CLINICO-HISTOPATHOLOGIC PRESENTATION OF A NIGERIAN CHILD WITH TORSION OF THE EPIDIDYMAL APPENDAGE: A CASE REPORT

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ABSTRACT

Introduction: Torsion of the appendix of the epididymis is a rare cause of acute scrotum. However, it can be distinguished from testicular torsion by its insidious onset and localizable tenderness. In addition, colour Doppler ultrasonography has characteristic findings in torsion of the testicular appendage.

Case presentation: We present a case of a 9-year-old male presenting with sudden onset left hemi-scrotal pain, and had clinical features mimicking testicular torsion with consequent immediate scrotal exploration. Intra-operative finding was in keeping with torsion of the epididymal appendage as against the earlier clinical presentation suggestive of testicular torsion.

Conclusion: Acute scrotum being a time dependent emergency may in some circumstance not allow time for Doppler ultrasonography which will differentiate torsion of epididymal appendix from testicular torsion, and thus preventing unnecessary scrotal exploration.

INTRODUCTION

The appendix testis and the appendix epididymis are the two testicular appendages that can undergo torsion mimicking testicular torsion (spermatic cord torsion). Torsion of the intra-scrotal appendages is responsible for about 60% of common urologic emergencies within the paediatric population.¹

The appendix testis or hydatid of Morgagni is a vestigial remnant of the Mullerian duct which is present in about 76 - 83% of testes, 1,2 while the less common epididymal appendage, or the appendix epididymis, is a developmental remnant of the Wolfian or mesonephric duct that sprouts from the head of the epididymis with an autopsy incidence of 20% - 24%. 1 Epididymal appendix is located on the superior pole just on the head of the epididymis (Figure 1), and it is a less common appendage to undergo torsion after appendix testis 1,2. Although such a structure is generally of not much clinical significance, its pedunculated configuration makes it prone to torsion with acute scrotal pain similar to that of testicular torsion in prepubertal boys, and a rare cause of an acute scrotum. 1,3,4

Physical examination in torsion of testicular appendage may reveal a normally appearing scrotum with intact cremasteric reflex and tenderness localized to the upper pole of the testis or epididymis with a palpable localizable mass in the area of maximum tenderness. However, reactive inflammation of surrounding structures may cause a more diffuse pain making torsion of epididymal appendix indistinguishable from testicular torsion.⁵

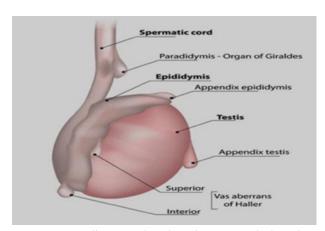


Figure 1: A diagram showing the anatomic location of the testicular appendages.

Colour Doppler ultrasonography is the imaging modality of choice for the evaluation of the acute scrotum in all age groups. While torsion of the intrascrotal appendages rarely requires surgical intervention, its typical clinical presentation often makes it challenging to distinguish from testicular torsion, a more serious diagnosis that often requires prompt scrotal exploration. Treatment of torsion of a testicular appendage therefore is usually non-operative, as symptoms usually resolve within few days to a week, and scrotal exploration is performed when there are doubts about the diagnosis. Scrotal exploration is

recommended without delay for scrotal scan to improve chances of testicular salvage in patients with very high risk of having testicular torsion as determined by the Testicular Workup for Ischaemia and Suspected Torsion (TWIST) score.⁵ TWIST score is a 7-point tool for evaluating acute scrotal pain. It consists of testicular swelling (2 points), hard testis (2 points), highriding testis (1 point), absent cremasteric reflex (1 point) and nausea/vomiting (1 point)⁷. A score of 5 and above is of great concern for testicular torsion for which delays should be avoided, and urgent scrotal exploration recommended.^{6,7}

We report a case of torsion of the epididymal appendage in a 9-year-old Nigerian male who presented with sudden onset features of acute scrotum that required prompt surgical exploration to accurately diagnose.

CASE PRESENTATION

A 9-year-old male presented with a sudden onset left scrotal pain few hours prior to presentation. The pain was dull in nature and relieved transiently by the use of analgesics. There was no history of fall or trauma to the scrotum. There was no history of dysuria, urinary frequency, pus or blood in the urine. He had no history

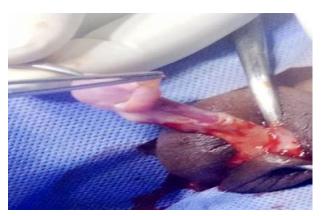


Figure 2: Showing inflamed torsed epididymal appendage at the superior pole of the testis.

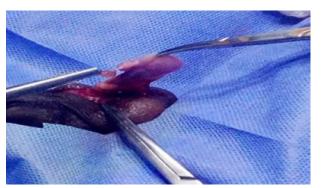


Figure 3: Showing the ligated detorsed epididymal appendage just before excision.

of recent urethral catheterization or previous episode of scrotal pain. There was no associated fever, nausea or vomiting.

At presentation, he was afebrile and well hydrated. His vital signs were normal while genital examination revealed a swollen left hemi-scrotum which was warm and tender. Cremasteric reflex was absent and Prehn sign was negative. He had a TWIST score of 5 (left testicular swelling; 2, absent cremasteric reflex; 1, hard left testis; 2) which necessitated immediate scrotal exploration.



