

A CASE OF ORGANIZED SCROTAL HEMATOCELE WITH HIGH SERUM CA19-9 LEVEL

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A 67 year-old male consulted our department for examination a painless left scrotal mass accompanied by a continuously high serum carbohydrate antigen 19-9 level (CA19-9, 563.2 IU/ml). The patient underwent radical orchiectomy of the mass. The histopathological diagnosis was an organized hematocele, and left testicular tissue was found in the cyst wall. There was no evidence of malignancy in the cyst wall or cyst contents, and immunohistochemical analysis showed no CA19-9-positive cells. However, the CA19-9 level lowered to the normal range immediately after surgery. The patient's CA19-9 level has remained normal, with no recurrence of tumor to date. Considering the clinical course, we suspected the resected mass to have been the cause of the high serum CA19-9 level, and to our knowledge, this is the first case report of organized hematocele in the scrotum with a high CA19-9 level.

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Key words : Carbohydrate antigen 19-9, Scrotal mass, Organized hematocele

INTRODUCTION

CA19-9 (carbohydrate antigen 19-9), a Lewis-(a) blood group-related antigen, was described by Koprowski et al. in 1979 as a tumor-associated antigen in a colorectal cancer cell line¹⁾. In 1983, Del Villano et al. first established an assay against the serum soluble antigen²⁾. The clinical correlation circulating in human serum is a mucin with a molecular weight of 10,000 daltons³⁾. CA19-9 occurs in many human tissues: it has been detected in salivary glands, thyroid, lung tissue, mammary glands, liver tissue, exocrine pancreas, ovaries, endometrial tissue, gastrointestinal tract, the prostate and the seminal vesicles as well as in fetal tissue³⁾. Therefore its organ specificity is unlimited³⁾.

In recent years, CA19-9 has been widely applied in clinical practice as a tumor marker for numerous types of malignancies⁴⁾. It is particularly useful in diagnosis, monitoring and follow-up of patients with carcinoma of the exocrine pancreas⁴⁾. In other gastrointestinal tumors, especially in colorectal carcinomas, and in gastric cancer, CA19-9 has also shown to be an efficient tumor marker⁴⁾.

We report a patient with a large scrotal mass diagnosed as an organized hematocele and having a significantly elevated CA19-9 level.

CASE REPORT

A 67-year-old man was diagnosed with a high serum CA19-9 level (563.2 IU/ml) during a health examination at our hospital and was admitted for further examination. His left scrotal mass was enlarged and he consulted our department for

treatment and for elucidation of the relationship between his high serum CA19-9 level and the mass. Approximately 15 years before this examination, he had undergone surgery for left scrotal hydrocele and experienced re-swelling of the left scrotum 6 months post operation. It continued to enlarge to approximately 20 cm in diameter. However he felt no pain and did not seek medical attention. On investigation, we could not distinguish the left testis or epididymis from the mass by palpation and the surface was regular. There were no other abnormal findings such as pigmentations on scrotal skin. No pulsation could be palpable from outside and the surface was regular. After admission, a high serum CA19-9 level was observed persistently, but the serum levels of other tumor markers (α -fetoprotein, carcinoembryonic antigen, β -human chorionic gonadotropin) were within their normal range. Left scrotal ultrasound showed a large hyperechogenic tumor with nearly homogenous structure and smooth margins. Pelvic computed tomography revealed a large unilocular cystic lesion measuring 8×6×6 cm in the left scrotum, and there was no apparent nodule in the cyst wall. There was no abnormal uptake in Gallium scintigraphy. Because we could not exclude the possibility of a malignant tumor, especially testicular carcinoma, the tumor was resected by radical orchiectomy. The tumor was a unilocular cyst filled with approximately 200 ml of dark red purulent fluid (Fig. 1). Pathohistological diagnosis revealed an organized hematocele. The lumen of the cyst was covered with fibrin, and infiltrations of many macrophages containing iron and giant cells. There was an insignificant number of inflammatory cells.

