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Acquired Intraabdominal Testis due to Adhesions after Necrotizing Enterocolitis

Isber Ademaj^{1*}, Hysni Jashari¹, Naser Gjonbalaj² and Arta Sylaj¹

¹Department of Pediatric Surgery HUČSK-Hospital and University Clinical Centre of Kosovo

² Department of Radiology HUČSK-Hospital and University Clinical Centre of Kosovo

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Corresponding author:

ISBER ADEMAJ

Department of Pediatric Surgery
University Clinical Centre, Prishtina Kosovo
E- mail: isberademaj@gmail.com

Abstract

Undescended testis (UDT) refers to stagnation of the testicle during descent, or absence of the testicle in the hemiscrotum. In about 90% of cases this is unilateral. The etiopathogenesis of UDT is well described in the literature, where in most cases stagnation occurs in the inguinal canal and very rarely in the abdomen. Stagnation of a testicle in the abdomen, in addition to the histopathological damage that may result, may be the target of adhesions with intestinal segments. Some very rare cases of intestinal obstruction due to adhesions between an intraabdominal testis and the intestinal segment have been reported. Our case report aims to document a very rare variant of recurrent (acquired) intraabdominal cryptorchidism due to adhesions after necrotizing enterocolitis. Any newborn

after a laparotomy has a significant risk of developing intraperitoneal adhesions, which are common complications of necrotizing enterocolitis, even without surgical treatment. In this report, we describe a case of previously palpable testis in the inguinal canal at neonatal age, which was pulled inside the abdomen at the seventh month of age through adhesions between testis and the segment of sigmoid colon after necrotizing enterocolitis. Pediatric surgeons should consider a patient undergoing laparotomy or necrotizing enterocolitis especially when treated surgically as a risk factor for recurrent intraabdominal cryptorchidism.

Keywords: Undescended testis, acquired cryptorchidism, necrotizing enterocolitis, adhesions, newborn, laparotomy.

Introduction

Undescended testis (UDT) refers to the absence of the testicle in the hemiscrotum, mainly due to stagnation of the testis during descent. It is among the common causes of surgical interventions in pediatric surgery.¹ It is estimated that 1-2% of male babies have UDT till the 12th months of age.² In most cases, stagnation occurs in the inguinal canal and very rarely in the abdomen where, in addition to the histopathological damage that may result, it may also be the target of adhesions with intestinal segments. Some very rare reports of intestinal obstruction have been documented due to adhesions between an intra-abdominal testis and the intestinal segment.

The most common cause of adhesions in newborns is necrotizing enterocolitis (NEC) with consequently ileostomies. Although obstructive ileus as a consequence of adhesions between bowel segments is well known, intestinal obstruction due to adhesions between testicular structures and bowel segments has been reported very rarely. Our case documents a very rare cause as an etiological factor for acquired intraabdominal cryptorchidism due to adhesions after NEC.

Recurrent cryptorchidism refers to a testis that was previously much lower, in the process of descending into the scrotum but re-ascended, or retracted into the upper line due to different factors. While the pathogenesis of congenital undescended testis is considered multifactorial including hormonal, genetic, and environmental influences, the etiology of recurrent cryptorchidism remains unclear.³⁻⁶ Acquired cryptorchidism has a reported prevalence of 1%–7% and peaks around 8 years of age.^{7,8} We have reviewed the literature and to our knowledge this is first case reported where adhesions due to NEC have been caused by acquired intraabdominal testis in infant age.

Case Presentation

In this presentation, we will describe a case of a testis previously palpable in the inguinal canal at neonatal age that was retracted intraabdominally after seven months of age due to adhesions between the testis and the sigmoid colon segment after NEC. He was operated on at neonatal age for a perforated bowel segment due to complicated NEC. An ileostomy was performed, which was closed three months later. At the time of the first and second operations (ileostomy and ileostomy

closing) the presence of the testis in the inguinal canal was noted. The patient, at the time of the first operation, had many adhesions released, which were more evident when the ileostomy was closed. While under anesthesia, the patient was examined for the undescended testis, and it was found to be in the inguinal canal and could reach the neck of the hemiscrotum with slight retraction.

After closure of the ileostomy, the patient was discharged on the 5th postoperative day and was examined in our outpatient clinic every 2 months, including for the left undescended testis with ultrasound and physical examination.

At the first outpatient visit at five months of age, the left hemiscrotum showed the absence of testicle which was slightly smaller than the right testicle and was evident in the inguinal canal, while the contralateral testicle was normal. At the second visit, two months later, it was observed that the left testis was receded into the internal inguinal orifice, and could be pulled nearer to the neck of the left hemiscrotum, which was possible in the previous visit.

