症 例 報 告

島根県中病医誌 48
45頁 ～ 49頁

Adhesive small bowel obstruction caused by intraabdominal testis associated with a tunica albuginea cyst: A case report

Kazuya OKAMURA[1)] Aiko GOBARA[1)] Emi ISHIBASHI[1)]
Kazumi ARAKI[1)] Yoshiko KANASAKI[1)] Koji YUASA[1)]
Koji KODAMA[1)] Akinobu KANAZAWA[2)] and Hideyuki ONUMA[3)]

Abstract：Diagnosing intraabdominal testis is important because it can cause small bowel obstruction (SBO). There are reports of SBO caused by malignant tumors in the intraabdominal testis, but only a few studies report SBO caused by factors other than malignant tumors. Herein, we report a case of intraabdominal testis complicated with a tunica albuginea cyst, which caused adhesive SBO due to abdominal trauma. Intraabdominal testis can cause recurrent SBO if not surgically removed. In our case, the SBO recurred a month after adhesiotomy was performed, and intraabdominal testis was removed. The characteristic imaging findings of intraabdominal testis should be known and a correct preoperative diagnosis should be made.

Key words：Small bowel obstruction (SBO), intraabdominal testis, adhesive, cyst, male

精巣白膜嚢胞を伴う腹腔内停留精巣による癒着性小腸閉塞の一例

岡村　和弥[1)]　河原　愛子[1)]　石橋　恵美[1)]　荒木　和美[1)]　金崎　佳子[1)]
湯浅　貢司[1)]　児玉　光史[1)]　金澤　旭宣[2)]　大沼　秀行[3)]

概　　要：腹腔内停留精巣は小腸閉塞を引き起こす可能性があり，診断することが重要である．腹腔内停留精巣に発生した悪性腫瘍により引き起こされる小腸閉塞の報告はあるが，悪性腫瘍以外の要因で引き起こされる小腸閉塞を報告する研究はわずかである．今回我々は，腹部外傷により癒着性小腸閉塞を引き起こした精巣白膜嚢胞を合併した腹腔内停留精巣の症例を報告する．腹腔内停留精巣は外科的に切除しない場合には再発性の腸閉塞を引き起こすことがある．我々の症例でも腸閉塞の癒着解除術の１か月後に腸閉塞を再発し，腹腔内停留精巣の切除術が施行されている．腹腔内停留精巣の特徴的な画像所見を把握し，正確な術前診断を行う必要がある．

索引用語：腸閉塞，腹腔内停留精巣，癒着，嚢胞，男性

Introduction

An undescended testis, also known as cryptorchidism, can be classified into prescrotal, intracanalicular, and intraabdominal types according to their location. Among these, intraabdominal testis is the rarest and can cause

１）島根県立中央病院　放射線科
２）島根県立中央病院　消化器外科
３）島根県立中央病院　病理組織診断科

１）Shimane Prefectural Central Hospital, Department of Radiology
２）Shimane Prefectural Central Hospital, Department of Gastrointestinal Surgery
３）Shimane Prefectural Central Hospital, Department of Diagnostic Pathology

small bowel obstruction（SBO）or recurrent SBO even without a malignant tumor, therefore it is necessary to determine characteristic imaging findings[1].

Intraabdominal testis may be associated with a tunica albuginea cyst in rare cases[2]. A tunica albuginea cyst is a benign extratesticular cyst that develops in the albuginea of the testis.

Herein, we report a rare case of SBO in which a tunica albuginea cyst associated with intraabdominal testis ruptured due to abdominal trauma, causing adhesive SBO.

Case report

A 74-year-old male patient presented with abdominal pain and vomiting. SBO was suspected by plain computed tomography（CT）at another hospital. Contrast-enhanced CT was performed in our hospital, and adhesive SBO was suspected（Fig. 1）. He had no history of surgery, but had an abdominal trauma. A solid mass with calcification and cyst was found near SBO, and the mass and small bowel were in proximity（Fig. 1）. Hence, SBO was assumed to be caused by adhesion. CT showed no left testis（Fig. 2A）, and the solid part of the intraabdominal mass had a similar shape and contrast effect as the right testis（Fig. 2B）. Additionally, since the vein originated from the mass（Fig. 3）and flowed into the left renal vein, it was considered as the left testicular vein. Therefore, the patient was diagnosed with adhesive SBO caused by an intraabdominal testis. Adhesiotomy was performed,

Fig. 1
Contrast-enhanced computed tomography（CT）image（coronal image, early phase）.
Contrast-enhanced CT revealed a distended small bowel and a solid mass with calcification and a cyst at the margin（arrow）.

