Cystic Hygromas in Adults: Reports of Two Cases

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ABSTRACT

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Cystic hygroma (lymphangioma) is a benign congenital malformation of the lymphatic system that occurs in infant or children younger than two years of age. Here we present two cases of 52 and 83 years old patients with complaints of axillary and cervical masses. Computerised tomography revealed cystic masses. After total excision of the masses pathological examination revealed cystic lymphangioma. After uneventfull postoperative period patients were discharged and no recurrence occurred since then.

The objectives of these reports are to discuss the clinical presentation, diagnosis and pathologic findings, and management of this malformation.

Key words: Cystic hygroma, lymphangioma, lymphatic system, servical tumor, axillary mass

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ÖZET

Erişkinlerde kistik higroma: İki olgunun sunumu

Kistik higroma (lenfanjioma) genellikle hayatın ilk iki yılında ortaya çıkar. Bu konjenital lezyon erişkinlerde nadir bir durumdur. İlk olarak boyun ve aksiller bölgede kitle olarak ortaya çıkan iki hastayı sunuyoruz. Hastalar 52 ve 83 yaşlarındaydı. Bilgisayarlı tomografi kistik kitle olarak rapor edilmişti. Iki hastada da kitleye total eksizyon uygulandı ve patolojik değerlendirmesi kistik lenfanjioma olarak rapor edildi. Postoperatif dönemde herhangi bir komplikasyon gelişmedi ve takiplerinde nükse rastlanılmadı. Erişkinlerdeki kistik higroma hastalığının başvuru şekli, patolojisi, tanı ve tedavi yöntemlerini değerlendirmeyi amaçladık.

Anahtar kelimeler: Kistik higroma, lenfanjioma, lenfatik sistem, boyun tümörü, aksiller kitle

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INTRODUCTION

Cystic hygroma (lymphangioma) is a benign congenital malformation of the lymphatic system that occurs as a result of sequestration or obstruction of developing lymphatic vessels. These lesions are usually discovered in infant or children younger than two years of age. Occurrence in adults is uncommon, and fewer than 100 cases of adult lymphangioma have been reported in the literature (1,2) . The objectives of these reports are to discuss the clinical presentation, diagnosis and pathologic findings, and management of this malformation.

CASE REPORTS

Patient 1: A 52-year-old woman was referred to our clinic with a ten-month history of a slowly enlarging mass on the left side of her neck. She noted discomfort and mild pain while turning her head to the left side. She was further asymptomatic. Physical examination revealed a semi-solid, smooth and mobil 8-9 cm mass in the supraclavicular fossa. Fine needle aspiration biopsy (FNAB) showed a cystic lesion, yellow, liquid with mature lymphocytes and histiocytes (Figure 1). Computed tomography of the neck revealed that 7x8x6 cm

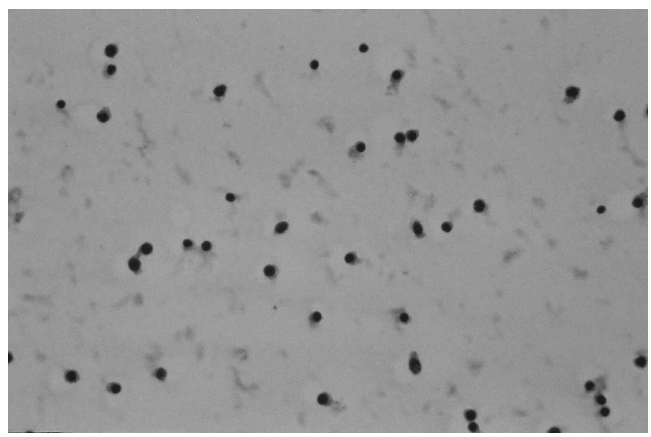

Figure 1. Grade 4 renal travmalı bir hastanın bilgisayarlı tomografisi stain; X100).

