CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 35 (2017) 29–32

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

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

Yuichi Akama[a,c,*], Kimiyoshi Shimanuki[a], Shuji Asahi[a], Yohei Watanabe[a], Kazuhide Ko[a,c], Ryotaro Takano[a,c], Hodaka Amano[a], Takanori Kawaguchi[b], Eiji Uchida[c]

[a] Department of Surgery, Aizu Chuo Hospital, Fukushima, Japan
[b] Department of Pathology, Aizu Chuo Hospital, Fukushima, Japan
[c] Department of Gastrointestinal and Hepato-Biliary-Pancreatic Surgery, Nippon Medical School Hospital, Tokyo, Japan

ARTICLE INFO

Article history:
Received 6 March 2017
Received in revised form 29 March 2017
Accepted 3 April 2017
Available online 5 April 2017

Keywords:
Splenogonadal fusion
Laparoscopic transabdominal preperitoneal hernia repair
Congenital anomaly

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.

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. Medical 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

* Correspondence to: 1-1 Tsuruga-machi, Aizuwakamatsu-shi, Fukushima, 965-8611, Japan.
E-mail address: y-akama@nms.ac.jp (Y. Akama).

http://dx.doi.org/10.1016/j.ijscr.2017.04.004

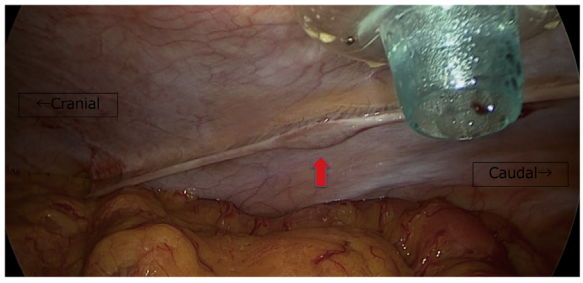

Fig. 1. A mass and cord of linear tissue observed by laparoscopy. The cord of linear tissue stretching in the cranial direction adhered to the peritoneum. At the top of the adhesion, the linear tissue was separated from the peritoneum and stretched into the omentum towards the spleen. A 5-mm laparoscopic 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).

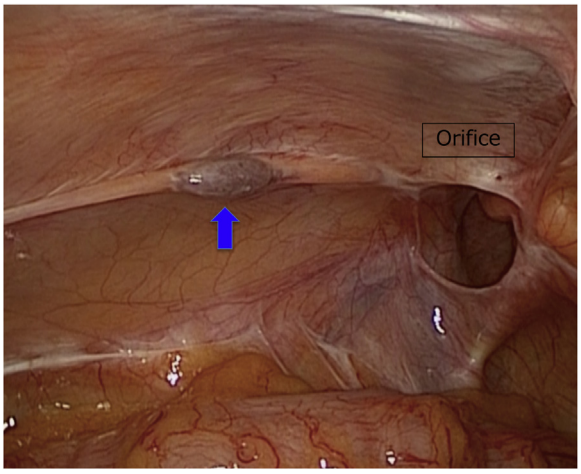

Fig. 2. A second mass observed along the cord of linear tissue by laparoscopy. The cord of linear tissue extended from the reddish-purple mass (blue arrow) to the left internal inguinal ring.

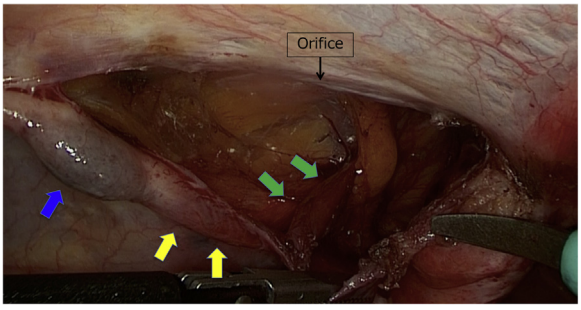

Fig. 3. A cord of linear tissue extends from the mass. The cord of linear tissue extending from the mass (blue arrow) adhered strongly to the peritoneum (yellow arrows), and passed transversely across the peritoneum and through the internal inguinal ring (green arrows).

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.

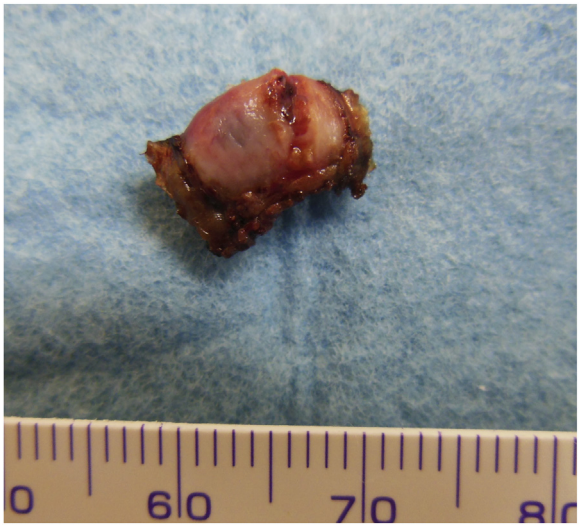

Fig. 4. The operative specimen. The mass measured 12 × 8 mm in diameter.

