SJIF Impact Factor: 5.464

WORLD JOURNAL OF ADVANCE HEALTHCARE RESEARCH

ISSN: 2457-0400
Volume: 4.
Issue: 2.
Page N. 23-26
Year: 2020

Case Report www.wjahr.com

ACQUIRED TESTICULAR AND EPIDIDYMAL LYMPHANGIOMA: A RARE CASE REPORT AND REVIEW

Muthanna S. Abduljawad*

FIBMS-Uro, F.MAS- Urosurgeon Chief Urologist-Department of Urology-Medicano Hospital- Erbil-Kurdistan Region-Iraq.

Received date: 02 January 2020 **Revised date:** 23 January 2020 **Accepted date:** 13 February 2020

***Corresponding author: Dr. Muthanna S. Abduljawad**

FIBMS-Uro, F.MAS- Urosurgeon Chief Urologist-Department of Urology-Medicano Hospital- Erbil-Kurdistan Region-Iraq.

ABSTRACT

Scrotal and intrascrotal lymphangiomas are rare pathologic conditions. Mismanagement and hence rapid recurrence occur due to its rarity and similar clinical and imaging findings with other common pathologic conditions.

INTRODUCTION

Lymphangiomas are rare benign slow-growing lesions of the lymphatic system. They result from an inadequate drainage of lymph from sequestrated lymphatic channels. Genital lymphangiomas are extremely rare. Few reported cases of lymphangiomas involving the scrotum, tunics, spermatic cords, epididymis and testis. Most common presentation is soft scrotal swelling. Due to the rarity of the condition it might be misdiagnosed as hydrocele.[1,2]

Most reported lymphangiomas (90% of cases) are congenital.[2] Acquired cases represent 10% and can occur due to inflammation, trauma or degeneration.[3]

They frequently affect the neck (75%) and the axilla (20%).[4] Intra-abdominal lymphangiomas (fewer than 5%) have been reported in the mesentery, gastrointestinal tract, spleen, liver and pancreas. Scrotal and intrascrotal involvements are extremely rare. Few published literatures discuss intrascrotal lymphangiomas whether congenital or acquired. The clinical presentations and the diagnostic imaging studies mainly the ultrasound are non-specific. Such factors make the final solution a dilemma. Management procedures involves primarily a wide surgical excision for the lesions and the surrounding tissues. Laser ablative procedures and intratunical injection of sclerosing agents are optional treatment however frequent recurrence have been observed.

The current study is aiming for presenting an intrascrotal lymphangioma in a 56 years diabetic patient who has been diagnosed clinically and by ultrasound as hydrocele; and operated as such with rapid recurrence.

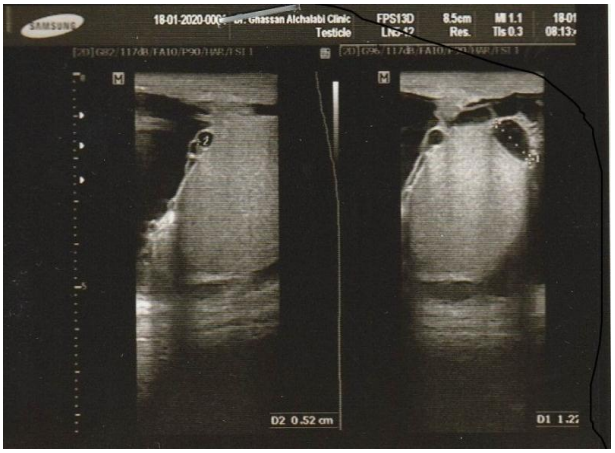
CASE REPORT

The current study was conducted in Medicano Private Hospital in Erbil, at the north of Iraq. Ethical approvements was concerned about and oral as well as written consents were obtained from the studied case.

The studied case was a 56 years old male presented on March, 3rd 2019 with a history of slowly progressive right sided scrotal swelling approximately for more than two years. He had a history of epididymoorchitis infection 3 years ago and had been managed by antibiotic coverage for 2 weeks. No history of trauma to the genetalia was reported by the case. As well, family history of alike lesion was negative.

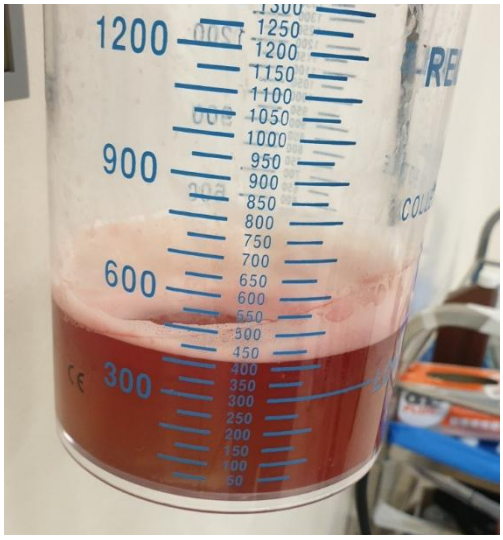
Medical history revealed having type II diabetic for 6 years prior to the consult and well controlled by Metformin 850 mg 1×1, not smoker, not alcoholic.

