Case Report

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NON-INTERCOMMUNICATING ABDOMINOSCROTAL HYDROCELE WITH MULTI-SYSTEM ANOMALIES: A CASE REPORT

Gadelkareem R.A.

MD Urology, Lecturer of Urology, Assiut Urology and Nephrology Hospital, Faculty of Medicine, Assiut University, Assiut, Egyptt.

ABSTRACT

Aim: Presentation of an abdominoscrotal hydrocele case with a very rare association to multiple anomalies and separate abdominal and inguinoscrotal sacs.

Case Report: A 21-year-old male patient presented with left-sided abdominoscrotal hydrocele, impalpable right testis, hydrocephalus and cerebral and musculoskeletal deformities. On abdominal ultrasonography, the abdominal and inguinoscrotal components were seen separated by a septum that was confirmed on surgery. Extended inguinal incision was used for repair and the post-operative course was uneventful.

Discussion: Abdominoscrotal hydrocele is a rare entity and its simultaneous association to multiple anomalies, like in the current case, is a rarer finding. Non-intercommunication state is another very rare criteria of abdominoscrotal hydrocele which was clinically, sonographically and surgically demonstrated in this case. Extended inguinal incision was indicated and enough to deal with both abdominal and scrotal components.

Conclusion: Abdominoscrotal hydrocele is mostly intercommunicating, but it is exceptionally non-intercommunicating and associated to multiple anomalies. Surgical excision via an extended inguinal incision may be indicated for voluminous cases and those with a solitary testis.

Key Words: Abdominoscrotal Hydrocele, Hydrocephalus, Septum

INTRODUCTION

Abdominoscrotal hydrocele is a simple hydrocele extends into the abdominal cavity forming two intercommunicating sacs (inguinoscrotal and abdominal) in the hourglass fashion[1, 2]. Old and recent reviews described this lesion as an unusual or uncommon entity[1-3]. Initially, it was reported among adults as single case reports [1-4]. However, few case series were published, especially among pediatrics[3].Regarding the nomenclatures, Prather[1] credited the initial term *"l'hydrocèle en bissac"* to Dupuytren in 1834[4] and the more acceptable term *"abdominoscrotal hydrocele"* to Bickle in 1919[5]. However, this latter term was coined, firstly, by Baitcheff in 1903[6].

Only three cases were published from Egypt, a century ago[7-9], and the case reported below is just the fourth one.

CASE REPORT

A 21-year-old male patient presented by a long-standing left inguinoscrotal swelling with a recently developed lower abdominal swelling. Also, he presented by other multiple anomalies and defects including reduced mental capabilities and epilepsy which were diagnosed and managed early in childhood and adolescence. Also, the mother gave a history of previous consultation for treatment of a small scrotal hydrocele and absent right testis, 5 years ago. Laparoscopy was done for the right testis which was diagnosed as atrophied intra-inguinal one. After a missing follow up, however, she returned for treatment because of the increasing swelling size and the patient's complaint of progressive scrotal heaviness.

On physical examination, there were:

1. Left inguinoscrotal mass that seemed to be continuous with another visible lower abdominal mass. The latter mass extended upwards to a level near to the umbilicus and slightly crossed the midline to the right side. The scrotal mass had a smooth surface and looked as a bilocular swelling at site of the deviated median raphe to the right (Fig. 1). The abdominal and scrotal masses were cystic with no cross-fluctuation between them

Corresponding Author:
Gadelkareem R.A., Rabea Ahmed Gadelkareem, MD Urology, Lecturer of Urology, Assiut Urology and Nephrology Hospital, Faculty of Medicine, Assiut University, Assiut, Egypt; E-mail: dr.rabeagad@yahoo.com

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could be elicited, but hardly the gross movement of the scrotal mass could be felt through the abdominal one. Scrotal mass was transilluminable. Both testes could not be palpated.

2. Other multi-system anomalies and lesions:
   2.1. Hydrocephalus: Huge head with exophthalmos, abnormal teeth and senile appearance.
   2.2. Cerebral lesions: Slurred and unclear speech and diminished mental capabilities.
   2.3. Musculoskeletal deformities: Dorsolumbar kyphosis, flat foot, short stature and under body built.

