

Acute Idiopathic Scrotal Edema: A 10-year period retrospective study

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ABSTRACT

Objective: To report a case series of children diagnosed of Acute Idiopathic Scrotal Edema in a secondary referred hospital over a 10-year period, including the description of its clinical manifestations, management and evolution of this condition.

Methods: A retrospective chart review was performed. It was found to include 11 patients, between 19 months and 12 years old, who presented scrotal edema and/or erythema with no self-explanatory trigger.

Results: On physical examination all patients were found to have sudden onset of scrotal swelling and/or erythema despite normal testicular and epididymal inspection. 73% of the patients experienced mild pain. In most cases, localization was unilateral. A personal history of atopic diseases and/or allergies was present in 82% of the patients. Scrotal ultrasound was performed in all patients with suspected acute scrotum. Marked bilateral scrotal wall thickening and increased peritesticular blood flow visible on color Doppler (Fountain's sign) were detected in all patients. No other abnormalities were observed except for one case in which there was a clinical suspicion of decreased flow in the right testis (not confirmed in subsequent studies).

Conclusion: Acute idiopathic scrotal edema is a benign self-limiting condition commonly seen in prepubertal boys. Given its clinical features it should be considered within the differential diagnosis of acute scrotum.

Keywords: Acute Idiopathic Scrotal Edema, acute scrotum, Fountain sign, atopy.

Introduction

Acute idiopathic scrotal edema (AISE) is a benign, self-limiting condition first described by Qvist in 1956 [1]. The etiology remains unclear. It is characterized by marked edema and/or erythema of the scrotum and the dartos without expansion to the underlying layers of *scrotum's* wall or to the endoscrotal structures. It can be confined to the superficial layers of the scrotum, although it may extend to perineum and inguinal canal. The erythematous area of the skin may be painful. Since the condition runs a self-limiting and benign course, it is probably underdiagnosed, especially mild cases. Due to its good evolution, AISE may be successfully managed with conservative treatment such as rest and anti-inflammatory drugs.

The majority of AISE cases appear in the pediatric age, mainly in children between 5 to 10 years old, even though some adult cases have been described [2]. Recurrence may occur. Color Doppler imaging sonography contributes to documentation of AISE and the exclusion of other basic causes of acute scrotum, mainly those that require immediate surgical intervention, such as testicular torsion. Color Doppler sonography shows scrotal wall hypervascularity with homogeneous testicular parenchyma and equal arterial supply to both testes. The pattern created by increased blood flow confined within the scrotal wall is known as the fountain sign as it resembles a fountain [3][4].

We aim to describe clinical findings, management, progress and treatment of patients diagnosed with AISE in our hospital. This case series highlights the importance of being able to recognize this condition as a correct diagnosis can avoid unnecessary surgical exploration of the scrotum.

Methods

A retrospective chart review of the electronic medical record was performed which included emergency discharge and in-patients summaries, of patients diagnosed of AISE during a 10-year period (from 2010 to 2019) in Hospital Universitario Dr. Peset, Valencia (Spain). Patients included were known to have sudden onset of swelling and/or erythema without testicular changes and after excluding other causes of an acute scrotum.

Epidemiological data, such as age at diagnosis, and past medical history of allergies or atopy (rhinitis, asthma, atopic dermatitis, angioneurotic edema, urticarial and drug allergy), enterobiasis, previous groin surgery, concomitant acute disease or secondary to a trauma injury were collected. Moreover, location, extension, clinical cutaneous and testicular features and presence of pain were described. A review of blood work and imaging results, treatment received, clinical progression and recurrence of the disease was also performed.

Informed consent and ethical approval were not required as the data was retrospectively collected from the patient's medical history record.

Results

In our hospital, 11 children met the diagnostic criteria for AISE described above. Table 1 summarizes the analyzed data. The patients ranged in age from 19 months to 12 years-old, with most patients in between 9 and 12 years-old (mean age of 8.7 years-old). Previous history of a similar episode was found in 2 cases.

Analysis of the data showed that 82% of the patients had past medical history of allergies and/or atopy. 27% were diagnosed of enterobiasis and all of them were found to live in the same geographical area, Valencia (Spain). One patient underwent an inguinoscrotal surgery due to a retractile testicle. One patient is currently undergoing further investigations in the outpatient clinic for eosinophilia. No past medical history of angioneurotic edema, drug allergy, concomitant acute disease or secondary to a trauma injury were identified.

