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Review

Abdominoscrotal hydrocele: A systematic review and proposed clinical grading

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KEYWORDS
Abdominoscrotal hydrocele;
Dupuytren;
Processus vaginalis;
Testicular dysmorphism

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 hydroceles 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.

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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 were 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).

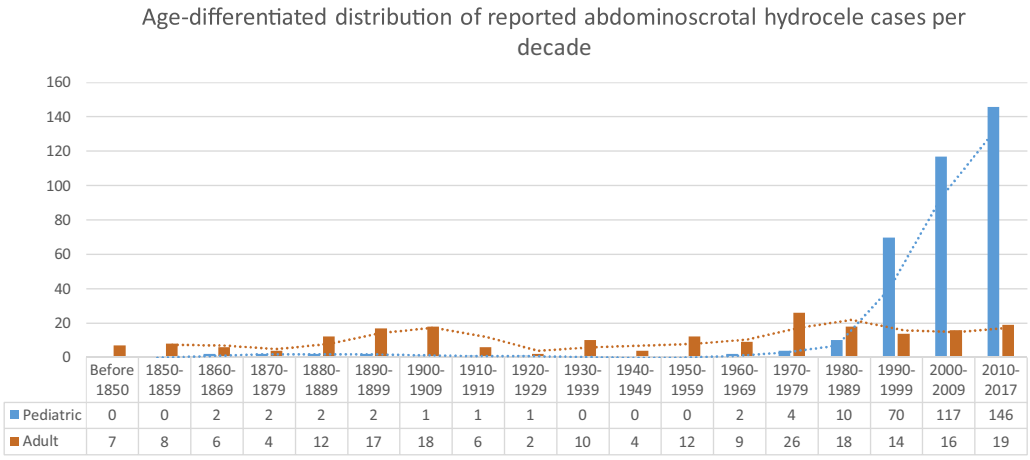
Evaluation of the quality of evidence to the measured outcome in the included studies provided 558 ASH cases have been described in full or incomplete form and 21 ASH cases reported in a variably deficient form.

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



	Before 1850	1850-1859	1860-1869	1870-1879	1880-1889	1890-1899	1900-1909	1910-1919	1920-1929	1930-1939	1940-1949	1950-1959	1960-1969	1970-1979	1980-1989	1990-1999	2000-2009	2010-2017
Pediatric	0	0	2	2	2	2	1	1	1	0	0	0	2	4	10	70	117	146
Adult	7	8	6	4	12	17	18	6	2	10	4	12	9	26	18	14	16	19

Figure 1 Chart of age-differentiated distribution of the reported abdominoscrotal hydrocele cases per decade.

Kocher in 1878 [11]. Competitive reporting of other pioneer events resulted in inaccurate crediting of events in many occasions [3]. This could be attributed to language differences and unavailability of rapid updating of the world literature.

Prather [3] followed the development of terminology from the initial term "*l'Hydrocele en bissac*" of Dupuytren to the widely accepted one "*abdominoscrotal hydrocele*". The latter term was recommended by Bickle in 1919 and most of the following authors credited the term ASH to him [3,12]. However, the oldest text used this term was a thesis belonged to Ivan K. Baitcheff in 1903 [13].

Another confusion was detected in two recent articles titled as ASH. They described hydroceles formed due to patent processus vaginalis (PPV) in cases of massive ascites [14,15]. However, it is a different pathology from ASH and Perrier specifically referred to this point in 1922 [16].

Incidence

Many reviews have been conducted for studying of ASH without reporting the correct total number of published cases [3,4,17]. By the year 2000, a random number of 200 cases was reported [18]. In 2017, Virgilio et al. [19] mentioned 230 cases as 130 in pediatrics and 100 in adults. According to the current review, however, the collected total number was 579 cases; 360 pediatrics, 208 adults, and 11 cases of non-reported age (Fig. 1). So, ASH is really more common than reported, but it is still rare among both pediatrics and adults. It has been mainly reported as case reports [3]. Many authors reported an incidence rate from their centers. The reported rates ranged as 0.18–3.1% of all types of hydrocele in pediatrics [20,21]. However, it is still difficult to estimate the global incidence rate. The average reporting rate was 1–2 cases per year which increased from a single case every many years to >60 cases in the last year only. Reporting was, initially, more among adults [3], but it has been reversed to be more among pediatrics [22] (Fig. 1). This manner promoted such impression that the incidence of ASH is higher than the reported values [23]. This could be attributed to the increased awareness by this entity, and ultrasonography that enabled the clinicians to detect the lesion even prenatally [24,25]. The noticed phenomenon of focal high regional incidence could be explained as just a coincidence matter [26].

