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Hüseyin Gümüş
Halil Kazanasmaz
Osman Kocaman
Halil Aslan
Harran University, Şanlıurfa-Turkey
huseyingumus2163@hotmail.com; kazanasmazhalil2@gmail.com;
drhakankocaman@yahoo.com; drhaslan1@hotmail.com

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ORCID ID	0000-0002-9326-2194	0000-0003-4671-4028
	0000-0002-8072-5292	0000-0001-8111-121X
CORRESPONDING AUTHOR	Hüseyin Gümüş	

CYSTIC HYGROMA OF THE CHEST WALL: A RARE LOCATION

ABSTRACT

Cystic hygroma, a congenital lymphatic malformation, is mostly localized to the cervical region and more rarely on the chest wall. Herein, we present a case with a cystic hygroma that was diagnosed immediately after birth and started mainly from the axillary region and extending to the anterior wall of the thorax, in the light of the literature.

Keywords: Chest Wall, Cystic Hygroma, Lymphatic Malformation, Newborn, Septum

1. INTRODUCTION

Cystic hygroma, also known as cystic lymphangioma, is a congenital malformation that occurs in the entire body, especially at the back of the neck, at the side of the neck, after the breakdown of lymphatic vessels in relation to the main collecting vessels. This pathology seen in 1/6000 gestations has localized and common forms [1]. Cystic hygroma, first described by Redenbacher in 1828, is the most common form of lymphangiomas, accounting for 5% of benign tumors in infancy and childhood [2 and 3]. The main pathology is the lack of connections between jugular vein and lymphatics. The fetal lymphatic system begins to appear in the fifth week of gestation. In the neck, the lymphatic vessels are expected to bind to the internal jugular venous around 7 weeks of gestation. If the connection does not occur until 11th-12th week of gestation, the cystic hygroma process may begin [4]. In our study, we aimed to present a giant septal cystic hygroma extending to the chest wall in the context of literature information.

2. RESEARCH SIGNIFICANCE

In our study, a case of cystic hygroma located in the anterior thorax wall, which is a very rare location, is presented. The cystic hygroma in the presented case has a rare location, the size of the mass and its septation made the case different. Radiological and microscopic demonstration of cystic hygroma made the case important.

3. CASE REPORT

The woman, who was the second live birth at the age of 19 years old and who was pregnant according to the last menstrual period, was referred to our hospital after the babies were delivered in cesarean

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section and at the external health center and the baby was found to have swelling in the chest area. It was understood that the patient had not gone through the pregnancies during the pregnancy and that axillary and abdominal mass was detected in US prenatal ultrasonography (US). On the physical examination of the patient, a soft, painless mass was palpated on the left axilla and thoracic anterior wall (Figure 1 and Figure 2).