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

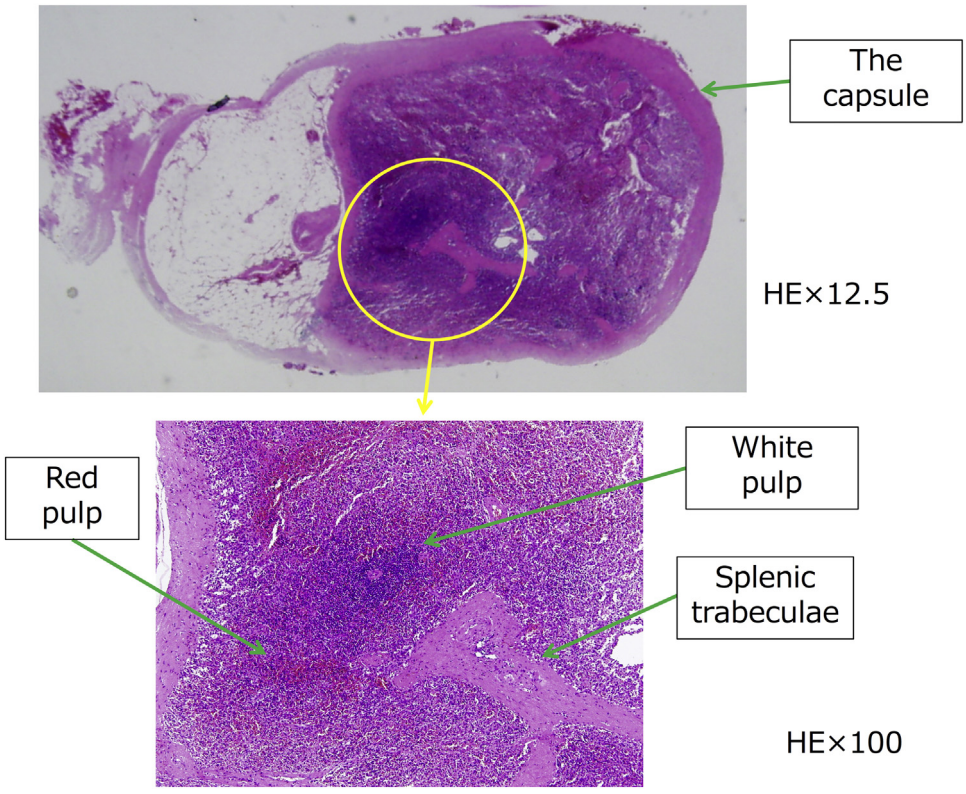

Fig. 5. Histological examination of the mass. Hematoxylin and eosin staining of the surgically obtained mass indicated that it was surrounded by a capsule and comprised red pulp, white pulp and splenic trabeculae. (A) ×12.5 magnification. (B) ×100 magnification.

Table 1
Overview of previously reported cases of SGF with inguinal hernia repair.

Author [Refs.]	Age, Sex	Side, Diagnosis	Surgery	Congenital disorder	Type (Continuous/ Discontinuous)	Postoperative course
Daniel [5]	26 yo, male	Left, indirect hernia	Bassini's procedure and orchiectomy Excision of tissue	Not described	Continuous	Uneventful
Sieber [6]	14 mo, male	Left, indirect hernia	Potts' procedure Excision of tissue	Mobius syndrome	Continuous	Uneventful
Nimkin [7]	5 yo, male	Left, indirect hernia	Hernia repair (unspecified) Excision of tissue	Not described	Continuous	Uneventful
Li [8]	7 yo, male	Left, not described	(unspecified) Excision of tissue	Not described	Continuous	Not described
Bosnali [9]	7 yo, male	Left, indirect hernia	Hernia repair (unspecified) Excision of tissue	None	Continuous	Uneventful
Babu [10]	21 yo, male	Left, indirect hernia	Hernia repair (unspecified) Excision of tissue	Not described	Continuous	Uneventful
Present case	76 yo, male	Left, direct hernia	TAPP Excision of tissue	None	Continuous	Uneventful

yo = years old; mo = months old; TAPP = laparoscopic transabdominal preperitoneal hernia repair.

occurring during this period can cause the surface of the developing genital ridge and the splenic anlage to fuse. Subsequent descent of the gonad in the 8th-10th weeks results in a concurrent descent of part of the spleen to result in the continuous form of SGF [13]. Although we did not observe direct contact between the spleen and the cord of linear tissue, the fact that this cord stretched into the omentum towards the spleen and moved when the scrotum was pulled suggests a diagnosis of continuous SGF. All previous cases have also reported diagnoses of continuous SGF; among these, 50% had additional congenital abnormalities, most commonly cryptorchidism [3]. Further, SGF is usually present on the left side (98%) and is most prevalent in males (95%) [14]. In the present study, the patient had no additional congenital anomalies.

