A rare variant of inguinal hernia: Cryptorchid testis at the age of 50 years. Etiopathogenicity, prognosis and management

Radwan Kassir a,⁎, Joelle Dubois a, Sid-Ali Berremila b, Sylviane Baccot a, Alexia Boueiil-Bourlier a, Olivier Tiffet a

a Department of General Surgery, CHU Hospital, Jean Monnet University, Saint Etienne, France
b Department of Pathology, CHU Hospital, Jean Monnet University, Saint Etienne, France

ARTICLE INFO

INTRODUCTION: Cryptorchidism is characterized by the extra-scrotal position of the testis. The surgical community has little to no knowledge of cryptorchid testis in adults apart from of pediatric surgeons. Therefore, we sought to describe this unusual cause of inguinal hernia.

PRESENTATION OF CASE: A 50-year-old man was referred with an inguinal hernia. Diagnosis of cryptorchidism was made during surgery, as the patient underwent an operation for repair of his left inguinal hernia. The testicle was non-viable and a left testicle was resected. Histopathology report confirmed a atrophic testis without testicular germ cell tumor (TGCT).

DISCUSSION: This is an extremely rare case of cryptorchidism revealed in an adult. The patient remained asymptomatic for 50 years. Most studies have concluded that there is a direct correlation between how long the testis was subjected to a cryptorchid position and TGCT incidence. The recommended age of surgical correction is before the age of 2 years. In our case, we did not find correlation between the time of surgery and risk of TGCT. Histopathology report confirmed the presence of leydig cells, seminiferous tubule and Sertoli cells without TGCT. Very little is known about link between cryptorchidism and TGCT.

The correct diagnosis of inguinal hernia is usually made during an inguinal hernia repair.

CONCLUSION: The surgeon must always be alert to the possibility of cryptorchid testis during a surgical exploration of an inguinal hernia. In suspected cases, laparoscopy ultrasonographic, CT scan and laparoscopy evaluation may be helpful in diagnosing of this atypical inguinal hernia before surgery.

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1. Introduction

Although inguinal hernia is frequent, the cryptorchid testis at the age of 50 years is an uncommon entity. The cryptorchid testis is characterized by the extra-scrotal position of the testis. This case demonstrates the utility to understand the surgical anatomy of inguinal hernias. We wish to present this extremely rare case of inguinal hernia, which is the first one that we have encountered in our practice, along with the accompanying prognosis and therapeutic issues and a review of the literature.

2. Presentation of case

We report the case of a 50-year-old man who admitted to the department of general surgery due to a mass in the left inguinal area. His medical history included renal transplantation (right iliac fossa). He had not known history of testicular disease. The patient did not have a problem of infertility. When the patient has cough, the mass does not change size. Physical examination revealed a inguinal hernia. Wrongly, we did not examine the scrotum in the preoperative period. The diagnosis was inguinal hernia. The rest of the examination was unremarkable. Surgical exploration revealed the presence of cryptorchid testicle in inguinal hernia (Fig. 1). The left testis with its overlying tunica vaginalis was found at the deep inguinal ring. A atrophic testis was found in the hernial sac with its blood vessels and vas deferens. As a result, it was felt that the testicle was non-viable and a left testicle was resected. The hernia sac was resected. The surgical correction was made (Fig. 2). The hernia was repaired by a synthetic mesh (Lichtenstein Hernioplasty). Histopathology report confirmed a atrophic testis (Figs. 3 and 4) and the presence of leydig cells, seminiferous tubule without testicular germ cell tumor (TGCT). The patient tolerated the procedure and the postoperative period was uncomplicated.

