

CYSTIC HYGROMA OF SPERMATIC CORD: THE HIDDEN MYSTERY

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ABSTRACT

Lymphangioma is a benign infiltrative malformation of lymphatic channels commonly occurring in neck, axilla, mediastinum and retroperitoneum. It is mostly congenital but may also occur secondary to trauma, infection, inflammation or degeneration. Cystic hygroma is a subtype of lymphangioma which exhibits large cystic (macro-cystic) spaces histologically. We report a rare case of a 17 year old boy who presented with a painless cystic inguino-scrotal swelling. After inguinal exploration, histopathological examination of excised cystic mass showed it to be a lymphatic malformation (cystic hygroma) of spermatic cord. Complete surgical excision of cystic hygroma with preservation of cord structures was performed. Any cystic inguinal swelling should alert the surgeon to consider the possibility of inguinal lymphatic malformation as a differential diagnosis. Complete surgical excision of lymphangioma is mandatory to prevent recurrence.

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INTRODUCTION

Lymphatic malformations represent morphogenic errors in the development of the lymphatic vessels. Most commonly, those with large cysts have been called hygromas and those with more tissue parenchyma have been called lymphangiomas¹. They are most commonly located in regions of confluence of major lymphatic channels, including the neck (75-90%), axilla (20%), mediastinum and retroperitoneum². The cause of cystic hygroma is believed to be developmental defect or primary multi-locular cystic malformation of dilated lymphatic channels. Cystic hygroma commonly occurs in posterior triangle of the neck as a large, deep diffuse swelling². They often cross the midline, reaching axilla and mediastinum. Such localization verifies the complexity and extent of the lymphatic system in the cervical region when compared to other regions of the body. They usually appear as solitary lesions. They are usually infiltrative, often separating fascial planes and tend to invest normal vital structures i.e., nerves, muscles, and blood vessels. They are fluctuant, painless and trans-illuminate well. The skin overlying the lesion is normal and usually there is no associated lymphadenopathy. These lymphatic anomalies may become infected or sustain intra-lesional hemorrhage. Predominantly macro-cystic lymphatic malformations may be treated with cautious intra-lesional sclerotherapy using ethanol, doxycycline, sodium tetradecylsulfate, and OK-432 (a killed strain of group-A streptococcus) under fluoroscopic guidance³.

Unfortunately, persistent malformation and recurrent swelling are common after sclerotherapy. Complete Surgical resection is the only way to “cure” these malformations⁴. The strategy should be to perform as thorough a resection as possible in a single anatomic region. Extensive lesions cannot usually be removed completely, even with multiple procedures. Re-enlargement of residual lesion is not uncommon if an anatomic region is not adequately resected⁵. Great care must be taken to avoid injury to vital structures. Proper pre-op evaluation of the extent of the lesion radiologically is important in planning the appropriate approach for the mandatory complete surgical excision⁶, to avoid the recurrence commonly associated with incomplete excision.

CASE REPORT

A 17 year old boy presented to us with a slow growing gradually progressive painless swelling in the left inguino-scrotal region since 4 years [Fig 1].



Fig 1 Left Inguino-scrotal ovoid swelling

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On clinical examination, a 7x5 cm ovoid, non-tender, soft cystic swelling was palpable in the left inguino-scrotal region which was fluctuant, brilliantly trans-illuminant, palpable separately from testicle but not palpable separately from cord structures, irreducible and did not have any impulse on coughing. There was no palpable inguinal lymphadenopathy. Ultrasound revealed a large encysted collection of 81x34 mm of 26 cc in close association with left spermatic cord in left inguino-scrotal region with patulous deep inguinal orifice. Differential diagnoses considered were encysted hydrocele of left spermatic cord, encysted hydrocele of irreducible hernial sac and lymphatic malformation of spermatic cord. Inguinal exploration was performed. An 8x4x3 cm multi-loculated cystic lesion was identified amidst spermatic cord structures containing brownish black fluid [Fig 2] and separate from testicle.

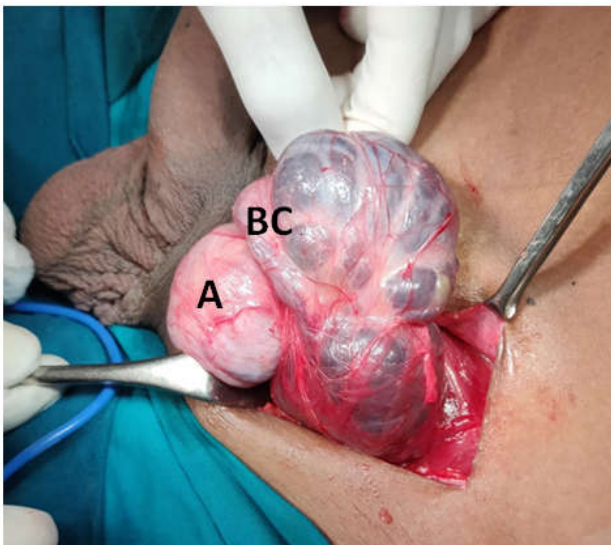


Fig 2 Multi-cystic mass in close association with spermatic cord [A- Testicle, B - Spermatic cord, C - Multiloculated cyst]

The cystic mass was found deep to the external oblique aponeurosis with a patulous deep inguinal ring but had no further intra-abdominal extension [Fig 3]. The entire extent of cystic mass was carefully dissected out in toto by separating and preserving the structures of the spermatic cord.

