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Case Report

Primary mucinous cystadenoma originating from testis: a case report and review of literature

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Abstract: Primary testicular mucinous cystadenoma (PTMC) is one of the rarest 'ovarian type' tumors originating from surface epithelium. We aimed to present a 21-year old case and try to analysis the clinical characteristics of this condition. In our case, a right radical orchiectomy was carried out and mucinous cystadenoma was finally diagnosed based on pathology. Additionally, recurrence was not observed with a follow-up of 6 months. Furthermore, except for our case, only five patients have been diagnosed as benign PTMC based on our search of all English literature. Our case would be the youngest based on literature research. However, it is still very limited about understanding of this disease.

Keywords: Mucinous cystadenoma, testis, case report

Introduction

Primary testicular mucinous cystadenoma (PTMC) is one of the rarest 'ovarian type' tumors originating from surface epithelium [1]. Up to now, only five patients have been diagnosed as benign PTMC based on our search of all English literature from Pubmed [1-5]. However, the clinical characteristics, diagnoses, treatments and prognosis of this disease are still uncertain. We tried to present another PTMC patient and analyze the clinical and pathological characteristics of PTMC through a systematic review of literature.

Case report

A 21-year old male got into our hospital with a chief complain of aggressively enlarging right testis for more than 1 year. The patient denied of any malaise in his testes, epididymis and scrotum, like pain, fever, mass, nodule or ulceration, and any etiological factors, such as infection or trauma. And because of this, the lesion was neglected.

On physical examination, the appearance of his perineum was normal, while an enlarged testis could be palpated in his right side of scrotum with a diameter of about 7 cm. The diameter of his left testis was evaluated to be about 5 cm. However, the rough shapes of bilateral testes were normal, and local trigger and mass was unconspicuous in scrotal contents, except for a slight softer for his right testis.

For further diagnosis, alpha fatal protein (AFP) and beta-human chorionic gonadotropin (β-HCG) in his blood were detected, which revealed that a normal level of 2.77 ng/ml for AFP while significantly increasing of 433.37 mIU/ml (normal <3.81) for β-HCG. Furthermore, ultrasonography of scrotum was subsequently conducted which reported that an anechoic mass measured to be about 54×35×52 mm could be found at the lower polar of right testis except for a few normal testicular tissue at its upper polar. Multiple divisions accompanying with spot blood flow signal could be also discovered in this mass, and a teratoma was subsequently suspected. Additionally, an enhanced abdominal computerized tomography was carried out and didn't reveal any specific anomalies except for similar description for the right testis. Other preoperative examinations were all in normal ranges apart from elevating lactate dehydrogenase (LDH) (320 IU/L) and hydroxybutyrate dehydrogenase (HBDH) (267 IU/L).

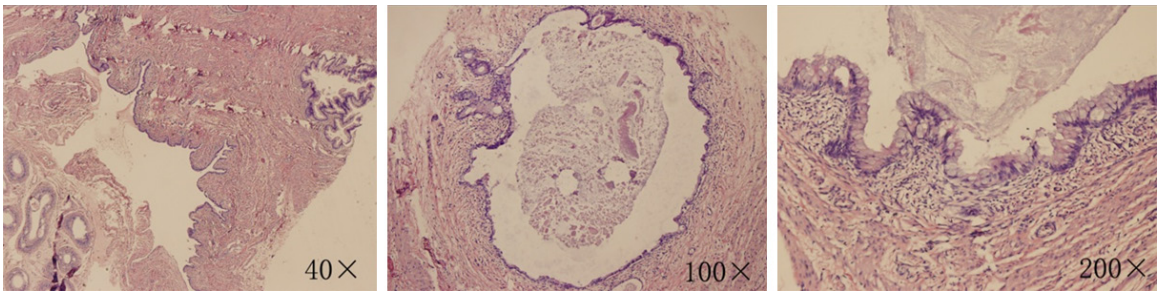

Figure 1. Different sizes of cystic structures could be observed in the right testis. The cystic cavities were surfacely covered with single or pseudostratified columnar epithelium, and filled with mucous secretions.

Table 1. Clinical and pathological characteristics of patients with primary testicular mucinous cystadenoma

NO./Age (Yrs)	Size (cm)	Side	Tumor markers				Follow-up (months)	IHC						
			AFP	HCG	CEA	LDH		MUC2	MUC6	MUC5AC	CK20	Chromogranin A	Vimentin	Ki67
1/35	9×6×6	R	-	-	-	NA	8	+	NA	-	NA	+	-	NA
2/55	4×2×2	R	-	-	-	-	5	+	-	+	+	-	NA	+
3/43	4.6×3.5×3.1	R	-	-	NA	NA	30	NA	NA	NA	NA	NA	NA	NA
4/39	3.0×2.5×3.09	R	-	-	NA	-	12	NA	NA	NA	+	+	NA	NA
5/54	3.1×2.3×2.0	L	-	-	NA	NA	3	NA	NA	NA	NA	NA	NA	NA
6/21	3.7×3×2.8	R	-	↑	NA	↑	6	NA	NA	NA	NA	NA	NA	NA

R: right; L: left; AFP: alpha fatal protein; HCG: human chorionic gonadotropin; CEA: carcino-embryonic antigen; LDH: lactate dehydrogenase; IHC: immunohistochemistry; CK: cytokeratin; NA: not available.

