

·Case Report·

Enterogenous cyst of the testis

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Abstract

Enterogenous cyst is a rare congenital lesion generally located in the mediastinum or the abdominal cavity. We reported the first case of testicular enterogenous cyst in a 55-year-old white male presented with testicular pain and a gradually enlarging left scrotal mass with a 2-week duration. (*Asian J Androl* 2006 Mar; 8: 243–245)

Keywords: enterogenous cyst; testis; testicular pain; congenital lesion

1 Introduction

Enterogenous cyst (EC) is a rare congenital lesion of presumed endodermal derivation resulting from a dysembryogenetic error that occurs in the third week of fetal life [1]. These cysts are generally located in the mediastinum, the abdominal cavity, or within the spinal canal, skull, but they have never been described in the testis. We reported the first case of testicular EC.

2 Case Report

A 55-year-old white male presented with testicular pain and a gradually enlarging left scrotum with a 2-week duration. The medical history was negative for previous testicular symptoms and remarkable for appendectomy at the age of 52 with a histological diagnosis of appendicular carcinoid. Physical examination revealed a hard,

well-circumscribed mass, closely adherent to the testis and the cord. The left testis was atrophic, with a firm consistency, and the contralateral scrotal content was normal. Scrotal ultrasound examination showed a complex mass with two cystic lesions measuring 14 mm and 7 mm in diameter, respectively, and a hypoechoic pseudonodular lesion measuring 7 mm and adhering to the upper pole of the testis (Figure 1A and B). Tumor markers, including human chorionic gonadotrophin, alpha-fetoprotein, lactic acid dehydrogenase and carcinoembryonic antigen, were negative. A 6-cm oblique incision was made in the inguinal area approximately 2 cm above the pubic tubercle, and the spermatic cord was isolated and occluded with a noncrushing clamp at the level of the internal ring. The testis and its investing tunics were delivered into a carefully draped-off field as gubernacular attachments were divided. An intraoperative biopsy of the hypoechoic lesion was carried out and the histologic diagnosis ruled out malignancy. Therefore, a resection of the mass and a cuff (1 cm) of testicular tissue were performed.

Macroscopic examination revealed a multilocular cyst filled with yellow-green slightly viscous fluid. Histologically, the cysts had an epithelial lining, a loose subepithelial fibrous stroma with a few scattered lymphocytes and plasma cells, and no muscular or neural elements.

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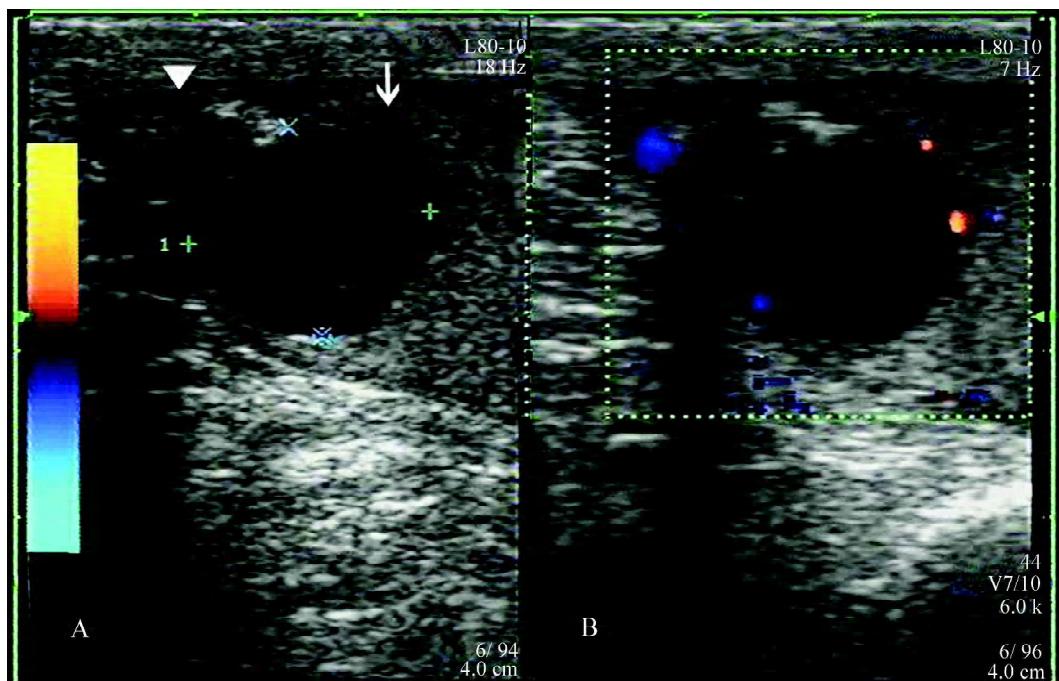


Figure 1. (A): Ultrasound shows a round fluid filled structure (white arrow) with debris, adjacent to a small well-defined (white triangle) hypoechoic area and hyperechoic marginal scar. (B): Color Doppler ultrasound shows poor vascularization of the lesion.