Old ASH cases were mostly from tropical areas like India and African countries and described by foreigner writers [27–29]. However, the delivered articles in the current review referred to 45 countries from which ASH cases have been reported (Supplementary Fig. 1). Among all these countries, India has the highest rate of ASH, especially among adults [30]. On the contrary, the concept of ASH was recent in Japan [31]. However, the rate of reporting from Japan increased to >46 cases in the last decade (Supplementary Fig. 1).

Age is commonly less than 5 years in pediatrics and second and third decades in adults [32]. It ranged from the fetal life up to 77 years [24,33]. According to the largest case series of pediatric ASH, 60% of cases were younger than 1 year [34]. Although the distribution of ASH incidence between body sides has been reported as equal in previous reviews [3,4], the current review revealed that the right side is predominant. The right side, left side, bilateral, and unknown side included 242 cases (42%), 146 cases (25%), 78 cases (14%), and 113 cases (19%), respectively.

Surgical anatomy and pathology

ASH is a congenital anomaly of the processus vaginalis [35]. Mostly, it starts since birth as an inguinoscrotal hydrocele that extends to the abdomen. However, it could be misdiagnosed and pass to adulthood [25]. ASH is an intercommunicating two-compartment cystic lesion; inguinoscrotal and abdominal ones. The isthmus is the part that connects the two sacs resulting in the hourglass or dumbbell-shape appearance and occupies the inguinal canal [2,3,17]. The inguinal canal and ring are, mostly, dilated due to the pressure effect. Although this dilatation has been commonly proposed as a result of the compression forces, inversely, it could be considered as an etiologic factor that facilitates the intraabdominal extension [36]. Volumes of the abdominal and scrotal sacs are, independently, variable. However, the abdominal sac is usually larger than the scrotal one [2,3].

The abdominal sac is located extra-peritoneal with variable relations to the anterior abdominal wall and peritoneum as retroperitoneal or properitoneal. The latter form is more frequent. Also, the coverings are formed mainly of the transversalis fascia [2,3]. It extends broadly, may cross the midline to the contralateral side and ascends

upwards variably to the level of the costal margins [32,37]. In most of the cases, it is a single sac, but bilocular and multilocular sacs have been described [38,39]. Its shape is usually rounded like a human head or a football [27,28]. Cylindrical abdominal sac was described by Kocher [11]. The scrotal sac is usually smaller but could be larger extending to near to the knee and hinder walking [40]. It is pyriform in shape and almost always unilocular including the testicle at its posterior aspect [36], but bilocular sacs have been reported [41]. Moreover, it may have normal tunica vaginalis and the sac occupies the abdominoinguinal cord only to be a variant named abdominoinguinal hydrocele (AIH) [17,36].

Free intercommunication between the two sacs is a cardinal feature [36]. It is the reference of the diagnostic clinical signs and imaging criteria [17]. Non-communication to the peritoneal cavity is another cardinal feature [36,42]. However, communication has been, occasionally, reported in pediatrics [43] creating a debate and considered as an element of misdiagnosis or confusion with the inguinal part [25]. So, standard ASH is a non-communicating intercommunicating two-sac hydrocele [3,19]. If the communication of ASH to the proper peritoneal cavity should be accepted, it is an exceptional variant i.e. ASH with congenital inguinal hernia. Non-intercommunication state between the two sacs is an extremely rare finding of ASH with a maximum of seven cases have been reported [6,44–46].

Physical, microscopic, and biochemical properties of the ASH fluid have been studied [3,42]. Clear aspect is common in the uncomplicated ASH and it correlates with positive transillumination test on physical examination and transparency on surgery [3,17]. Color has been reported in different degrees of yellow with a common description as straw-colored fluid [3,16–18,29,41,45]. In hematoceles, however, the fluid is turbid and discolored into red or brown bloody description [3,17,40]. Volume of fluid has been estimated variably from 75 ml to 20 liters [16,18]. Microscopic analyses suggested an exudative fluid formation with low cellular components [42].

