Spontaneous spermatic vein thrombosis in pediatric patients: a condition to be considered

J. Pérez-Ardavín, A. Serrano Durbá, I. Miró, M.A. Conca Baena, J.A. March-Villalba, A. Polo Rodrigo, C.C. Sangüesa, C. Domínguez Hinarejos

Pediatric Urology Unit. Urology Department. La Fe University Polytechnic Hospital. Valencia (Spain).

ABSTRACT

Spermatic vein thrombosis is a very rare pathology, with 25 cases published only, 6 of which in patients under 15 years of age. We present the case of a male patient, as well as a review of the literature.

A 12-year old boy presented at emergency with 3-day progressive testicular pain. Following abdominal Doppler ultrasound imaging, he was diagnosed with left spermatic vein thrombosis and nutcracker syndrome. Admission and enoxaparin treatment were decided upon. Patient evolution was satisfactory, with subsequent ultrasound imaging demonstrating the absence of thrombus. The patient is currently under follow-up and without treatment.

Various treatments are mentioned in the literature, with conservative management being the treatment of choice.

KEY WORDS: Venous thrombosis; Renal nutcracker syndrome.

TROMBOSIS ESPONTÁNEA DE LA VENA ESPERMÁTICA EN LA EDAD PEDIÁTRICA: UNA ENTIDAD A TENER EN CUENTA

RESUMEN

La trombosis de la vena espermática es una patología muy poco frecuente, con solo 25 casos publicados, 6 de los cuales menores de 15 años. De esta manera presentamos el caso de un varón, así como la revisión de la literatura.

Un paciente de 12 años de edad acudió a Urgencias por dolor testicular de aumento progresivo, de 3 días de evolución. Mediante ecografía doppler abdominal se diagnosticó de trombosis de la vena espermática izquierda y síndrome de cascanueces. Se decidió ingreso y tratamiento con enoxaparina. La evolución del paciente fue satisfactoria, en la ecografía posterior se observó la desaparición del trombo. El paciente se encuentra en seguimiento y sin tratamiento.

Existen varios tratamientos reflejados en la literatura, siendo el de elección el manejo conservador.

PALABRAS CLAVE: Trombosis venosa; Síndrome de cascanueces renal.

Corresponding author: Dr. Javier Pérez Ardavín. Pediatric Urology Unit. Urology Department. La Fe University Polytechnic Hospital. Valencia (Spain). E-mail address: ardavin.j@gmail.com

Work presented at the Pediatric Urology Group Meeting. 2018.

Date of submission: May 2019 *Date of acceptance:* December 2019

INTRODUCTION

Spermatic vein thrombosis is a very rare pathology. However, it falls within differential diagnosis of acute testicular pain, a group of pathologies that are highly frequent in children. We present the case of a pediatric patient, as well as a review of the literature.

CASE DESCRIPTION

This is the case of a 12-year old boy presenting at the emergency room with 3-day progressive left testicular pain, without temperature, voiding symptoms, or trauma history. At exploration, he had an enlarged testis, with diffuse pain, congestive appearance, and edema in testicular coverings.

Given the suspicion of subacute testicular pain in this pediatric patient, abdominal-scrotal Doppler ultrasound imaging was carried out. Scrotal ultrasound imaging demonstrated a normal right testis and a left testis with preserved vascularization, increased pampiniform plexus, and no reflux at Valsalva's maneuver. Therefore, exploration was carried on cranially, which showed the presence of a thrombus in the most distal portion of the spermatic vein, as well as increased venous wall thickness, so diagnosis of thrombophlebitis was established (Fig. 1). Superior mesenteric artery exploration identified a 13º aortomesenteric angle and a 3 mm gap (Fig. 2), so the patient was diagnosed with nutcracker syndrome. The remaining abdominal ultrasound imaging did not reveal any further pathologies. Blood test, urine sediment, and urine culture were performed, which demonstrated a mild leukocytosis and a positive urine culture with multi-sensitive *E. coli*.

Following admission, the patient received 1 mg/kg/12 h full-dose enoxaparin treatment, ceftriaxone antibiotic treatment, and analgesia. Pediatric hematology control

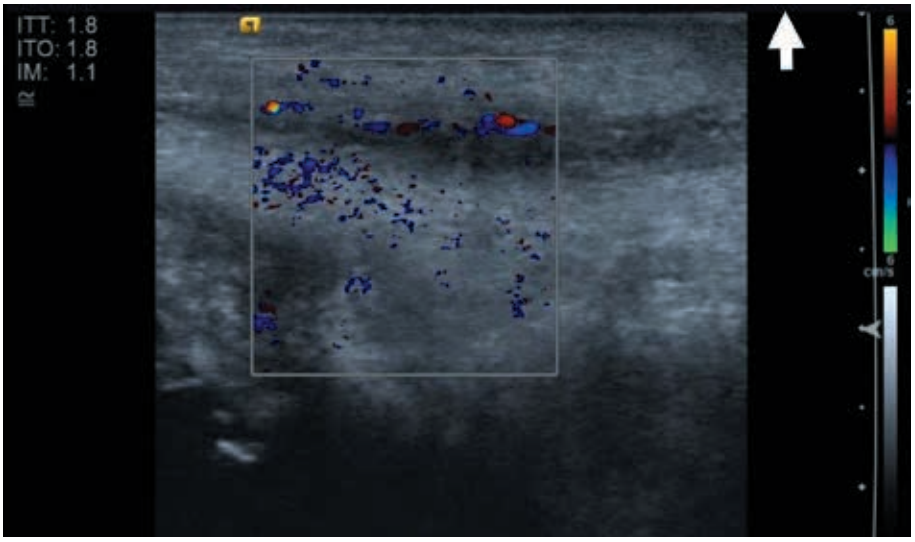

Figure 1. Distal spermatic vein ultrasound image showing absence of venous flow and thickened tunics, as typically seen in thrombophlebitis.

