

Primary borderline mucinous neoplasm of the testis: A case report and literature review

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ABSTRACT

Testicular/paratesticular neoplasms morphologically resembling surface epithelial tumors of ovarian type are rare neoplasms. The criteria for the diagnosis and nomenclature of these tumors parallels those used for ovarian homologues. Pathologists and urologists need to be wary of this uncommon entity, excluding metastatic mucinous adenocarcinoma and herniation of mucinous tumors into paratestis/scrotal sac by careful clinicopathological correlation. Herein, we present the first case of borderline mucinous tumor of testis to be reported from India.

Key words: Borderline, mucinous neoplasm, testis

INTRODUCTION

Surface epithelial tumors are the most common type of neoplasms arising in the ovary. Intriguingly, a similar spectrum of tumors can occur in the testis/paratestis. Borderline tumors of serous epithelial type are the most frequent subtype within this category. Mucinous epithelial tumors of the testis/paratestis are extremely rare.^[1,2] Although small case series and anecdotes are described in pathology literature, the awareness of this entity amongst urologists is limited.^[1] The paucity of literature and rarity of the tumor has prompted us to report a case of primary mucinous tumor of the testis with borderline features.

CASE REPORT

A 47-year-old man presented with a left testicular mass and dull aching scrotal pain of six months duration. He had no risk factors for testicular

cancer, such as cryptorchidism. Physical examination was unremarkable, except for a left testicular mass. No inguinal lymphadenopathy was found on palpation. Tumor markers, including CA-125, alpha-fetoprotein and β -human chorionic gonadotropin were within normal limits. Ultrasound examination revealed a 6 × 4 × 4 cm heterogeneous mass predominantly hyperechoic and almost replacing the entire left testicle except at the upper pole. The mass appeared to arise in relation to the tunica. The contralateral testis and bilateral spermatic cord were normal. There was no evidence of any mass lesion or any intraabdominal lymphadenopathy on contrast computed tomography (CT) scan of the abdomen and pelvis.

The patient underwent high left inguinal orchidectomy with hernioplasty. A 7.5 × 5 × 3.2 cm orchidectomy specimen was received in the pathology laboratory. Externally, there were no surface nodules but the specimen had a bosselated appearance. On sectioning, there were variegated areas of cystic change, hemorrhage and mucin collection. The tumor was arising in relation to tunica layers, compressing the native testicular parenchyma and epididymis. Microscopy revealed a mucinous tumor with predominantly cystic areas. Within the cystic areas there were papillary projections lined by mucinous cells of intestinal type epithelium. Some of the papillae showed complex, confluent architecture with focal stratification of the mucinous cells. The individual cells exhibited moderate nuclear atypia. At an occasional focus, mucin extravasation was seen into the cyst wall with associated inflammatory and fibrotic response. A diagnosis of borderline mucinous tumor of ovarian type surface epithelium with focal microinvasion was rendered. Intratubular germ cell neoplasia, unclassified (IGCNU) component was not seen. Even after extensive

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search no teratomatous component was appreciated. On immunohistochemical evaluation, the tumor cells were immunoreactive to pancytokeratin, cytokeratin 20 and carcinoembryonic antigen (CEA). Stains for Cytokeratin 7, CDX-2, CA125 and placental alkaline phosphatase PLAP were negative [Figure 1-3].

DISCUSSION

Tumors of ovarian surface epithelial type of the testis and paratestis are rare. The entire spectrum of histologic cell types has been described in the literature, including serous, mucinous, endometrioid, clear, transitional (Brenner), and squamous subtypes. Among these serous tumors are the most common, with borderline cases outnumbering carcinomas. Primary mucinous tumor of the testicle and paratesticular region are extremely rare and the largest series reported is of nine primary mucinous tumors of the testis and paratestis.^[2] In this study, there were six borderline mucinous tumors, two cystadenoma and one case of a mucinous carcinoma. Elliott *et al.*,^[1] in their recent case report and literature review have documented only 23 cases of primary testicular mucinous neoplasm in the English literature. These included 14 cases of primary intratesticular mucinous neoplasm, of which six were borderline tumors, four were cystadenomas, and three were either mucinous cystadenocarcinoma or mucinous carcinoma. To the best of our knowledge this is the first case to be reported from India.

The histogenesis and origins of these enigmatic tumors are debatable and speculative owing to the rarity of these tumors. The first case reported by Kellert^[3] described a mucinous cystadenoma in the paratestis of an 11-year-old boy. It was accompanied by an oviduct-like structure and was presumed to have arisen from occult ovarian tissue. Most of the authors believe that these tumors arise from the metaplasia of the mesothelium of the tunica vaginalis.^[2,4] Others have postulated that these tumors may arise from mullerian remnants, such as the appendix testis.^[5] A teratomatous origin of these mucinous tumors is also fathomed.^[6] This proposition is less likely in our case as it lacked other teratomatous components and did not display an intratubular germ cell neoplasia component which is known to accompany about 90% of the teratomas in adults.