By clinical examination large non-tender cystic scrotal lesion was detected on the right side. It was extending to the right subinguinal region. Trans-illumination test was positive. Essential laboratory evaluations, included general urine exam, erythrocyte sedimentation rate and renal function tests were normal. Ultrasound revealed large volume right sided hydrocele with internal septa. (Photograph 1).



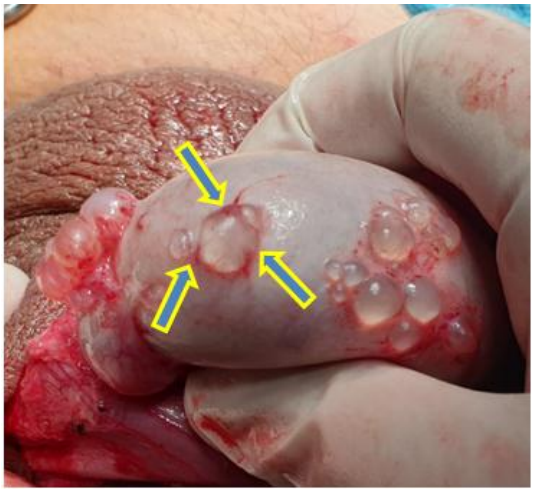
Photograph 1: Septated hydrocele and testicular saccules as revealed by ultrasound view of the Right testis.

A preliminary diagnosis of hydrocele was made, so that hydrocelectomy was decided after a full discussion of the condition with the patient. Operation was conducted under spinal anesthesia with a right sided oblique scrotal incision. Thick tunica vaginalis noted and incised longitudinally. A gush of large amount of pinkish fluid of approximately 400 ml drained completely and a sample sent for cytology. (Photograph 2).

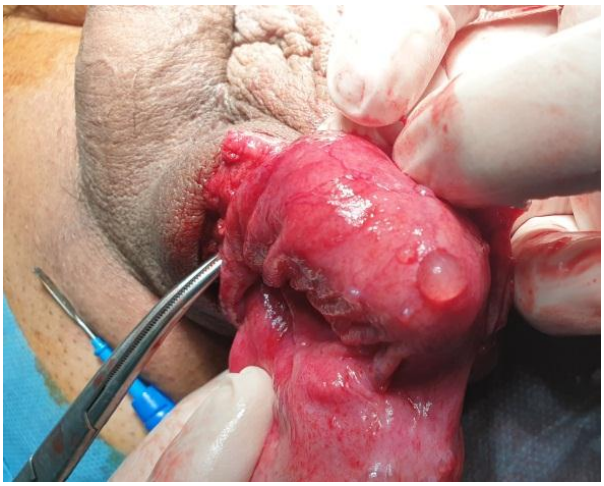


Photograph 2: Large amount of Pinkish fluid has been drained.

Multiple small cystic lesions has been seen projecting from the Tunica Albuginea on the testicular surface and the head of the epididymis. The tunica vaginlis was thickened, septated but no cystic lesions identified in it. (Photographs 3 and 4).



Photograph 3: Multiple thin walled cystic lesions from the tunica albuginea and epididymis



Photograph 4: Thickened septated Tunica Vaginalis.

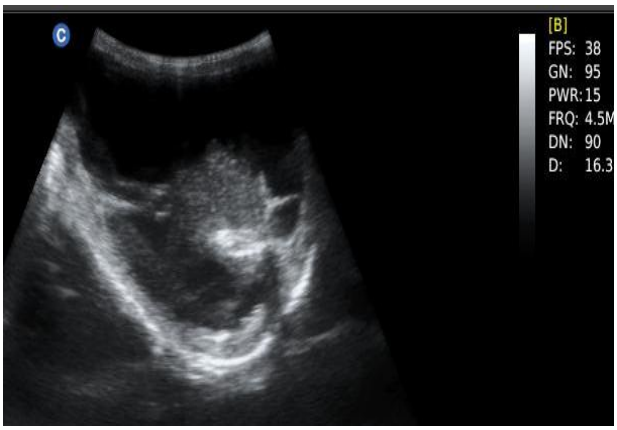
Aspiration of fluid from the cysts has been sent for cytological evaluation and then all cysts were punctured. Pieces from the tunica containing cysts excised for histopathology study. The patient discharged well.