At the third visit, when the child was nine months old, we could not palpate the testis in the inguinal canal. Ultrasound showed it to be close to the internal inguinal orifice, which was confirmed by pelvic MRI, and it was smaller than the contralateral testis (Figure 1).

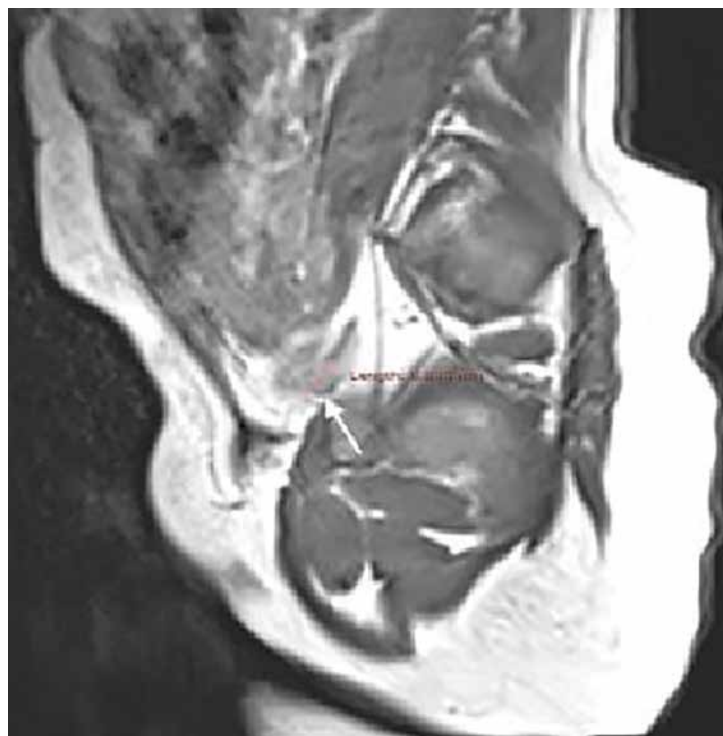


Figure 1: MRI of the pelvis demonstrating the presence of the left testis in the pelvis, near the left inguinal canal.

The child was scheduled two months later for laparoscopic exploration and was found to have testis attached by adhesions to the loop of the sigmoid colon (Figure 2).

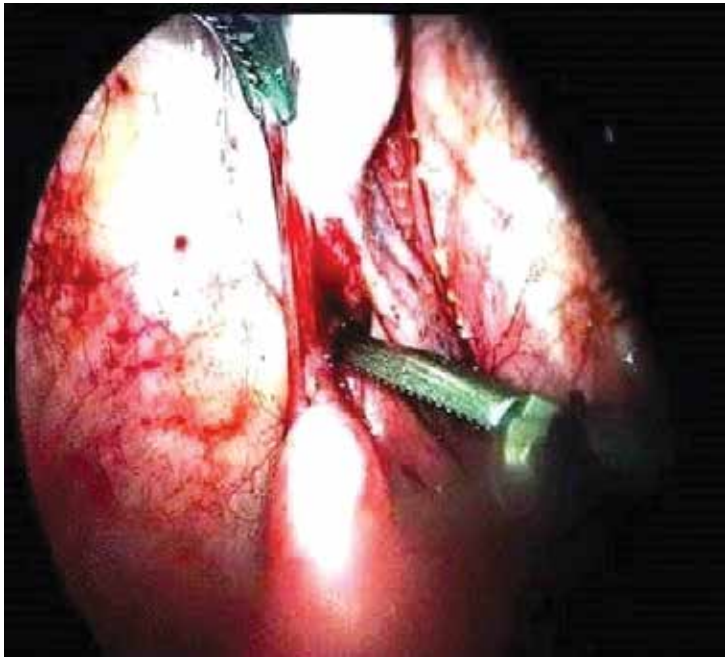


Figure 2: Intraoperative picture during the laparoscopy, showing the adhesions between the sigmoid loop and the left testis.

Adhesions and peritoneal attachments were released to increase length and the testis was brought to the neck of the hemiscrotum, where it was attached, with slight tension. The patient was discharged the following day.

Five months after laparoscopic orchiopexy, the testis remained in the neck of the left hemiscrotum with good vascularization but with very small progressive development (10x6mm) compared to the contralateral testis (11.5x7.5 mm) (Figure 3).

Discussion

Bowel obstruction due to adhesions is often seen in children after laparotomies. NEC is frequently the cause of adhesions, usually between bowel segments and surrounding intraperitoneal structures. The UDT can be involved in these adhesions, while still intraabdominal. Moreover, adhesions causing acquired intraabdominal testis, as in our case, have not been reported previously. Cryptorchidism is defined as the failure of the testis to reach the scrotum. It occurs in 3% of full-term and 30% of preterm male neonates. However, over 80% of these cases descent spontaneously by the 12th month of age.⁹⁻¹¹ After this age, it is necessary to operate, due to histological changes that start to occur.