lobulated cystic mass which lies down from hyoid bone to left supraclavicular fossa and obliterates jugular chain and posterior cervical triangle. Excision of the mass was performed under general anesthesia. Pathologic diagnosis was cystic lymphangioma which was supported with a thick fibrous wall, containing dilated blood vessels partly filled with erythrocytes and infiltrated with lymphocytes (Figure 2). Follow-up at 51

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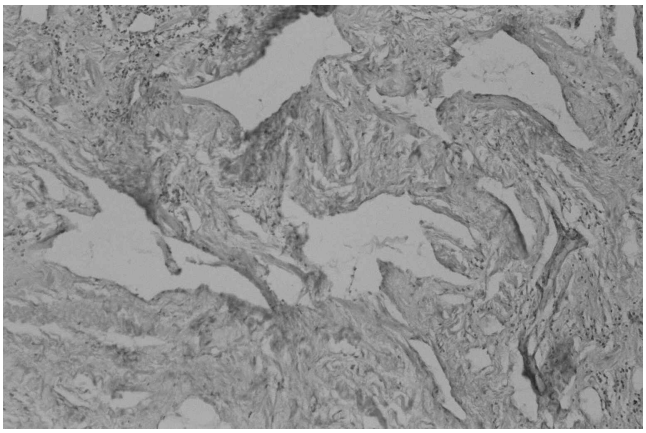

Figure 2. Histopathology of the excisional biopsy of patient 1. Lymphocyte infiltration between connective tissue, dilated lymphatic vessels with empty or full of erythrocyte (Hematoxylin -eosin stain; X100).

months revealed no recurrence.

Patient 2: The other patient was a 83-year-old man presented for a right axillary mass with four months history. He had no pain but noted discomfort during inspirium phase of the respiration. Physical examination revealed a mobile, soft mass in the right axillary fossa. Computed tomography of the thorax revealed a 10x8 cm mass in the right axillary fossa, settled the anterior part of the subscapularis and latissimus dorsi muscles, with close contact to the thoracic wall and ribs (Figure 3). Excision of the mass was performed under local anesthesia. Mass was scraped easily from thoracic wall by blunt dissection (Figure 4). It was full of transilluminant cystic material (Figure 5). Pathologic diagnosis was cystic lymphangioma. Follow-up at 5 months revealed no recurrence.

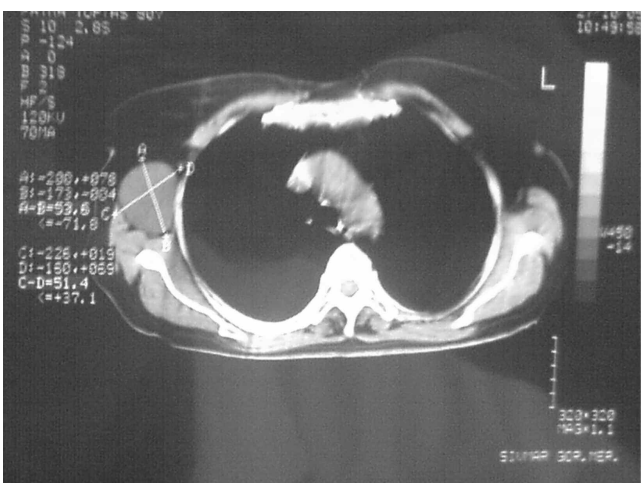

Figure 3. A cystic mass can be seen in the rihgt axillary region on the thorax CT of the patient 2.

