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Review

Abdominoscrotal hydrocele: A systematic review and proposed clinical grading

R.A. Gadelkareem *

Assiut Urology and Nephrology Hospital, Faculty of Medicine, Assiut University, Assiut, Egypt

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Abstract
Introduction: Abdominoscrotal hydrocele is a rare hydrocele variant in pediatrics and adults. Besides the historical concerns, controversies in etiology and management of abdominoscrotal hydrocele warrant studying.
Subjects and methods: A systematic review was conducted based on a multilingual search of the world literature of abdominoscrotal hydrocele through electronic engines (Google Scholar and MEDLINE/PubMed). The demographic and clinical characteristics are critically addressed and a clinical grading system is proposed.
Results: From the 487 delivered articles, 320 articles were eligible to this review including only 21 case series. They delivered 579 abdominoscrotal hydrocele cases. Abdominoscrotal hydrocele affects pediatrics more than adults with significantly increased rate of reporting in the last decades. Full or incomplete case descriptions were found in 558 cases versus 21 cases with deficient description. Abdominoscrotal hydrocele has been reported from 45 countries and India has the highest rate. Eight proposed hypotheses were differentiated for etiology and grouped according to the direction of fluid formation and hydrocele growth. Associated congenital anomalies include contralateral hydroceles and cryptorchidism. Complications result from compression, hemorrhage, infection, torsion, and coincident malignancy. A clinical grading system considering the increased anatomical, pathological or clinical complexities has been proposed and provided two categories; simple and complex abdominoscrotal hydrocele with further sub-classes.
Conclusions: Abdominoscrotal hydrocele is rare, but the number of the reported cases is far larger than the previously reported numbers. Etiology follows multiple hypotheses and management is speculative. Proposed clinical grading may support differentiation of severity of the associated cumulative risks.

Abbreviations: AIH, abdominoinguinal hydrocele; ASH, abdominoscrotal hydrocele; ASH-C, complex abdominoscrotal hydrocele/complex ASH; ASH-S, simple abdominoscrotal hydrocele/simple ASH; OPV, obliterated processus vaginalis; PPV, patent processus vaginalis.

* Correspondence to: Assiut University, Elgamaa Street, Assiut 71515, Egypt.
E-mail address: dr.rabeagad@yahoo.com

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Introduction

Most of the detectable publications of abdominoscrotal hydrocele (ASH) proposed the same definition as a hydrocele extends to the abdominal cavity forming two intercommunicating compartments (inguinoscrotal and abdominal) [1–4]. Observable confusions in the historical aspects, controversies in incidence and pathogenesis, and differences in clinical presentation and management warranted systematic studying. This is a review of demographic and clinical aspects of ASH. A new clinical grading system is also proposed. Such an instrument may enhance evaluation of the risks associated with the clinical, anatomical, and/or pathological ASH complexities to improve management and prognosis.