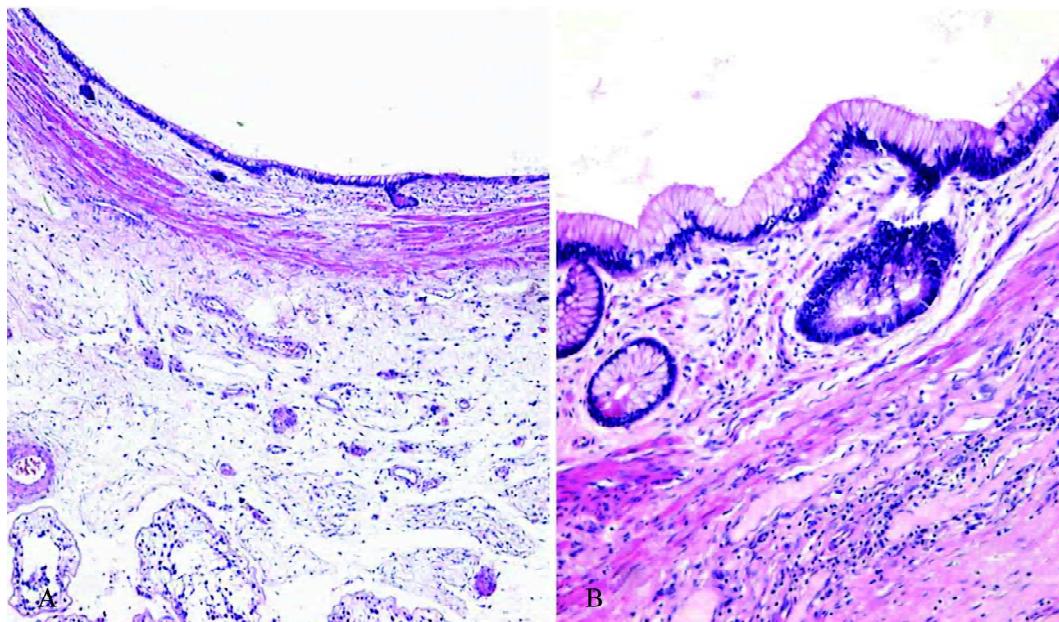


Figure 2. (A): Histopathologic appearance of the cyst wall shows epithelium, subepithelial connective tissue and smooth muscle ($\times 25$). (B): The cyst is lined by columnar non-ciliated epithelium secreting mucus with Paneth cells ($\times 200$).

phocytes and a well-circumscribed smooth muscle layer (Figure 2A). The inner wall of the cysts was lined by columnar non-ciliated epithelium secreting mucus and resembling intestinal epithelium. Isolated Paneth cells were interspersed among mucus-secreting cells (Figure 2B). The adjacent seminiferous tubules showed compression atrophy. Immunohistochemically, the epithelium of the cysts showed strong staining for the nuclear transcription factor CDX-2 and cytokeratin 20, but was negative for cytokeratin 7. The final pathologic diagnosis was EC.

During 1-year follow-up the patient was well with no evidences of disease recurrence.

3 Discussion

ECs are considered congenital anomalies. Hypotheses regarding their development have included intrauterine volvulus with subsequent ischaemia and infarction, persistence of intrauterine diverticulum, and incomplete vacuolization of the solid alimentary tract. Cysts of foregut and hindgut origin are rare and are often associated with vertebral anomalies suggesting incomplete separation of the foregut and notochord [2]. Genital development may be considered to begin in the 3-week-old embryo with the primordial germ cells located in the wall of the yolk sac. The formation of the genital ridges starts during the fourth or fifth week when primordial germ cells migrate from the yolk sac along the dorsal mesentery to populate the mesenchyme of the posterior body wall near the 10th thoracic level and originate the genital ridges.

Although the exact mechanism for the testicular localization of the EC in this case is not known, we suggest that the embryogenetic history of the testis, particularly during the fourth week, and the migrations of cells into it by way of the abdominal region must play an important role. The clinical presentation of EC after more than 50 years could be related to one of the major complications of cystic lesions (hemorrhage, infection) leading to swelling of the EC and symptoms. However, in the absence of any histological evidences of hemorrhage

or infection in the surgical specimen, the most likely cause of the presentation could be undiagnosed testicular trauma.

The preceding histological diagnosis of appendiceal carcinoid, another rare lesion, represents an interesting and unusual finding. As there appears to be no linkage between these apparently unrelated conditions, the two presentations may be a coincidence.

Differential diagnosis should be made from teratoma and mucinous cystadenoma.

Teratomas occur more frequently in the first and second decades of life, almost always have components other than mucinous epithelial-lined cysts and are accompanied by intratubular germ cell neoplasia (IGCNU).

Although enteric-type features have been described in mucinous cystadenomas, a muscular coat is normally absent. Conversely, ECs contain structures of all three germ layers (epithelium, blood vessels, fibrous tissues and smooth muscle) [3].

Because of its benign nature, EC might be amenable to conservative treatment. Extensive sampling of adjacent nonlesional tissue is indicated because an association with IGCNU would warrant the diagnosis of teratoma. The exclusion of IGCNU by examination of biopsy specimens of the adjacent testis permits local excision.

Acknowledgment

Written consent was obtained from the patient for publication of this study.

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