Testicular infarction in a male patient with cystic fibrosis: Testicular remnant preservation

Masoud Bitaraf; Mohammadreza Modaresi²,3; Abdol-Mohammad Kajbafzadeh¹*
¹Pediatric Urology and Regenerative Medicine Research Center, Section of Tissue Engineering and Stem Cells Therapy, Children’s Hospital Medical Center, Tehran University of Medical Sciences, Tehran, Iran.
²Pediatric Pulmonary Disease and Sleep Medicine Research Center, Pediatric Center of Excellence, Tehran University of Medical Sciences, Tehran, Iran.
³Cystic Fibrosis Research Center, Iran CF Foundation (ICFF), Tehran, Iran.

Abstract

With improvements in management of Cystic Fibrosis (CF), nowadays more patients reach adulthood and desire to have their own biological child. Although relatively 97% of men with CF suffer from obstructive azoospermia, they are expected to have regular spermatogenesis. Subsequently by utilizing sperm retrieval and assisted reproductive techniques their wish can become true. In this case report we present a case of testicular infarction in a man with CF, which is the first reported case to the best of our knowledge, in an attempt to highlight the importance of monitoring genitourinary system in these patients to prevent possible sequelae for the sake of a better life quality.

Keywords: testis; infarction; cystic fibrosis; fertility; spermatozoa.

Abbreviations: CF: Cystic Fibrosis; CFTR: Cystic Fibrosis Transmembrane Conductance Regulator; CBAVD: Congenital Bilateral Absence of the Vas Deferens; ICSI: Intracytoplasmic Sperm Injection; PESA: Percutaneous Epididymal Sperm Aspiration; MESA: Microsurgical Epididymal Sperm Aspiration; TESA: Percutaneous Testicular Aspiration; TESE: Testicular Surgical Biopsy and Testicular Sperm Extraction.

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Introduction

The autosomal recessive disorder, Cystic Fibrosis (CF) due to CF Transmembrane Conductance Regulator (CFTR) gene mutations is mainly characterized by pulmonary, gastrointestinal and sweat glands involvement. Male genitourinary system is also affected in forms of Congenital Bilateral Absence of the Vas Deferens (CBAVD), testicular and epididymal cysts, spermatozoa maturation arrest and reduced mature sperm count [1].

Due to the improvements in management of CF, increasing numbers of patients reach adulthood; hence, escalating quest for parenthood. Despite the reported rate of 97% infertility in male patients mainly due to obstructive azoospermia, the emergence of assisted reproductive techniques such as Intracytoplasmic Sperm Injection (ICSI) is found promising and signifies tests preservation through identification of and observation for potential threats [2]. Here we present hit her to the first case of testicular infarction in a male with CF.

Case description

In January 2002, an 18 year old Persian male, known case of CF, was referred to our clinic by his gastroenterologist due to painless swelling and stiffness of his left testicle. History was negative for trauma, fever, dysuria and discharge. On physical
exam, left testicle was swollen and firm but with no erythema or tenderness. Right testicle appeared normal. Also his secondary sex characteristics were normal. Scrotal gray scale ultrasonography and tumor markers were requested, the results of which were within normal range. Testicular Doppler ultrasonography depicted focal testicular necrosis evident by hypo vascularity and no evidence of tumor or testis torsion.

The gentleman refused to undergo radical orchiectomy as next step in management. Accordingly, on March 2002, after obtaining a written informed consent explaining the procedure and asking for permission of data publication in various forms of study, testicular biopsy was performed after cord clamp under a short anesthesia with mask ventilation. Frozen section was sent to pathology. On macroscopic examination, a fresh soft grayish tissue measuring 2 X 1 cm and 2 X 0.3 cm submitted in two blocks was reported. Microscopic examination reported no evidence of malignancy and histopathologic findings were compatible with necrotic changes and organizing arterial branches thrombosis.

The patient recovered from surgery and was discharged on same day. On follow up visit in August 2002, Left testicle appeared atrophic on physical exam (Figure 1). He was last visited in 2012 while he had no genitourinary complaint and was not in a relationship. It should be noted that he lost his follow-up since then.

**Discussion**

By survival improvement among men with CF, the propensity to father their biological child has become a concern. It is a very delicate issue with multiple factors to be taken into consideration aside from the feasibility of conceiving their partner. Factors including the life expectancy and long-term health condition of the potential father and the indisputable fact that conceived child carries the CFTR gene mutation with its inevitable impacts on their life must be considered and counseled before any intervention [2].

Although considering CFTR protein role in spermatogenesis, spermatozoa maturation and epididymis, ductus deferens and seminal vesicles formation, regular spermatogenesis is expected due to normal Testosterone, LH and FSH levels; providing sources to retrieve sperm through a variety of procedures including Percutaneous Epididymal Sperm Aspiration (PESA), Microsurgical Epididymal Sperm Aspiration (MESA), Percutaneous Testicular Aspiration (TESA), Testicular Surgical Biopsy And Testicular Sperm Extraction (TESE) with tubular microdissection [1,2].

Testicular infarction can occur due to any insult resulting in lack of blood flow to testicles. It could be secondary to testicular torsion, an arterial or venous thrombosis, trauma orepididymo-orchitis. Also there have been few reports of testicular infarction secondary to protein s deficiency. It can also occur idiopathically [3,4]. An association between CF and protein s deficiency has been reported in literature [5].

Our report is limited by the facts that we could not perform radical orchiectomy and poor compliance of patient did not allow further work ups to identify the underlying etiology of his testicular infarction. Albeit it raises the question about new genitourinary manifestations of CF or at least disease associated conditions leading to genital sequelae.

**Conclusion**

We strive to highlight the importance of regular genitourinary evaluation in men with CF from the onset of puberty with special consideration of possible factors that lead to testis loss since it is apparent that there should be a normally functioning, healthy testicle to produce sperm cells. It could even be a note worthy suggestion that early sperm retrieval be considered in affected adolescents. And last but not least, regarding natural course of CF and the possibility of recurrence of infarction in contralateral testis, we recommend against radical orchiectomy in such cases for purpose of preserving every potential of spermatogenesis.

**Highlights**

- 97% of males with CF suffer obstructive azoospermia.
- Regular spermatogenesis is expected in males with CF.
- Testicular infarction in a male with CF is reported.
- Sperm retrieval and ART can yield biological child for CF patients.
- Monitoring genitourinary system in CF patients from adolescence matters!
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References