Laparoscopic Diagnosis and Treatment of Splenogonadal Fusion Associated with Intra-Abdominal Cryptorchidism

İntraabdominal Kriptorşidizmle Bağlantılı Splenogonadal Füzyonun Laparoskopik Tanı ve Tedavisi

Bakhtiyar Hüseynov¹, Deniz Abat¹, Şeyda Erdoğan², Yıldırım Bayazıt¹

¹Çukurova Üniversitesi Tıp Fakültesi Üroloji ve ²Patoloji Anabilim Dalları, Adana

ABSTRACT
Splenogonadal fusion is a rare congenital anomaly in which there is a fusion between spleen and gonad. According to the literature, 37% of them had unnecessary orchiectomy. We present a patient with splenogonadal fusion in undescended testis, who was preserved from unnecessary orchiectomy.

Key Words: Laparoscopy, Infertility, Orchiopexy, Orchiectomy, Testis, Splenogonadal fusion, Spleen.

ÖZET
Splenogonadal füzyon dalak ile gonadın füzyon olduğu nadir görülen konjenital anomalilerden biridir. Literatüre göre bu olguların %37’sine gereksez orşiektomi yapılmaktadır. Burada inmemiş testisle birlikte splenogonadal füzyonu olan ve gereksez orşiektomiden kaçınılan bir hasta sunulmaktadır.

Anahtar Kelimeler: Laparoskopi, İnfertilite, Orşiopeksi, Orşiektomi, Testis, Splenogonadal füzyon, Dalak.

INTRODUCTION
Splenogonadal fusion is a rare congenital anomaly and approximately 150 cases have been reported in the literature. Splenogonadal fusion was first described by Bostroem in 1883¹. Most cases presented with a testicular mass and unfortunately 37% of them were treated with unnecessary orchiectomy². We present a patient with splenogonadal fusion in undescended testis, who was preserved from an unnecessary orchiectomy.

CASE REPORT
A twenty - seven years old infertile man was admitted in our urology clinic for undescended left testis. On genital examination, atrophic right testis was palpable in the scrotum, whereas left testis was not palpable. Sonographic evaluation revealed an 8 mm atrophic left testis in the inguinal region. Testicular tumor markers were normal. Laparoscopic exploration was planned. Laparoscopic surgery was performed via three 5 mm ports and left testis was found in the abdomen. It was adjoined with colon and surrounding tissue. There was a dark purple tissue fused with the testis and it was excised and examined with frozen section. Histopathological evaluation confirmed splenogonadal fusion, discontinuous type (Figure 1). The procedure was completed with left orchiopexy.
DISCUSSION

Splenogonadal fusion is a rare congenital anomaly and approximately 150 cases have been reported since the first description by Bostroem in 1883\(^2\). Two forms of splenogonadal fusion have been described by Putschar and Manion in 1956, continuous and discontinuous\(^2\). In continuous type, gonad and spleen connected with a cord like structure that may be totally splenic, beaded with multiple splenic nodules, or composed of fibrous tissue\(^4\). In discontinuous type, gonad and splenic tissue was fused and there is no anatomic connection between ectopic or principal spleen\(^3\). This condition seems predominantly in males with a male to female ratio of about 16:1. Splenic anlage is formed between the layers of the dorsal mesogastrium by mesenchymal cells between 5\(^{th}\) and 6\(^{th}\) week of gestation. At 6\(^{th}\) week of gestation, germ cells migrate to the gonadal ridge which is lies just lateral to the mesogastrium. When stomach is rotates to the left side, splenic anlage rotated with it, and at this point splenic anlage can be connecting with gonadal ridge. Two theories attempt to explain how the spleen fuses with the gonad; simple adhesion or mild inflammation\(^5,6\).

The most associated defect is of the limbs and the other related anomalies include micrognathia, microgastria, cardiac anomalies, cleft palate, anal anomalies, spina bifida, craniosynostosis, thoracopagus, diaphragmatic hernia, lung anomalies and facial muscle agenesis\(^6,7\). These defects are usually associated with continuous type, however discontinuous type is rarely associated with congenital anomalies\(^5\). Diagnosis can be confirmed with color Doppler sonography, spiral computerized tomography and technetium-99m sulfur colloid scintigraphy. Preoperative diagnosis is very difficult especially for discontinuous type due to this condition is very rare and only a few cases were diagnosed preoperatively using by technetium-99m sulfur colloid scintigraphy\(^2,5\). Splenogonadal fusion is generally found during groin exploration for an undescended testis or hernia. The most common presentation is that of a testicular swelling\(^5,8\). Unnecessary orchiectomy usually was performed because of suspicion that it could be testicular neoplasm\(^5\). Other presentations included acute scrotum secondary to scrotal splenic enlargement, traumatic rupture of the scrotal spleen and bowel obstruction related to the continuous form of splenogonadal fusion\(^2,9\). Patient with scrotal mass if have any congenital defect, should be assessed radiologically for splenogonadal fusion\(^5\).

Alivizatos et al. performed inguinal orchiectomy for a left testicular mass in a 20-year-old patient. Histopathology revealed no tumor and the diagnosis was splenogonadal fusion\(^10\). However, it is possible to save the testicle in some cases and unnecessary orchiectomy can be prevented by intraoperative use of frozen section examination. Irkilata et al. presented 3 cases of splenogonadal fusion and performed orchiectomy in first case since testicular neoplasm could not be ruled out. They preserved the testicle in the latter 2 cases by using imaging studies and frozen section\(^8\). M Karaman et al. preserved the testicle by using frozen section in 2 cases who presented with acute left inguinal pain and swelling, and a left scrotal mass respectively\(^7\). A Khairat et al. completely excised the splenic tissue from the testicle intraoperatively and performed orchiopexy\(^3\). K. C. Balaji et al. advocated that familiarity with this entity allows for intraoperative diagnosis and they reported all the 7 patients in their series were diagnosed during surgery\(^9\).
Intrascrotal testis has a higher risk than intrabdominal testis for orchiectomy. Frozen examination may confirm the diagnosis both in an intrascrotal and in an intraabdominal testicle. Laparoscopic approach as a minimally invasive treatment modality for orchiopexy can provide some advantages like image magnification that helps diagnosis and may decrease probability of an unnecessary orchiectomy in intraabdominal testicle.

When an unusual tissue attached to testicle was detected during laparoscopy, frozen section examination may confirm the diagnosis and prevents an unnecessary orchiectomy.

CONCLUSION

Despite splenogonadal fusion is a very rare entity, to suspect and research with radiologic tools preoperatively and / or with frozen section intraoperatively may prevent unnecessary orchiectomy.

REFERENCES


Yazışma Adresi / Address for Correspondence:
Dr. Yıldırım Bayazı
Çukurova Üniversitesi Tıp Fakültesi
Üroloji Anabilim Dalı
ADANA
e-mail: ybayazit@yahoo.com
geliş tarihi/received :12.08.2012
kabul tarihi/accepted:17.10.2012