Pathogenesis

Pathogenesis of ASH is controversial with many postulated hypotheses for formation of its abdominal sac [25]. These hypotheses based on many factors. However, they could be classified according to the principle of the direction of ASH growth with different underlying mechanisms.

Mechanisms of upward extension of a scrotal hydrocele

Dupuytren's hypothesis. Dupuytren [1] postulated that the abdominal sac is an upward extension of a scrotal hydrocele through the inguinal canal under an excessive intrinsic pressure. It has been advocated by many authors [17,47], while it has been argued against by others based on the presence of many tense ordinary hydroceles which do not expand outside the confines of the scrotal tunica vaginalis [3,45]. In concordance, Jamal et al. [36] postulated that maternal factors like early strong uterine contractions, intrauterine fetal position or compressions during labor push-up a scrotal hydrocele to assume an intraabdominal extension. However, postulation of Jamal et al. [36] may not be acceptable, because the processus vaginalis is already still patent in 80–94% of fetuses during this age period [48]. So, no confined scrotal hydrocele would be formed to be compressed.

Jacobson's hypothesis. Jacobson postulated that ASH is an intraabdominal extension of an infantile hydrocele {obliterated processus vaginalis (OPV) at the internal inguinal ring} [25]. This hypothesis gained a wide acceptance among authors with many modifications [3,5,22,42]. Brodman et al. [49] explained the development of ASH on the basis of Laplace's law and the effect of intra-hydrocele pressure. Intraabdominal extension occurs when the pressure difference is greater than 4–6 cm H_2O supported by the inexpansible musculofascial boundaries of the inguinal canal [18,50]. Another modification resumed an old suggestion proposed by Tanzer [6]. Besides the increased pressure, Wlochynski et al. [38] suggested a co-existing congenital inguinal defect like a wide internal ring or a pre-existing diverticulum of tunica vaginalis. Postulation of a pre-existing diverticulum has been argued against by occurrence of ASH in very young infants with no time for formation of a diverticulum [43,50].

Roller's hypothesis. Roller [41] suggested that two co-existing hydroceles like an encysted hydrocele of the cord and an ordinary hydrocele may form ASH, but he did not explain how intercommunication occurs. This hypothesis may explain the cases of ASH with separated sacs [46]. However, intercommunication should be explained where recanalization of the interseptum in response to the high pressure from either or both sides may be suggested. Finding of a thin septum between the two sacs may support this concept [45,46]. Also, the sudden or rapid development of the scrotal or both sacs could be explained by this hypothesis [51]. This is especially in those patients who attributed their lesions to trauma or efforts [52]. Moreover, the AIH cases may support this concept. If the idea of recanalization has been denied, this form of non-intercommunicating hydroceles should be considered as a separate ASH variant [44,51]. In concordance, Cabot suggested a bottle-neck connection between an encysted hydrocele and tunica vaginalis [53].

Downward extension of an intraabdominal hydrocele

Macewen's hypothesis

Macewen [52] postulated that a preformed peritoneal sac as a persistent patency of the deepest portion of the funiculo-vaginal sheath inside the pelvis is responsible for the abdominal sac which distends forming an intraabdominal hydrocele and extends downwards. This hypothesis may explain the retroperitoneal variety, while it is not the matter with the most common properitoneal variety [6,54]. Moreover, it is not supported by the embryological bases or by the natural history of development of the scrotal sac before the abdominal one [6,50].

Hypothesis of Sasidharan and colleagues

Similar to Macewen's [52] hypothesis, Sasidharan et al. [24] proposed a downward extension of an early formed intraabdominal hydrocele due high intraabdominal pressure during the fetal life, but they did not explain how the intraabdominal hydrocele is formed. Also, this hypothesis is contradicting to the natural history of ASH [50]. However, it may correlate with ASH with small inguinal sac or AIH [51].

Guibe's hypothesis

Hydrocele of an undescended testis parallels to the concept of Sasidharan et al. [24], but in certain variants like ASH developed with abdominal testicular ectopia. Considering this undescended testis as the etiological factor for ASH is a matter of debate [55,56].