In addition to a teratoma with predominant mucinous component, the other plausible differential diagnosis to be excluded is a metastatic carcinoma. Metastatic mucinous tumors to the testicle are more common than primary mucinous testicular tumors. More than two-thirds of cases occur in men over the age of 50, and 10% of patients present initially with a unilateral testicular mass.^[7,8] Metastases from the colon, stomach and very rarely from the pancreas represent 53% of metastatic tumors to the testicle.^[2] These may produce cystic lesions in the testicle mimicking a primary mucinous tumor. However, a metastatic mucinous adenocarcinoma

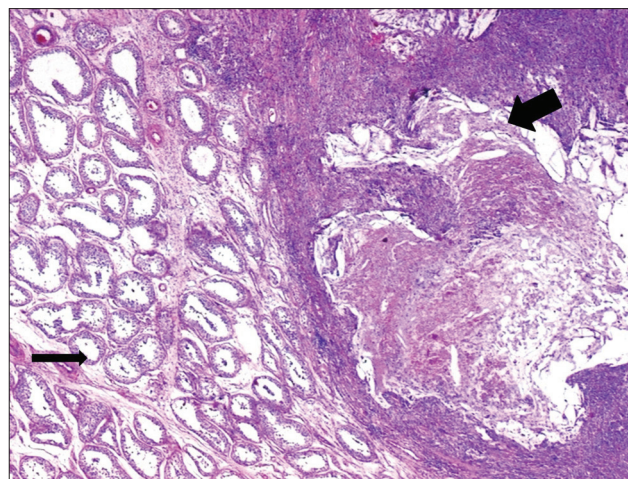


Figure 1: Microscopic section from the testis showing the compressed seminiferous tubules of the native testis (thin arrow) and the pools of extravasated mucin along with cholesterol clefts (thick arrow) (H and E, original magnification $\times 40$)

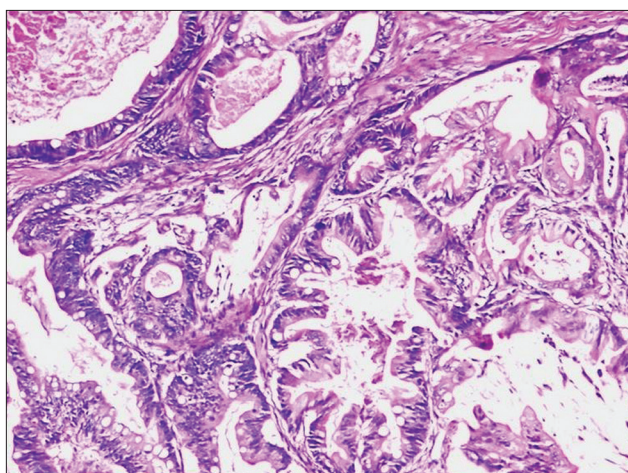


Figure 2: Borderline mucinous tumor of the testis: complex glandular and tubular structures lined by mucinous epithelium, which is focally stratified (H and E, original magnification $\times 200$)

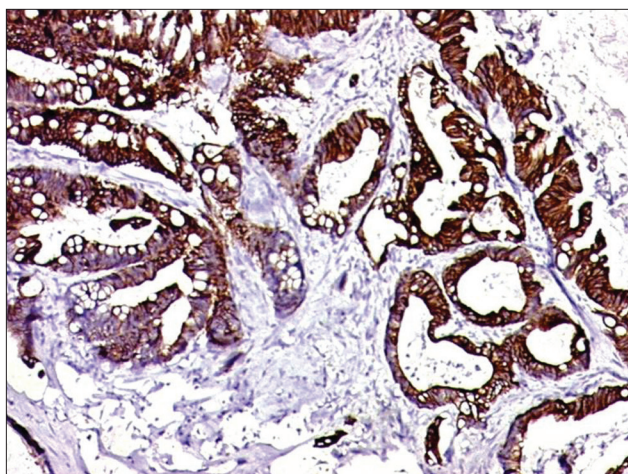


Figure 3: The epithelium of borderline mucinous tumor of the testis immunoreactive to cytokeratin 20 (Indirect immunoperoxidase $\times 200$)

would more likely have multifocal deposits, an interstitial growth pattern within the testicular parenchyma with

conspicuous lymphovascular space invasion, which were clearly lacking in our case. Also, the presence of borderline features in a mucinous neoplasm would be extremely rare in a metastatic setting.^[2] Immunohistochemistry can be an adjunct but may be of limited use in distinguishing primary mucinous tumor of the testis from a metastatic malignancy. In the present case, the tumor cells were CK 20-positive and CK7-negative, the classical pattern seen in most of the lower gastrointestinal tract tumors,^[9] but immunostains for CDX-2 came negative thus almost nullifying the possibility of colorectal origin. A gastric or pancreatic tumor is usually CK 7+/CK 20- which was not so in our case. It, however, needs to be emphasized that the immunohistochemical profile of primary mucinous ovarian type carcinoma of the testis overlaps with those of pancreatic and gastric adenocarcinoma and hence it is most appropriate to exclude metastasis by adequate radiological studies.^[10]

Another differential diagnosis which needs to be entertained by the urologist is spread of low-grade mucinous tumors of the appendix along peritoneal surfaces into a hernia sac, presenting as a paratesticular mass. The distinction depends primarily on identifying the neoplasm as contents of a hernia sac.^[11]

Although there is too little data to predict survival rate and prognostic factors in these tumors an outcome similar to ovarian counterparts has been shown in the cases reviewed by Elliott^[1] *et al.* The malignant counterpart obviously had poor outcome as compared to cystadenomas or borderline cases which had a lower metastasis and recurrence rate. Although there was a focus of microinvasion in this mucinous neoplasm, the predominant borderline features in our patient is expected to confer a favorable prognosis as he has undergone adequate surgical resection. Presently, the patient is on follow-up and has not shown any local recurrence/residual disease, metastasis or elevated tumor markers in the last six months after surgery.

In conclusion, we have presented a rare case of mucinous borderline tumor of the testis which histologically resembles its ovarian counterparts. The literature is still embroiled in controversy regarding the genesis of these fascinating tumors and it is imperative that urologists and pathologists be aware of this rare entity in their clinical practice.

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