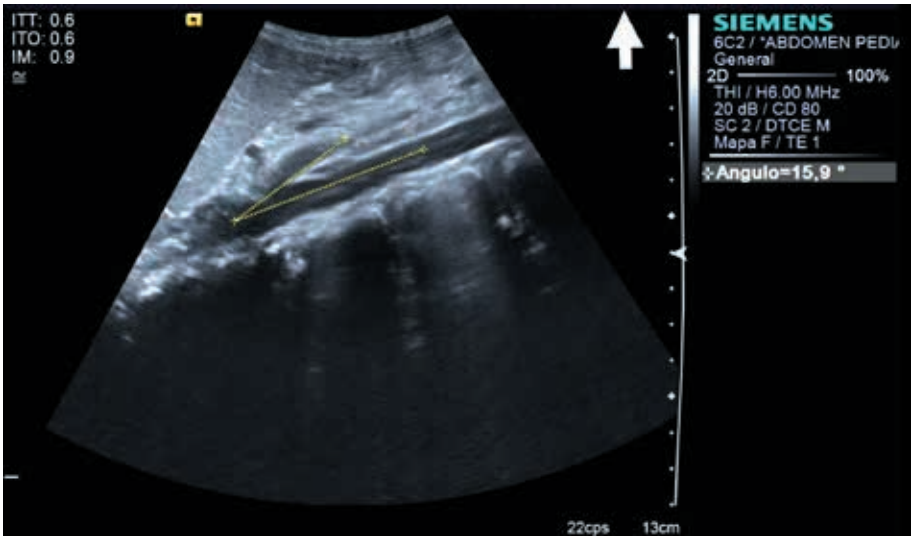

Figure 2. Abdominal ultrasound image showing the origin of the superior mesenteric artery from the aorta, with a 15.9º angle. The left renal vein can be observed between both structures.

was requested at admission for anticoagulant treatment follow-up purposes.

After 2 treatment days, a new Doppler ultrasound imaging was performed, which demonstrated that the thrombus found in the previous test had disappeared. Therefore, the patient was discharged, with pediatric urology and hematology external consultation control. A study on the potential causes of thrombosis was carried out. 500 mg/8 h amoxicillin-clavulanic acid oral antibiotic treatment was prescribed for 7 days, and 30 mg/24 h enoxaparin treatment was prescribed for 3 months.

Evolution was satisfactory, and the patient presented no new events. Blood test demonstrated the presence of a factor V Leiden heterozygous mutation. The patient is currently under follow-up, without symptoms and without treatment. He has been warned about potential alarm signs and is under conservative treatment.

DISCUSSION

Spermatic vein thrombosis is a very rare condition. According to the literature, there are some 28 cases described, with the longest series including 15 patients[1]. In pediatric patients (under 16 years of age), spermatic vein thrombosis is even less frequent, representing 6% of the total cases. However, this is the most relevant group clinically speaking, with the longest series including 3 patients only[2] (Table 1).

Spermatic vein thrombosis is more frequent on the left side, as in our case, with four right spermatic vein thrombosis cases only: one newborn requiring catheterization owing to cardiac abnormalities[3], one patient with factor V Leiden mutation[4], one patient with inguinal hernia[5], and one patient with bilateral thrombosis owing to extreme exercise[6].

Exercise is the most frequent etiology, with increased abdominal pressure and reduced testicular venous return[7].

Table 1.

Study	*Age*	*Etiology*	*Diagnosis*	*Treatment*
12	Newborn	Idiopathic	Ultrasound imaging	Orchiectomy
3	Newborn	Catheterization	Ultrasound imaging	Heparin
6	8	Schönlein	Exploration	Exploration
2	7	Idiopathic	Venography	Exploration
2	10	Idiopathic	Venography	Anti-inflammatories
2	15	Exercise	No	Exploration

However, many other causes have been reported to explain thrombosis according to Virchow's triad: factor V Leiden mutation[4], blood vessel lesions (catheterizations[3]...), Henoch-Schönlein purpura[6], and any etiology that may increase venous hypertension[8]. They all have one thing in common – alteration of one of the three triad criteria: vessel lesion, coagulability disorder, or blood stasis. In our case, both factor V Leiden heterozygous mutation and nutcracker syndrome – two risk factors described in the literature[4,9] – were present.

Diagnosis should be suspected in case of testicular pain, with scrotal Doppler imaging being the most widely used test. However, other tests such as venography[2] have been described in the literature, especially during the 80s. In adults or in doubtful cases, contrast abdominal CT-scan is the gold standard technique.

Treatment varies according to the hospital and patient evolution. In our case, conservative treatment with enoxaparin allowed the thrombus to be removed, as demonstrated at control 3 days later. In some cases, the thrombus may even disappear spontaneously following anti-inflammatory administration for pain control[10]. In other patients, surgical exploration is required, especially in case of ultrasound findings of poor scrotal vascularization, with sperm cord study[6], thrombectomy[11], or orchiectomy in case of parenchymal bleeding or destructuration[12].

The most severe complication stemming from this pathology has been pulmonary thromboembolism (PTE) in an adult patient[13], so open or laparoscopic spermatic vein ligation may be required in some cases[7]. According to algorithms, if the thrombus surpasses the internal inguinal ring, the spermatic vein should be ligated[14].

In our view, ultrasound thrombus control 48-72 hours following the event should be carried out to check whether it has been removed or not. If it has not, surgery – namely laparoscopic spermatic vein ligation – should be considered.

CONCLUSION

Spermatic vein thrombosis is a rare condition, but it should be considered in acute scrotum differential diagnosis in pediatric patients, especially in the presence of risk factors.

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