Downward direction of fluid and upward extension of hydrocele
Hypothesis of Saharia and colleagues

This hypothesis postulated presence of a flap-valve mechanism, where the intraperitoneal fluid flows downwards through the PPV which acts as a one-way valve [57]. It relied up on that PPV pumps down the peritoneal fluids during the episodes of increased intra-abdominal pressure only and compression by the hydrocele overdistension or other factors like inflammations [23]. It is a modification of Jacobson's hypothesis and has been adopted by many authors [58]. Luks et al. [20] considered the isthmus as the site of the one-way valve mechanism which may make it confused as the PPV. However, this postulation should be cautiously considered because of the commonly reported complete OPV in most of the pediatric ASH [22,26,42]. If the PPV exists, continuous downwards flow with fluid-forming intraperitoneal pathology must be present, while it is really not [50]. This concept was, firstly, described by Saharia et al. in 1979 [57], but without referring to it as their own idea.

Hypothesis of Khorasani and colleagues

Khorasani et al. [34] adopted hypothesis of Saharia et al. [57] and hypothesis of imbalance between supply and drainage/resorption of hydrocele fluid [25]. However, their own added postulation was that the reverse of fluid imbalance as a further step after closure of the processus vaginalis to explain the spontaneous resolution of ASH [34].

Some authors believe that the state of increased hydrocele pressure with OPV explains the development of adult ASH, while the state of PPV explains the development of pediatric ASH [38,43,57]. However, the subject is more complex enough to consider ASH as a multi-variant pathology with different underlying etiologies. Also, cases with detectable PPV are far less than those with OPV even in the very young ages. Moreover, adult ASH cases with unusual patterns like ASH inverse, AIH, and non-intercommunicating ASH are anatomically different [45,46,55]. So, single-hypothesis etiology is not a logic explanation.

Clinical presentations

Natural history of ASH is different among pediatrics and adults. In the former, ASH is an observable growing lesion in short periods with the possibility of spontaneous regression [34,59]. In the latter, however, it is long-standing with no reported spontaneous regression [3,6,46]. Clinically, ASH starts as a painless and progressively increasing scrotal or inguinoscrotal swelling followed by another abdominal swelling without a definite timing for the start or detection [25,31,54].

Physical examination usually reveals a scrotal mass that extends through the inguinal region with another lower abdominal mass. The latter looks like a midline mass with a slight deviation to the ipsilateral side and extends upwards, variably, to the costal margins [32,37]. It has been commonly resembled to a distended urinary bladder or a pregnant uterus [3,47]. The scrotal sac is a non-tender, cystic mass occupying the inguinoscrotal region, with impulse on coughing, reducible or partially emptied on lying supine, and having positive transillumination test, except in hematocele cases [2,54]. Sometimes, inguinoscrotal sac could be irreducible [60]. Fluid thrill and cross fluctuation tests have been described as diagnostic [54]. Pathognomonic signs of the ASH include free cross-fluctuation between the scrotal and abdominal sacs and "Springing back ball" signs. The latter is more characteristic and was suggested firstly by Wlochynski et al. [21,38]. It is elicited by compression of the scrotal component making the abdominal one to expand, but when compression is released the scrotal component increases again. It has been propagated by many authors [34,43]. Keihani et al. [61] introduced the new term "hourglass transillumination" for ASH. They combined this complementary sign to the "springing back ball" sign for confirmation of diagnosis. Rectal examination could be done to evaluate the mass consistency and ballottement in pediatrics or adults [41,45,62].

Associated congenital anomalies

Contralateral simple hydrocele is a common finding with pediatric ASH [25], but it could be encountered in adults [7]. The type is either communicating (congenital), vaginal, or infantile hydroceles [18,43]. Contralateral congenital inguinal hernia is another association with ASH [26].

Cryptorchidism or testicular ectopia is one of the commonest congenital anomalies with ASH (Supplementary Table 1) and its incidence seems to be more than reported [23]. Testicle is located in the inguinal canal and being ipsilateral [58] or contralateral [63], either, in pediatrics or in adults [18,64]. ASH may be associated with bilateral inguinal [65] or abdominal testes [66]. In certain cases, the abdominal sac contains the testis and associated with only inguinal extension where it was named "hydrocele en bissac inverse" [51,55]. Transverse testicular ectopia has been reported once with ASH [67]. Surprisingly, acquired up migration of the normally descended testis and displacement into the abdominal sac has been reported with a relatively smaller inguinoscrotal sac [51]. This phenomenon has not been fully explained, but it may be attributed to spontaneous rupture of ASH [68]. Epididymal anomalies with ASH could be explained as a complication of high pressure or an intrinsic embryological defect [69].