Neither systemic symptoms nor fever were described. All the 11 patients showed cutaneous manifestations of sudden swelling and/or edema (2 to 4 hours), but only in 2 cases a local temperature increase was registered (Figure 1). Mild testicular pain was reported in 73% of the patients. Predominant left-sided location was found in 73% of the patients. In 3 of the patients the swelling and/or edema progressed until the ipsilateral pubic and groin area and/or perianal region. Only in one case, it extended to the penis. On physical examination, none of the patients revealed testicular abnormalities.

In reference to the complementary exams carried out, it is important to outline that all patients underwent a testicular ultrasound, which confirmed diagnoses. Ultrasound confirmation avoided surgical exploration. The most common finding was thickening of subcutaneous tissue and increased Doppler flow. It was present in 100% of the patients. In two patients, reactive lymphadenopathies were described. In another one, reactive hydrocele was reported. No testicular abnormalities were observed except for one case (19 months old) in which there was a decreased flow in the right testis. Only in one case blood work was performed. It showed eosinophilia without increased acute-phase reactants, nor leukocytosis or left deviation. Blood and urine cultures were considered negative.

After diagnosis was confirmed, 82% of the patients were discharged with anti-inflammatory drug therapy. In one patient a papular lesion on the groin area was considered as a possible entrance door for infection, so oral amoxicillin-clavulanic acid and mupirocin ointment were prescribed. Two patients were admitted and treated with anti-inflammatory drugs. In one of them intravenous amoxicillin-clavulanic acid was administered. The 19-month-old child in whom a decrease Doppler flow of the right testis was identified was transferred to the referral hospital for surgical assessment. Upon arrival, a second ultrasound was performed in which no abnormalities were observed, therefore there was no need for surgical intervention. All patients presented a favorable clinical evolution free from complications.

Discussion

AISE is a self-limiting condition characterized by edema and/or erythema of the skin and dartos fascia. It may be unilateral or bilateral, without involvement of deeper layers or testes. It is much more common among the pediatric population, especially prepubertal children. The incidence is highly variable, ranged between 2.5 and 30% of all diseases included under the diagnosis of acute scrotum [5][6]. The exact etiology of AISE remains unclear. Etiologic theories range from infectious, allergic or inflammatory explanations [4]. In our review, a personal history of allergy and/or atopy was described in 82% of the patients. This hypothesis may be justified by the similarity between AISE and angioneurotic edema, the absence of response to empirical antibiotic therapy, then presence of peripheral eosinophilia (found in the only patient where a complete blood count was performed) and the improvement with antihistamines reported in the available literature [3][5].

The patients usually describe a sudden onset of clinical features associated to mild localized pain. Location is initially referred to as unilateral but it may extend and affect both hemi-scrotum, anterior abdominal wall, groin and perianal area and more rarely the penis. On physical examination, patients always show swelling and/or non-itchy erythema. Testicular palpation may be difficult if there is intense scrotal edema. The cremasteric reflex is usually difficult to assess [8]. Taking into account that it is a local inflammatory reaction, there is no increase in acute-phase reactants nor fever. In addition, blood and urine cultures are negative.

Being able to differentiate between AISE and other conditions that cause acute scrotum such as testicular torsion, acute epididymitis, inguinoscrotal hernia and testicular trauma is crucial as they tend to require surgical evaluation and/or more aggressive examinations or treatments. Although the diagnoses are achieved based on the physician's clinical suspicion, a scrotal ultrasound is recommended in order to rule out the different entities that cause acute scrotum. The most constant ultrasound findings are marked bilateral scrotal wall thickening and increased peritesticular blood flow visible on color Doppler, also known as Fountain's sign (present in 100% of our patients) (Figure 2). Associated findings include reactive hydrocele, reactive lymphadenopathies and preserved echo-structure and size of testes and epididymis.

Treatment includes anti-inflammatory drugs, which provide pain relief and the resolution of clinical manifestations in 2 to 3 days [6]. In some cases, when cutaneous infection is suspected, antibiotics are associated while awaiting clinical improvement [9]. In our casuistic, all patients received anti-inflammatory medications and antibiotic treatment was associated in two of them. The use of antihistamines was not necessary, as described per other case series [10]. No sequelae have been described even though recurrent episodes have been reported in 10-20% of the cases [6][11]. (18% of the reviewed patients).

On this basis, we conclude that clinical suspicion of this condition is essential. This would prevent physicians from requesting unnecessary tests and performing aggressive treatments. Moreover, it may be useful to search for allergies and/or atopy during the anamnesis since there seems to be a significant association between them.

Conflict of interest

No financial or nonfinancial benefits have been received or will be received from any party related directly or indirectly to the subject of this article.