Figure 3: Ultrasonographic image of the left and the right testis, 5 months after laparoscopic orchiopexy.

UDT is associated with increased risk of torsion, trauma, infertility, malignancy, and also the psychological impact of testicular absence later in adulthood.^{3,4} The risk of tumors (seminoma) in cryptorchid testes is 10 to 40 times higher than in descended ones.^{12,13} Furthermore, acquired cryptorchidism as well as congenital UDT can be associated with development of the same adverse histologic changes seen in primary cryptorchid testes requiring surgical correction.¹⁴ In this case, our patient had a mild hypotrophy probably due to hypoperfusion provoked by vascular stretching through the sigmoid segment. About 10% of testicular torsion occurs in UDT. Bowel obstruction has been documented from adhesions between the intraabdominal testis and bowel.^{15,16} Our case clearly shows the correlation between NEC and acquired intraabdominal testis, where adhesions due to NEC attached the testis to the sigmoid colon segment and retracted the testis inside the abdominal cavity. In our case, the adhesions were responsible for testicular retraction from the inguinal canal into the abdominal cavity.

In about 20% of cases, undescended testicles cannot be palpated on regular examination. A quarter of these cases can be palpated when the child is under anesthesia, and 6% of non-palpable cases are intraabdominal.⁹ It is well known that retractile testes are at risk of becoming true ascended testes, therefore yearly examination is recommended in these cases.^{17,18}

The case we have described here was positioned in the inguinal canal in the first five months, then ascended into the abdomen due to retraction from adhesions attached to the sigmoid colon, resulting in the intraabdominal testis. This case highlights the importance of intraoperative exploration of the internal inguinal ring during adhesiolysis in the case of inguinal testis because of the potential risk of adhesions between the bowel segment and testis that could cause acquired intraabdominal testis. Cryptorchid testes undergo parenchymal changes if surgical correction is delayed beyond 2 years of age.¹⁹ Our case showed slow development and was observed to be slightly hypotrophic.

Exploration of the pelvic cavity by imaging is essential for non-palpable testis, especially when it has previously been observed in the inguinal canal. It is especially important because of the risk of becoming torqued, atrophied or even retracted intraabdominally. If, during surgical intervention, the testis is found to be severely hypoplastic, gonadectomy must be considered.¹⁹

Preoperative diagnosis allowed us to discuss with the patient's parents the possibility of performing orchiopexy, knowing from the imaging studies that testis was slightly hypotrophic.

Any neonate requiring laparotomy is at significant risk of developing adhesions.²⁰ In the case of an inguinal testis, it is important to explore the region near the inguinal canal to ensure that no potential adhesions are left that could attach the testis and later retract it into the abdominal cavity, as highlighted by this case study.

Our case emphasizes a very rare presentation of acquired intraabdominal testis. It shows the importance of thorough intraoperative exploration of the internal inguinal ring during laparotomies. This is especially pertinent in the case of adhesions following pathologies such as NEC, or when a testis is in the inguinal canal or even absent. However, few cases have been reported showing correlation between cryptorchidism and intestinal obstruction in children.²¹ Reviewing the literature, to our knowledge, acquired intraabdominal testis in children caused after NEC adhesions has not been reported previously.

Conclusion

Our case emphasizes a very rare etiological factor of acquired intraabdominal testis. This case should be evaluated in the context of the etiopathology of UDT. This case provides valuable information for pediatric surgeons. In light of this case, when a neonate or infant with inguinal testis undergoes laparotomy, especially after potentially adhesion-producing diseases such as NEC, this should be considered as a risk factor for acquired intraabdominal testis.

Thus, it is imperative that when an infant with UDT in the inguinal canal undergoes laparotomy, especially for diseases with a high risk of adhesions, measures are taken to prevent adhesions near the inguinal canal, in order to minimize the potential for resulting acquired intraabdominal testis.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Conflicts of Interest

The following authors have no financial disclosures: (Isber Ademaj, Hysni Jashari, Naser Gjonbalaj and Arta Syla).

Authors Contributions

Isber Ademaj carried out design of the study, data collection, drafted the manuscript and was a major contributor in writing the manuscript supervision and analysis of the literature. Hysni Jashari and Arta Sylja performed the literature review and was involved in writing the manuscript. Naser Gjonbalaj performed radiological examination of the pelvic MRI. All authors read and approved the final manuscript.

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