

Ectopic Epididymis in Testicular Appendices – Report of Two Cases –

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We report two cases of ectopic epididymal ducts and efferent ductules in the testicular appendices (TAs) of adult men with normally descended testes. In both cases, a sessile TA was incidentally found at the upper pole of the right testis during the scrotal hydrocelectomy. Microscopically, a few closely arranged tubules were detected within the TA. In the first case, the tubules were lined with a pseudostratified columnar epithelium with numerous, long microvilli, and were surrounded by a smooth muscle coat. In contrast, in the second case, the tubules had a wavy luminal surface, because ciliated columnar cells alternated with groups of cuboidal cells. In both cases, strong CD10 immunoreactivity was observed in the luminal border of the lining epithelium. Surgical pathologists should be aware of the presence of both ectopic epididymal ducts and efferent ductules that can occur in TAs, in order to avoid misinterpretation as transected, functional reproductive structures.

Key Words: Parovarian cyst; Choristoma; Epididymis; Wolffian ducts

Ectopic epididymal tissue is very uncommon and can be associated with both hernia sacs and neoplasms.¹⁻³ Furthermore, ectopic efferent ductules have never been described in the literature. Recently, a case of a 7-year-old boy with cryptorchidism, who had a testicular appendix (TA) containing ectopic epididymal tissue, was reported.⁴ The TA is a small sessile globular structure located at the upper pole of the testis, just below the head of the epididymis. It is considered to be a vestigial remnant of the Müllerian duct.⁵ To the best of our knowledge, neither ectopic epididymal ducts nor efferent ductules have been reported in the TAs of adult men without cryptorchidism. We herein report two cases of ectopic epididymal ducts and efferent ductules occurring in the TAs of adult men with normally descended testes.

CASE REPORTS

Case 1

A 60-year-old man presented with right scrotal swelling. He

had normally descended testes bilaterally and denied any feelings of pain or any urinary symptoms. He also denied any history of trauma, drug use or past medical problems. Physical examination was significant for a child-fist sized mass in the right scrotum. Laboratory examination revealed an elevation of total cholesterol to 220.0 mg/dL, whereas others were all within normal range. Scrotal ultrasonography revealed a hypoechoic fluid collection measuring 10 cm in diameter, suggestive of a hydrocele. The patient underwent right scrotal hydrocelectomy, and during the operation, a sessile structure was found at the upper pole of the right testis and was excised.

Case 2

A 49-year-old man presented with right scrotal swelling. He also had normally descended testes bilaterally and denied any history of trauma, drug use or past medical problems. Laboratory tests were all within normal range. Scrotal ultrasonography revealed a 12-cm diameter hydrocele in the right scrotum. During the scrotal hydrocelectomy, a small sessile structure was detected and excised.

