
- lesion and there was no any recurrence found on follow-up after one year of surgery [10].

87 88 89	In our case, there was no history of any predisposing factors. The presence of blood in the removed cyst suggests the possibility of minor trauma before coming to the hospital for the check-up. The size of cystic hygroma of this case was medium size when compared to other two cases.
90 91 92	The most recently, Copley at al reported one case of spontaneous cystic hygroma in the axilla of a 59-year-old female, which was managed by ultrasound guided aspiration but was unsuccessful due to recurrence and then was removed successfully by total excision surgically.
93 94 95	Detailed imaging procedures like ultrasound, MRI and CT scan will help the diagnosis preoperatively and histopathological examination of surgically removed specimen will confirm the diagnosis of cystic hygroma.
96 97 98 99 100 101	<b>4. CONCLUSION</b> Cases of cystic hygroma in adult male especially in the axillary region is a rare occurrence and becomes the sixth reported case.
102 103 104	ETHICAL APROVAL: It is not applicable.
105 106 107 108	<b>REFERENCES:</b> 1. Naidu SI, McCalla MR. Lymphatic malformations of the head and neck in adults: a case report and review of the literature. Ann Otol Rhinol Laryngol. 2004 Mar;113(3 Pt 1):218-222.
109 110 111 112	2. Gow L, Gulati R, Khan A, Mihaimeed F. Adult-onset cystic hygroma: a case report and review of management. Grand Rounds. 2011;11(1):5–11.
112 113 114 115	3. Bloom DC, Perkins JA, Manning SC. Management of lymphatic malformations. Curr Opin Otolaryngol Head Neck Surg. 2004;12(6): 500-504.
116 117 118	4. Manikoth P, Mangalore GP, Megha V. Axillary Cystic Hygroma. Journal of post graduate medicine. 2004; 50 (3) 215-216.
119 120 121	5. Fonkalsrud EW. Lymphatic disorders. In: Grosfeld JL, O'Neil JA Jr, Caron JA. Pediatric surgery. 6 th ed. Mosby Elsevier, Chicago 2006: pp 2137-2145.
122 123 124	6. Stacey e. Mills, Sternburg's Diagnostic surgical pathology 4 <sup>th</sup> ed. Volumes 1-3. Lippincott Williums & Wilkins. 2004 pp 72, 1098,1259-60,1632,1754,2617, 2662.
125 126 127	<ol> <li>Gelal F, Yucel K, Tugsel E, Guney S.Axillary Cystic Lymphangioma Presenting in Pregnancy. Tr J of Medical science 1998: 28: 571-572.</li> </ol>
128 129 130 131	8. Güner A, Ayd>n. A, Çelik F. Cystic Hygromas in Adults: Reports of Two Cases. Olgu Sunumlar / Case Reports. 2006; 2:101-103
132 133 134	9. Michail O, Michail P, Kyriaki D, Kolindou A, Klonaris C, Griniatsos J. Rapid development of an axillary mass in an adult: a case of cystic hygroma. South Med J. 2007;100(8):845-9.

135 10. McCaffreya F, Taddeob J. Surgical management of adult-onset cystic hygroma in the axilla

- 136 Int J Surg Case Rep. 2015; 7: 29–31
- 137
- 138 11. Copley PC, Ali L, Mirza S. Spontaneous lymphocoele: an unusual cause of an axillary mass.
- British medical journal, Case Rep. 2016 Feb 11;2016.