Besides these genital anomalies, sporadic urinary and extragenitourinary congenital anomalies have been reported in association with ASH [18,22,36,46]. Until now, no documented explanations for these associations could be proposed. Speculatively, however, it seems to be just a matter of co-existence until being proven otherwise [46].

Complications
Compression effect-induced complications

Testicular dysmorphism and impaired spermatogenesis. Testicular dysmorphism (Supplementary Table 1) is a relatively common complication of ASH [21,42]. It is defined as flattened and elongated or fusiform testis and it has been reported as 69.1–90% of ASH cases [22,70]. It is often reversible after early surgical intervention in pediatrics [25]. The proposed etiological mechanisms included: (1) increased hydrostatic pressure of tense ASH on testicular parenchyma or vessels. It is the commonest proposal which was suggested by Bayne et al. [42] depending on the increased hydrocele pressure; (2) intrinsic developmental changes of the testis, either as ASH complex or co-existing anomaly [70]. Impaired spermatogenesis or azoospermia has been reported in ASH associated with testicular ectopia [67]. However, the definite effect on fertility is unclear [42]. Unilateral or bilateral testicular atrophy is another complication. Atrophied testes have been usually removed during surgery for ASH [7,71].

Urinary compressions. Hydronephrosis is another relatively common complication [49,72]. It is usually unilateral [58] rather than bilateral [50,73]. Bilateral hydronephrosis could be caused by bilateral ASH [18] or, surprisingly, by unilateral ASH [50,73]. Hydronephrosis with ASH is explained by the compression of ureters at the pelvic prim. It is a simple compression, so it resolves after hydrocelectomy [74]. Ureteral displacement and urinary bladder compressions have been reported [35]. Also, urethral compression may occur [40].

Vascular compressions. Lymphedema is a relatively frequent presentation with ASH in the recent pediatric cases. It results from compression of veins and lymphatics [75].

Infections

Pyocele of ASH is uncommon and has been reported only in few instances [76,77].

Torsion

Torsion of the abdominal sac has been reported only twice (0.35%) [78]. Tate [79] reported a case of ASH with repeated attacks of acute abdomen. Although it was not proved on exploration, attacks of incomplete torsion of abdominal sac could be a considerable explanation.

Hematocele

Hematocele or hemorrhagic ASH is a significant pathological variant raising the suspicion of an underlying malignancy [80,81]. Also, abdominoscrotal hematocele is, either a benign complication or genuine. It may be an acute or old event discovered on aspiration or surgery [40,54,80]. Causes include spontaneous rupture [54], trivial or unnoticed trauma [80], strangulated hernia [82], or most seriously underlying malignancy [66,83].

Inguinal hernia

Co-existence of indirect inguinal hernia with ASH occurs in two forms; firstly, extra-vaginal hernia predisposed by the dilated internal inguinal ring. It is, either, due to tense hydrocele, where hernial sac is situated posterior to the tunica vaginalis or a congenital dilatation as postulated by Wlochynski et al. [38,84]. Secondly, intra-vaginal hernia protrudes through a PPV which was illustrated as rare types of inguinal hernia. These types of hernia could be proved to be a variety of ASH by location of the retained testis within the sac of hernia which is filled with hydrocele fluid rather than herniated intestine [82].

Malignancy

Malignancy with ASH has been reported 5 times only (0.86%). It is either testicular [53] or paratesticular [66]. The latter has been reported only twice; mesothelioma by Velasco et al. [66] and embryonal rhabdomyosarcoma by Matsumoto et al. [83]. Pathological examination for malignancy exclusion has been tried [58], but without detectable evidences that ASH is responsible to neoplasms. It looks that it is just an association, relying up on the very low incidence, especially, the paratesticular neoplasms which have direct contact.

Investigations

In the old era, cases of ASH were suspected clinically [17]. However, many times it represented an intra-operative surprise [44,53].

