Case Report

Intrathoracic Testicular Ectopia in Congenital Diaphragmatic Hernia

Mehran Hiradfar and Sadegh Sadeghipoor, Department of Pediatric Surgery, Dr Sheikh Pediatric Hospital, Mashhad University of Medical Sciences, Iran.

Congenital diaphragmatic hernia (CDH) is a defect in the diaphragm through which intra-abdominal and retroperitoneal organs may pass. However, the presence of the testis in the thoracic cavity is rare. Here, we describe a case of left-sided Bochdalek CDH with herniation of the left testis through the defect into the thorax, which was managed successfully by primary orchiopexy. [*Asian J Surg* 2006;29(4):303–5]

Key Words: Bochdalek, congenital diaphragmatic hernia, testis, thorax

Introduction

Congenital diaphragmatic hernia (CDH) is a defect in the diaphragm with an incidence of one in 2,000–5,000 births.[1] Intra-abdominal and retroperitoneal organs (kidney and adrenal gland) may pass through the defect. Presence of the testis in the thoracic cavity is rare, but has been reported in CDH.[2] Here, we describe a case of left-sided CDH (Bochdalek) with herniation of the left testis through the defect into the thorax, which was managed successfully by primary orchiopexy.

Case report

A male baby was delivered by caesarean section at a gestational age of 38 weeks; he weighed 2,300 g and was diagnosed with left-sided CDH. Because of respiratory distress, endotracheal intubation was performed and he was put on conventional mechanical ventilation. Physical examination revealed nonpalpable undescended testis on the left side. Abdominal exploration through a transverse left upper quadrant incision was performed on the 3rd day of life. The stomach, all of the small intestine, parts of the large intestine and the spleen had passed through the posterolateral defect of the left diaphragm and were in the left hemithorax. During reduction of these organs through the defect, we also found the left testis in the left hemithorax. It had a large and broad gubernaculum, and its long cord crossed the defect and was attached to the left gutter of the abdomen. It could easily be brought out from the incision up to the umbilicus (Figure 1A). The diaphragmatic defect was repaired and the testis was pulled through an inguinal incision and was fixed successfully in the left hemiscrotum without tension on the cord (Figure 1B). The long cord was fixed with a couple of absorbable sutures in the left gutter of the abdomen with careful attention to its elements. The postoperative course was uneventful. The size of the testis was acceptable on physical and ultrasound examination 2 years later.

Discussion

The diaphragm is formed by several fused embryonic components, including the septum transversum, pleuroperitoneal membranes, oesophageal mesentery and body wall mesoderm. The septum transversum separates the pericardial and the peritoneal cavities as it fuses dorsally with the mesodermal tissue surrounding the foregut. As

Address correspondence and reprint requests to Dr Mehran Hiradfar, Flat 4, No. 20, 10th Milad Street, Sadjad BLVD, 9187765661 – Mashhad, Iran.
E-mail: M-Hiradfar@mums.ac.ir • Date of acceptance: 4 July 2005

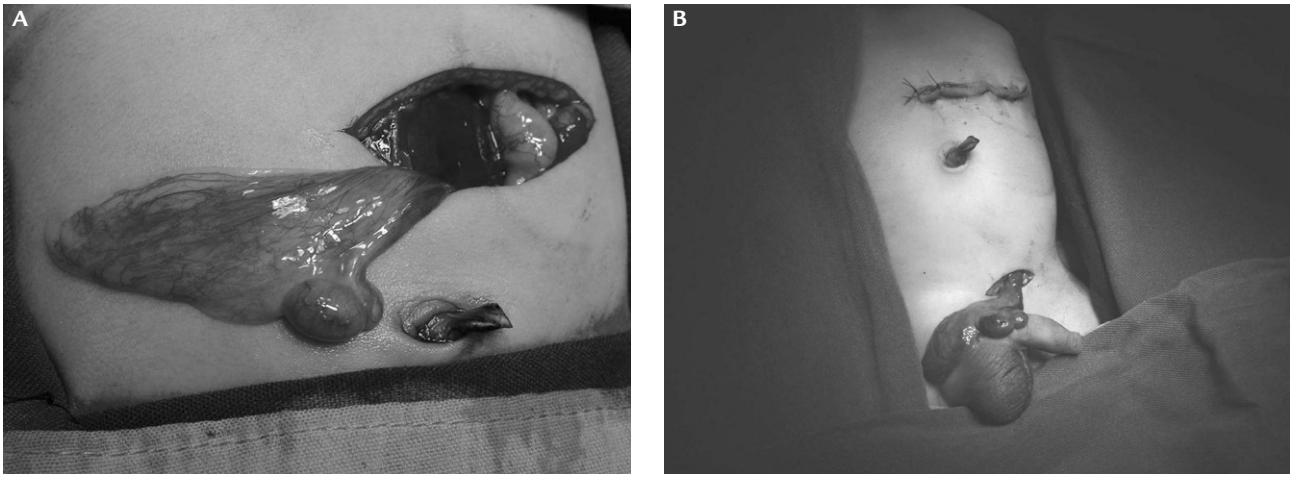

Figure 1. (A) Left testis and a very large gubernaculum after bringing out through the left upper quadrant incision. (B) Testis after pulling down through the inguinal region.