Subjects and methods

A comprehensive multi-lingual search of the world literature was carried out on Google Scholar (Web and Books) and MEDLINE/PubMed for publications of ASH. Time frame was unlimited. Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines were the reference of methodology. Employed English keywords were abdominoscrotal hydrocele, hydrocele en bissac, bilocular hydrocele, abdominal hydrocele, intraabdominal hydrocele, hydrocele of Dupuytren, hourglass hydrocele, and their further combinations. Also, the corresponding terms in other languages were used and included Hydrocele bilocularis, Hydrocele bilocularis abdominalis/intraabdominalis, Biloculären intraabdominalen Hydrocele, Hydrocele biloculaire (German/Dutch), l'hydrocèle/hydrohematocele en bissac, l'hydrocèle de Dupùytren (French), Ematocele biloculare (Portuguese), Hidrocele abdominoescrotal (Spanish), Idrocele addominoscrotale (Italian), and Abdominoskrotal hidrosel (Turkish). Moreover, the related articles from the bibliography of the delivered articles were further employed for extended searching or delivering of reported cases. Inclusion criteria considered the articles with texts (structured or not, but describe one ASH case at least) which were published as journal articles, text books, reports in scientific meeting, theses, or reports in scientific websites. Review articles without new reported cases, articles of incorrect/confused diagnoses, and articles written in sites other than those in the inclusion criteria were excluded and not considered for counting the total number of ASH. Then, articles with duplicated publications were excluded. Texts of languages other than English were translated into English using online translation programs; Online PROMT-Online Translation, Google Translation, and Microsoft Translation (powered by PROMT-Online, Google™ and Bing™). The measurable outcome was defined as the description (demographic and clinical characteristics) of one original ASH case or more. Full form of description should cover the following items: demography, clinical presentation, investigations, diagnosis, associated anomalies, complications, and treatment. Incomplete description misses one or two items only. Deficient description misses >2 items. The Methodological Index for Non-Randomized Studies (MINORS) instrument [5] was used for evaluation of the quality of evidence. According to the number of the reported cases, articles were distributed into case reports and cases series (≥5 cases). All articles were subjected to data extraction and stratification. Detailed description of the demographic (age, country, incidence, and style of case reporting; single case or case series), clinical (symptoms, signs, investigations), pathological (anatomical side, site, type, associated anomalies or co-morbidities, physical and biochemical criteria of hydrocele fluid, and complications), and surgical (incisions, approaches, techniques, complications) characteristics were studied. These data were discussed and employed for construction of a clinical up-grading system for stratifying the ASH varieties in a novel clinical perspective considering the potential risks of the cumulative anatomical, pathological, and clinical complexities of ASH.

Results

Search processes provided 487 articles. Of them, 143 articles just mentioned the subject without case reporting and were excluded. Furtherly, 24 articles were excluded, because they reported duplicated cases, had translated texts from language to another for previously-reported ASH cases, or only reviewed the subject without new cases. The other 320 articles fulfilled the inclusion criteria and comprised of 278 journal articles, 14 text books, 17 meeting reports, 6 thesis texts, 5 scientific websites articles reported new ASH cases (Supplementary List of References). Distribution according to the number of cases per article revealed 254 single case reports, 45 multiple case reports (article included 2–4 cases), and 21 case series (Supplementary Table 1).

The total number of the delivered ASH cases was 579 cases distributed over the period 1777–2017. Demographic characteristics including the age category distribution over time demonstrated the changes of reporting rates among both the pediatrics and adults (Fig. 1). Geographical distribution of reported ASH cases per country showed marked variations (Supplementary Fig. 1). Inaccurate crediting of pioneer events was detected such as first time of reporting pediatric and bilateral cases of ASH.

Detailed demographic and clinical characteristics, surgical interventions and therapeutic outcomes of the ASH cases have been studied. Distribution of the delivered cases to the constructed clinical grading system was illustrated by a chart (Supplementary Fig. 2).

Evaluation of the quality of evidence to the measured outcome in the included studies provided 558 ASH cases have been described in full or incomplete form and 21 ASH cases reported in a variably deficient form.

Discussion

History: terminology, reporting and crediting

ASH is a hydrocele that extends into the abdominal cavity due to unknown mechanisms forming two intercommunicating sacs. Since its vivid description by Dupuytren [1], many terms and hypotheses have been proposed to characterize ASH until formulation of the commonly used current term [3].

The rarity of ASH reporting created a confusion in crediting the first publisher of this entity. Tanzer [6] credited the first case to Lister in 1856 [7]. Also, Prather [3] revised the old German, French and English literatures in an excellent review and credited Dupuytren [1] as the first publisher. However, Sabatier [8] cited this case of Dupuytren in 1824. Then, Dupuytren [1] presented it again in 1834 to be named after him as Dupuytren's hydrocele [3]. However, the first case of ASH should be credited to Parcival-Pott in 1777 [9]. First pediatric case should be credited to Syme in 1861 [10], instead of

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