Cystic lymphangioma with adrenal origin: 2 case reports

Adrenal kaynaklı kistik lenfanjioma: 2 olgu sunumu

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Abstract

Cystic lymphangioma is a rare benign vascular lesion that originates from lymphatic endothelial cells. Adrenal cysts are very rare with occurrence of 0.06-0.18%. Pathogenesis of adrenal lymphangioma is still unclear; however, the most favored theory is anormal development of lymphatic vessels or/and ecstasia. These lesions mostly occur in the axilla and neck region anatomically but also intra-abdominal with the occurence of 5%. Adrenal gland lymphangiomas are generally asymptomatic. They are often diagnosed incidentally on radiological tests or during surgery. In this study we present 2 cases of cystic lymphangioma with adrenal origin, a rare presentation of cystic lymphangioma hence only found in form of cases in the literature.

Key words: Adrenal gland, cystic lymphangioma, surgical treatment

Özet

Kistik lenfanjioma, lenfotik endotelyal hücrelerden köken alan benign nadir lezyonlardandır. Adrenal kistler %0.06-0.18 oranında çok nadirdirler. Adrenal lenfanjioma patogenezi tam olarak bilinmemekle beraber en yaygın teori lenfatik damarların anormal gelişimi ya da ektazisi sonucu olduğu yönündedir. Bu lezyonlar sıklıkla anatomik olarak aksilla ve boyun bölgelerinde olmakla beraber %5 oranında intraabdominal de olabilirler. Adrenal bez lenfanjiomaları genellikle asemptomatiktir. Çoğunlukla radyolojik incelemeler esnasında insidental olarak ya da cerrahi sırasında tanı alırlar. Bu çalışmada kistik lenfanjiomanın çok nadir bir formu olup literatürde yalnızca vaka sunumları şeklinde bildirilmiş olan adrenal kaynaklı kistik lenfanjiomalı 2 olgu sunacağız.

Anahtar Kelimeler: Adrenal bez, kistik lenfanjioma, cerrahi tedavi

Introduction

Cystic lymphangioma is a rare benign vascular lesion that originates from lymphatic endothelial cells[1]. Through the first time described in 1965, limited cases have been reported in the literature to date[2]. These lesions mostly occur in the axilla and neck region anatomically but also intra-abdominal with the occurence of 5%[3,4].

In this study we present 2 cases of cystic lymphangioma with adrenal origin, a rare presentation of cystic lymphangioma hence only found in form of cases in the literature.

Case 1

A 19 years old woman referred to our centre for further management with suspicion of liver hydatid cyst due to loss of appetite, right-sided abdominal pain and ultrasound finding of a thick-walled cystic lesion in the 6th segment of the liver with 8x6 cm dimensions. The physical examination and blood work at our centre were unremarkable. Contrary to the ultrasound finding the patient's contrast-enhanced abdominal computerized tomography revealed a 59x99 mm slightly lobular purely cystic lesion with small wall septations and millimetric calcifications located on the

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right adrenal gland. The patient was operated on with radiologic diagnosis of right adrenal hydatid cyst and possible simple adrenal cyst. Intraoperative exploration revealed a well-defined cystic lesion of the right adrenal gland and it was removed ***(Figure 1)***. Postoperative recovery was uneventful; the patient started per oralfeeding in the same evening and discharged on day 2. Postoperative check up was unremarkable after a week and the pathologic findings revealed a cystic lymphangioma with adrenal origin. There wasno recurrence in the 27 months follow up period.

Case 2

A 28 years old male presented with back pain. His lumbar MRI (magnetic resonance imaging) revealed an intraabdominal mass with 20x18 cmdiameter. An abdominal MRI for further evaluation of the mass revealed a cystic mass withmesenteric origin. The patient was referred to our centre for further management. The patient's physical examination and blood work was unremarkable. The patient was operated on with radiologic diagnosis of mesenteric mass. Intraoperative exploration revealed a cystic mass of right adrenal gland that filled the middle abdominal cavity, displacing transverse colon to lower quadrant while deviating the right kidney and adrenal gland into the right upper quadrant ***(Figure 2)***. The cyst ruptured during the dissection and it was aspirated. The content was sent for cytologic examination. The right adrenal gland was removed with the cyst. Postoperative recovery was uneventful as patient started oral feeding on day 1 and discharged on day 3. The follow up examination a week later was unremarkable. Pathology revealed a cystic lympangioma with right adrenal gland origin. The patient had no recurrence in 18 month follow up period.