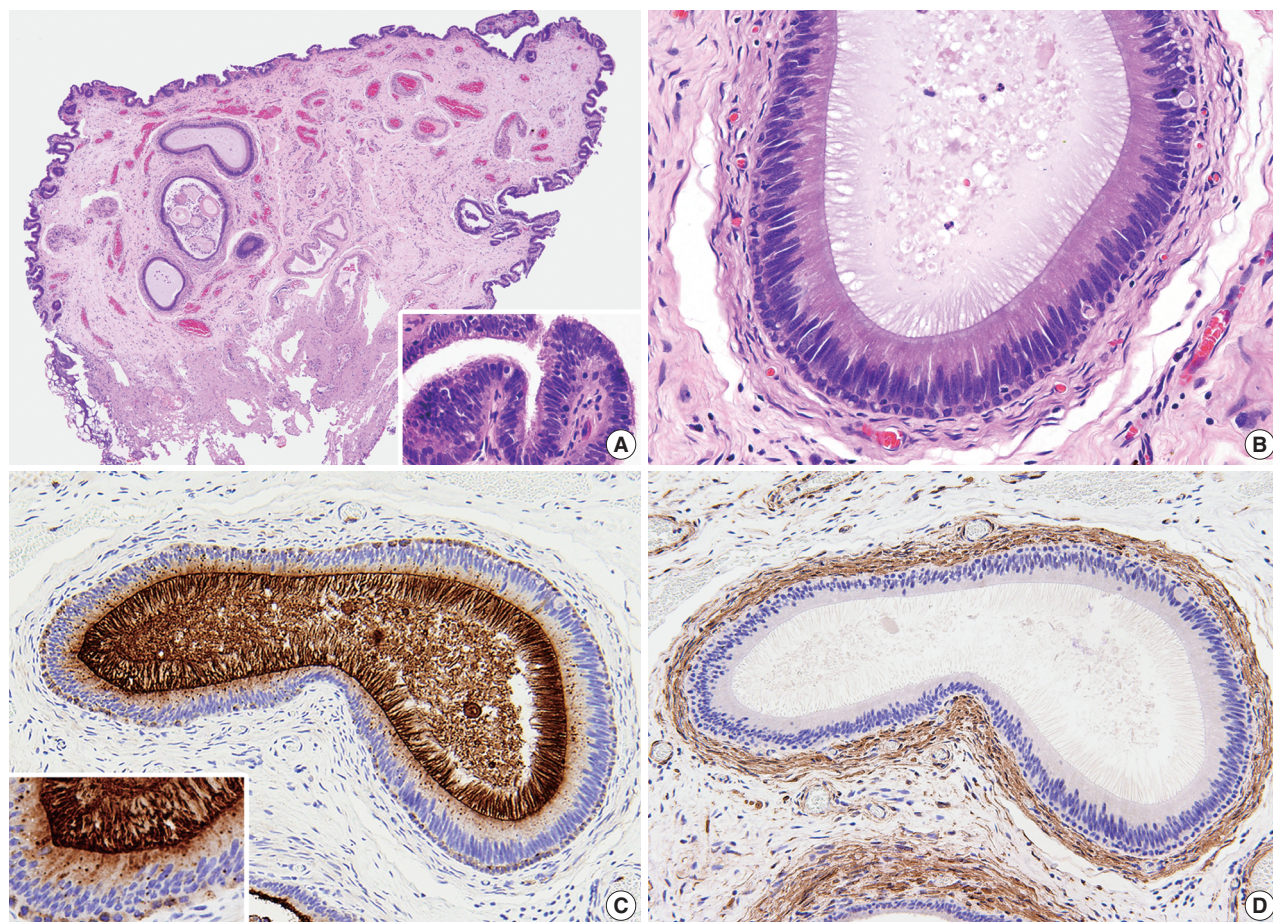


Fig. 1. Case 1. (A) A few closely arranged tubules are detected within the sessile testicular appendix, covered with a pseudostratified columnar epithelium (inset). (B) The individual tubule shows a smooth luminal surface and is lined with tall columnar cells with numerous, long microvilli and elongated nuclei, as well as short basal cells. The smooth muscle coat surrounds the tubule. (C) Strong CD10 immunoreactivity is detected in the luminal border, microvilli and the cytoplasmic granules of the tall columnar and short basal cells (inset), as well as in the luminal secretions. (D) Smooth muscle actin immunostaining highlights the smooth muscle coat which continuously surrounds the tubule.

Pathologic findings

The sessile structures (i.e., TAs) measured $0.5 \times 0.4 \times 0.4$ cm in case 1 and $0.4 \times 0.3 \times 0.3$ cm in case 2, respectively. Histologically, they consisted of fibrovascular cores of loose connective tissue, containing a number of blood vessels, fibroblasts, lymphocytes, and a few lymphatic vessels. The excised structures were covered with a pseudostratified columnar epithelium, some of which were invaginated into the stroma (Figs. 1A, 2A). Within the cores, a few closely arranged tubules were observed. In case 1, the tubular lumina showed smooth inner surfaces and were filled with either amorphous eosinophilic secretions or proteinaceous concretions with histiocytes. The tubules were lined with the pseudostratified columnar epithelium, which contained two cell types: tall columnar cells and short basal cells. The tall columnar cells had elongated nuclei and numerous,

long microvilli, whereas the short basal cells had spherical nuclei and lay close to the basement membrane (Fig. 1B). In addition, the tubules were surrounded by a smooth muscle coat, consisting of circular layers of smooth muscle cells. Thus, the tubules and their associated smooth muscle coat were identical to those of the epididymal ducts of normal adult men. In case 2, in contrast, the tubular lumina had an uneven or wavy appearance, because groups of ciliated, columnar cells alternated with groups of non-ciliated, cuboidal cells. Moreover, when compared to the structure of case 1, the columnar cells of case 2 had much shorter and less abundant cilia (Fig. 2B). These findings were identical to those of efferent ductules of normal adult men.

