

Complete separation of the testis and epididymis presenting as inguinal hernia

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ABSTRACT

Complete separation of the epididymis from the testis is a rare phenomenon. In all reported cases, both structures remained undescended. We recently managed a 2-month-old boy with bilateral inguinal hernia and complete dissociation of the epididymis from the testis on the left side. At exploration, a right indirect inguinal hernia was confirmed while on the left side, an indirect inguinal hernia was associated with an undescended abdominal testis completely separated from a normally descended epididymis. This could reflect an essential role of the epididymis in the process of testicular descensus. The diversity of congenital abnormalities possibly encountered in the inguinoscrotal region during infancy and childhood necessitates the acquisition of insightful knowledge in the pathological anatomy of this area and adequate surgical skill to avoid undesirable intraoperative confusion with its negative impact on proper diagnosis and appropriate management.

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Epididymal malformations have long been reported in association with undescended testis,¹ and less frequently in cases of ipsilateral inguinal hernia or hydrocele particularly when the condition is communicating with the tunica vaginalis testis.² Complete dissociation between testis and epididymis, an extreme variant of these abnormalities, is considered rare.^{1,3,4} We report an infant with bilateral inguinal hernia and a unique variety of complete separation between testis and epididymis.

Case Report. A 2-month-old Saudi boy, a full term product of spontaneous vaginal delivery with 3170-gram birth weight, 50 cm body length, 33 cm head-circumference and Apgar score of 8 at one minute and 9 at 5 minutes, was referred with the diagnosis of bilateral inguinal hernia. On physical examination the referral diagnosis was confirmed. The boy had normal external genitalia with well-

developed hemiscrotums that seemed to contain both testes. Differences in consistency and shape of the left hemiscrotal mass had not been appreciated. There were no other positive findings. At exploration, right herniotomy was performed. Examination of the left groin revealed a bigger hernia sac that was dissected from what was initially believed to be the spermatic cord, recognizable by the silky sensation of ductus deferens. As dissection of the hernial sac progressed, another cord-like structure was found at the level of the deep inguinal ring. An attempt to anatomize this structure led to a normal-looking abdominal testis. A gubernaculum was not seen. After the hernial sac was closed, the left hemiscrotum was explored to reveal an ovoid mass measuring 12 x 20 mm, and likewise it was surrounded by a thick muscular coat, that perhaps made its clinical distinction from a normal testis difficult (**Figure 1**). A gubernaculum was attached to

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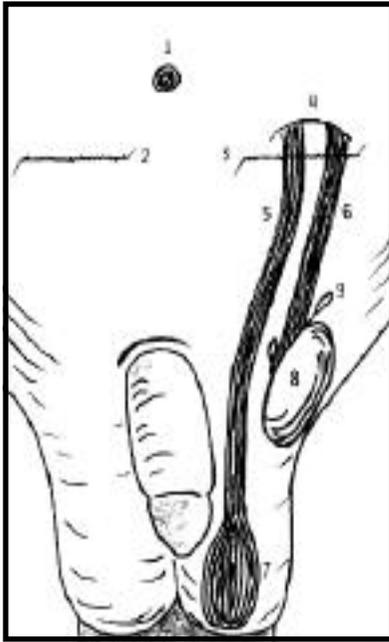


Figure 1 - Diagrammatic representation of intraoperative findings post bilateral herniotomy and left-sided orchido and epididymopexy. 1 - umbilicus, 2 & 3 - bilateral skin crease incisions, 4 - deep inguinal ring, 5 & 6 - cord structures, 7 - epididymis, 8 - testis and 9 - appendices testicularum.



Figure 2 - Micrograph depicting a cluster of epididymal ducts surrounded by concentric layers of smooth muscle (Hematoxylin & Eosin x 40).

the most distal point of the mass. A biopsy specimen of the mass was sent to the histopathology. The mass was put back to the bottom of the scrotum and fixed on a gauze peanut by absorbable suture. The paraffin section disclosed architecturally disorganized aggregate of epididymal ducts surrounded by concentric layers of smooth muscle. The adjacent connective tissue was rich in blood vessels. No gonadal tissue was identified (**Figure 2**). Funiculolysis of the abdominal testis enabled orchidopexy at the level of scrotal neck (**Figure 1**).

Discussion. The embryogenesis of testis, epididymis and vas deferens is a complex cascade of events: the testis evolves from the sex cord of the gonadal ridge while the epididymis and the vas evolve from the mesonephric ducts and Wolffian duct.⁵ These structures, although different in their embryological origins, develop in close relationship to unite later by fusion of the efferent ducts with the rete testis. It is postulated that Y-chromosome, androgens, inducers and suppressor substances facilitate this complex sequence of events. The descent of the testes, the last stage in the embryological gonadal development, is induced by chorionic gonadotropin, androgen hormones and supported by the gubernaculum.^{6,7} We found the gubernaculum attach to the normally descended

epididymis, indicating the likely role of the epididymis for the descent of the testes, thus contradicting the traditional teaching that the gubernaculum is connected to the testis. An abnormal epididymis or epididymal-testicular interaction may play a role in inhibiting the descent and final maturation of the gonad as observed in our case.⁸ In fact, cryptorchidism is often associated with epididymal abnormalities in 36% of cases, ranging from simple segmental atresia of the vas to complete separation of the epididymis from the testis.¹ However, the complete separation is considered rare.^{3,4} In most patients with complete separation, a normal proximal undescended testis is identified with a more distally located undescended epididymal-vasal complex or an atrophic tissue mass.^{3,4} To our knowledge, this case represents the first variant reported, where an abdominal testis is associated with a completely descended epididymis. Outcome and advantage of attempts to establish the continuity of the ductal system of both testis and epididymis remain subject to speculation and future research. The complete dissociation between testis and epididymis typically represents an incidental discovery at the time of exploration. The nearly missed diagnosis, as in our case, a high rate of malignant degeneration in cryptorchid testis, and a high rate of surgically missed cryptorchid testis found in patients with malignant testicular tumors,

calls for a thorough physical examination on all boys with inguinoscrotal ailment.⁹ Whether the malignant change in undescended testis is proportional to the degree of epididymal malformations, remain the issue for upcoming studies. To avoid missing the diagnosis of undescended testis with its undesirable sequelae, appropriate preoperative investigations as well as extensive operative search for the testes should be carried out. The diversity of congenital abnormalities, encountered in the inguinoscrotal region, makes meticulous dissection and knowledge in the pathological anatomy of this area, an essential prerequisite for all inguinoscrotal procedures, to minimize the risk of missing the proper diagnosis with its negative impact on appropriate management.

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