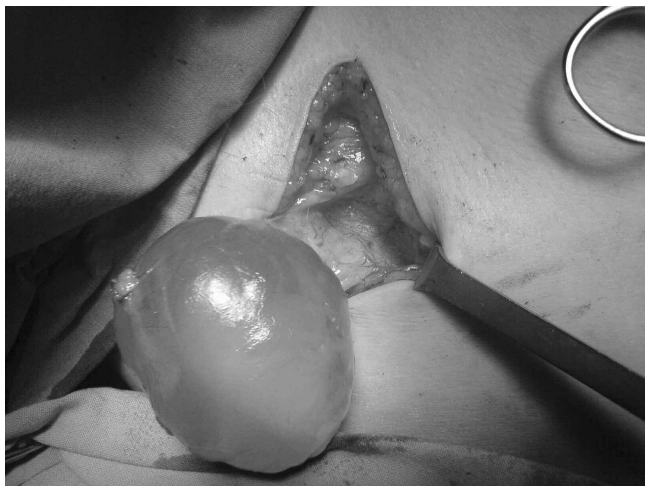

Figure 4. Peroperative image of patient 2.

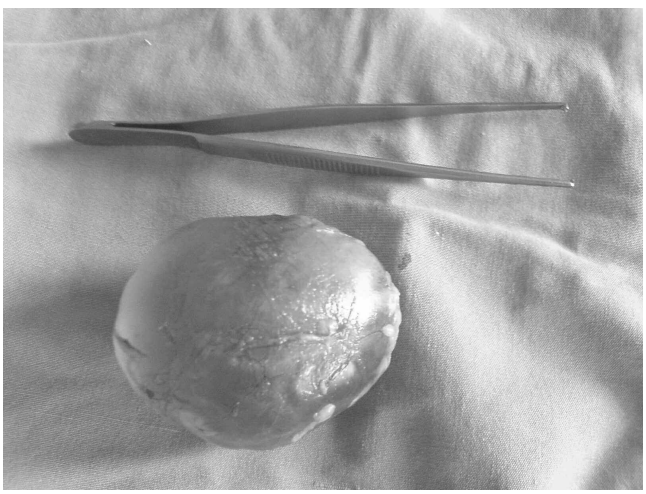

Figure 5. The view of the operative specimen of the second patient.

DISCUSSION

Lymphangioma is a benign congenital malformation of the lymphatic system. There are three histological subtype. Capillary lymphangioma (composed of small lymphatics), cavernous lymphangioma (composed of larger lymphatics), cystic lymphangioma (cystic hygroma- composed of large macroscopic lymphatic spaces with collagen and smooth muscle). Cavernous lymphangioma is the most common subtype. But cystic lymphangioma occurs approximately 1 in 12000 births and 95% occurring by the second year of life. Although the lesion can occur anywhere, the most common sites are in the posterior triangle of the neck (75%), axilla

(20%), mediastinum (5%), groin, retroperitoneal space and pelvis (3). Adult patients are usually asymptomatic in the adults. On physical examination; soft, painless, mobile, transilluminable cystic mass were detected. Lymphangiomas are best visualized by magnetic resonance imaging (MRI); the high water content allows lymphangiomas to appear hyperintense on T2-weighted images (4). The other imaging methods are doppler ultrasonography and computed tomography (CT). This disease may be associated with Turner syndrome, Noonan syndrome, cardiac anomalies, trisomy syndromes and fetal hydrops. Differential diagnoses are hemangioma, mucosel and meningomyelocele (5). Infection within the cysts (usually caused by streptococcus or staphylococcus species) may occur. This complication can cause rapid enlargement which may result in airway obstruction. Bkeeding into the cyst is the another complication of the lymphangioma. These tumours do not resolve spontaneously. Percutaneous aspiration is not preferred because of the risk of bleeding, infection and recurrence. These tumors are not sensitive to radiotherapy. Injection of sclerosing agents like alcohol, bleomycin and OK-432 (a lyophilized mixture of streptococcus pyogenes and penicilin G potassium), with favorable results have been reported (6). Complete surgical excision is the preferred treatment. It can be performed under general or local anesthesia. Sometimes, this may be impossible due to the infiltrating nature of the hygroma within and around neurovascular structures, muscles, blood vessels. In this condition, unroofing, partial cystectomy and drainage of the cystic content can be performed. In this kind of treatment, recurrence rate of 10-15 % was reported (7). Baer and Davis reported an 89-year old with lymphangioma, the oldest such patient reported in the literature (8). Thirty-two patients with cervical lymphangioma were treated at the Mayo Clinic; this is the largest series of the literature (9). Aneeshkumar et al. suggested that trauma could trigger formation of lymphangioma (10). Mhoon et al. reported a scrotal enlargement after trauma (11). There was bleeding into a scrotal cystic lymphangioma. Although neck and axilla are the most common sites, different places have been also reported. Ates at al12 reported on right adrenal gland, Shaffer at al13 reported thoracic lymphangioma, Nakazato et al.reported a case in mediastinum (14), Chung et al. reported a case in the breast (15) and Solomou et al. reported a splenic lymphangioma (16).

