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Incidentally detected splenogonadal fusion in a laparoscopic transabdominal preperitoneal hernia repair operation: A case report



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ABSTRACT

INTRODUCTION: Splenogonadal fusion (SGF) is a rare congenital malformation in which the spleen is connected to the gonad. Few SGF cases have been reported in the English scientific literature, and we are unaware of any previous case reports of SGF with inguinal hernia by laparoscopic transabdominal preperitoneal hernia repair (TAPP). Here, we report a case of SGF that was incidentally detected during a TAPP procedure, with an uneventful postoperative course without complications.

PRESENTATION OF CASE: A 76-year-old male presented with a 10-year history of left inguinal swelling. He was diagnosed with a left inguinal hernia, and we performed TAPP. Laparoscopy revealed the left inguinal hernia and two reddish-purple masses, one located close to the left inguinal ring. A cord of soft tissue extended cranially from the mass to the spleen, and passed through the left internal inguinal ring caudally. We cut the cord for mesh placement and to make an accurate diagnosis of the mass. Pathological and intraoperative findings indicated a diagnosis of continuous SGF.

DISCUSSION: We observed two important clinical issues in this case. First, the potential for incidental diagnoses of SGF may be increasing. Second, to our knowledge, this is the first case report of a patient with SGF identified by TAPP. Such a therapeutic strategy for incidentally detected SGF has not been described; here we report a successful experience.

CONCLUSION: To our knowledge, this is the first report of a patient with SGF diagnosed by a TAPP procedure. The postoperative course was uneventful using our method.

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1. Introduction

Splenogonadal fusion (SGF) is a rare congenital anomaly that results from an abnormal connection between the primitive spleen and gonad during gestation. Presentation usually occurs as a scrotal mass discovered incidentally during orchiopexy or inguinal hernia repair [1]. To our knowledge, no case report of SGF from a laparoscopic transabdominal preperitoneal hernia repair (TAPP) has been reported. Here, we report the first case of SGF that was incidentally detected during a TAPP procedure.

2. Presentation of case

A 76-year-old man presented with a 10-year history of left inguinal swelling and a one-month history of occasional pain. Med-

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ical history was unremarkable except for an anterior approach hernia repair for a right inguinal hernia at the age of 64. Physical examination revealed left inguinal swelling, which was easy to reduce. Laboratory data on admission was unremarkable. The patient had no imaging tests except for plain film of the chest and abdomen, which showed no significant abnormalities.

Intraoperatively, a left direct inguinal hernia was observed, while the right was normal. Two reddish-purple masses (Fig. 1, red arrow; Fig. 2, blue arrow) were identified along a cord of linear tissue, which extended toward the spleen in the cranial direction (Fig. 1) and adhered strongly to the peritoneum, passing transversely through the left internal inguinal ring caudally (Figs. 2 and 3. The cord of linear tissue moved when the scrotum was pulled. One of the masses (Figs. 2 and 3, blue arrow) was located close to the left inguinal ring. It was oval-shaped and the length of the longest axis was approximately 10 mm. Checks revealed that the vas deferens and testicular arteriovenous vessels were normal.

To place the mesh and accurately diagnose the masses, we cut the cord of linear tissue and removed the mass located closest to the left inguinal ring, which was examined by a pathologist. The

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Fig. 1. A mass and cord of linear tissue observed by laparoscopy. The cord of linear tissue stretching in the cranial direction adhered to the peritoneum. At the top of the adhesion, the linear tissue was separated from the peritoneum and stretched into the omentum towards the spleen. A 5-mm laparoscopic port is shown inserted at the left lateral region at the same level as the umbilicus. A reddish-purple mass was observed along the cord (red arrow).

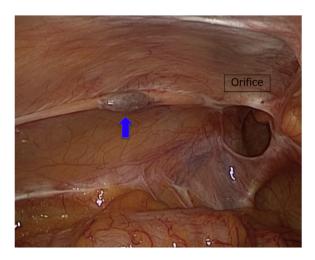


Fig. 2. A second mass observed along the cord of linear tissue by laparoscopy. The cord of linear tissue extended from the reddish-purple mass (blue arrow) to the left internal inguinal ring.

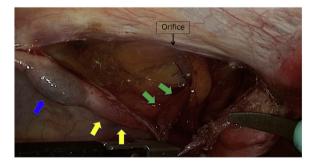


Fig. 3. A cord of linear tissue extends from the mass. The cord of linear tissue extending from the mass (blue arrow) adhered strongly to the peritoneum (yellow arrows), and passed transversely across the peritoneum and through the internal inguinal ring (green arrows).

operative specimen was a 12×8 mm diameter mass (Fig. 4). Histological examination showed that the mass was surrounded by a capsule and comprised red and white pulp, and splenic trabeculae (Fig. 5), characteristic components of splenic tissue. Thus, pathological diagnosis was splenic tissue. The cord contained fibrous tissue. Therefore, pathological, combined with intraoperative, findings confirmed the diagnosis of SGF.

The patient's postoperative course was uneventful, and he was discharged from hospital two days after surgery without any problems. At the 6-month follow-up, the patient had no problems and the groin was not enlarged.



Fig. 4. The operative specimen. The mass measured 12×8 mm in diameter.

3. Discussion

We observed two important clinical issues in this case. First, the incidence of SGF may be increasing. Second, to our knowledge, this is the first case report of a patient with SGF identified by TAPP.

The potential for incidental diagnoses of SGF may be increasing. Intraoperatively, it is easier to recognize SGF by TAPP than by open inguinal hernia repair due to the ease of identifying the abdominal linear and splenic tissues with laparoscopy. In contrast, identification of SGF is difficult by open repair in the absence of splenic tissue in the inguinal canal or in the case of direct hernia. Introduction of new operating techniques over the past decade has significantly increased the prevalence of endoscopic hernia surgeries. This may lead to an increased potential for incidental SGF diagnoses. Interestingly, more than 70% of reported SGF cases are in patients younger than 20 years, with approximately 50% being younger than 10 years of age [2]. Although the TAPP procedure is optimized for adults, the laparoscopic percutaneous extraperitoneal closure (LPEC) procedure, which is becoming a more prevalent surgery, is a laparoscopic procedure that has been optimized for younger patients.

To our knowledge, this is the first case report of a patient with SGF diagnosed by TAPP. Previously, there have been cases of preoperative, but not postoperative, diagnosis of inguinal hernia; in many of these, swellings observed in the groin were splenic tissue [1,3,4]. Moreover, a few studies have reported combined SGF and inguinal hernia. We identified six case reports [5–10] of open inguinal hernia repair in patients with SGF (Table 1); the first of these was by Daniel [5] in 1957. In all previous cases, as in our case, the mass was removed and diagnosed by pathology; we found no cases of preoperatively diagnosed SGF, which is difficult due to the rarity of this disease. It is therefore typically diagnosed incidentally during surgery [11]. As in the current study, the postoperative course was uneventful in all cases.

In 1956, Putschar and Manion [12] classified SGF into two types, continuous and discontinuous. The continuous type is characterized by the presence of a cord of ectopic or fibrous tissue that courses from the upper pole of the orthotopic spleen to the testis. In the discontinuous type, ectopic splenic tissue is attached to the gonad, but there is no connection to the orthotopic spleen. The spleen develops from a mass of mesenchymal cells located between the layers of the dorsal mesogastrium, while the gonad develops from the primitive genital ridge that lies just lateral to the mesogastrium. During the 5th week of gestation, the stomach is displaced to the left of the median plane and rotates around its axis. An insult

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