Urology Case Reports 42 (2022) 102032

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: www.elsevier.com/locate/eucr

Complete testicular-epididymal dissociation presenting as adult chronic orchialgia

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ARTICLE INFO

Keywords:
Testicular epididymal fusion abnormality
Epididymis
Fusion abnormality
Testicular dissociation
Testicular torsion
Orchialgia

ABSTRACT

Complete testicular epididymal dissociations are exceedingly rare conditions where the epididymis and the vas deferens are completely dissociated from the testicle. We present the case of a 46-year-old male with a history of chronic, intermittent and severe left testicular pain who was found to have a complete testicular epididymal dissociation at the time of surgical exploration and bilateral orchidopexy. Microsurgical approximation of the tail of the epididymis to the tunica albuginea of the testis with reapproximating the muscularis of the spermatic cord to the epididymal appendage was performed with subsequent relief of symptoms.

1. Introduction

Testicular epididymal fusion abnormalities are rare anatomical variants, usually incidentally found at pediatric orchidopexy for cryptorchidism. Normal fusion of the testicle and epididymis is via the efferent ducts.[1] Fusion abnormalities occur when there is disruption in the attachment of epididymis and testis. Epididymal body separation alone is considered normal.[1] Fusion abnormalities can be classified into head, tail, and complete non-fusion.[1] The most severe form is complete testicular epididymal non-fusion, where no contact exists between the testicle and epididymis; This is exceedingly rare.[1] In adults with normally descended testicles, fusion abnormalities may be identified during surgical exploration for orchialgia, such as for testicular or epididymal torsion.

Herein we present a 46-year-old male with chronic orchialgia who was found at exploration to have congenital, complete testicular epididymal dissociation. This is the first reported case of an adult with no pre-existing testicular conditions having this finding. Testicular epididymal dissociations should be on the differential diagnosis for chronic orchialgia.

2. Case presentation

A 46-year-old male presented with chronic intermittent, "stabbing" left orchialgia, present since childhood. The pain was positional, worse laying on his side. During episodes the testicle felt "balled up", and manual massage resulted in improvement. There was no association with urination or sex. The patient had not attempted nor desired paternity. Medical, surgical, social and family histories were noncontributory. Exam revealed normal testicles, 18 cc bilaterally, palpable vasa and no varicoceles. The left epididymal head was full and tender to palpation.

Urinary testing was normal. Doppler ultrasound showed a 4 mm left testicular appendix, and a 4 mm left epididymal head cyst. The patient's history was consistent with intermittent testicular or epididymal appendage torsion. We attempted to perform ultrasound during a painful episode but were unsuccessful in capturing these episodes.

The patient had failed several oral pain medications, including nonsteroidal's and neuropathic agents. He was interested in a definitive surgical repair. After discussion of treatment options, patient opted for exploration, excision of left epididymal appendage and bilateral orchidopexy.

In the operating room, the left testicular unit was delivered. It was immediately apparent that the epididymis and vas deferens were completely dissociated from the testicle, consistent with complete congenital testicular epididymal dissociation (Fig. 1). The epididymal tubules were grossly dilated. Since the patient did not desire paternity, we did not biopsy or aspirate these.

We hypothesized that there may have been intermittent epididymal torsion or epididymal strangulation by the testicular vasculature. The epididymal appendage was excised. Under the operative microscope, the epididymal cyst was dissected, excised, and a tunical flap placed. We

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https://doi.org/10.1016/j.eucr.2022.102032
Received 1 February 2022; Accepted 15 February 2022
Available online 17 February 2022

