Patient Report

Immunohistochemistry: Additional armamentarium in the management of polyorchidism

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Polyorchidism is an infrequent developmental abnormality, resulting in supernumerary testes, often complicated by inguinal hernia, hydrocele, maldescent testis, varicocele, spermatic cord torsion or neoplasms.1 To our knowledge 123 cases have been reported to date, including 18 (14.6%) of complete testicular and adnexal duplication (CTAD).2

Approach to CTAD is still controversial, some authors recommend orchiectomy of the smaller of the homolateral testes, because of a possible risk of subsequent malignancy,1 others recommended scrotal orchidopexy after biopsy to assess developmental stage and germ cell count in both the homolateral testes.3 A recent report on left CTAD found immunoexpression of the Sertoli cell marker CD99 (MIC-2), and included evaluation of the mean spermatogonia index (MSI)4 and c-kit protein (stem cell factor receptor), which is known to assist germ cell survival and proliferation, by preventing apoptosis.5

Case report
A 11-year-old boy had long-standing, asymptomatic para-testicular mass inside the left hemiscrotum. Physical examination identified a double, superimposed, non-adherent scrotal mass. Color Doppler ultrasonography indicated normoechogenic homolateral testes and epididymides, with individual blood vessel networks. The right testis measured 40 × 19 mm; on the left, the lower testis was 21.8 × 15.2 mm and the upper one, 17.5 × 12.7 mm (Fig. 1). At surgery, left scrotal exploration indicated two separate testes within a single tunica vaginalis. Both testes had their own epididymis, vas deferens and vessels. The two vasa joined together within the operative field to form a common vas (Fig. 2). After biopsies of the left testes a double orchidopexy was performed. HE histology and immunocytochemistry using c-kit and CD99 rabbit antibodies were carried out.

On light microscopy the upper left testis was found to have post-pubertal seminiferous tubules, with mature Sertoli and germ cells, including round and/or long spermatids (Fig. 3a). In contrast, the lower left testis had empty or variously depleted seminiferous tubules, because spermatogonia and Sertoli cells were detached from the basal lamina, and thickened peritubular layer often occurred (Fig. 3b). As regards CD99 expression, a strong cytoplasmic immunostaining of Sertoli cells was common in both left testes. On the basis of CD99 immunostaining, MSI in 50 transverse sections of seminiferous tubules was counted to be 7.7 ± 2.0 in the upper testis and 3.5 ± 1.2 in the lower one. On Student’s t-test the MSI difference between the study testes was found to be significant (P<0.001). Moderate c-kit immunostaining of endotubular cells in the upper left testis (Fig. 3c) contrasted with a weak, focal c-kit positivity in the lower one (Fig. 3d).

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The postoperative course was uneventful and follow up to 24 months indicated normally echogenic left testes.

Discussion

The present case exemplifies type 3 polyorchidism, according to Thum’s classification. It is the rarest type of polyorchidism, implying epididymal and incomplete deferential duplication with superimposed homolateral testes. To the best of our knowledge the present case is the 19th reported CTAD, depending on a longitudinal division of genital ridge and mesonephric ducts, so that each homolateral testis had its own excretory ducts, allowing an active spermatogenesis. Color Doppler sonographic features substantiate the preoperative diagnosis of CTAD, denoting normal parenchymal texture, blood vessels, and epididymal-deferential ducts, without other scrotal lesions. Recently some authors have claimed a high accuracy for the diagnosis of supernumerary testis, with new imaging techniques such as sonography and magnetic resonance imaging rendering exploration unnecessary. On the basis of the present histology and pediatric age, we believe that a double homolateral orchidopexy is reliable to prevent both the risk of spermatic cord torsion and venous kinking. This is based on the findings that duplicated testes frequently have a ‘bell clapper’ deformity, predisposing to a possible intermittent testicular torsion and subsequent acquired and evitable damage.

Histology of the biopsied left testes indicated an advanced germ cell maturation of the upper testis, while germ cell depletion and Sertoli cell hyperplasia of the lower one occurred. CD99 immunostaining of the upper testis confirmed germ cells to be more represented in the upper than in the lower testis. In the former, spermatogonial density and MSI are significantly higher. Diffuse c-kit immunolabeling suggested an active spermatogenesis of the upper testis, as compared to the lower one. In the latter, c-kit immunolabeling was occasionally observed inside seminiferous tubules, indicating hypospermatogenesis and germ cell apoptosis.

Histology and immunocytochemistry in superimposed homolateral testes of uncomplicated scrotal CTAD indicated germ cell failure in the lower duplex testis. The reduced c-kit cell expression there substantiates such a feature, as a possible consequence of an enhanced apoptosis and a reduced cell survival.

Histological lesion might be due to recurrent hypoxia by intermittent testicular torsion in the duplicated testes as a consequence of anomalous insertion of gubernaculum tesnis. In this way, scrotal orchidopexy should be performed early to avoid tissue damage due to chronic blood vessel obstruction and/or spermatic...
cord torsion. Light microscopy and CD99 immunocytochemistry are very useful in assessing germ cell development and predicting spermatogenetic potential. c-kit immunostaining also confirms germ cell survival and viability.

In conclusion, such morphological tools for uncomplicated scrotal CTAD could be used as predictors of future fertility and tubular–interstitial damage. The latter may be prevented by early orchidopexy, the surgical approach of choice. Follow up on periodic ultrasonography can also be done to detect possible testicular and scrotal complications.

References