After introduction of ultrasonography for medical imaging and first use for ASH diagnosis [35], it has become the traditional tool. It is sufficient in simple cases and describes the hourglass anechoic lesion with fluid contents [22]. Also, it describes the testes and their abnormalities like testicular dysmorphism and ectopia [21,43]. Dynamic examination or graded compression during ultrasound examination may delineate intercommunication [34,85]. However, computed tomography is indicated to prove intercommunication or investigate the complex cases [73]. Magnetic resonance imaging is indicated for vascular complications like hematocele, lymphedema and for suspicion of malignancy [76,81,86].

Differential diagnosis

ASH differential diagnoses follow two states; Firstly, lesions which lead to abdominoscrotal swellings such as inguinal hernia which is the most common differential diagnosis [83], cord lymphangioma, and abdominoscrotal spermatocele [44]; Secondly, lesions which lead to cystic abdominal masses with co-existing ordinary hydroceles such as bladder diverticulum, hydronephrosis, urinoma, ascites, and cystic tumors [16,28,39].

Classification and grading

Crude classifications like characterizations into adult and pediatric, or hydrocele and hematocele ASH have been encountered on reviewing ASH. However, specified e.g. anatomical classifications have been also suggested. The commonest one belonged to Parekh et al. [30]. They considered ASH as a spectrum of complete and incomplete forms which were furtherly differentiated, like an inguinal hernia, into direct and indirect. Regarding the relations of the abdominal sac to the peritoneum and anterior abdominal wall, another old classification proposed properitoneal, interstitial, and superficial forms. However, Prather [3] suggested the addition of the retroperitoneal form to the latter classification, while presence of superficial and interstitial forms was doubtful. So, this old classification has been reduced to properitoneal and retroperitoneal forms only.

Here, a step-wise clinical grading system was proposed considering the cumulative increased complexities of the clinical presentations and complications, besides, the anatomical aspects. So, ASH could be differentiated into simple ASH (ASH-S) and complex ASH (ASH-C) classes with further sub-classes (ASH-S_n and ASH-C_n):

Simple ASH (ASH-S)

ASH-S_1: Unilateral intercommunicating ASH with its abdominal component is properitoneal and below the umbilicus and free contralateral scrotal compartment.
ASH-S_2: Unilateral intercommunicating ASH with its abdominal component is retroperitoneal and below the umbilicus and free contralateral scrotal compartment.
ASH-S_3: Unilateral intercommunicating ASH and its abdominal component is extending to the Retzius space, reaching above the umbilicus, multilocular or interstitial ASH.
ASH-S_4: Unilateral intercommunicating ASH with a documented PPV.

ASH-S_5: Unilateral intercommunicating AIH variety.
ASH-S_6: Unilateral non-intercommunicating ASH (or AIH) components.
ASH-S_7: Unilateral intercommunicating ASH or AIH with a contralateral simple hydrocele or congenital hernia.
ASH-S_8: Bilateral ASH or AIH.

Complex ASH (ASH-C)

When ASH-S is associated with any disorder of the followings, it should be classified as an ASH-C through following grades:

ASH-C_1: ASH with congenital anomalies other than contralateral ordinary hydroceles and congenital hernia.
ASH-C_{1A}: ASH with genital anomalies as cryptorchidism and epididymal anomalies.
ASH-C_{1B}: ASH with urinary anomalies such as hypospadias and vesicoureteral reflux.
ASH-C_{1C}: ASH with extra-genitourinary anomalies as hydrocephalus or macrocephaly.
ASH-C_{1D}: Bilateral ASH with associated anomalies and/or combinations of any of the above congenital anomalies.
ASH-C_2: ASH with compression-induced complications.
ASH-C_{2A}: ASH with testicular or epididymal dysmorphism or atrophy, or dyspermatogenesis.
ASH-C_{2B}: ASH with urinary compressions like hydronephrosis.
ASH-C_{2C}: ASH with vascular compressions like lymphedema.
ASH-C_{2D}: Bilateral ASH and compression-induced complications and/or combinations of them.
ASH-C_3: ASH after previous hydrocelectomy (misdiagnosed and recurrent ASH).
ASH-C_4: ASH with co-morbidities contraindicating interventions or threatening patient's life.
ASH-C_5: ASH with acute complications like pyocele, rupture, torsion, etc.
ASH-C_6: ASH with ipsilateral complicated inguinal hernia.
ASH-C_7: Abdominoscrotal hematocele.
ASH-C_8: ASH with testicular or paratesticular malignancy.