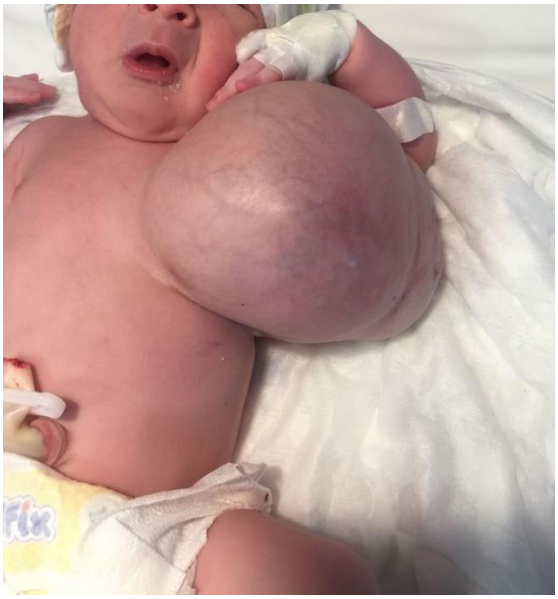

Figure 1. The appearance of the cystic hygroma in the examination

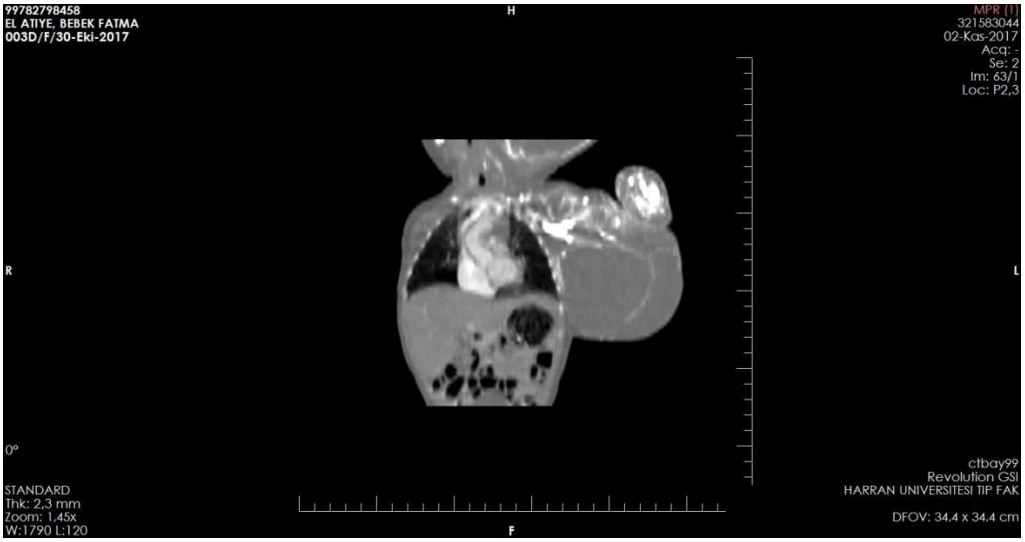

Figure 2. Cystic hygroma in computerized thorax tomography imaging

The axillary and thoracic anterior wall US showed multilocular anechoic cystic lesion starting from under the skin on axillary and thoracic anterior wall and extending to deep tissues, separated by echogenic septa. Abdominal US findings of the case showed normal findings. Lymphangioma (cystic hygroma) was considered as a preliminary diagnosis when radiological findings and physical examination findings were considered together. After intravenous contrast material administration, computerized thorax tomography (CT) was performed with axial thickness of 5mm. Within the section, a cystic multilocular lesion (lymphangioma), extending inferiorly from

the left shoulder to the left lateral chest wall and measuring the size of approximately 122x93mm and septal enhancement, was observed in the multiple septal coronal plane in the near neighborhood (Figure 2). On the basis of this, the case was made by the pediatric surgery clinic all around the cystic masses and the axillary structures were preserved and total excision was performed to the cystic masses. During surgery, it was determined that the lesion consisted of multiple, variable dimensions and septal cystic components. After macroscopic examination, 13x8x5 cm sized cysts were sampled for tissue alignment. Microscopic examination revealed eosinophilic-protein lymphatic fluid deposits in the lumen with irregular vascular spaces embedded with flattened endothelial cells embedded in a fibroblastic and collagenous stroma (Figure 3), and calm-flattened endothelial flooring in large magnification (Figure 4). The case was reported consistent with cystic lymphangioma.