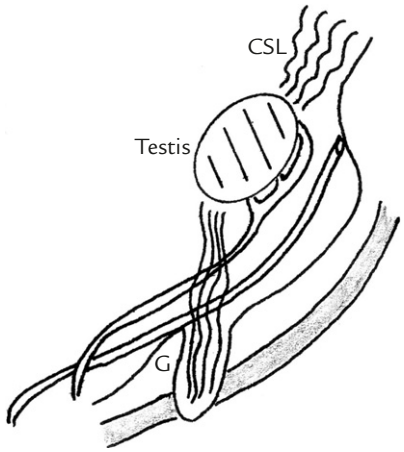

Figure 2. Testis is suspended by the cranial suspensory ligament (CSL) and gubernaculum (G). *Adapted with permission from Reference 3*.

this occurs, the pleuroperitoneal canal remains in continuity, connecting the pleural spaces and peritoneal cavity. Closure of the pleuroperitoneal canal by the pleuroperitoneal membrane completes the formation of the primitive fetal diaphragm. Left-sided CDH is a failure of complete closure of pleuroperitoneal membrane around 8–10 weeks of gestational age.[1]

The normal mechanism of testicular descent is a complex interaction of hormonal and anatomical factors, and the exact process remains unclear. During the indifferent stage of sexual differentiation, two opposing ligaments, the gubernaculum and cranial suspensory ligament (CSL), suspend the gonads[3] (Figure 2). The CSL extends from the cranial end of the testis to the dorsal abdominal wall near the last rib. In the transabdominal phase of descent at 8–15 weeks, the swollen male gubernaculum anchors the testis near the groin while the abdominal cavity enlarges. At the same time, the CSL regresses with testosterone effect.[3] Some researchers believe that regression of the CSL is only needed in rodents for normal descent of the testis,[4–6] but others also ascribe a role for regression of the CSL in humans.[3–5]

Although we cannot fully explain the mechanism of this type of ectopia, anatomical intimacy of the CSL to the CDH may point to a problem in this region that may have resulted in both CDH and abnormal regression of the ligament. On the other hand, as most CDH cases are not associated with this type of ectopia, hypertrophy and proliferation of the mobile gubernaculum may have allowed migration of the testis to the thorax.

Undescended testis can occur in CDH because of ineffective increase in abdominal pressure during the fetal period. However, it cannot explain unilateral cryptorchidism, as the defect in intra-abdominal pressure should affect both testes equally. Orchiopexy in intrathoracic testicular ectopia cases is usually performed with some delay as a second operation.[2] Due to the presence of a long cord in this case, tension-free and atraumatic orchiopexy was possible in the first operation without adding much to the operative time. This strategy prevents a difficult future orchiopexy and inadvertent trauma to the cord and testis and intra-abdominal organs in a second operation.

References

1. Robert M. Congenital diaphragmatic hernia and eventration. In: Ashcraft K, ed. *Pediatric Surgery*. Philadelphia: Elsevier–Saunders, 2005:304–19.
2. Hisano K, Kurisu S, Okada M. Laparoscopic orchiopexy of intraabdominal undescended testis associated with congenital diaphragmatic hernia. *Surg Today* 1998;28:1224–6.

3. Tomiyama H, Sasaki Y, Huynha J, et al. Testicular descent, cryptorchidism and inguinal hernia: the Melbourne perspective. *J Pediatr Urol* 2005;1:11–25.
4. van der Schoot P, Elger W. Androgen-induced prevention of the outgrowth of cranial gonadal suspensory ligaments in fetal rats. *J Androl* 1992;13:534–42.
5. van der Schoot P, Emmen JM. Development, structure and function of the cranial suspensory ligaments of the mammalian gonads in a cross-species perspective; their possible role in effecting disturbed testicular descent. *Hum Reprod Update* 1996;2:399–418.
6. Huston J. Undecended testis, torsion, and varicocele. In: O'Neill JA, Jr, ed. *Pediatric Surgery.* St Louis: Mosby, 1998:1087–109.