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An inguinal hernia with cryptorchidism with a Leydig cell tumor in an elderly man: A case report



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ABSTRACT

INTRODUCTION: Cryptorchidism is common in children but is rare in the elderly. It often presents with a constellation of signs and symptoms similar to routine inguinal hernias. We present the case of an elderly man with cryptorchidism containing a Leydig cell tumor and provide clinical insights.

PRESENTATION OF CASE: An 84-year old man was admitted with an incarcerated right lower quadrant hernia. Both testes were absent on palpation of the scrotum. After reduction of the hernia, computed tomography scan revealed a round lesion in the hernia sac, which was suspected to be the ectopic testis. Laparoscopic exploration was performed in combination with an open anterior approach. The hernia orifice was the right internal inguinal ring, and the inguinal canal was obliterated by adhesions because the spermatic cord did not pass through it. The ectopic testis was resected with the hernia sac, and the hernia repaired with a KUGEL™ patch (Bard, USA).

DISCUSSION: Laparoscopic exploration was useful to delineate the anatomy of this unusual inguinal hernia. The open anterior approach was necessary to dissect the ectopic testis and the hernia sac. Pathological findings revealed tumor cells with clear cytoplasm in the resected testis, diagnosed as a Leydig cell tumor. **CONCLUSION:** The combination of laparoscopic and anterior approaches facilitated the surgical treatment of an unusual inguinal hernia with cryptorchidism. The resected ectopic testis should undergo thorough histopathologic examination.

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1. Introduction

Cryptorchidism is common in childhood and often presents simultaneously with an inguinal hernia. However, it is rare to be initially diagnosed in the elderly, because cryptorchidism is usually diagnosed and treated at a young age. We present an elderly patient with an unusual inguinal hernia with cryptorchidism, treated with a combined laparoscopic and open anterior approach. The resected specimen contained a Leydig cell tumor in the resected ectopic testis. This work has been reported in conformity with the SCARE criteria [1].

2. Presentation of case

An 84-year-old man presented with repeated episodes of vomiting and abdominal pain. Physical examination showed an incarcerated hernia in the right lower quadrant. The hernia was easily reduced under mild sedation. The hernia orifice was palpable in the right lower abdomen. The external inguinal ring was not dilated, and both testes were not palpable in the scrotum. The patient had been diagnosed with “bilateral testicular deficiency” in childhood and had received no further examination or treatment. Preoperative blood tests showed no abnormalities.

Computed tomography (CT) scan of the abdomen after hernia reduction revealed that the hernia sac was located in the right lower quadrant and extended outside of abdominal musculature superiorly. The hernia orifice seemed to be located at the inguinal ring, but the anatomic details remained unclear. The hernia sac contained a round mass, thought to be an ectopic testis (Fig. 1). The patient complained of intermittent abdominal pain, and was considered to be at risk of recurrent incarceration. It was considered that the mass thought to be an ectopic testis should be resected along with

Abbreviations: CT, computed tomography; TEP, totally extraperitoneal; TAP, transabdominal preperitoneal.

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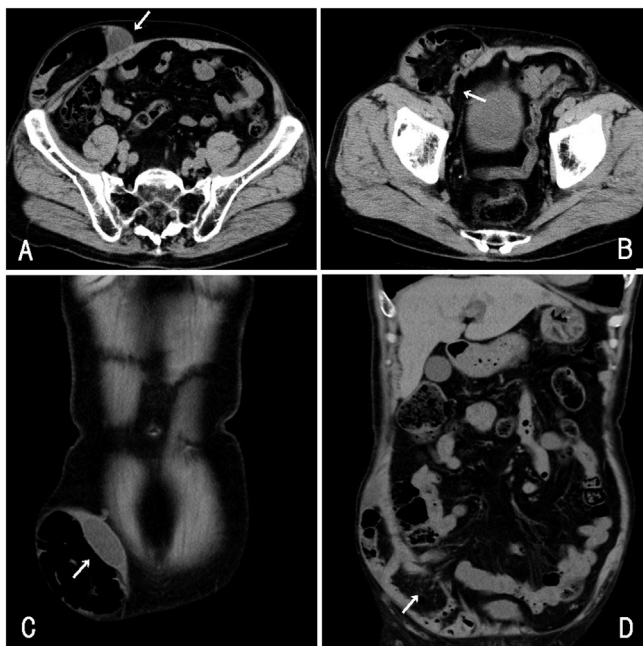


Fig. 1. Computed Tomography (CT) scan findings.
 A. The hernia sac extends to the subcutaneous layer on the external oblique aponeurosis, and contained a round mass (arrow).
 B. The hernia protrudes from the lateral side of the right epigastric vessels (arrow).
 C. The round cystic mass is positioned superiorly in the hernia sac (arrow).
 D. The hernia orifice is seen at the right internal inguinal ring (arrow).