Figure 4: A micro photograph of a polypoid tissue partly lined by pseudostratified columnar epithelium.

Intra-operative finding was an inflamed tiny polypoid tissue in 630Ú anticlockwise torsion at the superior pole, on the epididymis of the left testis (Figure 2). The appendage was detorsed, ligated at its base and excised (Figure 3). Scrotal incision was closed with subcuticular vicryl 2/0 stitches, and patient was discharged on post-operative day one. He was seen at surgical outpatient clinic at 2nd week, one year post-surgery with no complaints and satisfactory wound healing. Microscopy of excised surgical specimen revealed sections of polypoid tissue partly lined by pseudostratified columnar epithelium (Figure 4). Some

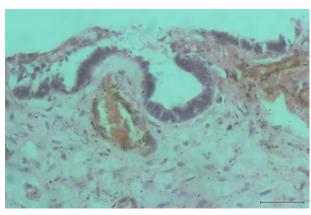


Figure 5: A micro photograph showing ducts lined by pseudostratified epithelium within the underlying loose fibromyxoid stroma. Also seen are congested blood vessels.

of these ducts lined by pseudo stratified epithelium are within the underlying loose fibromyxoid stroma with congested blood vessels. Features are in keeping with an epididymal appendage (Figure 5).

DISCUSSION

The actual cause of torsion of testis or its appendage is unknown but may be related to trauma or prepubertal enlargement, and for unexplained reasons it has been found to be commoner during winter.⁷

Torsion of epididymal appendage and hydatid of Morgagni commonly causes pain similar to that of testicular torsion.⁴ Torsion of the epididymal appendage classically presents in children aged 7-14 years and rarely occurs in adulthood.⁸ Patients classically present with gradual onset of scrotal pain in the presence of a preserved cremasteric reflex, which may be associated with scrotal swelling and erythema. In addition, patients may also have other non-specific symptoms such as nausea, vomiting, or abdominal pain.⁸

Physical examination in torsion of testicular appendage may reveal a normally appearing scrotum with intact cremasteric reflex and tenderness localized to the upper pole of the testis or epididymis with a palpable localizable mass in the area of maximum tenderness.9 Furthermore, while only present in about 20% of cases, the palpation of a para-testicular nodule at the superior aspect of the testis, a so-called "blue-dot" sign, is pathognomonic for this condition. The combination of the blue-dot sign with clear palpation of an underlying normal, non-tender testis, presence of a cremasteric reflex are of significant clinical value for the exclusion of testicular torsion on clinical grounds alone.^{1,8} Nevertheless, the clinical picture for a torsed epididymal appendage can often be misdiagnosed as epididymitis or epididymo-orchitis, making an assessment with sonology crucial to discerning the ambiguity.

Colour Doppler ultrasonography is the imaging modality of choice for the evaluation of the acute scrotum in all age groups. The torsed appendage usually appears as an enlarged oval avascular mass, with heterogeneous echo texture, posterior enhancement close to the upper pole of the testis, and typically shows normal blood flow to the testicle on the affected side. ^{1,8,10} While the detection of an epididymal appendage is not pathognomonic for torsion, its detection with a diameter greater than 5.6 mm is highly suggestive of torsion, with almost 90% sensitivity. ^{8,11} In the event of infarction of the torsed peduncle, the appendage may detach and wander inside the scrotum, appearing as minute mobile particles. Findings

consistent with a thickened scrotal wall and a reactive hydrocele may also be present. However, these findings are non-specific and may be found in individuals with epididymitis. Epididymal tumours may also mimic epididymal appendage torsion but the absence of mass-like lesions and vascular flow in the epididymis on the scrotal ultrasound distinguishes torsion from an epididymal tumor. ¹¹ In addition, a delay in performing Doppler ultrasonography may lead to increased vascular flow in the epididymis. This may lead to an erroneous diagnosis of epididymitis and unnecessary antibiotic treatment. ^{8,11}

Accurate and rapid evaluation of the acute scrotum is critical for patient management. Treatment of torsion of a testicular appendage is usually conservative therapy. Conservative management includes bed rest, scrotal elevation, use of ice pack, and analgesics such as non-steroidal anti-inflammatory drugs. Symptoms are usually self-limiting. However, in order to improve testicular salvage rates in testicular torsion, it is important to promptly offer scrotal exploration to patients with suspicion of testicular torsion without delay for scrotal ultrasonography.6 Hence, the TWIST score was developed, and it has been validated to identify patients at high risk of testicular torsion among patients with acute scrotum.6 Our patient had a TWIST score of 5 which raised a suspicion of testicular torsion, and symptoms have been present for hours unlike the natural history that is typically gradual pain in torsion of the epididymal appendage. Hence, the decision not to delay scrotal exploration for scrotal ultrasonography as surgical intervention through appendage excision can shorten the average duration of symptoms. 1,4

CONCLUSIONS

Torsion of the epididymal appendage is an uncommon cause of an acute scrotum in the paediatric population which may be distinguished clinically from testicular torsion. Given its often challenging and atypical presentation, high index of clinical suspicion and Doppler ultrasonography are needed to guide accurate diagnosis and obviate unnecessary surgical exploration as presentation may mimic testicular torsion. Nevertheless, in clinically equivocal cases, prompt emergent scrotal exploration is necessary to reach a lucid diagnosis.

Conflict of Interest Statement

The authors affirm that they have no conflict of interests to declare.

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