Abdominal ultrasound examination revealed a marked left inguinoscrotal anechoic mass with clear fluid content and thin smooth capsule. This mass extended through the left inguinal canal into the abdominal cavity as another large cystic component with measured dimensions as 15 cm x 10 cm x 8 cm at the widest dimensions. A unique finding of a thin septum was noted at the level of the internal inguinal ring separating the two components (Fig. 2). The report described the left testis with absent right one. Preoperative routine laboratory work up and surgical fitness were unremarkable.

Surgery was done through an extended inguinal incision like that described by Roller [10]. The left inguinal canal was opened, the widened internal ring was incised, and the incision extended upwards by cutting the lower abdominal wall muscles. There was a glistening and transparent hydrocele occupying the inguinoscrotal region with intra-abdominal component. A well-defined ring constriction was obvious at the level of the internal inguinal ring (Fig. 3) which mostly corresponds to the sonographically described septum. Abdominal sac was pro-peritoneal reaching above the level of the umbilicus. It had few adhesions with the surroundings and was dissected by sharp and blunt dissections. It was pear-shaped when delivered from the wound (Fig. 3). A fibrous band connection was found at the inferomedial aspect of the sac. Mostly, this band represented the obliterated processusvaginalis and it was ligated and transected. Another finding that confirmed the non-intercommunication state between the two sacs was that the abdominal one remained full after accidental rupture of the inguinoscrotal one. Then the ruptured scrotal sac was drained, dissected, and excised, except the part adherent to the testis. This residue of the tunica was managed like in simple hydrocelectomy where it was reduced and everted around the testis. Drained fluid was clear, greenish yellow, and about 2 to 2.5 liters. The left testis looked larger than normal, fusiform, and was slightly soft in consistency. These slight testicular dysmorphism findings could be attributed to the prolonged compression effect of hydrocele. The lower abdominal wall and inguinal canal were repaired, a drain was inserted in a dependent part, and the scrotum was compressed within a bandage. Post-operative recovery course was uneventful.

DISCUSSION

Abdominoscrotal hydrocele is an anomaly of the processusvaginalis which originates from a simple (mostly infantile) hydrocele [11, 12]. Its association to other anomalies like cryptorchidism and contralateral herniais encountered frequently[13]. However, the simultaneous association to multiplegenital and extra-genitalanomalies, like the current case, is unusual. Currently, no specific explanation for this association could be speculated. So, it is considered as just a matter of co-existence until studying of more similar cases could prove otherwise.

The scrotal and abdominal sacs are connected via the isthmus which is a narrow segment corresponds to the inguinal canal giving them the hourglass appearance[1, 9]. Abdominal sac is usually the larger and may reach up to the costal margins[9, 14, 15]. Its relations to the peritoneum were classified into pro- and retro-peritoneal, but like the current case, it seems to be commonly pro-peritoneal[1, 2]. Also, as I found in this case, its coverings were described to be formed mainly of the transversal is fascia [2].

Free intercommunication between the two components is a constant pathological finding. It is the underlying basis of all the diagnostic clinical and imaging criteria in old and recent literature[2, 16, 17]. However, there were four reported cases with a separation between the abdominal and scrotal components; all were among adults[18-21]. In one of those cases, there was a wide distance between the sacs [21]. In another case, however, the abdominal and scrotal components were adherent, but could be separated surgically [20]. In the current case, absence of cross fluctuation, detected septum by ultrasound and the gross finding of a ring constriction at the level of the internal inguinal ring referred to an abdominoscrotal hydrocele with separate components. This finding was confirmed after delivery of the abdominal component and evacuation of the scrotal one, while the former was remaining full. The septum was very thin and the two components were adherent and non-separable surgically. Roller's suggestion [10]of an encysted hydrocele of the cord with a vaginal hydrocele as etiology of abdominoscrotal hydrocele is accepted for cases like the current one. However, to withstand the intercommunicating form in this hypothesis, I suggest a possibility of septum recanalization under the effect of increasing internal pressure.

Non-communication to the peritoneal cavity is a cardinal feature in most of the reported cases, especially in adults[1, 16]. The present case finding of an obliterated and fibrosed processusvaginalis correlates to this common finding.

Tense compressing mass effect of the internal pressure is the reference of many complications as testicular dysmorphism, lymphedema and hydronephrosis[12, 14]. In the current

case, there was only one large-volume left testis with hardly detectable dysmorphism.