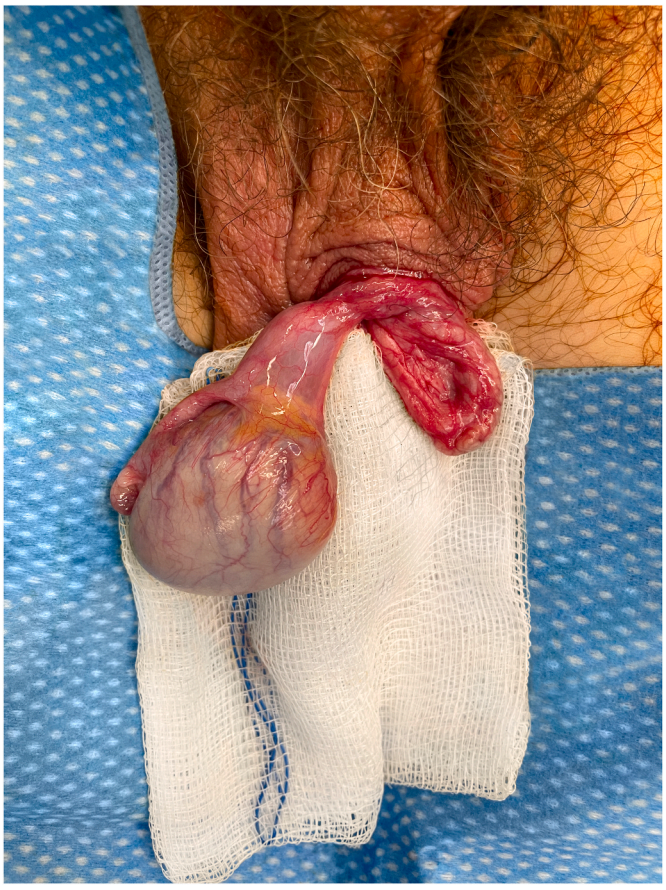

Fig. 1. Left epididymis and the vas deferens completely dissociated from the testicle, demonstrating a complete congenital testicular epididymal dissociation.

then turned our attention to the epididymal dissociation. We elected to orthotopically pex the epididymal tunic to the testis, and then reinforced our anastomosis by approximating the muscularis of the spermatic cord to the epididymal appendage, both using 4.0 Vicryl. In this way, there were no gaps at the level of the testicle, epididymis or spermatic cord for possible herniation or torsion. At the end of the procedure, Doppler confirmed an excellent testicular arterial pulse. A 3-point orchidopexy was performed bilaterally.

At 6-week follow-up, the pain had changed from sharp spasms to a dull ache. At 4-month follow-up, the patient was pain free. We discussed semen analysis testing and cystic fibrosis screening, but patient declined both as he did not desire paternity.

3. Discussion

This is the first reported case of complete testicular epididymal non-fusion in an adult with descended testes. Our patient may have had a bell-clapper deformity, where testicular and epididymal hypermobility resulted in intermittent torsion of either the testicle or epididymis. There is scant data looking at the incidence of bell-clapper deformity, with subsequent torsion (testicular or epididymal) in men with testicular epididymal fusion abnormalities. One series of 50 testicular torsion cases found that 12% had epididymal fusion dissociations, although how many of these were complete dissociations was not stated.[2]

Testicular epididymal dissociations can only be diagnosed at surgical exploration, explaining why they are usually identified at orchidopexy. As in our case, preoperative exam and ultrasound are of limited utility. It is important that clinicians are aware of this condition, since if identified at surgery, reconstruction can be undertaken. As in this case, micro-surgical approximation of the epididymis to the testicle may help alleviate orchialgia.

Both cryptorchidism and fusion abnormalities may result in fertility issues, from production and obstructive causes, respectively. Cryptorchidism has been associated with azoospermia in 13% of unilateral and 89% of bilateral cases.[3] Fusion abnormalities can result in obstruction from a lack of continuity in sperm passage. Additionally, non-fused testicles may have smaller volumes when compared to fused testicular-epididymal units, indicating some potential hypospermatogenesis.[4] While both can contribute to fertility issues, the full interplay of these causes is poorly understood.

There have been two reports of fusion abnormalities resulting in orchialgia due to bell-clapper deformities. The first was a 31-year-old male with chronic orchialgia and delay in conception. At exploration, a fusion abnormality was identified, with only the head of the epididymis attached to the testicle. The unit was found to be mal-rotated, he was diagnosed with intermittent torsion, and managed successfully with orchidopexy.[5] The second case was a 17-year-old male with intermittent orchialgia, found to have complete testicular epididymal non-fusion with a bell-clapper deformity. Orchidopexy was successful in alleviating the pain.[2] Neither patient had a history of cryptorchidism.

Due to the rarity of testicular epididymal fusion abnormalities in adults, more data is needed to understand the impact of these on fertility and orchialgia. As imaging modalities improve, it is possible that these may be identifiable with imaging. For now, it is important that fusion abnormalities remain on the differential diagnosis for males presenting with testicular or epididymal pain, or infertility.

4. Conclusion

Testicular epididymal fusion abnormalities are exceedingly rare and may be identified at exploration for chronic orchialgia, even with a normal pre-operative ultrasound. Reapproximating the epididymis to the tunica albuginea of the testis, with subsequent reapproximating the muscularis of the spermatic cord to the epididymal appendage, may result in resolution of pain.

Declaration of competing interest

This research did not receive any specific grant from funding agencies in the public, commercial or not-for-profit sectors.

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