A right inguinal radical orchiectomy was then carried out. An ashen mass with multiple cysts sized about 3.7×3×2.8 cm could be observed from section of specimen. The mass was filled with clear liquid and the border between mass and normal testicular tissue was explicit. Further, the pathology of this mass reported that mucinous cystadenoma without invasion of spermatic cord (**Figure 1**).

Above all, no recurrence was observed in the follow-up of 6 months.

Literature review

To date, less than 30 patients have been diagnosed as testicular or epididymal mucinous tumor from the research of English literature [6], while only five patients were reported as PTMC. The medium age of these patients were 43 years (range: 35-55 years). Therefore, our case was the youngest one. Furthermore, lesions in four patients were located at the right side, while only one case caught a left lesion. And, all patients were reported with satisfactory results without recurrence. However, some differences in tumor markers and immunohistochemistry could be found. The clinical and pathological characteristics of these patients were summarized in **Table 1**.

Discussion

There are several kinds of benign cystic neoplasms have been found in testes, such as ovarian-type epithelial tumors, Brenner tumor, cystadenoma of rete testis, mature cystic teratoma and adenomatoid tumor, from which PTMC, belongs to ovarian-type epithelial tumors, is extremely rare [3]. Further, according to histological manifestation of mucinous tumors locating at testes, it can be classified into three types of cystadenomas, borderline tumors and carcinomas [6].

Up to now, including our case, only six cases with PTMC were presented which made our understanding on this disease very limited. Three mainstream opinions were speculated for its origination: mullerian, mesothelial inclusions and germ cell derivations [5]. However, it is still controversial though more publications support a mullerian origin. From summarizing of all cases with PTMC, radical orchiectomy would be the most effective choice for this con-

dition, and currently satisfactory results had been achieved. However, the effectiveness of radiotherapy and chemotherapy was still absent, which made it worried for patients with bilateral lesions, recurrence or metastasis though no cases were reported.

Furthermore, radiological investigations can only be helpful to distinguishing primary or metastatic tumors, while ultrasonographic examination is often nonspecific for differentiating it from malignant tumors like teratoma [1]. This would be the reason that teratoma was suspected before surgery for our case.

The testicular mucinous tumors of ovarian-type epithelium present a similar morphological appearance with those of other locations, and may be histologically difficult to distinguish from other types of ovarian-type epithelial tumors, especially the serous subtype [7]. However, from the summarizing of pathological characteristics, markers for immunohistochemistry were not sensitive and specific enough to distinguishing these two subtypes.

In conclusion, PTMC is a very rare condition with benign behavior. However, to date, its etiology, epidemiology, diagnosis, treatment and prognosis are poor understood. We reported a youngest case with a satisfactory result at the follow-up of 6 months. Further studies focusing on its characteristics need to be carried out.

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Disclosure of conflict of interest

None.

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References

[1] de Lima MM Jr, de Lima MM, Granja F. Primary testicular mucinous cystadenoma: case report and literature review. Can Urol Assoc 2015; 9: E814-816.

[2] Nokubi M, Kawai T, Mitsu S, Ishikawa S, Morinaga S. Mucinous cystadenoma of the testis. Pathol Int 2002; 52: 648-652.

[3] Naito S, Yamazumi K, Yakata Y, Shono T, Hakariya H, Nakayama T, Nakashima M, Sekine I. Immunohistochemical examination of mucinous cystadenoma of the testis. Pathol Int 2004; 54: 355-359.

[4] Shimbo M, Araki K, Kaibuchi T, Kuramochi H, Mori I. Mucinous cystadenoma of the testis. J Urol 2004; 172: 146-147.

[5] Alasio TM, Borin J, Taylor K, Bar-Chama N, Unger PD. Intratesticular mucinous cystadenoma: immunohistochemical comparison with ovarian and colonic tissue. Arch Pathol Lab Med 2005; 129: 399-402.

[6] Funada S, Yoshida T, Ito M, Kono F, Segawa T. Primary borderline mucinous tumors of the testis: a case report and literature review. Case Rep Oncol Med 2015; 2015: 863745.

[7] Maruschke M, Schmidt W, Casper J, Hakenberg OW. Ovarian type surface epithelial carcinoma of the testis with delayed metastatic spread. Urol Int 2008; 81: 119-121.