We incidentally detected SGF by TAPP, which required cutting the cord of fibrous tissue for mesh placement. It is possible that changing the operative procedure to open inguinal repair, especially anterior approaches such as the Lichtenstein method, which do not require cutting of the cord for mesh placement, may have proved easier and more efficient. In any case, as with previous case reports [5–10], it was necessary to cut the cord to remove the mass for accurate diagnosis, since this could not be accomplished pre- or intraoperatively. Fortunately, the postoperative course was uneventful and we can report a successful experience; however,

additional studies with more cases are needed to determine the best way to treat incidentally detected SGF with inguinal hernia by TAPP.

4. Conclusion

We report a case of SGF which was incidentally detected by TAPP. To our knowledge, this is the first report of this condition. The patient made favorable progress up to the 6-month postoperative follow-up, with no evidence of recurrent hernia. Further studies and more cases are needed to optimize a therapeutic strategy for incidentally detected SGF with inguinal hernia by TAPP.

Conflict of interest

The authors declare no conflict of interest associated with this manuscript.

Source of funding

The authors received no funding for this paper.

Ethical approval

This is not a research study. Ethical approval is not required.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Authors contribution

Y. Akama, K. Shimanuki, S. Asahi, and Y. Watanabe performed the surgery and wrote the paper.

K. Ko, R. Takano, and H. Amano cared for the patient and generated the figures and table.

T. Kawaguchi pathologically diagnosed the specimen.

E. Uchida contributed to manuscript revision and had final approval of the article.

All authors discussed the results and commented on the manuscript.

Registration of research studies

This report is not research study.

Guarantor

Yuichi Akama.

References

[1] A.M. Carragher, One hundred years of splenogonadal fusion, Urology 35 (1990) 471–475.
[2] X.C. Shen, C.J. Du, J.M. Chen, Z.W. Zhang, Y.Q. Qiu, Splenogonadal fusion, Chin. Med. J. 121 (2008) 383–384.
[3] H.C. Irkilata, E. Aydur, I. Yildirim, Y. Kibar, M. Dayanc, A.F. Peker, Splenogonadal fusion in adults: presentation of three cases and review of the literature, Urol. Int. 81 (2008) 360–363.
[4] P.M. Lakshmanan, A.K. Reddy, A. Nutakki, A surprising content of congenital hernia: complete splenogonadal fusion band, BMJ Case Rep. (2014), http://dx.doi.org/10.1136/bcr-2014-203640.
[5] D.S. Daniel, An unusual case of ectopic splenic tissue resembling a third testicle, Ann. Surg. 145 (1957) 960–962.
[6] W.K. Sieber, Splenotesticular cord (splenogonadal fusion) associated with inguinal hernia, J. Pediatr. Surg. 4 (1969) 208–210.
[7] K. Nimkin, P.K. Kleinman, J.S. Chappell, Abdominal ultrasonography of splenogonadal fusion, J. Ultrasound Med. 19 (2000) 345–347.
[8] W.F. Li, M.X. Luan, Z. Ma, Y.J. Chen, Splenogonadal fusion: report of four cases and review of the literature, Exp. Ther. Med. 6 (2013) 816–818.
[9] O. Bosnali, I. Cici, S. Moralioglu, A. Cerrah-Celayir, Continuous-type splenogonadal fusion: report of a rare case, Turk. J. Pediatr. 56 (2014) 680–683.
[10] N.V. Babu, P.B. Kumar, Rare content in congenital inguinal hernia: splenogonadal fusion band, Int. Surg. J. 3 (2016) 394–396.
[11] K. Bal, M. Ermete, U. Balci, C. Dincel, Splenogonadal fusion: a very rare congenital anomaly in the differential diagnosis of a testicular mass, Turk. J. Urol. 40 (2014) 62–64.
[12] W.G. Putschar, W.C. Manion, Splenicgonadal fusion, Am. J. Pathol. 32 (1956) 15–33.
[13] D.R. Varma, G.R. Sirineni, M.V. Rao, K.M. Pottala, B.V. Mallipudi, Sonographic and CT features of splenogonadal fusion, Pediatr. Radiol. 37 (2007) 916–919.
[14] G. Alivizatos, A. Skolarikos, O. Sopilidis, N. Ferakis, M. Chorti, Case report splenogonadal fusion: report of a case and review of the literature, Int. J. Urol. 12 (2005) 90–92.