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<http://www.pediatricurologycasereports.com>**Cystic lymphangioma: A rear finding during hydrocele surgery in children “case report”****Thayer Mohammed Ameen Aboush, Zaidoon Moayad Altaee, Mahmood Mosa Mahmood***Department of Pediatric Surgery, Alkhansaa Teaching Hospital, Mosul, Nineveh, IQ, Iraq***ABSTRACT**

Cystic lymphangioma is a benign, unencapsulated tumor caused by congenital malformations of the lymphatic system. It is a rare type of hematoma that unusually causes scrotal swelling. A 3-year-old boy presented with a painless swelling in his right hemiscrotum that gradually increased over a year. The swelling was clinically evaluated as a hydrocele, but a multi-localized cystic mass filled with hemorrhagic fluid was detected on surgical exploration. A complete excision was made and histopathological evaluation revealed cystic lymphangioma. As a result, cystic lymphangioma mimicking hydrocele can be seen in children. Although it is a rare type of tumor, this pathology should be suspected during the surgical operation in the groin area.

Key Words: Lymphangioma, scrotum, hydrocele, surgery, child.

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Introduction

Lymphangioma is a benign unencapsulated hematoma and caused by congenital malformations of the lymphatic system. It mostly affects the lymphatic system of the neck and axilla, sometimes the mediastinum, retroperitoneum and thigh. However, it is extremely rare for it to involve the scrotum and perineum [1,2,3]. Here, we present a 3-year-old boy who was operated with the presumptive diagnosis of hydrocele in the right hemiscrotum, who had diagnosed with cystic lymphangioma after pathologic examination.

Case report

A three-year-old boy presented with a painless swelling in the right hemiscrotum, which gradually increased in size over a period of a year. No history of trauma was reported and the child was fine at an early age. He has no previous medical or surgical history, and no similar family history has been reported.

The swelling was a soft cyst in nature and there was no tender in the examination of the right hemiscrotum. The swelling was irreducible and the transillumination test was positive. On palpation, the right scrotal skin appeared normal, there were no signs of redness or laceration, and it was not attached to the underlying lesion. The right testicle could not be distinguished from swelling. The left hemiscrotum examination was normal. Hydrocele was considered according to the

first examination findings. Scrotal ultrasonography (US) revealed congenital hydrocele, thus he was scheduled for surgical intervention. Right transvers lower skin crease incision was done and it was revealed that right testicle and the lesion were separated from each other. The cystic lesion persisted even after pulling the testicle cord throughout the surgical incision. Therefore, a scrotal incision was made and a 7cmx5cm multiloculated cystic mass filled with hemorrhagic fluid adhering to the subcutaneous tissue of the right hemiscrotum was seen (Fig. 1). The mass was completely excised and the layers were closed according to its anatomy without drainage (Fig. 2). The sample was sent for histopathological evaluation.



Fig. 1. The multiloculated cystic mass filled with hemorrhagic fluid that attached to the subcutaneous tissue of the right hemiscrotum.

The patient was discharged on the same day with oral antibiotics and analgesics. The postop period was completely uneventful. Histopathological results confirmed that the lesion was a cystic lymphangioma rather than a hydrocele. The patient was followed up one year later without a history of recurrence and no other complications were reported.



Fig. 2. Macroscopic view of the completely removed mass.

Discussion

Lymphangiomas are rare benign unencapsulated hamartomas caused by congenital malformations of the lymphatic system [1,2,3]. Different terms are used to describe these lesions, such as cystic hygroma, congenital lymphatic hamartomas, or cystic lymphangiomas [3]. Although most lymphangiomas are congenitally caused by atresia or inadequacy of efferent lymphatic vessels and venous channels, they may arise especially due to infection, inflammation or degeneration and can also develop after trauma [3,4].

Lymphangiomas were first described by Whimster in 1976. He evaluated them as congenital developmental anomalies of lymphatic ducts that were abnormally connected to the lymphatic system and appear as a mass lesion [2,3,5]. Lymphangiomas account for approximately 26% of all benign vascular tumors in children [5]. 50% of these are present at birth, and 90% occur in the first two years of life [6]. Approximately 95% of all lymphangiomas are located in the neck and axillary region. The remaining 5% can usually be found in the mediastinum, retroperitoneum,

mesentery, internal organs, bone, spleen and groin. Perianal and scrotal localization is extremely rare [7,8]. We also presented a lymphangioma in scrotal region.

Lymphangiomas are classified histologically according to the size of the vessels. Therefore, they are divided into capillary (a rare type originating from subcutaneous tissue), cavernous (located in the mouth and tongue), cystic or hygromatous [2,3,7]. Scrotal lymphangioma typically arises from the scrotal wall (lymphangioma circumscriptum) and the tunics. It may involve testis, epididymis, spermatic cord, and colles fascia, as reported in present case. Each type necessitates a different surgical approach [1, 4].

The most common manifestation of cystic lymphangioma is a gradually growing painless mass. Sometimes it may present with sudden onset of pain and rapid increase in size due to hemorrhage within the cyst that could be caused by post-traumatic infection or excessive formation of lymph [4,6,9].

There are many scrotal pathologies such as hernia, hydrocele, hematocele, varicocele, epididymal cyst, spermatocele, spermatic cord lipoma, teratoma, dermoid cyst, epidermoid cyst, and infection that has similar clinical findings with scrotal lymphangioma. Rarely, acute presentation is confused with testicular torsion [1-3,6,8,9]. US examination is very useful as an initial diagnostic tool and can assist in determining the type and extension of the lesion, if any. If the lesion is filled with homogeneous fluid (no echo), it appears as a cystic mass, but the presence of internal echoes reflects bleeding or infection [1,3,9,10]. Doppler can be used in combination with US to distinguish lymphangiomas from other lesions. It may indicate that there is no blood flow in the cystic mass [6,8]. Computed

tomography (CT) and magnetic resonance imaging (MRI) are other important radiological diagnostic tools in cases with suspected pelvic or retroperitoneal extension [1,4,7,9].

Lymphangiography is not recommended as a diagnostic test for lymphangioma because it has no communication with the lymphatic system [4,10]. Tc-99m antimony sulfate lymphoscintigraphy is used to detect complicated lymphocutaneous fistulae and lymphocele [9].

On the other hand, diagnostic aspiration can be used to diagnose the primary lesion as well as recurrent cases [1].

The definitive diagnosis of lymphangioma is made by histopathological examination [1,6]. In surgical treatment, total excision of the mass is preferred with preservation of the intrascrotal structures, which is the gold standard treatment option. In addition, orchietomy may be required for lesions that cannot be separated from spermatic cord structures, and scrotal skin excision in cases of lymphangioma circumscriptum [1]. Although medical treatments such as sclerosing injection, cryotherapy, laser and fulguration can be used, the recurrence rate after such treatment is high [3,4,8]. In the case we presented here, a multicystic mass was found in the patient who was operated with a clinical and radiological diagnosis of hydrocele. The mass was totally excised while preserving the testis. The diagnosis was only made histopathologically. Therefore, although rare, cystic lymphangiomas should be suspected in inguinal surgery.

Compliance with ethical statements

Conflicts of Interest: None.

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Consent: Patient confidentiality is maintained and written consent for the publication of patient details and clinical pictures in this journal has been obtained from the patient's parents or closest relative and can be given as required.

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