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Familial testicular torsion in siblings of different age groups: A case report

Murad Zetawi^a, Wesam Altawil^{b,*}, Mohannad Ashour^a, Mohammad Abughali^a,
Anas Ghousheh^a

^a Department of Urology at The Specialty Hospital, Amman, Jordan

^b Global Health Division at Civilian Research and Development Foundation Global, Amman, Jordan



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ABSTRACT

The acute scrotum (AS) is one of the urologic emergencies, where testicular torsion (TT) represents 20–30% of it. In the peri-puberty period, TT is not uncommon, yet there are very few reports of familial testicular torsion worldwide.

This case report highlights a man who experienced TT and had a family history of TT.

TT should be considered one of the leading causes of AS. We avoid any delay in the diagnosis of TT, where the signs, symptoms, family history of TT, physical examination, and color Doppler ultrasound (CDU) are helpful in earlier diagnosis and management to salvage the twisted testicle.

1. Introduction

AS is a medical emergency. It is characterized by pain, swelling, and/or reddening. Although 20–30% of instances of AS are caused by TT, as a urological emergency, TT needs immediate surgical intervention. TT incident is estimated to be 3.5/100000 males younger than 25-year-old per year, with two peaks: the highest is between 10 and 14-year-old (7.7/100000 males), and the second is below 1 year of age (7.6/100000 male infants).¹ A salvage rate of 90%, 50%, and fewer than 10% were noticed if the detorsion of the twisted testis had been done within 6, 12, and 24 hours of TT onset, respectively.²

Testicular tumors, horizontally positioned testicles, a lengthy intra-scrotal segment of the spermatic cord, high or close insertion of the tunica vaginalis to the spermatic cord, trauma, and recent exercise in addition to low temperatures are additional risk factors.³ Therefore, TT in the period of peri-puberty is not uncommon, but only a few reports of familial TT are in the literature.

2. Case report

In this case report, we are presenting a 25-year-old man who presented to the emergency room (ER) in February 2021 with intermittent left scrotal pain of moderate intensity of 3 days duration. He was also diagnosed previously to have left varicocele which he thought that it was the cause of his left scrotal pain, so he delayed seeking medical

advice. Therefore, after taking the patient's history of the presenting illness and as we know from the family history that one of his brothers (14-year-old) had right TT before four months, and according to the physical examination findings of a swollen, firm, tender, and high-riding left testis, the CDU was done and showed decreased blood flow of the left testis, where the decision had been made to do urgent scrotal exploration to find left TT that was unsalvageable, so left orchidectomy with contralateral testicular fixation were done.

By September 2021, we also diagnosed his other two brothers, 20- and 18-year-old, to have right TT 5 and 8 months after the older brother, respectively.

His first brother (14-year-old) presented to the ER in October 2020 with right AS of 3 hours duration that started suddenly, while upon physical examination there was swollen tender right testis high in the scrotum, and after the CDU the results were suggestive of TT, so he underwent urgent scrotal exploration and we found twisted right testis. Right testicular detorsion, and bilateral testicular fixation were done.

The second brother (20-year-old) complained in June 2021 of severe right AS of 5 hours duration that started suddenly, and upon physical examination, he had swollen tender right testis, and CDU he was found to have a decreased blood flow of the right testis, for which he underwent scrotal exploration and found to have right TT, so detorsion of the right testes with good testicular color and consistency, with bilateral testicular fixation were done.

While the third brother (18-year-old) presented in September 2021

Abbreviations: AS, Acute scrotum; CDU, Color Doppler Ultrasound; ER, Emergency room; TT, Testicular torsion.

* Corresponding author.

E-mail address: wesamaltawil@hotmail.com (W. Altawil).

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with sudden severe right AS of 2 hours duration, and according to physical examination findings of tenderness and swelling of the right testis, and CDU findings, he was diagnosed to have right TT, so immediately scrotal exploration was done with detorsion of the right testis and fixation of both testicles.

Thus, 3 out of the 4 brothers managed to save their testicles, and on the follow-up, 2 of the salvaged testicles have minimal shrinkage in testicular size. And the father and his sons were completely satisfied with the surgical interventions they received.

Moreover, on asking the father and the brothers to get more details, we found that all of his sons had TT while sleeping, all of them diagnosed previously to have retractile testicles, and all of them are football players. Also, there is relative relation between the parents as they are cousins, and their grandparents were also cousins. While, according to the father, there is no known history of TT in their previous generations.

3. Discussion

Family history is an important part of the patient's history in addition to the physical examination and CDU in making an accurate and quick diagnosis in patients with AS due to TT. In particular, the family history of TT is found to be of high benefit to ensure timely diagnosis and improve patient management. It was confirmed by our literature search that familial TT is relatively infrequently reported, especially for TT in siblings, which is an uncommon occurrence. Therefore, a family history is rarely sought when a patient presents with an AS.

However, autopsy studies indicate that roughly 12% of males have bell clapper malformation, and up to 80% of males who had been diagnosed to have TT have this malformation also.⁴ Although these percentages suggest a hereditary propensity toward TT, the mode of transmission still needs further studies as it is not yet been determined,⁵ which could be difficult to determine due to the low number of familial cases of TT in addition to the fact that TT occurrence is multifactorial.

4. Conclusion

TT should be ruled out in any case of AS. As it was noticed in our case report, TT can run in families, as we depend on the positive family history in addition to the history of presenting illness, clinical signs and symptoms, the physical examination, and the CDU for quicker diagnosis and management that end up with salvaging 3 out of 4 twisted testicles. In addition, for the diagnosis and treatment of TT, asking for a family

history of TT will be helpful as it could be considered an alarming sign for a higher possibility of TT, so will lead to earlier diagnosis and management with a higher rate of salvaging the twisted testicles. Family counseling on the high risk of developing TT, its signs and symptoms, and how early asking for medical advice had better outcomes and higher salvage rates should be part of case management in families with a strong propensity for TT.

Informed consent

Verbal informed consent was obtained from the patients who are 18-year-old and more, and for the patients who are younger than 18-year-old, verbal informed consent was obtained from their father.

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Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

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