Histopathology study result revealed multiple cystic structures lined by thin attenuated single layer of epithelial cells rest on thin ibrovascular connective tissue wall contain few lymphoid cells. Fluid aspiration and cytology result revealed that the fluid is a cellular containing neither keratin nor sperms.

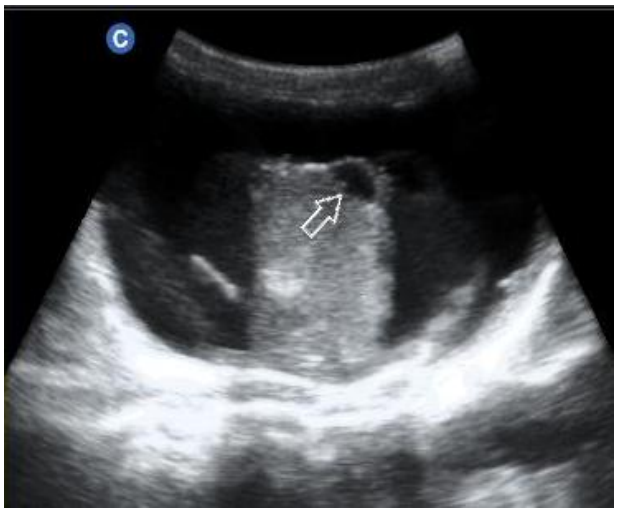
Based on the histopathology study a scheduled follow up was organized to report any abnormality. Neither subjective nor clinical abnormality was reported after one month follow-up of the patient.

After 4 months the patient consulted again for a complain of recurrence of the scrotal swelling. Ultrasound evaluation revealed moderate to severe right sided hydrocele fluid with multiple small cystic lesions scattered on the testicular surface. Abdominal ultrasound was performed in addition to the scrotal ultrasound

during the patient's regular visits to role out involvement of other intra-abdominal organs. (Photographs 5 and 6).



Photograph 5: Scrotal ultrasound revealing recurrent hydrocele with septations.



Photograph 5: Scrotal ultrasound image revealing recurrence of cystic lesion.

Based upon the histopathology result and the clinical outcome, the possibility of redo surgery or injection of sclerosing agent or regular checkup was discussed with the patient.

DISCUSSION

Lymphangiomas are either congenital occurring due to benign proliferation of the lymphatic vessels as hamartomas; or acquired as a result of interruption of previously normal lymphatic channel due to traumatic or inflammatory or degenerative effect on the lymphatic vessels.[2]

Histologically they are divided into 3 types; capillary, cavernous and cystic lesions. Cystic lymphangioma is the most common variant manifested as thin-walled cystic lesions.[5,6] Capillary lymphangioma is a lesion composed of small accumulated lymphatics; cavernous lymphangioma is composed of larger lymphatics. cystic lymphangioma (cystic hygroma-composed of large macroscopic lymphatic spaces with collagen and smooth muscle or merely a thin wall cyst.

Cystic or cavernous lymphangiomas demonstrate large, irregular vascular spaces lined with a single layer of flattened endothelial cells within a fibroblastic or collagenous stroma, which may contain lymphocytes.[8,9]

Clinical presentation and diagnosis

Scrotal and intrascrotal lymphangioma is extremely rare. There are no specific clinical features for this condition. Due to its rarity it can be misdiagnosed as hernia, hydrocele, varicocele or epididymal cysts.[10]

Ultrasound evaluation findings are nonspecific. It might be manifested as multi-cystic extra testicular scrotal mass filled with homogeneous echo free fluid might be present in the inner layer of the Tunica vaginalis, the testicular body surface or the epididymis.[11] Positive echoes in the fluid implies infection or haemorrhage. Such findings that can be observed in intra scrotal pathologic conditions such as hydroceles, spermatoceles, pyoceles.[10] In some instances multiple small intrascrotal cystic lesions can be observed in the tunica, spematic cord or the epididymis.

Management

Medical treatment includes injection of sclerosants such as 1% or 3% sodium tetradecyl sulfate, doxycycline or ethanol, Pulsed Nd-Yag laser fulguration, laser or cryotherapy but is associated with high recurrence rates. Recurrence is common with incomplete excision.[12]

Follow up is very important including thorough Doppler scrotal ultrasound of the scrotal contents and ultrasound of the abdomen and CT scan or MRI of abdomen if ultrasound is suspicious for an intra-abdominal involvement.[12]

Upon reviewing the history of the case presented in our study, the history of epididymo-orchitis is a point of importance as it's considered as a causative factor for cystic lymphangioma.[6,12]