Discussion

Adrenal cysts are very rare and have reported occurrence of 0.06-0.18%[2]. Pathogenesis of adrenal lymphangioma is still unclear; however, the most favored theory is anormal development of lymphatic vessels or/and ecstasia[2,5,6]. Though most lesions occur in the axilla and mediastinum they can develop in any part of the body[7].

Adrenal gland lymphangiomas are generally asymptomatic. They are often diagnosed incidentally on radiological tests or during surgery. However they can manifest with non-specific symptoms like fever, abdominal pain or palpable mass. Both patients in our study presented with abdominal pain (on the affected side). Diagnosis of giant lesion in the second patient delayed because of the patient that he thoughthe was gaining weight until symptoms increased.

Also adrenal cystic lymphangiomas have no pathognomonic radiological finding, new imaging methods provide helpful information that contribute to diagnosis[8]. MRI is more specific than CT (computerized tomography) and generally helps differentiating malign or benign adrenal lesions[8]. Despite clinical and radiological studies, immunohistochemical and histopathologic examinations are often used for definitive diagnosis[2,9].

Since there is no study except the case reports in the literature there is no common diagnosis and treatment algorithm ,but there are 3 factors determining the treatment strategy in endothelial-based cystic lesions. These are; functional status of the cyst, possibility of incidental malignancy and potential complications like hemorrhage and infection[10].

In summary, cystic lymphangiomas with adrenal origin are very rare, mostly asymptomatic, benign, vascular lesions that diffucult to diagnose by the available imaging techniques. Whether incidental or symptomatic these lesions

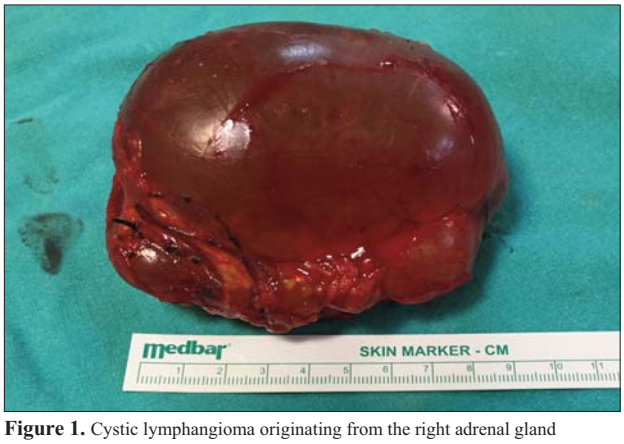

Figure 1. Cystic lymphangioma originating from the right adrenal gland

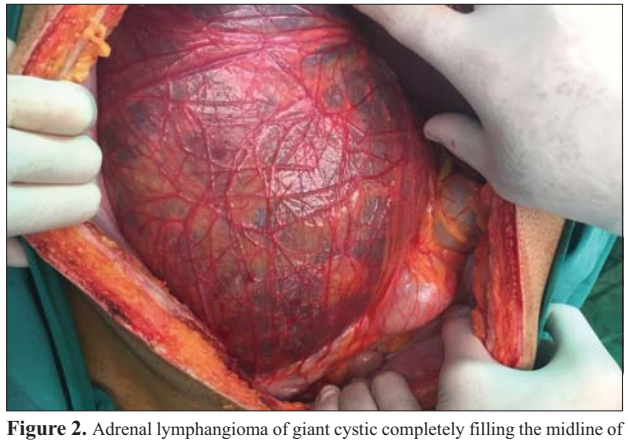

Figure 2. Adrenal lymphangioma of giant cystic completely filling the midline of the abdomen

should be planned for excision either by open or laparoscopic access in order to relieve the symptoms and/or rule out adrenal malignancy. No recurrence have been reported in long term follow up of patients treated appropriately.

Conflicts of interest

The author declare that they have no financial or non-financial conflicts of interest related to the subject matter or materials discussed in the manuscript.

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