

## Right Testicular Infarction Secondary to Spontaneous Testicular Vein Thrombosis in a Child: Case Report and Literature Review



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<b>OBJECTIVES</b>	To describe a case of right testicular vein thrombosis in a child and to review the literature describing similar cases and their management.
<b>METHODS</b>	An extensive literature review of the English language literature was conducted using the following databases: PubMed, ScienceDirect and Google Scholar.
<b>RESULTS</b>	Six cases were reported previously and summarized in this article.
<b>CONCLUSION</b>	A 14-year-old boy who suffered from spontaneous right testicular vein thrombosis was treated with right orchiectomy and oral anticoagulation. We also reviewed and summarized the previously described cases of pediatric testicular vein thrombosis. <i>UROLOGY</i> 130: 144–147, 2019. © 2019 Elsevier Inc.

**T**esticular vein thrombosis (TVT) is an exceedingly rare entity in the pediatric population.<sup>1-4</sup> Thrombosis of the testicular vein leads to progressive infarction of the affected testis. It has been shown to have similar recurrence rates and mortality rates when compared with lower limb deep vein thrombosis (DVT).<sup>5</sup>

Patients typically present with gradually worsening scrotal and inguinal pain and swelling that mimics testicular torsion, epididymitis or incarceration of inguinal hernia. Diagnosis requires a very high index of suspicion combining clinical information and intra-operative findings. Management in adults consists of excising the unviable testis, treating the underlying cause and commencement of anticoagulant therapy. The literature is very limited in the pediatric population and there is no standardized management.

We present a case of right TVT in a 14-year-old boy that is, to our knowledge, the first case of spontaneous right testicular vein thrombosis described in a pediatric patient and the first pediatric case to be treated with oral anticoagulant therapy. We also reviewed and summarized previously published cases of pediatric TVT.

### CASE REPORT

A 14-year-old, obese, adolescent boy with no past medical, surgical or family history of venous thromboembolic disease (VTE), presented to the emergency department with a 4-day history of dull and constant right inguinal and right scrotal pain with fever (38.5° C). He denied any history of trauma to the genitalia. On the day of presentation, he reported that the pain had gradually worsened, becoming severe with swelling of the right scrotum. On examination, both testes were in the bottom of the scrotum. The right scrotum appeared swollen and erythematous. The right testis was firmer than left testicle and no palpable knot was felt through the right spermatic cord. Examination of the remainder of the genitalia, inguinal regions, and abdomen was unremarkable.

Laboratory investigations revealed leukocytosis ( $15.0 \times 10^9$ ). Color Doppler ultrasonography revealed coarse heterogeneous echotexture with no vascularity of the right testicle (Fig. 1). The right testicle measured  $30 \times 29$  mm. The left testicle measured  $26 \times 15$  mm with normal vascularity and echotexture.

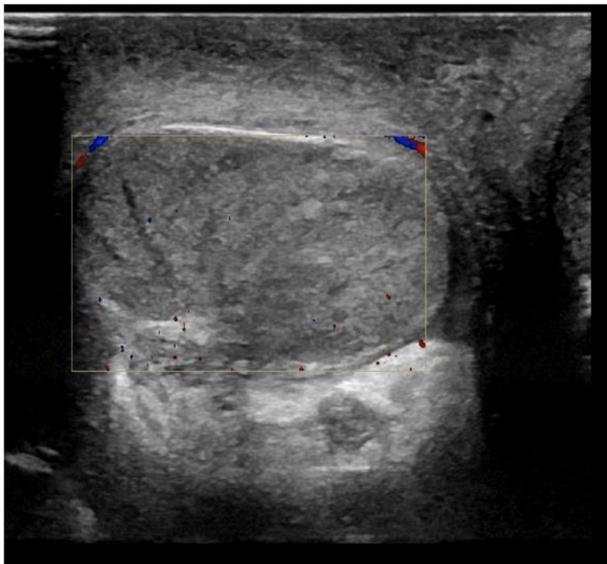
He underwent emergent scrotal exploration that revealed a discolored, grossly edematous, and nontorted right testicle with dark bluish discoloration of the spermatic cord extending up to and beyond the external inguinal ring (Fig. 2). The right testicle and thrombosed spermatic cord at the level of the external ring were excised. Postoperative course was unremarkable. Given the atypical presentation and operative findings, a vascular pathology was highly suspected.

Level of evidence: Level 5

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**Figure 1.** Color Doppler ultrasound of the right testis demonstrating absent flow to the right testicle and heterogeneous echotexture.

Postoperatively, CT-angiogram of the abdomen and pelvis revealed no enhancement of the right testicular vein in a segment proximal to the internal inguinal ring (Fig. 3), representing thrombosis of the testicular vein.

The hematology service was consulted and a thrombophilia screen (antithrombin III deficiency, protein C, protein S deficiency, Factor V Leiden, Lupus Anticoagulant) was negative. Testing for the JAK-2 V617F gene mutation was also negative. Histopathological examination showed venous thrombosis in the spermatic cord and hemorrhagic infarction of the testicle (supplemental Figure 1).



**Figure 2.** Intraoperative finding of thrombosis of the right testicular vein extending up to the external inguinal ring.