Stratification of the ASH cases according to this grading system may demonstrate the high risks that are associated with complexities of congenital anomalies and complications (Supplementary Fig. 2).

Treatment

Treatment of ASH has been eventually changed from the primitive methods of puncture and aspiration with or without sclerotization to the most recent minimally invasive total excision [11,40,87]. Broadly, the methods of treatment of ASH are traditional surgical interventions, minimally invasive intervention, and conservatism [3].

Traditional surgical interventions could be classified into; puncture or tapping drainage, incisional drainage, and excision [3]. Currently, aspiration of ASH fluid is a common initial step before or after skin incision to facilitate surgical dissection and excision [21].

Surgical approach is through the standard inguinal, extended inguinal, abdominal, or scrotal incisions [23,26,57]. Different abdominal incisions have been used [20,62,67]. Scrotal approach is recommended in pediatrics to decrease surgical damages [21,88]. Abdominal and combined incisions have been employed for large abdominal component [27,28]. In the recent literature, the tendency toward inguinoscrotal and scrotal approaches is more noticeable [22]. However, inguinal incision is still predominant, because of its effectiveness [32].

Surgical excision of the sacs takes different forms; total excision of an intact sac to prevent recurrence [43], even with the cord and testis i.e. en-bloc [89]. Orchiectomy could be warranted for reasons like testicular atrophy [3], surgical difficulties [54,71], and malignancy [83]; partial excision of the scrotal sac and leaving the abdominal one for drainage [34]; excision of the abdominal sac and partial excision of the scrotal one is the commonest form with tunical eversion like in vaginal hydrocele [43]. Some authors may just fenestrate the scrotal sac [64]. Others may schedule hydrocelectomy as a staged procedure through two incisions [90]. Depending on the less supported rationale of PPV, Matsukawa [91] advocated such a technique of just closure of PPV. Post-operative wound drainage is a classic step [3]. If it is neglected, hematoma could form and lead to testicular necrosis by compression [79]. Repair of bilateral cases could be synchronous or sequential [21,26,43].

First trial of laparoscopic ASH management was reported in 2004 [92]. In 2006, Kinoshita et al. [31] reported laparoscopic evaluation of ASH before inguinal repair, then Horst et al. [93] in 2007 described a retropertinoescopic dissection-assisted inguinal excision of ASH. Bouhadiba et al. [87] in 2007 described the first complete laparoscopic ASH excision. Also, Abel et al. [59] described laparoscopic marsupialization of the abdominal component before inguinal repair of the scrotal component. Besides the treatment purposes, laparoscopy plays confirmatory roles [63,65]. Recently, progressively increasing use of novel laparoscopic techniques in ASH management has been noticeable [94–96].

Conservative treatment has been described with spontaneous resolution in asymptomatic and uncomplicated pediatric ASH or in cases having contraindications to surgery [97,98]. Moreover, in the most recent and largest series of ASH, Khorasani et al. [34] reported observation as a reasonable first step management for uncomplicated pediatric ASH. However, Ceccanti et al. [99] reached to less convincing outcomes. The phenomenon of spontaneous resolution created a question that why ASH develops in adults while it is a congenital lesion with spontaneous resolution probability? The answer may come from the discussed pathogenesis; ASH is a multi-variant lesion with different etiologies.

Conclusions

ASH is a rare lesion reported mainly in single case reports. It affects the pediatrics more than the adults where it seems to be a congenital lesion. It has different etiological hypotheses and multiple clinico-pathological variants. Physical examination and ultrasonography are usually sufficient for diagnosis, but more diagnostic tools could be indicated. It could be associated with many other congenital anomalies like simple hydrocele and cryptorchidism and many complications ranging from the compression effect to coincident malignancies. In spite of the recent trends in treatment toward the less invasive approaches like the novel laparoscopic techniques and conservation, still surgical excision via the inguinal incision is the standard approach. Clinical grading of ASH may differentiate

the cumulative risks of the increased complexities of the associated anomalies and complications.

Author's contribution

Rabea Ahmed Gadelkareem, MD, developed the manuscript idea, reviewed the literature, analyzed the data, completed writing, revised manuscript, responded to reviewers' comments, submitted the manuscript and its revision with final approval of the manuscript.

Conflict of interest

The author has no conflicts of interest to disclose.

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Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at https://doi.org/10.1016/j.afju.2018.01.006.

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