3. Discussion

An undescended testicle, sometimes called a cryptorchid testicle, can be found in 3% of the term newborns and 0.5–1.0%...
of adults. Cryptorchidism is more commonly seen in premature males and associated to genetic disorders in 10% of the cases. The causes of cryptorchidism are: prematurity, spina bifida, hormonal disorders, testicular absence or retractile testes. Jensen et al. concluded that smoking more than 10 cigarettes a day during pregnancy increased the risk of cryptorchidism. Kaffanovskaya et al. concluded that the second inguinoscrotal stage of testicular descent is clearly androgen-dependent. The diagnosis of cryptorchidism is made by physical examination. The diagnosis of cryptorchidism should be considered when non palpable testes and inguinal hernia are present. However, each patient may experience symptoms differently. Nonetheless, for inguinal hernia, the clinical presentation varies, depending on the contents of the hernial sac and the degree of herniation. Because of its varied presentation, clinical examination is often inconclusive.

In our case, the diagnosis of cryptorchidism is not diagnosed before operation because we did not examine the scrotum in the preoperative period and in our practice, sonographic examination is not systematically executed for hernia. The correct diagnosis of inguinal hernia is usually made during an inguinal hernia repair, although ultrasonography and computerized tomography have been used to identify an inguinal hernia. Laparoscopy is useful for both diagnosis and treatment of atypical inguinal hernia.

The complications of unrepaired cryptorchidism are mainly: testicular cancer, inguinal hernia, testicular torsion and infertility. The testicles begin to lose the process of spermatogenesis if they are not in the scrotum because the scrotum is a “cooler location”. This process explains the link between cryptorchidism and infertility. Very little is known about the link between cryptorchidism and TGCT tumorigenesis. In our case, despite its age, histopathology report confirmed the absence of tumor. Because the incidence of testicular cancer generally increases in cryptorchid testes, careful follow-ups are essential.

Usually, cryptorchidism resolves without any intervention before the age of 6 months. Surgical repair for cryptorchidism will be carried if the testicles have not descended. The prognosis of cryptorchid testes is related to the precocity of the management. Most studies have concluded that there is a direct correlation between how long the testis was subjected to a cryptorchid position and TGCT incidence. Pettersson et al. demonstrated that Individuals who had corrective surgery after the age of 13 had an incidence rate of 5.4%, where as those who were treated before 13 had an incidence rate of 2.23%. In our case, we did not find correlation between the time of surgery and risk of TGCT. Histopathology
report confirmed the presence of Leydig cells, seminiferous tubule and Sertoli cells without TGCT. The treatment of cryptorchidism improves the risks of infertility and gonadal neoplasia. Surgical repair for cryptorchidism will result in earlier detection of an eventual tumor.

It is difficult to understand the surgical anatomy of inguinal hernias, but once the surgical exploration is performed, surgical repair is simple. It is controversial whether a contralateral orchidopexy is needed as. Furthermore, the incidence of testicular cancer generally increases in fixed testes. In our case, the contralateral orchidopexy was not performed. An alternative is hormonal treatment with a series of injections HCG (human chorionic gonadotropin) which stimulate the testes. Kjaer et al. concluded that no age dependency of HCG effects was found, but the position of the testes before treatment influenced the success rate. This treatment is recommended if the testis is very close to the scrotum.

Our case is particularly notable because of the unusual presentation of cryptorchid testis as an incarcerated inguinal hernia at the age of 50 years. The patient remained asymptomatic 50 years. To our knowledge, the present case represents the first case of cryptorchid testis revealed at the age of 50 years with an inguinal hernia.

**Conflict of interests**

No conflict of interests.

**Funding**

None.

**Ethical approval**

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.

**Author contributions**

Radwan Kassir was contributed in writing of this article. Joelle Dubois was contributed in conceptualized and designed the paper. Sid-Ali Berremila was contributed in data collections. Sylviane Baccot and Alexia Boueil were reviewed the paper. Olivier Tiffet was contributed in conceptualized, designed, reviewed and revised the paper.

**Key learning points**

- The surgeon must always be alert to the possibility of cryptorchid testis during a surgical exploration of an inguinal hernia.

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