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TABLE 1: clinical and paraclinical data of patients with a diagnosis of AISE (n=11)

	AGE	HISTORY OF ALLERGIES/ ATOPY	PERSONAL HISTORY	PAIN	LOCATION	BLOOD WORK	ULTRA-SOUND	TREATMENT	RECURRENCE
CASE 1	12 y/o	AD, urticaria	None	No	Bilateral Scrotum	No	Fountain's sign	NSAIDs	No
CASE 2	11 y/o	AD, asthma, rhinitis, conjunctivitis	Oxyuriasis	Yes	Unilateral (left) Scrotum, perineum, pubis	No	Fountain's sign	NSAIDs + ABX	No
CASE 3	11 y/o	AD, urticaria	None	Yes	Bilateral Scrotum	No	Fountain's sign	NSAIDs	No
CASE 4	6 y/o	Urticaria	None	Yes	Unilateral (right) Scrotum, penis	No	Fountain's sign	NSAIDs	No
CASE 5	19 months old	AD, urticaria, asthma	None	No	Bilateral Scrotum	No	Fountain's sign, decrease Doppler flow of the right testis	Admitted NSAIDs + ABX	No
CASE 6	11 y/o	Urticaria, rhinitis	Oxyuriasis	Yes	Unilateral (left) Scrotum, pubis, perianal	No	Fountain's sign	NSAIDs	No
CASE 7	9 y/o	AD, rhinitis	Oxyuriasis	Yes	Unilateral (right) Scrotum, groin y perianal	Yes (eosinophilia)	Fountain's sign	Admitted NSAIDs	2º episode (1st 5 y/o)
CASE 8	5 y/o	Asthma	None	Yes	Unilateral (left) Scrotum	No	Fountain's sign	NSAIDs	No
CASE 9	5 y/o	AD, urticaria, asthma	Right hydrocele y phimosis	Yes	Unilateral (left) Scrotum	No	Fountain's sign	NSAIDs	No
CASE 10	12 y/o	None	Retractile right testis, appendectomy	Yes	Unilateral (right) Scrotum	No	Fountain's sign	NSAIDs	2º episode (1st 11 y/o)
CASE 11	12 y/o	None	None	Yes	Unilateral (left) Scrotum	No	Fountain's sign	NSAIDs	No

AISE: acute idiopathic scrotal edema; **y/o:** years old; **AD:** atopic dermatitis; **NSAIDs:** Nonsteroidal anti-inflammatory drugs; **ABX:** antibiotics

Figures:



Figure 1: Edema and erythema of the left hemiscrotum. It also extended to the left inguinal and pubic area.

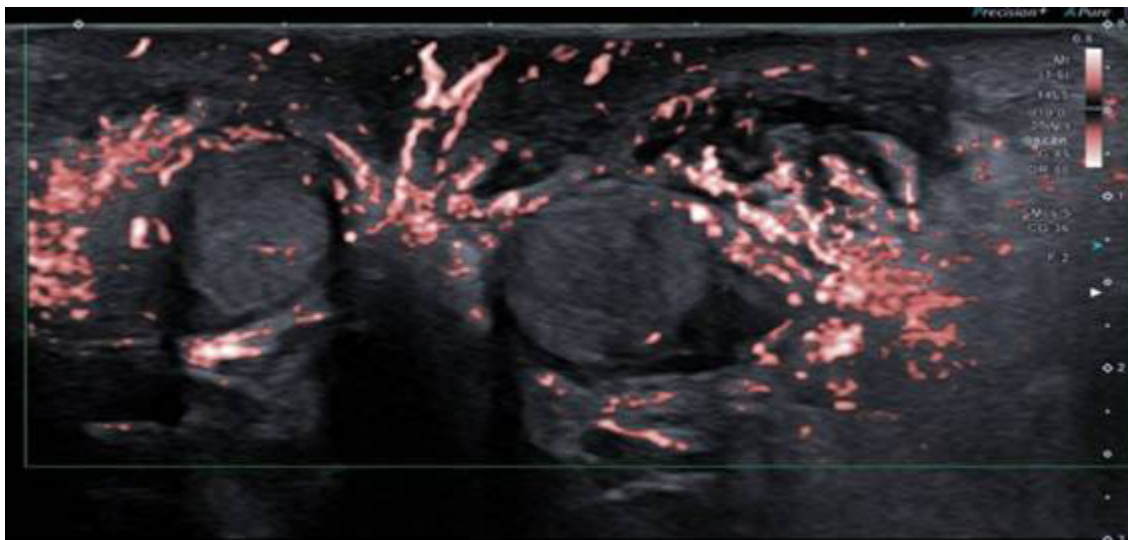


Figure 2: Increased peritesticular blood flow on Color Doppler, also known as Fountain's sign.