UIJ UroToday International Journal®

www.urotodayinternationaljournal.com
Volume 2 - April 2009

Splenogonadal Fusion: A Case Report

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Submitted on October 13, 2008 - Accepted for Publication on December 4, 2008

ABSTRACT

Splenogonadal fusion is a rare congenital anomaly in which there is fusion between the spleen and gonad, epididymis, or vas deferens. The abnormal splenic tissue may or may not be continuous with the orthotopic spleen. The diagnosis is most often made when an extratesticular scrotal mass is noted or a lesion is discovered during orchiopexy.

We reviewed the medical record of a 26-year-old male medical student with splenogonadal fusion diagnosed during surgical exploration for cryptorchidism.

KEYWORDS: Splenogonadal, Cryptorchidism, Undescended testicle, Congenital anomalies

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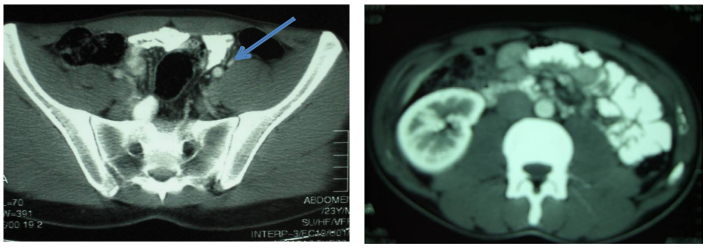
INTRODUCTION

There have been approximately 120 cases of reported splenogonadal fusion since the first description of this entity in 1883 by Bostroem [1]. Close proximity of the spleen and gonad during early embryological development allows for fusion, whether continuous or discontinuous, of these seemingly unrelated organs. The continuous type of splenogonadal fusion describes the gonad attached to the anatomic spleen. The discontinuous type consists of gonadal fusion with an accessory spleen or ectopic splenic tissue. We present a case of continuous splenogonadal fusion presenting as an undescended testicle. This case was unique in that the patient had left cryptorchidism and agenesia of the left kidney.

CASE

The patient was a 23-year-old male who was referred for left inguinal pain. On physical examination, the left testicle was not palpable in the scrotum or inguinal canal. The right testicle, epididymis, and spermatic cord were normal. The preoperative CT scan revealed intrapelvic left testis (Figure 1a) and right solitary kidney (Figure 1b). The left testicle was explored through an inguinal incision. The inguinal canal was empty. The testicle was found inside the abdomen. A reddish brown cord of tissue was fused to the left testicle. The aberrant tissue was continuous with a structure in the left upper quadrant. This was felt to be consistent with the spleen. The reddish brown cord was dissected from the spleen. Macroscopic examination identified an 18 x 2 x 0.5-cm reddish cord-like structure which contained 3 nodules (0.9 cm, 2.7 cm, 3.7 cm). Histological examination revealed that the bigger nodule was an atrophic testis (Figure 2). We identified complete aplasia of the seminal epithelium, small seminiferous tubules with thick walls, and mild hyperplasia of the Leydig cells. A few seminiferous tubules contained psammoma bodies. The morphological and immunohistochemical findings (negative stain for placental alkaline phosphatase (PLAP)) excluded in-situ carcinoma. The



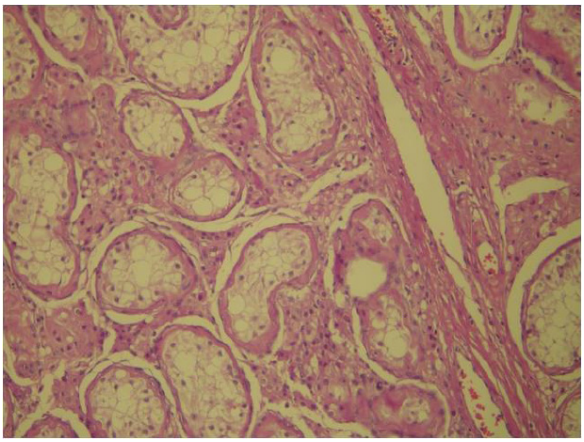
left Figure 1a. CT scan of intrapelvic left testis
doi:10.3834/uij.1944-5784.2009.02.03.f1
right Figure 1b. Right solitary kidney
doi:10.3834/uij.1944-5784.2009.02.03.f3

other 2 nodules consisted of splenic tissue.

DISCUSSION

This case presents an unusual case of splenogonadal fusion. This case is unique in that it involves only genitourinary anomalies. There have been 2 forms of splenogonadal fusion described: continuous and discontinuous. The continuous form occurs when the anatomic spleen is connected by a discrete cord to the gonad, as in our case. The discontinuous form consists of a fused splenogonadal structure that has lost continuity with the main spleen. The continuous type seems to be predominant [2]. Splenogonadal fusion has been associated with other congenital anomalies. Most cases that had multiple anomalies were of the continuous variety. Only 3 cases of the discontinuous type have had associated defects. The most common defect is of the limbs [1,3]. Other associated anomalies include micrognathia, cardiac defects, cleft palate, anal defects, spina bifida, and facial muscle agenesis. There have been 7 cases reported with bilateral cryptorchidism [1]. One case with associated bilateral cryptorchidism also had an association with an adrenal cortical rest. This case, however, was a case of discontinuous splenogonadal fusion [4]. As mentioned previously, there have been cases associated with cryptorchidism. In searching electronic databases, there were no cases identified of splenogonadal fusion and unilateral kidney agenesia [1,5,6].

Figure 2. Histological examination revealed that the bigger nodule was atrophic testis
doi:10.3834/uij.1944-5784.2009.02.03.f2



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TO CITE THIS ARTICLE: Charalambous SN, Triantafyllidis AK, Papanikolaou AI, Brouskelis NK, Rombis VE. Splenogonadal fusion: a case report. *UIJ*. 2009 Apr;2(2). doi:10.3834/uij.1944-5784.2009.02.03