Immunohistochemically, in case 1, the luminal border and microvilli of the tall columnar cells were strongly positive for CD10 (1:500, clone 56C6, Novocastra, Newcastle Upon Tyne, UK), as presented in Fig. 1C. The cytoplasmic granules of both

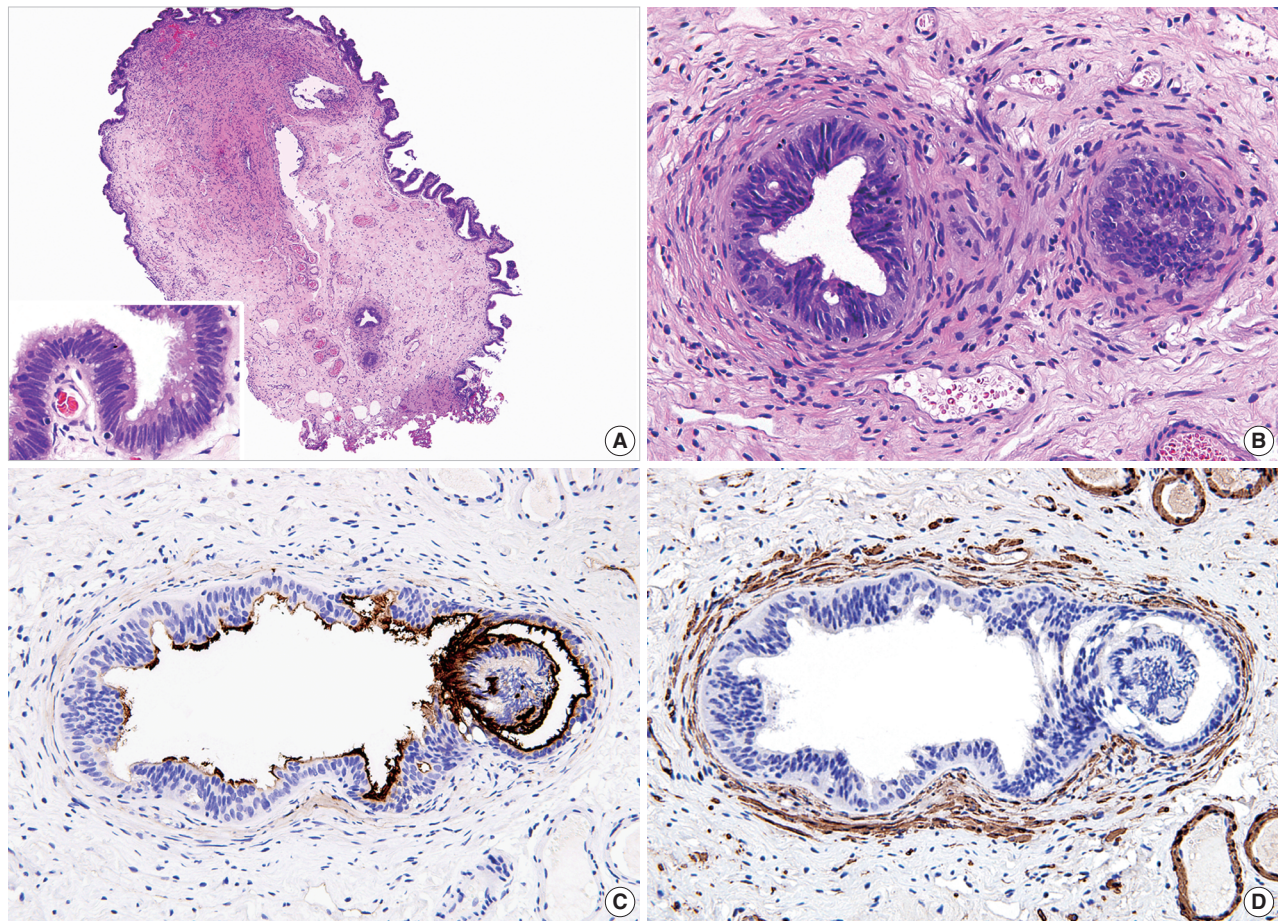


Fig. 2. Case 2. (A) Two tubular structures are detected near the base of the testicular appendix, with a pseudostratified columnar epithelial lining (inset). (B) One of the tubules shows an uneven luminal surface caused by groups of cuboidal cells, alternating with groups of columnar cells. (C, D) Similar to case 1, CD10 (C) and smooth muscle actin (D) highlight the luminal border and surrounding smooth muscle coat, respectively (polymer method).

tall columnar and short basal cells also displayed strong CD10 immunoreactivity (Fig. 1C inset). Smooth muscle actin (1:4,000, clone 1A4, Dakocytomation, Glostrup, Denmark) immunostaining highlighted the smooth muscle coat that surrounded the tubules (Fig. 1D). In case 2, the luminal border of the tubules and the smooth muscle coat also showed strong CD10 (Fig. 2C) and smooth muscle actin (Fig. 2D) immunoreactivities, respectively.