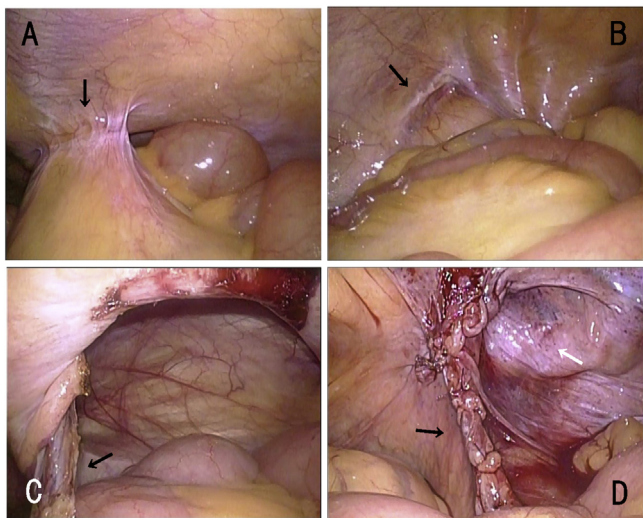


Fig. 2. Laparoscopic findings.
 A. The mesentery of the small intestine is adherent (arrow) to the hernia orifice.
 B. There is no hernia in the left internal inguinal ring (arrow).
 C. After dissection of the adhesions, a large hernia orifice was found lateral to the right inferior epigastric vessels (arrow).
 D. After hernia repair, the peritoneum was sutured next to the right epigastric vessels (black arrow) following resection of the hernia sac. The hernia orifice was closed (white arrow) with a Kugel patch which was laid onto the pre-peritoneal space.

a hernia repair. Hernia repair was then performed under general anesthesia.

A laparoscope was initially inserted through the umbilicus, and the hernia was evaluated intraperitoneally. The hernia orifice was found at the right internal inguinal ring, and was diagnosed as an inguinal hernia. Adhesions from the mesentery of the small intestine to the area alongside the internal inguinal ring were seen (Fig. 2A). The right inguinal canal was obliterated by adhesions

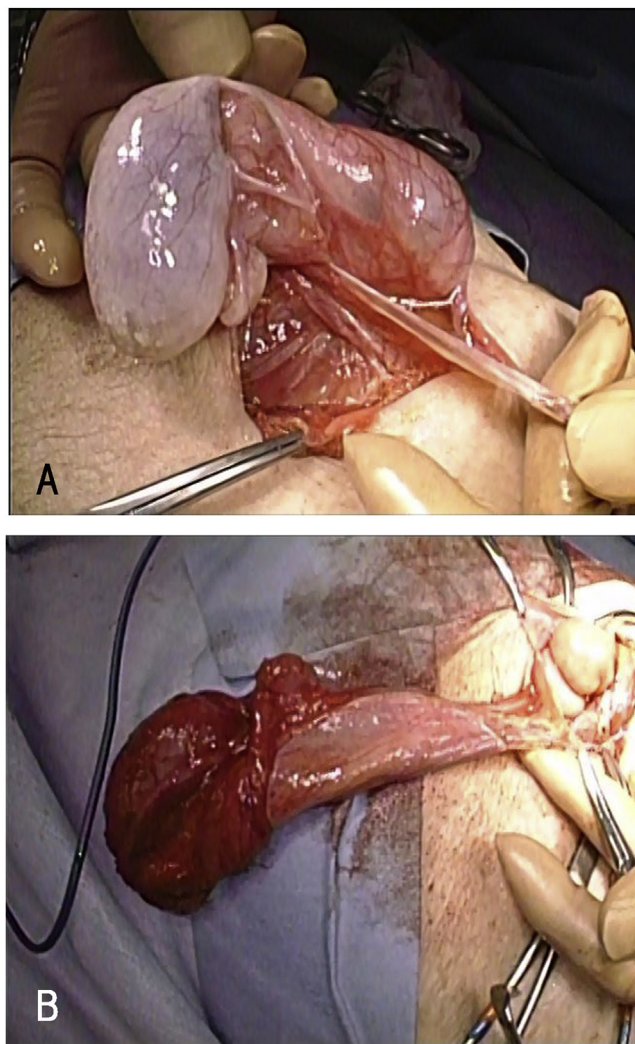


Fig. 3. The ectopic testis.
 A. The ectopic testis was found inside of the cystic lesion in the distal portion of the hernia sac.
 B. The spermatic cord is connected to the atrophied testis.

because the spermatic cord was not passing through it, and the hernia sac extended outside of the abdominal wall musculature superiorly. The ectopic testis could not be seen in the hernia sac from the intraperitoneal view. There was no hernia at the left internal inguinal ring (Fig. 2B), and the left testis was not seen in the abdominal cavity.