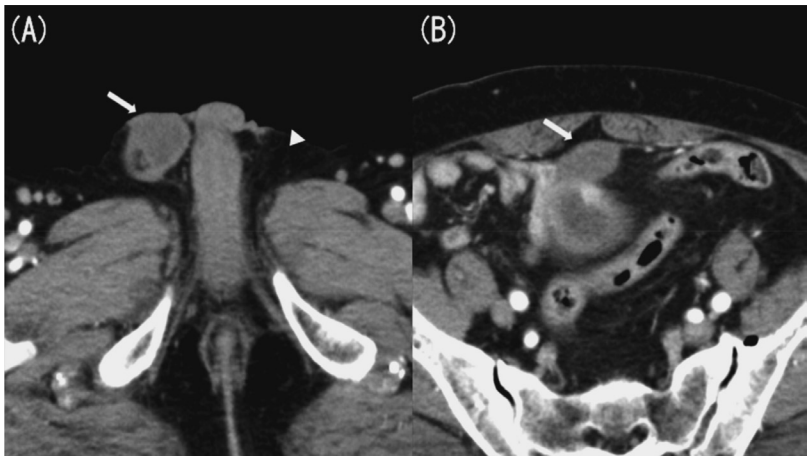

Fig. 2
A: Contrast-enhanced computed tomography（CT）image（axial image, early phase）at the left inguinal region level.
B: Contrast-enhanced CT image（axial image, early phase）at the mass level.
CT showed no left testis in the inguinal region, and the solid part of the mass had the same morphology and enhancement effect as the right testis（A, B, arrow）. The spermatic cord was absent in the left inguinal canal（A, arrowhead）.

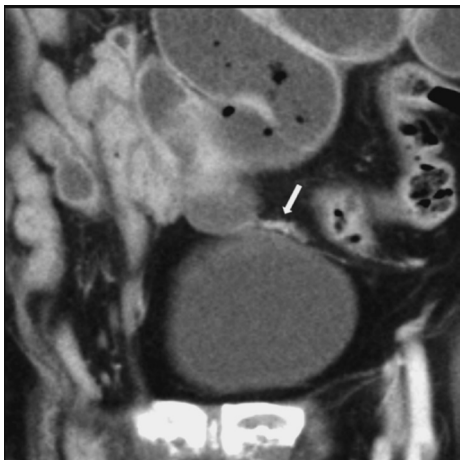

Fig. 3
Contrast-enhanced computed tomography（CT）image（coronal image, late phase）.
CT showed that the testicular vein originating from the intraabdominal mass（arrow）.

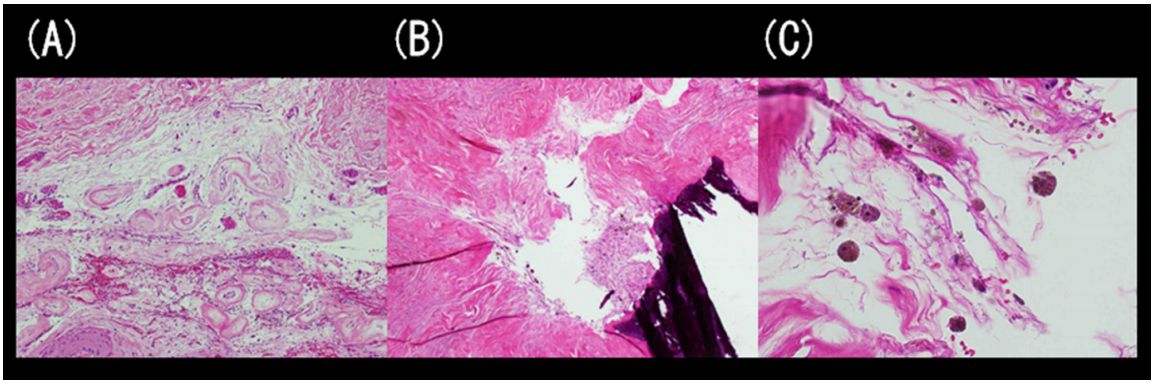

Fig. 4
Pathological examination:
A: Intraabdominal testis in hematoxylin and eosin stain with low magnification.
B: A tunica albuginea cyst in hematoxylin and eosin stain with low magnification.
C: Hemosiderin-phagocytic macrophages in hematoxylin and eosin stain with high magnification.
The pathological examination revealed testicular tissue within the mass (A). The mass was associated with a tunica albuginea cyst (B), and the cyst contained hemosiderin-phagocytic macrophages (C).

but the intraabdominal testis was not resected. A month later, SBO recurred. Reoperation was performed, and the mass was resected together with an adherent small bowel. Pathological diagnosis revealed an intraabdominal testis with a tunica albuginea cyst (Fig. 4A, 4B), with no malignant tumor. Calcification was observed in the tunica albuginea cyst, and hemosiderin-phagocytic macrophages were observed within the cyst (Fig. 4C), suggesting the possibility of a previously ruptured cyst. Abdominal trauma was assumed to cause leakage of the tunica albuginea cyst contents, leading to inflammation and adhesions. Thereafter, adhesive SBO recurred but was relieved in a short period by conservative treatment with an indwelling ileus tube. No recurrence of SBO has occurred in the subsequent 2-year follow-up.