The commonly reported method of treatment is the surgical excision of the sacs(hydrocelectomy), early as can as possible[3]. The gold standard surgical approach is through the inguinal incision [13].It is predominantly used because of its effectiveness in dealing with the both components in adults and pediatrics [15, 22]. Extended inguinal (inguinoabdominal) incision could be employed to facilitate dissection of the large abdominal sac [17]. In the old literature, abdominal and combined abdominal and inguinal or scrotal incisions were employed[8, 9]. Also, dissection of the abdominal sac was claimed to be easy, but some difficulties were reported in few cases of that era[2].

To the best of my knowledge, this case is the first case of abdominoscrotal hydrocele with this number of different anomalies and deformities; the second one to be associated to head deformities after the case of Delmonaco et al. who reported abdominoscrotal hydrocele with macrocephaly [23]; and the fifth one regarding the presence of non-intercommunicating components.

CONCLUSION

Abdominoscrotal hydrocele may be associated to multi-system anomalies and deformities, simultaneously. It is commonly intercommunicating two-component hydrocele, but exceptionally, it could be non-intercommunicating. Ultrasonography is sufficient for confirmation of the diagnosis. Extended inguinal incision could be employed, especially in cases with solitary testis and voluminous cases.

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Ethical approval

This case study was approved by the ethical committee of the Faculty of Medicine in Assiut University. Informed consent was taken from the patient's mother.

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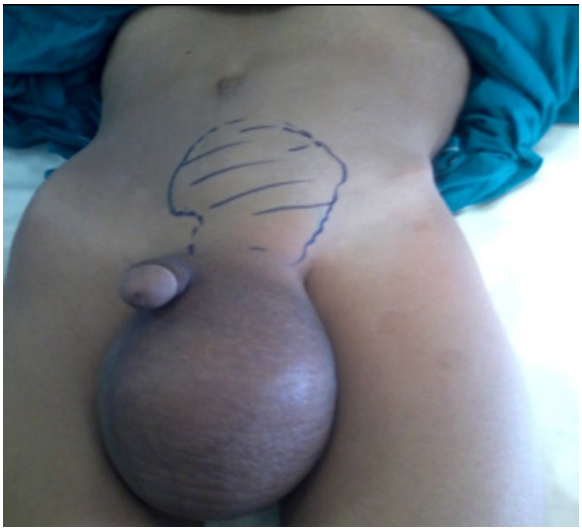

Figure 1: Clinical appearance of left-sided abdominoscrotal hydrocele;caudo-cephalic view.

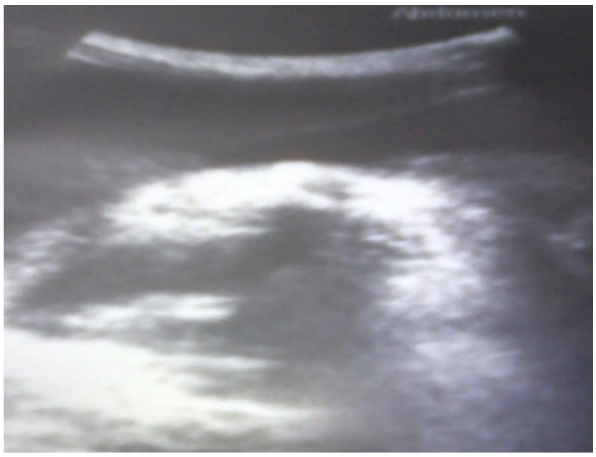

Figure 2: Very thin septum on ultrasonography at level of inguinal ring.

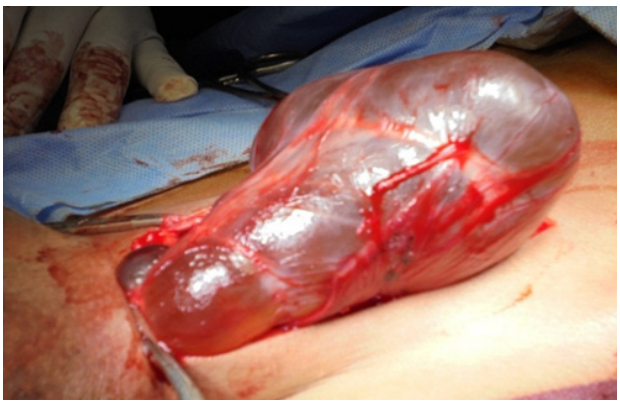

Figure 3: Pear-shaped abdominal component delivered through the extended inguinal incision: Note the signs of adhesion, site of the ring constriction and septum at the level of internal ring.

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