A 5 mm port was inserted in the right lower quadrant and adhesions around the right inferior epigastric vessels were divided (Fig. 2C). It was difficult to resect the hernia sac with the ectopic testis laparoscopically, and a 6 cm incision was made above the inguinal ligament transversely along a skin fold. The hernia sac which extended to the subcutaneous layer on the external oblique aponeurosis, was dissected. The ectopic testis was found inside the cystic lesion (likely a hydrocele) in the distal portion of the hernia sac (Fig. 3A), with attached spermatic cord. This finding established the diagnosis of an inguinal hernia with cryptorchidism. The ectopic testis was atrophied (Fig. 3B), and resected with the hernia sac after ligation of the spermatic cord. The resected specimen was sent for histopathological analysis. The hernia was repaired with a (10 × 15 cm) KUGEL™ patch (Bard, USA) which was laid on the pre-peritoneal space.

Pathologic findings revealed atrophic and hyalinized seminiferous tubules with a thickened basement membrane (Fig. 4A). Germ

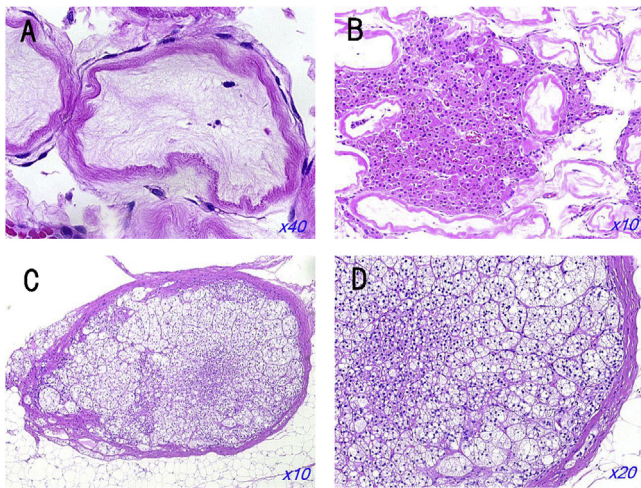


Fig. 4. Pathological findings.

- A. Atrophic and hyalinized seminiferous tubules with thickened basement membranes are seen, without germ cells ($\times 40$).
 B. Hypertrophy of Leydig cells ($\times 10$).
 C. The lesion has small, round nucleoli with clear vacuolated polygonal cytoplasm, diagnosed as a Leydig cell tumor ($\times 10$).
 D. Clear, vacuolated cytoplasm is caused by abundant intracytoplasmic lipid, and reminiscent of that seen in tumors of the adrenal cortex ($\times 20$).

cells were completely deficient, and Leydig cells were prominent and hypertrophied (Fig. 4B). In other areas, large, polygonal cells with round nuclei and clear cytoplasm were seen, diagnosed as a Leydig cell tumor (Fig. 4C, D). Other findings of malignancy, such as pronounced nuclear, cellular polymorphism, and abnormal mitoses, were not seen. The epididymis and the spermatic cord were normal.

3. Discussion

Review of this patient's management provides two important clinical insights. First, the combination of a laparoscopic and anterior approach was helpful for the surgical management of this inguinal hernia with cryptorchidism. The laparoscopic approach was useful to delineate the unusual anatomy of this hernia. It was difficult to resect hernia sac with the ectopic testis laparoscopically, and an open anterior approach was required. Second, the resected specimen contained a Leydig cell tumor in the atrophied testis, which underscores the need for a complete histopathologic examination of the resected ectopic testis.

An interparietal hernia was considered in the differential diagnosis of this patient. These are classified into three types according to the anatomical position of the hernia sac; preperitoneal, interstitial, and superficial hernia [2]. The incidence of interparietal hernia is from 0.08% to 1.6% [2], and a relationship with the etiology of cryptorchidism is presumed [3]. This patient's hernia sac was located above the external oblique fascia and was similar to a superficial hernia [4]. However, the inguinal canal was not opened and the hernia sac extended superiorly. The hernia did not pass through the inguinal canal.

