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CASE REPORT

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Primary Mucinous Cystadenocarcinoma of the Testis: A Case Report and Literature Review

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Abstract

Primary mucinous cystadenocarcinoma (MCA) of the testis is extremely rare, only 10 cases having been reported to date. Metastases of mucinous adenocarcinomas that have originated in different sites can mimic primary MAC and must be included in the differential diagnosis. We report a case of primary MCA of the tunica vaginalis testis in a 28-year-old patient who presented with a painless mass on the left side of the scrotum. We present the clinical and pathological characteristics to contribute to the further understanding of these rare tumors. Mucinous cystadenocarcinoma of the testis is extremely rare, particularly in individuals younger than 40 years. Histological examination, immunohistochemical analysis, and clinical examination to exclude metastases from other organs are necessary for a definitive diagnosis. (**International Journal of Biomedicine. 2023;13(1):168-171.**)

Keywords: mucinous cystadenocarcinoma • ovarian-type surface epithelial neoplasm • testicular adenocarcinoma

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Introduction

Ovarian-type surface epithelial tumors very rarely present as primary neoplasms of the testis.(1) The mucinous subtype is particularly rare, only approximately 30 cases having been published to date,(2) only 10 of which were primary MCA of the testis.(3-12)

Case Report

A 28-year-old man presented with painless swelling of the left testis that he had first noticed 1 month before. On physical examination, there was no erythema, and no lymph nodes were palpable. Ultrasound examination showed a mainly hypoechoic, heterogenous, cystic lesion on the lateral surface of the left testis. Laboratory examination showed a serum alpha-fetoprotein concentration of 3.27 ng/mL (normal range: 0–20 ng/mL) and the β-human chorionic gonadotrophin of 0.112 mU/mL (normal range: 0.5–2.67 mU/mL). Other hematological and biochemical findings were within normal reference ranges. Computed tomography of the abdomen and pelvis confirmed the ultrasound findings and did not detect any lesions in the peripheral lymph nodes or other organs. A chest radiograph was normal.

A biopsy of the nodule was obtained, resulting in a histopathological diagnosis of the primary mucinous cystadenocarcinoma of the testis. The patient accordingly underwent total radical left orchiectomy. Grossly, the tumor consisted of irregular tissue with mucinous elements. Microscopically, it consisted of glandular formations and cystic spaces lined with ciliated, cylindrical, epithelial cells. Some malignant goblet cells with intracellular mucin were noted, as were wide mucinous pools with scattered neoplastic cells. The tumor stroma was composed of a dense inflammatory, predominantly mononuclear, infiltrate and proliferation of wide blood vessels containing erythrocytes. The patient was discharged on Day 4 of the hospital stay and referred to an oncology center. Immunohistochemical analysis was positive for CK 20, carcinoembryonic antigen, and MUC2 and negative for CK7, vimentin, thyroid transcription factor 1, and WT1.

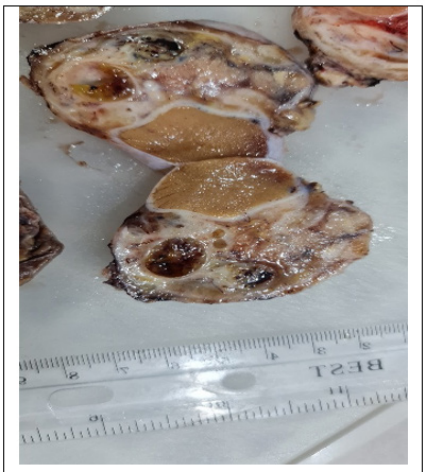

Figure 1. *Photograph of the cut surface of the testis with spermatic cord showing a mucinous cystic mass replacing the testicular parenchyma.*

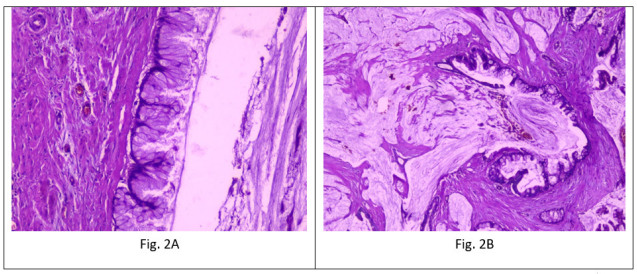

Figure 2. *Photomicrographs showing a moderately differentiated, cystic, malignant neoplasm with mucinous differentiation and irregular glands lined with single or multiple layers of columnar epithelium containing mucin. Papillary growth was also observed in some areas. Stain: H&E; Magnification: A, 40×; B, 200×*