Discussion

The testicles descend from the abdominal cavity to the scrotum during fetal life in males and are positioned within the scrotum at birth. However, it may remain on a descending path and is called an undescended testis. The incidence of undescended testis is 2% – 4% at birth and decreases to 0.8% by 1 year of age[3]. Genetic, maternal, and environmental factors are thought to cause undescended testis, but the exact cause is unknown[4]. Additionally, low birth weight and premature babies are considered at risk[4]. The undescended testis is classified into prescrotal, intracanalicular, and intraabdominal types, with the intraabdominal type being the rarest. Undescended testis usually requires surgery because it is predisposed to trauma, torsion, infertility, and malignancy[5]. The risk of developing testicular cancer is 4–8 times higher in undescended testis, and several studies report teratoma before puberty and seminoma after puberty[6,7].

Intraabdominal testis has a higher incidence of tumors, and cases of bowel obstruction caused by malignant tumor infiltration or mechanical exclusion have been reported[8,9]. However, few studies report bowel obstruction caused by factors other than malignant tumors. Only a few studies report adhesions, adhesion-associated internal hernia, undescended testis torsion, and enlargement-induced mechanical retraction[1,10,11,12]. No previous studies report adhesive SBO triggered by abdominal trauma, similar to this case.

We diagnosed adhesive SBO caused by an intraabdominal testis by CT. We confirmed that the solid mass in the abdominal cavity had a similar shape and contrast effect to the testis, and that the testis did not exist in the left inguinal region. Additionally, preoperative diagnosis is possible by confirming dilated testicular arteries and veins, and a lack of spermatic cord in the inguinal canal by CT if a testicular tumor develops in an intraabdominal testis[13,14,15]. The testicular vein originated from the mass and flowed into the left renal vein in this

case, and the spermatic cord was absent in the left inguinal canal. Therefore, intraabdominal testis can be diagnosed even in cases without a tumor. However, making an imaging diagnosis for intraabdominal testis may be difficult if intraabdominal testis is not recognized to be included in the differential diagnosis of intraabdominal masses in males. The intraabdominal testis can be correctly diagnosed based on this recognition if the characteristic imaging findings are confirmed as described above.

Moreover, intraabdominal testis has been reported to cause recurrent intestinal obstruction and is likely to have recurrent intestinal obstruction episodes if not surgically resected[1)]. Surgical removal is desirable if the surgeon finds that the intraabdominal testis causes SBO during surgery[1)]. In our case, adhesiotomy was also performed, but the intraabdominal testis was not resected, and intestinal obstruction recurred; thus, reoperation was performed. A radiologist must correctly diagnose SBO caused by an intraabdominal testis and recommend surgical resection.

Furthermore, the intraabdominal testis in our case was associated with a tunica albuginea cyst. A tunica albuginea cyst is a benign extratesticular cyst that develops in the tunica albuginea of the testis. It has uniform margin and interior and is 1–2 cm in size, rarely exceeding 4 cm[16)]. It may be unilocular or multilocular[17)]. It was previously presumed to be related to trauma and inflammation, but now the theory of congenital occurrence is dominant[16)]. It can be diagnosed by the cyst location in the scrotum by ultrasound or magnetic resonance imaging although it may need to be differentiated from a simple testicular cyst[16)]. A unilocular cystic mass of 1.5 cm in diameter was found outside the testis in our case（Fig. 5A, 5B）. The large-size mass may be differentiated by CT. There have been no reports of imaging findings that clearly show their positional relationship although studies report the association between tunica albuginea cyst and intraabdominal testis. Ultrasound is used for diagnosis in the scrotum, but the diagnosis may be difficult if it is associated with an intraabdominal testis; thus, it is useful to confirm extratesticular cyst by CT.

This case was an extremely rare case in which a tunica albuginea cyst ruptured due to abdominal trauma. When a tunica albuginea cyst ruptures, the cyst collapses and surrounding inflammatory changes are seen on imaging findings. However, no such change was observed in this case. If some time has passed after rupture, imaging findings that suggest rupture may not be obtained. In addition, there are no study reports of adhesive SBO caused by intraabdominal testis triggered by abdominal trauma. Therefore, if an intraabdominal testis is not complicated with a tunica albuginea cyst, it is unlikely that abdominal trauma causes SBO. However, an intraabdominal testis can cause SBO even without abdominal trauma, so it is necessary to accurately diagnose it.