DISCUSSION

In the present case, histologic findings of the ectopic tissues observed in TAs were identical to those of normal epididymal ducts and efferent ductules (Fig. 3). To the best of our knowledge, neither ectopic epididymal ducts nor efferent ductules occurring in the TAs of adult men with normally descended testes

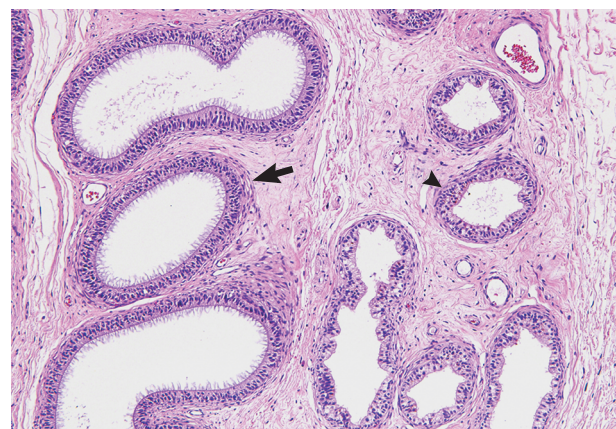


Fig. 3. The junction between the epididymal ducts (left) and the efferent ductules (right) of a normal adult man. The epididymal ducts (arrow) have smooth luminal surfaces and abundant, long microvilli. In contrast, the lumina of the efferent ductules (arrowhead) show an uneven or saw-tooth appearance, which is caused by groups of cuboidal cells alternating with groups of columnar cells. Some of the columnar cells possess cilia, but they are inconspicuous.

have ever been reported in the literature. There was a previous case report of a 7-year-old cryptorchid boy, describing a cluster of tubular structures found in the TA as epididymal heterotopias;⁴ however, the tubules shown in this report did not seem to be epididymal ducts, but were more likely efferent ductules. Although both the epididymal ducts and efferent ductules are lined with a pseudostratified columnar epithelium, there are a few differences between them. Thus, the epididymal duct epithelium consists of tall columnar principal cells and short basal cells and forms an elongated, smooth luminal surface; in contrast, the lumina of the efferent ductules have an uneven or wavy appearance, because the columnar cells alternate with groups of cuboidal cells. Moreover, the principal cells of the epididymal ducts have numerous, long modified microvilli, whereas the columnar cells of efferent ductules have much shorter and less abundant cilia. Similarly, the tubules shown in photomicrographs of the previous report of the 7-year-old boy were lined with alternating groups of low columnar and cuboidal cells with inconspicuous cilia, giving an uneven luminal surface.⁴ Therefore, these histologic findings are more likely to be ectopic efferent ductules, like those observed in the case 2 presented herein.

In general, both ectopic epididymal ducts and efferent ductules should be distinguished from glandular inclusions, occasionally found in inguinal hernia sacs and spermatic cords. Although glandular inclusions resemble either normal epididymis or vas deferens, they are considered a vestigial remnant, arising from the embryonic Müllerian duct.⁶⁻⁸ In contrast, the epididymal ducts and efferent ductules arise from the Wolffian ducts and are immunoreactive for CD10, whereas glandular inclusions do not show CD10 immunoreactivity.⁸ Therefore, in both cases presented here, strong CD10 immunoreactivities excluded the possibility of glandular inclusions.

TA is present in about 80-90% of normal adult men.⁹ In order to avoid the possibility of its torsion and potential infarction over time, careful exploration and subsequent excision have been suggested in every operation for inguinal hernia, cryptorchidism or hydrocele.¹⁰ Therefore, it is very important for surgical pathologists to recognize ectopic tissues in TAs, because they can be misinterpreted as portions of transected functional reproductive structures, thereby raising significant concerns regarding reproductive capability. The awareness of the presence of ecto-

pic epididymal ducts and efferent ductules may prevent such mistakes.

In summary, we presented two cases of ectopic epididymal ducts and efferent ductules in the TAs of adult men without a history of cryptorchidism. These ectopic tissues occurring in TAs have clinical importance for their potential for confusion with transected functional reproductive structures.

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