Discussion

Primary ovarian-type surface epithelial tumors of the mucinous subtype are very rare. The first case, a cystadenoma in an 11-year-old boy, was reported in 1959.[13] These tumors are classified as benign, borderline, and malignant.[14] They are almost identical to their ovarian counterparts, with some key differences: they are not as large and more often present as unilocular cystic tumors.[3] Ovarian-type testicular tumors generally present as unilateral, painless, scrotal masses, often accompanied by a hydrocele. They can be para- or intratesticular, the former generally arising from the tunica vaginalis of the testis.[15] Gross examination typically shows cystic masses with gelatinous material.[4] Mucinous tumors vary microscopically according to whether they are benign or malignant. Benign tumors generally have cystic spaces and tumor glands with endocervical-type epithelium, whereas borderline, malignant, and mucinous carcinomas characteristically feature intestinal-type epithelium.[16] Our patient's tumor had these gross and histological features. Immunohistochemical staining of primary mucinous cystadenocarcinoma of the testis can reportedly be positive for cytokeratins, carcinoembryonic antigen, cancer antigen 125, epithelial membrane antigen, carbohydrate antigen 19-9, and cytokeratin CAM 5.2.[6] These tumors have a higher incidence in older individuals than do other testicular tumors, typically presenting in the fifth and sixth decade of life.[3,17] However, our review of cases of primary mucinous cystadenocarcinoma of the testis showed a similar incidence across age groups. Based on our research and experience, at 28 years of age, our patient is the youngest yet reported.

The origin of mucinous tumors of the testis remains unclear. Many hypotheses concerning their histogenesis have been proposed. One widely accepted theory is that these tumors originate from Müllerian remnants in the appendix testis or extra testicular scrotal tissue. Another theory suggests that they may originate in Müllerian metaplasia of the tunica vaginalis.[15,18] Others have argued that intratesticular lesions may arise from mesothelial inclusions or represent monodermal teratomas.[17]

Because mucinous tumors of the testis are rare, metastasis of mucinous tumors to the testis is more common than primary mucinous cystadenocarcinoma. It is, therefore, important to exclude the possibility of a metastasis mimicking the primary mucinous cystadenocarcinomaof the testis. It has been estimated that 53% of mucinous tumors in the testis are metastases from other sites. Metastatic testicular tumors most commonly originate from mucinous carcinomas of the colon, stomach, pancreas, prostate, and appendix.[7,20-23] Metastases from other sites can be distinguished from primary testicular mucinous cystadenocarcinoma by their multifocality, growth in the testicular interstitium, and prominent vascularity.[4,7,19] Another important point of differentiation is that testicular metastases can be bilateral.[15] Immunohistochemical positivity in primary ovarian epithelial mucinous carcinomas is not completely specific and may overlap with positivity in mucinous metastases from other sites.[9] However, it has been reported that immunohistochemical staining for CK7, MUC2, MUC5AC, and MUC6 may be helpful in the differential diagnosis of metastatic mucinous tumors of the testis.[7] A combination of immunohistochemical studies and clinical correlations is therefore needed to make the correct diagnosis.

Other differential diagnoses include primary testicular tumors, such as mesothelioma, adenocarcinoma of the rete, appendix testis, epididymis, and germ cell tumors.[6,16,24] Mesothelioma of the tunica vaginalis can be excluded by histological differences, such as a lack of psammoma bodies, low cellularity, intestinal type, and ciliated serous morphology, as well as the immunohistochemical profile.[6,24] The possible diagnosis of adenocarcinoma of the rete testis, appendix testis, or epididymis can be eliminated by the parenchymal location of the tumor and the specific histological features.[6] These lesions are located in the extra scrotal region, and their histological characteristics include tubular and tubule-papillary structures lined by cuboidal cells.[25]

Germ cell tumors can be excluded by the presence of a ciliated serous component, psammomatous calcification, and a lack of teratomatous elements,[5,26] the latter being a feature of most reported cases and the present case. Germ cell tumors

also differ in the age of onset, generally presenting at younger ages.[27] Another differential diagnosis is mucinous tumors of the appendix or peritoneal surfaces in an inguinal hernia sac, the distinction relying on the location of the neoplasm within a hernial sac and the presence of a prominent, extracellular mucinous component.[4,17]

The prognosis and treatment of patients with primary mucinous cystadenocarcinoma of the testis differ depending on the benign or malignant nature of the tumor. The most common treatment is radical orchiectomy and follow-up.[2] The prognosis of primary mucinous cystadenocarcinoma of the testis remains unclear because of their rarity. Our literature review yielded 10 previous cases of primary mucinous cystadenocarcinoma of the testis; 4 of these patients had developed distant metastases, and 2 had died within months of the diagnosis.[3,4,7,9]

Conclusion

Mucinous cystadenocarcinoma of the testis is extremely rare, particularly in individuals younger than 40 years. Histological examination, immunohistochemical analysis, and clinical examination to exclude metastases from other organs are necessary for a definitive diagnosis.

Informed written consent was obtained from the patient to publish this case report and any accompanying medical images.

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Competing Interests

The authors declare that they have no competing interests.

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