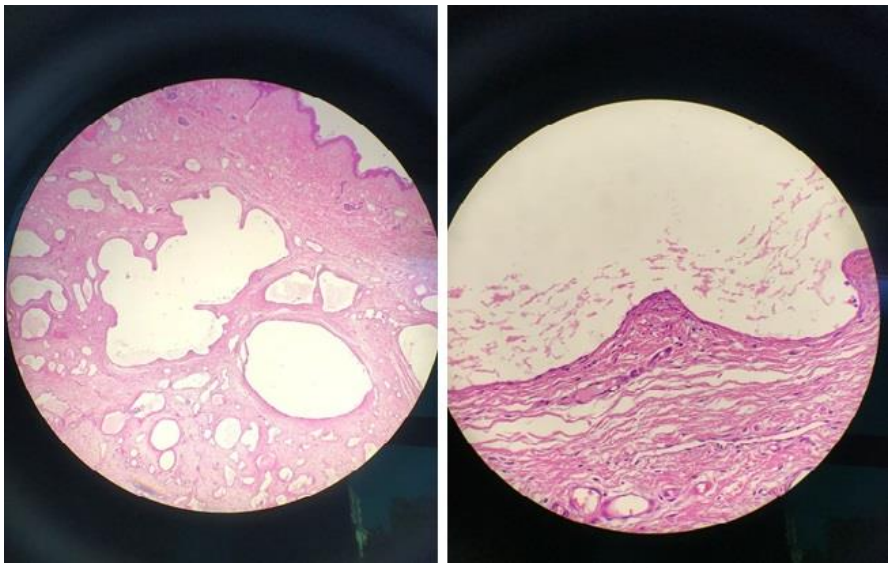

Figure 3. Irregular lymphatic spaces in microscopy

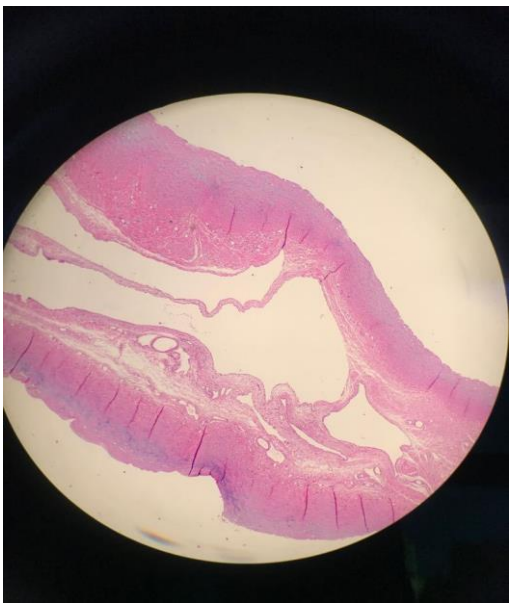

Figure 4. Calm-flattened endothelial tiling at large magnification in microscopy

4. DISCUSSION

Lymphangiomas, which are congenital lymphatic malformation, can occur in any place where the lymphatic system develops, mostly in the head, neck, and axillae. These lesions surround the anatomical structures and occasionally invade them, but they do not carry the potential for malignancy [5]. Lymphangiomas histologically are divided into 4 types: cystic hygroma, cavernous lymphangioma, capillary (or simple) lymphangioma, and vasculolymphatic malformation. All four types of lymphangiomas have endothelial cell-lined lymphatic channels separated by connective tissue stroma. Cystic hygromas consist of large, dilated lymphatic vessels, while cavernous and capillary lymphangiomas consist of smaller lymphatic channels [5]. Vasculolymphatic malformations consist of lymphatic and vascular elements. The lymphangiohemanjiomas are an example of this group. This is due to the fact that cystic hygromas reach larger sizes than other lymphangiomia types because they are surrounded by loose connective tissue that allows them to grow due to their anatomical location [5]. In this case, it was found that the lesion consisted of numerous cystic components with varying sizes and septa during the operation. In addition, microscopic examination revealed that it was lined with flattened endothelial cells and there were irregular lymphatic cavities in their lumen with eosinophilic- proteinaceous lymphatic fluid deposition. This case was found to be cystic hygroma, a subtype of congenital lymphatic malformation lymphangiomas.

75% of the cystic hygromas are located on the lateral side, 20% in the armpit, 5% in the mediastinum, retroperitoneal region and more rarely in the chest wall [3]. In this case, the type which is located rarely on the chest wall is seen. Cystic hygromas may be septated or non-septated as they are presented. The presence of septa with large size is a poor prognostic marker and often accompanies chromosomal anomalies [9]. Among these, Turner syndrome is frequently seen in 40-80% of cases. Most female fetuses with cystic hygroma are probably Turner syndrome. However, neck hygromas may also be associated with malformations such as trisomy 21, Noonan syndrome, and Robert's syndrome. Sometimes cystic hygromas can also be seen familial [10]. No accompanying chromosomal anomaly or malformation was found in this case. Cystic hygromas can be seen in spontaneous regression especially in cases that do not show septation. In these cases, regression occurs when the increased pressure in the lymphatic system can overcome incomplete obstruction [6]. In this case, surgical treatment was planned since the cysts showed septation. Cystic hygromas located in the neck region in the newborns may extend to the thoracic anterior wall. Cystic hygromas usually appear as asymptomatic soft packs. They may show slow growth but may develop sudden growth when into the cyst hemorrhage, inflammation, concomitant respiratory tract infection or in presence of trauma [7].

The differential diagnosis includes lipomas, hemangiomas, bronchial cysts, thyroglossal cysts, dermoid cysts, thymic cysts, laryngoceles, thyroid glands, primary neoplastic diseases (neuroblastoma, rhabdomyosarcoma), retropharyngeal abscess and secondary lymphadenopathy to the infection [8]. In this case, a more rare septated type is found, which is located in the underarm and chest wall. According to the anatomical location of the lesion, it is important to remove it surgically early. Non-surgical treatment options include radiotherapy, aspiration, sclerosing agent injection (bleomycin, OKT-432) and CO2 laser [3, 11 and 12]. In this case, surgical treatment was performed. During the surgical treatment, infection, hemorrhage, hematoma, postoperative seroma were reported. Facial, hypoglossal, glossopharyngeal nerve injury, Brachial plexus

injury were reported as complications of surgical treatment [4, 13, 15 and 16]. In our case, no complications developed during the surgical treatment. In conclusion, this case is presented because the CH located in the chest wall and septal is a rare case. It is also important to remember that, although this lesion is benign, it can lead to serious complications.

INFORMED CONSENT

Informed consent from patient's family was received.

CONCEPT

H.G, H.K., H.A., O.H.K., Data Collection or Processing: H.G, H.K., H.A., O.H.K., Analysis or Interpretation: H.G, H.K., Literature Search: H.G, H.K., H.A., O.H.K., Writing: H.G, H.K.

CONFLICT OF INTEREST

No conflict of interest was declared by the authors.

FINANCIAL DISCLOSURE

The authors declared that this study received no financial support.

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