CASE REPORT

Be cautious of “complex hydrocele” on ultrasound in young men

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Summary
Hydrocele is the most common benign cause of painful scrotal enlargement and only very rarely can be reactive to an underlying testicular tumor. We present the case of a healthy young man, complaining of mild left scrotal discomfort and swelling. Physical examination revealed a non-tender fluctuant left scrotum and serum tumor markers were normal. Scrotal ultrasonography (US) showed a normal right hemiscrotum and testicle and a fluid collection among thickened irregular septations in the left hemiscrotum, a finding which was considered as a complex hydrocele. Intraoperatively the presumed “complex hydrocele” was in fact a multicystic testicular tumor. We proceeded with orchietomy through the scrotal incision and pathology revealed a mixed germ cell tumor of the testis consisting of cystic teratoma, in situ germ cell neoplasia unclassified (IGCNU) and Sertoli cell tumor. This is the first reported case of this type of testis tumor presenting as complex hydrocele. The aim of this case presentation is to underline the need for an accurate preoperative diagnosis in cases of suspected scrotal pathology in young males.

KEY WORDS: Hydrocele; Testis tumor; Scrotal ultrasonography; IGCNU; Sertoli cell tumor.

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INTRODUCTION
Hydrocele is usually caused by either the presence of a patent processus vaginalis or an imbalance in the secretion and absorption of fluid within the tunica vaginalis. Increased fluid secretion may be a result of inflammation, whereas poor absorption commonly results from thickening of the hydrocele sac or impaired lymphatic drainage. So-called reactive hydrocele is present in 10% of testicular tumors. Seminoma is the most common type of testicular tumor (40%) while non-seminomatous germ cell tumors are more rare (1). A mixed germ cell tumor consisting of cystic teratoma, Sertoli cell tumor and in situ germ cell neoplasia unclassified (IGCNU) is very rare. Scrotal ultrasound (US) plays a pivotal role in the diagnosis of hydrocele and other scrotal pathology. However, there are cases where scrotal ultrasonography might fail to reveal a hidden testicular pathology (2-5). We present a case of a hydrocele with complex ultra-sonographic features which during elective surgery turned out to be an unusual testicular tumor.

CASE PRESENTATION
A 24-year-old male presented with a complaint of mild left scrotal discomfort of 3 months’ duration. His medical history was unremarkable with no recall of scrotal trauma or other urological symptoms. Physical examination revealed a painless, non-tender, moderately enlarged left hemiscrotum and a non-palpable ipsilateral testis. Complete blood count and urinalysis were normal. Tumor markers were within normal range with alpha-fetoprotein (a-FP) of 0.96 ng/ml and β-human chorionic gonadotropin (β-HCG) < 1.20 mIU/ml. Scrotal US showed a large amount of fluid collection in the left scrotal sac containing multiple thick echogenic septations, a finding described by the radiologist as a “complex hydrocele” (Figure 1). Scrotal US also revealed normal size and echogenicity of both testicles. The sonographic findings were consistent with a “complex” hydrocele of the left testis with inflammatory changes in the surrounding scrotal layers. An elective left hydrocelectomy was performed under the presumptive diagnosis of a complex hydrocele. Scrotal exploration ruled out the presence of a hydrocele, instead it revealed the presence of a multicystic testicular tumor distorting the testicular parenchyma. Following intraoperative consensus and informed parental consent we proceeded with orchietomy via the scrotal incision (Figure 2). Histopathology revealed the presence of a mixed germ cell tumor consisting of cystic teratoma, Sertoli cell tumor and IGCNU (Figure 3). Following multidisciplinary team (MDT) discussion and negative axial tomography staging the patient received prophylactic chemotherapy and is free of recurrence at 6-month follow-up.

DISCUSSION
Although in most cases hydrocele presents as a benign painless swelling, rarely a reactive hydrocele could be
Figure 1.
Scrotal ultrasound (US). Arrows depict the anechoic fluid inside the thick septations of the scrotal sac. (A, B, C). Image (D) shows a normal in size and echogenicity left testis. Images (E, G, F, H) demonstrate a right testis with homogeneous echo texture and echogenicity.