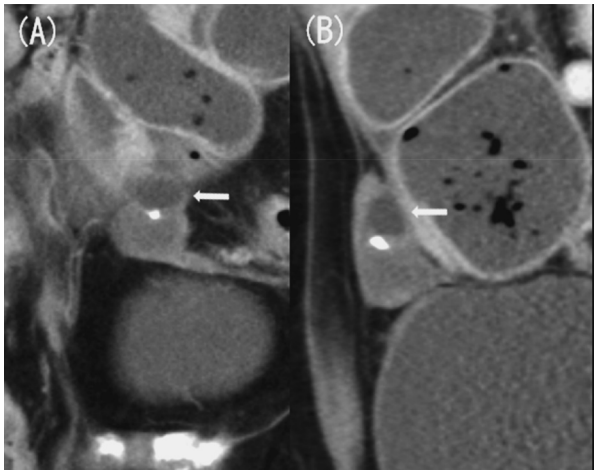

Fig. 5
A: Contrast-enhanced computed tomography（CT）image（coronal image, late phase）.
B: Contrast-enhanced CT image（sagittal image, late phase）.
A unilocular cystic mass was found outside the testis（arrow）.

In conclusion, an intraabdominal testis associated with a tunica albuginea cyst causes an adhesive SBO due to abdominal trauma in this case. It is critical to diagnose SBO correctly caused by an intraabdominal testis based on typical imaging findings.

References

1）Satoskar S, Kashyap S, Ziehm J, et al.: Cryptorchidism as an obscure cause of adhesive small bowel obstruction in an adult, a case report. Int J Surg Case Rep, 2021; 86: 106319.
2）Iwata K, Ito A, Hirabayashi N: Cyst of the tunica

albuginea associated with intraabdominal testes: a case report. Acta Urol Jpn, 1997; 43(12): 891-894.

3) Hutson JM, Balic A, Nation T, et al.: Cryptorchidism. Semin Pediatr Surg, 2010; 19(3): 215-224.

4) Kurahashi N, Kishi R: A review of epidemiological studies about the incidence and etiological factors of cryptorchidism relevance to endocrine-disrupting chemicals. Nippon Eiseigaku Zasshi (Jpn J Hyg), 2003; 57(4): 636-644.

5) Nik H, Kishen R, Azuddin MK: A rare cause of mechanical obstruction: a case report of cryptoorchidsm in adults. Int Surg J, 2020; 7(11): 3790-3791.

6) Manecksha RP, Fitzpatrick JM: Epidemiology of testicular cancer. BJU Int, 2009; 104(9b): 1329-1333.

7) Gupta A, Kureel SN, Bhalla S, et al.: Yolk sac tumor in intraabdominal undescended testis in a child with normal serum alpha-fetoprotein: a rare case report. Afr J Urol, 2020; 26(1): 1-5.

8) Ciftci F, Benek S, Kezer C, et al.: A rare cause of mechanical intestinal obstruction: a neoplastic intra-abdominal testis. Int Surg, 2016; 101(3-4): 167-170.

9) Takemura S, Yui S: Seminoma originating in an undescended testis in the abdominal space presenting as ileus: a case report. J Jpn Soc Coloproctol, 2012; 65(1): 16-21.

10) Bassiouny IE, Abbas TO, Alansari AN, et al.: Unique presentation of intra-abdominal testis: small bowel obstruction. ISRN Urol, 2011; Artile ID 579153: 3 pages.

11) Kim CW, Min GE, Lee SH: Small bowel obstruction caused by cryptorchidism in an adult. Ann Surg Treat Res, 2017; 93(5): 281-283.

12) Sangwan MK, Sangwan V, Garg MK, et al.: The torsed intraabdominal testis presenting with a mechanical large intestinal obstruction-A maiden case report. IJCR, 2019; 4(3): 82.

13) Altinkilic B, Lommel D, Franke FE, et al.: Seminoma in an intra-abdominal testis was misdiagnosed as a kidney tumor and associated with ipsilateral renal agenesis. Scand J Urol Nephrol, 2003; 37(2): 181-183.

14) Karcaaltincaba M, Kaya D, Ozkan OS, et al.: Preoperative multidetector computed tomography diagnosis of a seminoma originating from an undescended testis by "testicular vascular pedicle" sign. J Comput Assist Tomogr, 2006; 30(5): 794-795.

15) Jeong MJ, Kim SH, Kim JY, et al.: Radiological clues for diagnosing intra-abdominal seminoma in undescended testis on computed tomography. Radiol Case Rep, 2022; 17(6): 2273-2276.

16) Matsumura N, Sugimoto K, Hayashi T, et al.: Cyst of the tunica albuginea testis: a case report. Acta Urol Jpn, 2015; 61(2): 71-74.

17) Alvarez DM, Bhatt S, Dogra VS: Sonographic spectrum of tunica albuginea cyst. J Clin Imaging Sci, 2011; 1: 5.

(受付日：2023年10月11日，掲載決定日：2023年12月15日)