In addition, the combination of a Spigelian hernia and cryptorchidism has been reported, based on embryology [5]. The gubernaculum plays a key role in the descent of the testis [6,7], and malposition of attachments to the abdominal wall may affect the development of a Spigelian hernia with cryptorchidism [5]. This hypothesis may be applicable in this patient. A Spigelian hernia was excluded because of the location of the hernia [8,9], which develops in the anterolateral area of the abdomen between the anterior superior iliac spine and the umbilicus in proximity to the external

margin of the rectus abdominis muscle [8]. It is defined as a hernia through the Spigelian fascia as the part of the transversus abdominis aponeurosis extending from the semilunar line to the lateral edge of rectus abdominis [9].

Laparoscopic hernia repair is commonly used and includes totally extraperitoneal (TEP) and transabdominal preperitoneal (TAP) repairs. We usually perform the TEP repair for inguinal hernia, but we selected the TAP approach for this patient, because of the anatomical abnormality and the position of the ectopic testis being unclear on the preoperative CT scan. We felt that there could be difficulties with the TEP procedure. Indeed, the TAP approach was useful to delineate the unusual anatomy of this hernia, and laparoscopic orchiectomy is indicated in patients with an intra-abdominal testis [10]. In the present patient, the ectopic testis could not be identified laparoscopically and an additional open anterior approach was required to resect the hernia sac with the ectopic testis.

Approximately 7% of pediatric patients with inguinal hernia also have cryptorchidism, and inguinal hernia occurs in 90% of patients with cryptorchidism [11]. Cryptorchidism is a common congenital condition that is usually diagnosed and treated during childhood. It is subsequently an uncommon diagnosis in adults and is quite rare in elderly patients [10]. Cryptorchidism is known to be associated with neoplasms in the undescended testes and orchiectomy is recommended in adult patients. The undescended testis develops cancer 35–48 times more than a normally testis, and tumors tend to occur in middle-aged men [12]. The most common type of testicular tumor occurring in a cryptorchid testis is seminoma, but other germ cell tumors also occur [13].

Leydig cell tumors are common neoplasms arising from gonadal stroma, accounting for 1–3% of all testicular tumors in adults [14,15]. The Leydig cells are prominent in cryptorchid testes, and several reports in the literature indicate that cryptorchidism may predispose to the development of a Leydig cell tumor [15]. However, Leydig cell tumors with cryptorchidism are rare compared to other testicular tumors, such as germ cell tumors [13]. Microscopic examination of Leydig cell tumors have a variety of appearances. A diffuse, sheet-like pattern of cells is most common, although small nests, ribbons, and cords also may be present in a fibrous stroma [15]. Rarely, the tumor cells are spindle and arranged in fascicles [16]. Several cases were reported with clear cytoplasm, reminiscent of that seen in tumors of the adrenal cortex, as observed in this patient (Fig. 4C, D) [15]. Clear and vacuolated cytoplasm is caused by abundant intracytoplasmic lipid in the tumor, and microcystic formation can be present [17]. About 10% of Leydig cell tumors have malignant potential [15,18]. The diagnosis of a malignant Leydig cell tumor is not always easy, because no definite histological criteria exist to define malignancy. Immunohistochemical staining (Ki-67, p53, Bcl2) may be useful for the diagnosis of malignant Leydig cell tumors [18].

4. Conclusion

A combined laparoscopic and anterior approach facilitated surgical repair of this unusual inguinal hernia with cryptorchidism. The laparoscopic approach was useful to delineate the anatomical abnormality, but an additional open anterior approach was required to reliably correct the complex anatomy in this unusual hernia. The ectopic testis should be resected with the hernia repair, and the resected specimen must be thoroughly examined for the presence of tumors.

Conflicts of interest

The authors declare that they have no competing interests.

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Ethical approval

This paper is not a research study, so I assume we do not need the ethical approval.

Consent

Written informed consent was obtained from the patients or relatives for publication of this case report and any accompanying images.

Author contribution

TZ performed surgery, wrote the paper, made literature review, and drafted the manuscript. JO advised the management of this patient as an expert surgeon. TK and YM treated the patient and assisted surgery. NH advised pathological findings as an expert pathologist. AL reviewed as a native speaker, and revised the manuscript.

Guarantor

The manuscript has been read and approved by all of the authors and is not under consideration for publication elsewhere. Dr. Ohki who is the director of Yuki Hospital, is the Guarantor.

Availability of data and materials

The dataset supporting the conclusions of this article is not included within any repository.

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