Figure 2.
Macroscopic appearance of the multicystic mass, depicting the atrophic left testicular parenchyma. It is worth mentioning the absence of hydrocele.

the first and only sign of a testicular tumor (1, 2). Our case highlights the diagnostic pitfalls of a non-tender scrotal enlargement clinically and radiologically indicative of a complex hydrocele, which proved to be a testicular tumor in the operating room.

A “complex” hydrocele on US is characterized by the finding of multiple thick echogenic septations and calcifications surrounded by fluid with layering echogenic debris (3, 5). Further imaging might be necessary in cases scrotal US is ambiguous about the diagnosis of hydrocele. Magnetic resonance imaging (MRI) emerges as a valuable problem-solving imaging modality in cases of inconclusive testicular parenchyma and the presence of a green caseous material led to the decision for orchietomy. Histopathology revealed an epidermoid cyst (8). Iqbal et al. reported two cases of testicular cancer which were initially diagnosed as hydroceles and operated via a scrotal approach. Both patients underwent hydrocele repair, and concomitant testicular biopsy revealed embryonal cell carcinoma of the testis which led to radical orchietomy (9).

A metastatic to the testis pancreatic adenocarcinoma was also misdiagnosed as complex hydrocele in a 69-year-old patient who presented with painful swelling of the left scrotum and underwent left hydrocelectomy (10). Testicular teratomas can sometimes have a predominantly cystic appearance closely resembling a hydrocele. Lin et al. presented a case of a 3-year-old boy with a transilluminating scrotal mass initially diagnosed as a communicating hydrocele. Due to impalpable testes they proceeded with ultrasonography which depicted a heterogeneous cystic mass with subsequent orchietomy revealing mature teratoma (11). There was also a recent report of a case of adult germ cell testicular tumor with again sonographic features suggestive of hydrocele (12).

Recently, Trens et al. cited an infrequent case of mesothelioma of the tunica vaginalis tests, secondarily diagnosed during hydrocele surgery. Contrary to our case, the ultrasonographic findings were that of a simple left hydrocele (13). In our case histopathology revealed the presence of a mixed germ cell tumor consisting of cystic teratoma, IGCCNU and gonadal stromal tumor (Sertoli cell). This is the first reported case of misdiagnosis of this type of testis tumor as complex hydrocele. At MDT discussion of the case one of the proposed reasons for the missed diagnosis was the fact that the extended cystic part of the tumor gave a very hypoechoic image on US mimicking

Figure 3.
Histological images. (A) Hematoxylin - Eosin stain (x100): Sertoli cell tumor composed mostly of solid nests. (B) Hematoxylin - Eosin stain (x100): In situ germ cell neoplasia unclassified (ISGCU); proliferation of malignant germ cells resembling primitive gonocytes confined to the basilar aspect of the seminiferous tubules. (C) Hematoxylin - Eosin stain (x40): Gastrointestinal type epithelium and endometrial foci in teratoma of the testis.
the hypoechoic fluid accumulation seen in a typical hydrocele. On the other hand, it was noted that complex hydroceles usually result from previous failed hydrocelectomies or infected hydroceles and very rarely in de novo cases (14).

Irrespective of the aetiology, misdiagnosis of a testicular tumor as a complex hydrocele has significant implications in surgical planning as a scrotal incision used for hydroceles is contraindicated in cases of suspected testicular malignancy, and also for patient counselling (2, 9, 15). As a result of this misdiagnosis our patient was listed for a non-urgent elective procedure of hydrocele repair, whereas if a testicular tumor was suspected he should have taken priority and have had the procedure done within days of diagnosis.

A high level of suspicion should be maintained in any case of presumed hydrocele with discordance between testicular size, morphology and echotexture on scrotal US especially in younger men. It is important to underline the need for preoperative counseling for the risk of orchietomy if unanticipated pathology is realized during surgery. In cases of ultrasonographic diagnostic dilemmas, further imaging with scrotal MRI should be the next diagnostic step in cases of complex scrotal conditions.

CONCLUSIONS

What initially appeared to be a complex hydrocele on ultrasound, was finally diagnosed as a testicular tumor. In this setting, we aimed to underline the need for diligent preoperative ultrasound evaluation of the scrotum in young adults.

REFERENCES