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Olgu Sunumu / Case Report

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

İntraabdominal Kriptorşidizmle Bağlantılı Splenogonadal Füzyonun Laparoskopik Tanı ve Tedavisi

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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 füzyone olduğu nadir görülen konjenital anomalilerden biridir. Literatüre göre bu olguların %37'sine gereksiz orşiektomi yapılmaktadır. Burada inmemiş testisle birlikte splenogonadal füzyonu olan ve gereksiz 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 1883[1]. Most cases presented with a testicular mass and unfortunately 37% of them were treated with unnecessary orchiectomy[2]. 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.

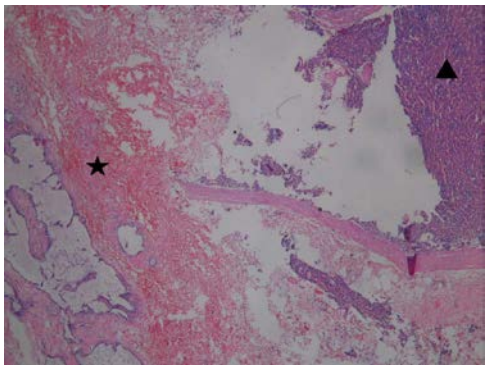

Figure 1.

DISCUSSION

Splenogonadal fusion is a rare congenital anomaly and approximately 150 cases have been reported since the first description by Bostroem in 1883[1]. Two forms of splenogonadal fusion have been described by Putschar and Manion in 1956, continuous and discontinuous[3]. 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 5th and 6th week of gestation. At 6th 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[2]. 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[6].

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. Irkılata 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[2]. A Khairat et al. completely excised the splenic tissue from the testicle intraoperatively and performed orchiopexy[5]. 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 intra-abdominal 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

1. Bostroem E. Demonstration eines Praparated von Verwachswung der Mils mit dem lenken Hoden. Gesell – schaft deutscher Naturforscher und Aerzte, Verhandlungen der 56 Versammlung, Freiburg. 1883; 149.
2. Karaman MI, Gonzales ET JR. Splenogonadal fusion: report of 2 cases and review of the literature. J Urol. 1996; 155:309-11.
3. Putschar WGJ, Manion WC. Splenic – gonadal fusion. Am J Pathol. 1956; 32:15.
4. Ziylan O, Sanli O, Atar A, Tefik T., Karadeniz M., Ander H. Splenogonadal fusion: An unusual cause of an ascending testis and laparoscopic solution. J Endourol. 2009; 3(Suppl1): A351.
5. Khairat AB, Ismail AM. Splenogonodal fusion: case presentation and literature review. J Pediatr Surg. 2005; 40:1357-60.
6. Varma DR, Sirineni GR, Rao MV, Pottala KM, Mallipudi BVP. Sonographic and CT features of splenogonadal fusion. Pediatr Radiol. 2007; 37:916-9.
7. Duncan WL Jr, Barraza MA. Splenogonadal fusion: a case report and review of literature. J Pediatr Surg. 2005; 40: 5-7.
8. Irkilata HC, Aydur E, Yildirim I, Kibar Y, Dayanc M. Splenogonadal fusion in adults: presentation of three cases and review of the literature. Urol Int. 2008; 81:360-3.
9. Balaji KC, Caldamone AA, Rabinowitz R, Ortenberg J, Diamond DA. Splenogonadal fusion. J Urol. 1996; 156:854-6.
10. Alivizatos G, Skolarikos A, Sopilidis O, Ferakis N, Chorti M. Splenogonadal fusion: report of a case and review of literature. Int J Urol. 2005; 12:90–2.

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