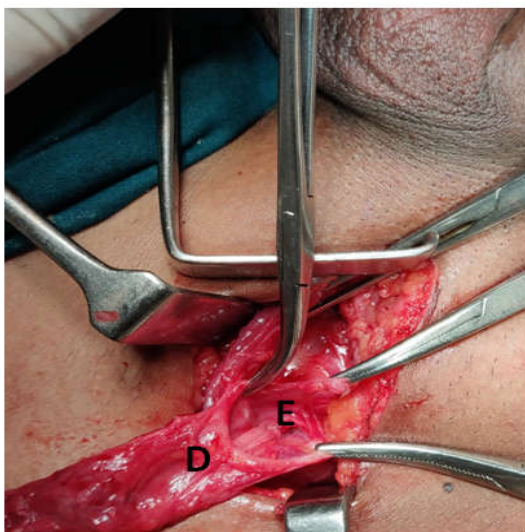


Fig 3 Cyst wall with patulous deep inguinal ring [D - cyst wall; E - deep inguinal ring]

Gross examination of cystic mass showed multiple variable size cystic cavities with smooth inner lining [Fig 4] and was filled with brownish black color clear fluid suggestive of lymphatic fluid.

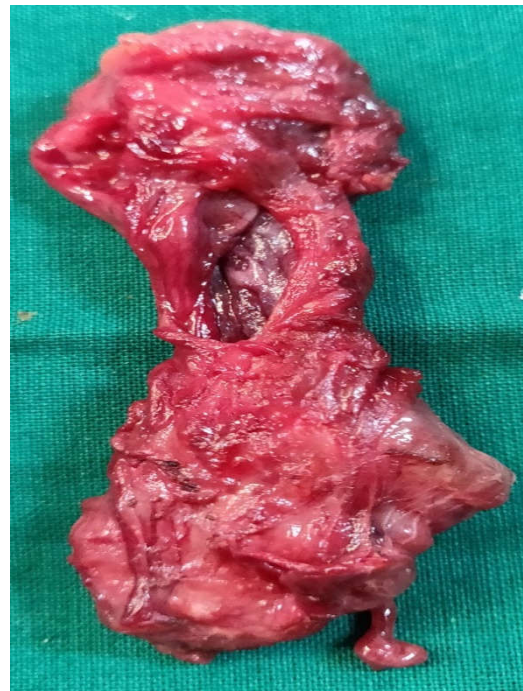


Fig 4 Gross photograph of cystic mass showing multiple variable size cystic cavities with smooth inner lining

On microscopic examination, sections showed many cystically dilated spaces lined by flattened endothelium, consistent with diagnosis of cystic hygroma [Fig 5]. The post-operative period was uneventful and patient was discharged on 4th post-operative day. 6 months post-operatively, the follow-up USG showed no evidence of recurrence.

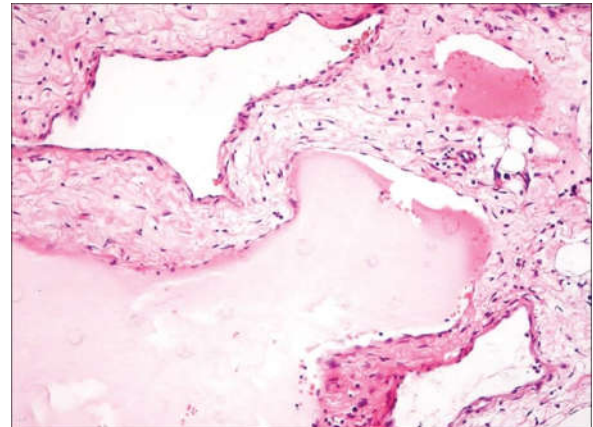


Fig 5 Microscopy showing large, irregular dilated lymphatic channels lined by attenuated bland endothelial cells s/o cystic hygroma

DISCUSSION

Lymphangiomas are malformations of the lymphatic system that occur as a result of the failure of lymph to drain from sequestered lymphatic vessels with consequent dilatation of the ducts and formation of a cystic mass. Majority of these lesions are congenital but they may also occur secondary to trauma, infection, inflammation or degeneration. Most commonly, those with large cysts have been called hygromas and those with more tissue parenchyma have been called lymphangiomas¹. The International Society for the Study of

Vascular Anomalies (ISSVA) has accepted the terminology proposed by Mulliken in 1982 more appropriately describing these lesions as 'lymphatic malformations'. They are subdivided into macro-cystic, micro-cystic and combined varieties. Most cases present as a progressively slowly enlarging soft mass. 95% of these lesions occur in the neck and/or axilla². Inguino-scrotal presentation of cystic hygroma is very rarely reported in literature. Intra-lesional sclerotherapy can be tried, but recurrence is high³. Draining fluid from cystic hygroma provides only temporary relief, so it should be completely removed surgically⁴. Ultrasonography is very accurate in identifying the cystic nature of the swelling and marking the extent of the lesion. The final diagnosis is usually made only after the histopathologic examination. A cystic or septate mass associated with spermatic cord discovered intra-operatively should not be dismissed as a hydrocele of cord, since cystic hygroma predictably recurs if incompletely resected⁵. So proper pre-operative diagnosis of the spermatic cord hygroma and its extent utilizing USG is essential for planning the appropriate approach for the mandatory complete surgical excision of hygroma to prevent recurrence⁶.

CONCLUSION

Any cystic inguinal swelling should alert the surgeon to consider the possibility of inguinal lymphatic malformation as a differential diagnosis. Cystic hygroma of cord may mimic a variant of hydrocele of cord & may pose an initial diagnostic dilemma.

Proper pre-operative diagnosis of spermatic cord hygroma & delineating its extent & cystic nature utilizing USG is essential for planning the appropriate surgical approach. Surgery should be mandatory complete excision of cystic mass with preservation of structures of spermatic cord.

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