**Figure 3.** Coronal view. Venous phase of the CT angiogram demonstrating the left spermatic cord with enhancement of the left testicular vein (white arrowhead) and no enhancement of right testicular vein in the excised right spermatic cord (white arrow).

The patient and his guardians were counseled about the option of starting oral anticoagulants to reduce the risk of progression or recurrence of VTE elsewhere in the body. They agreed, and a novel oral anticoagulant agent, rivaroxaban, was commenced for duration of 6 months. He was also followed up in the urology outpatient department 1 month and 4 months later and had no complaints or postoperative complications. He did not experience any side effects associated with the oral anticoagulant and was also planned for long-term follow-up with the hematology service.

## DISCUSSION

The incidence of VTE in the pediatric population is estimated to be 0.7-2.1 cases per 100,000 children.<sup>6</sup> Testicular vein thrombosis is very rare entity in the pediatric population with only a handful of cases reported in the literature (Table 1).<sup>1-4</sup> To our knowledge, this is the first case of spontaneous right testicular vein thrombosis described in a pediatric patient. In adults, only 20 spontaneous cases of TVT have been described.<sup>7</sup>

As outlined in this case and in previous cases,<sup>1-4</sup> clinical presentation is typically in the form of acute scrotal and inguinal pain associated with scrotal and spermatic cord swelling. In some cases, the pain may be persistently present for few days before presentation.<sup>1</sup> The presumptive diagnosis is usually testicular torsion, acute

**Table 1.** Cases of pediatric testicular vein thrombosis:

Author	Article	Year	Case(s)
Coolsaet and Weinberg <sup>1</sup>	Thrombosis of the Spermatic Vein in Children	1980	3 cases reported. —7-year-old boy, intraoperatively found to have of thrombosed left pampiniform plexus. Selective venography showed an incompetent spermatic vein with a filling defect in distal branches of the vein. —10-year-old boy, similar to the case above. —15-year-old boy, thrombosis of the left pampiniform plexus. Thrombosed veins excised and histology confirmed the diagnosis.
Hagstrom et al. <sup>2</sup>	Prevalence of the factor V Leiden mutation in children and neonates with thromboembolic disease.	1998	1 case. Details of case not specified.
Diana et al. <sup>3</sup>	A case of Pediatric Henoch-Schonlein Purpura and Thrombosis of the Spermatic Veins	2000	1 case. —8-year-old boy, presented with classical Henoch-Schonlein Purpura and treated with corticosteroids. After 2 weeks of treatment thrombosis of the spermatic vein developed and was treated with low-molecular-weight heparin for 4 weeks. Follow-up ultrasonography demonstrated resolution of the thrombosis.
Maas et al. <sup>4</sup>	Acute scrotum in a neonate caused by renal vein thrombosis	2011	1 case. —Neonate, 36 weeks gestation, developed discoloration of the scrotum and spermatic cord. Color duplex sonography demonstrated poor perfusion of the left testicle and hyperechogenic left testicle. Left orchidectomy performed. Developed hematuria post-operatively and ultrasound of the upper tract demonstrated renal venous thrombosis.

epididymo-orchitis or an incarcerated inguinal hernia.<sup>7,8</sup> A thrombosed pampiniform venous plexus is usually an incidental finding during scrotal exploration for a nonviable testis that may lead to suspecting TVT as the cause of testicular infarction.<sup>1,3,7,9</sup>

Management of testicular infarction consists of surgical excision of the nonviable testicle. However, controversy exists when considering anticoagulant therapy in TVT due to insufficient data in the literature.<sup>5</sup> The goal of anticoagulation is to prevent further progression of the thrombus, reduce risk of recurrence, improve patency of the vein, and prevent thromboembolism.<sup>6</sup> Cases of pulmonary embolism secondary to TVT have been reported in the literature.<sup>5,10</sup> Lenz et al conducted a case-control study comparing matched adult patients suffering from TVT with lower extremity DVT.<sup>5</sup> In total 39 cases of TVT were identified, the largest study of its kind and they concluded that recurrence of thrombosis and mortality rates were similar to cases of DVT and warrant anticoagulation.<sup>5</sup>

Coolsaet and Weinberg report conservative management with anti-inflammatory medication with complete resolution of symptoms in a child diagnosed by selective venography.<sup>1</sup> The child had a prior history of left orchidectomy for a similar complaint when

he was younger. No long-term follow-up data was available.

In the case described by Diana et al, the patient already had an established diagnosis of Henoch-Schönlein Purpura and management was with corticosteroids and low-molecular weight heparin postscrotal exploration.<sup>3</sup>

To date, there are no pediatric cases of idiopathic testicular vein thrombosis in which anticoagulation was used for treatment. Radulescu reviews the management and considerations of anticoagulation of VTE in children in a separate article.<sup>6</sup>

We elected to recommend anticoagulation based on the available literature, suspected provoked nature of the thrombosis, the atypical site of thrombosis, and to avoid the potential complications described above. It is also important to bear in mind the risk to benefit ratio of starting anticoagulation and that the pediatric population is special in that drug administration generally needs to be supervised by the guardians of the child.

## SUPPLEMENTARY MATERIALS

Supplementary material associated with this article can be found in the online version at <https://doi.org/10.1016/j.urology.2019.03.012>.

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