Based upon the clinical findings and examination with ultrasound evaluation a diagnosis of hydrocele had been made. This is due to nonspecific clinical and diagnostic findings that aid in the diagnosis of cystic lymphangioma. We assume that for this reason the scrotal exploration and hydrocelectomy procedure that was performed by us for the patient resulted in an early recurrence.[4,13-14]

The unusual peroperative observation of multiple cystic lesions scattered on the testicular surface raised the suspicion of pathology other than simple hydrocele and necessitated biopsy taking and further post op evaluation especially after obtaining the result biopsy result a cystic lymphangioma. The patient has been advised for a regular checkup as a standard follow up and expectant

treatment and recheck ultrasound of the scrotum and abdomen with the possibility of intrascrotal injection of sclerosing agent or radical orchiectomy and excision of the involved scrotal skin as a final curable solution.[15,16]

CONCLUSION

Although cystic lymphangioma is a rare disease, it represents a frustrating situation for the patient and the doctor. It should be put as a differential diagnosis during evaluation of scrotal swelling. A thorough and comprehensive ultrasound evaluation is mandatory and the urologist should put in mind a possibility of intrascrotal cystic lymphangioma when the ultrasound result states a multiseptated intrascrotal intrascrotal lesion before proceeding with a direct scrotal exploration surgery.

REFERENCES

1. Schwartz R A. Lymphangioma. Medscape, Available online on URL: https://emedicine.medscape.com/article/1086806-overview, 2019.
2. Muceli A., Stewart K M. Lymphangioma. Bookshelf, NCBI, Available online on URL: https://www.ncbi.nlm.nih.gov/books/NBK470333/, 2019.
3. Koshy A, Tandon R K, Kapur B M, Rao K V, Joshi K. Retroperitoneal lymphangioma. A case report with review of the literature. Am J Gastroenterol, 1978; 69(4): 485–490.
4. Bhavsar T, Vasa D S, Harbinson S, Innis S. Retroperitoneal cystic lymphangioma in adult: A case report and review of literature. World journal of gastrointestinal physiology, 2010; 1(21607159): 171-176.
5. De Perrot M, Rostan O, Morel P, Le Coultre C. Abdominal lymphangioma in adults and children. Br J Surg, 1998; 85: 395–397.
6. Ratan S K, Rattan K N, Sehgal T, Maggu S. Spermatic cord lymphangioma in a 7 year old child masquerading as a hydrocele. Br J Urol Int, 2003; 10: 4046.
7. Bahl S, Shah V, Anchilia S, Vyas S. Adult-onset cystic hygroma: A case report of rare entity. *Indian journal of dentistry*, 2016; 7(27134456): 51-54.
8. Hara H, Mihara M, Anan T, Fukumoto T, Narushima M, Iida T, Koshima I. Pathological Investigation of Acquired Lymphangiectasia Accompanied by Lower Limb Lymphedema: Lymphocyte Infiltration in the Dermis and Epidermis. Lymphat Res Biol, 2016 Sep; 14(3): 172-80.
9. Stewart CJ, Chan T, Platten M. Acquired lymphangiectasia ('lymphangioma circumscriptum') of the vulva: a report of eight cases. Pathology, 2009; 41(5): 448-53.
10. Pradeep Kajal, Kanal Nain Rattan, Vivek S Malik, Viipin Garsa. Scrotal Lymphangioma- A rare Cause of Scrotal Swelling. APSP Journal of Case Report, 2013; 4(5): 4-7.
11. Loberant N, Chernihovski A, Goldfeld M. Role of Doppler sonography in the diagnosis of cystic lymphangioma of the scrotum. J Clin Ultrasound, 2002; 30: 384-387.
12. Hurwitz R S, Shapiro E, Hulbert W C, Diamond D A, Casale A J, Rink R C. Scrotal cystic lymphangioma: the misdiagnosed scrotal mass. J Urol, 1997; 158: 1182–1185.
13. Thomas AM, Leung A, Lynn J. Abdominal cystic lymphangiomatosis: report of a case and review of the literature. Br J Radiol, 1985; 58: 467–469.
14. Loberant N, Chernihovski A, Goldfeld M. Role of Doppler sonography in the diagnosis of cystic lymphangioma of the scrotum. J Clin Ultrasound, 2002; 30(6): 384–7.
15. Olabanji JK, Oladele AO, Famurewa OC, Adejuyigbe O, Ademola SA. etroperitoneal and Genital Lymphangioma Therapeutic Challenges in a Developing Country. Libyan J Med, 2009; 4(1): 44-45.
16. Okoro PE1, Anyaeze CM, Ngaikedi C. Recurrent lymphangioma: what are the treatment options?. African Journal of pediatric surgery, 2009; 6(10.4103/0189-6725.48576).