As a conclusion, adult type lymphangiomas (cystic hygromas) are uncommon causes of the cervical and axillary region masses. This is a benign situation which can be treated with different methods. Surgical removal is the procedure of choice. Although this disease occurs during antenatal or early neonatal period, it should be remembered for the adult patients who have cervical, axillary, groin or retroperitoneal masses.

KAYNAKLAR

1. Sherman BE, Kendall K. A unique case of a large cystic hygroma in the adult. Am J Otolaryngol 2001; 22: 206-210.
2. Suk S, Sheridan M, Saenger JS. Adult lymphangioma: a case report. Ear Nose Throat J 1997; 76: 881-883.
3. Woods D, Young JE, Filice R, Dobranowski J. Late-onset cystic hygromas: the role of CT. Can Assoc Radiol J 1989; 40:159-161.
4. Terezhalmy GT, Riley CK, Moore WS. Lymphangioma (lymphatic malformation). Quintessence Int 2001; 32: 495.
5. Manikoth P, Mangalore GP, Megha V. Axillary cystic hygroma. J Postgrad Med 2004; 50:215-216.
6. De Santis M, Calo GF, Trombini P, Romagnoli R. Percutaneous sclerosing treatment with ethanol of a large cystic lymphangioma of the neck in an adult. Radiol Med 2003; 105: 127-130.
7. Ravitch MM, Rush JrBF. Cystic hygroma. Pediatr Surg 1969; 1:273-279.
8. Baer S, Davis J. Cystic hygroma presenting in adulthood. J Laryngol Otol 1989; 103: 976-977.
9. Schefter RP, Olsen KD, Gaffey TA. Cervical lymphangioma in the adult. Otolaryngol Head Neck Surg 1985; 93: 65-69.
10. Aneeshkumar MK, Kale S, Kabbani M, David VC. Cystic lymphangioma in adults: can trauma be the trigger? Eur Arch Otorhinolaryngol 2005; 262: 335-337.
11. Mhoon JM, Redman JF, Siebert JJ. Scrotal enlargement in boys with a history of scrotal trauma: two unusual findings. South Med J 2002; 95: 251-252.
12. Ates LE, Kapran Y, Erbil Y, Barbaros U, Dizdaroglu F. Cystic lymphangioma of the right adrenal gland. Pathol Oncol Res 2005; 11: 242-244.
13. Shaffer K, Rosado-de-Christenson ML, Patz EF Jr, Young S, Farver CF. Thoracic lymphangioma in adults: CT and MR imaging features. AJR Am J Roentgenol 1994; 162: 283-289.
14. Nakazato Y, Nakata Y, Tokano T, et al. Cystic lymphangioma of the mediastinum. Am Heart J 1995; 129: 406-409.
15. Chung SY, Oh KK, Kim DJ. Mammographic and sonographic findings of a breast cystic lymphangioma. J Ultrasound Med 2003; 22: 307-309.
16. Solomou EG, Patriarheas GV, Mpadra FA, Karamouzis MV, Dimopoulos I. Asymptomatic adult cystic lymphangioma of the spleen: case report and review of the literature. Magn Reson Imaging 2003; 21: 81-84.