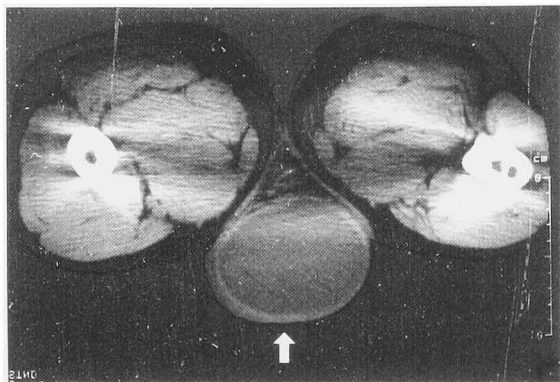
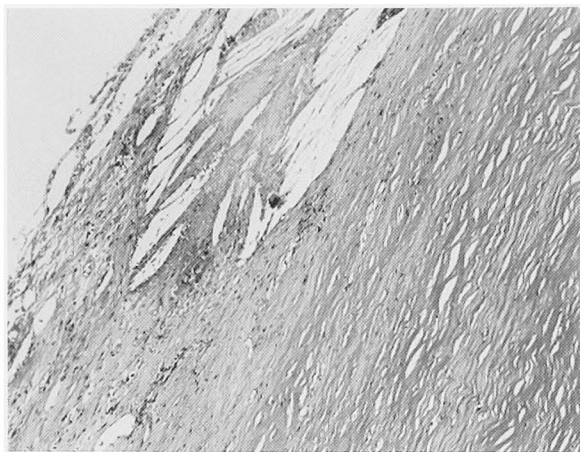
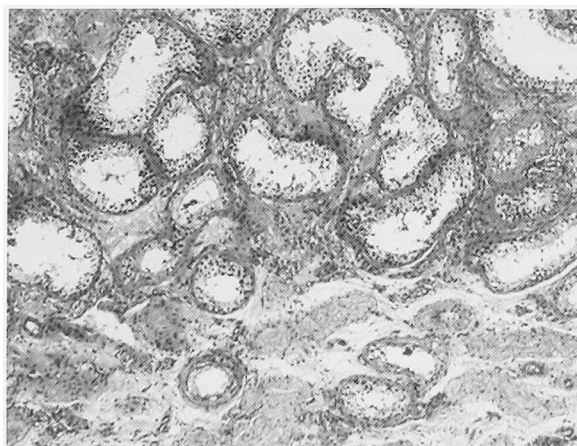


Fig. 1. Plain pelvic CT scan showing a cystic lesion in left scrotum (arrow).



A



B

Fig. 2. Histopathological examination of the tumor revealing organized hematocoele. The cyst wall is almost constituted of tunica vaginalis testis, and its intralumen is covered with fibrin precipitates and epithelial cells can not be found (A, H-E $\times 100$). Several choroiderol clefts can be seen within the fibrins. In a part of the cyst wall, left testicular tissue can be seen (B, H-E $\times 100$). Some seminiferous tubules are extant while others are atrophic.

A distorted left testis with some atrophic changes was seen in a part of the cyst wall (Fig. 2). We could see

no evidence of malignant cells in the cyst wall or fluid, and immunohistochemical analysis revealed no CA19-9-positive cells. The postoperative course was uneventful and soon after surgery, the serum CA19-9 level decreased immediately. Within 2 months, it lowered to within the normal range and to date, we have observed no re-elevation of serum CA19-9 level nor recurrence of the mass.

DISCUSSION

Unspecific elevations of CA19-9 have been described in various benign conditions⁴⁾. Very high levels have been detected, particularly in acute and chronic inflammations of exocrine pancreas⁴⁾.

Recent studies have shown that CA19-9 antigen is a specific ligand of E-secreting molecule that appears in the membrane of the endothelial cells of blood vessels by inflammatory stimulations of cytokines. The adhesion of cancer cells to the endothelium of blood vessels is an essential part of the mechanism of metastasis through blood circulation, and CA19-9 antigen plays an important role in this mechanism⁵⁾. It is suspected that a CA19-9-antigen-producing malignant tumor can spread more easily to other organs compared to non-antigen producing tumors⁵⁾.

In the field of urology, several cases of transitional cell carcinoma of the urinary tract with a high serum CA19-9 level have been reported⁶⁾, but Ohshio et al. reported that CA19-9 antigen is present in the normal mucosa of renal pelvis and normal tubules⁷⁾. Ito et al. also reported a case of hydronephrosis caused by a renal stone with a high serum CA19-9 level⁸⁾. Some cases of CA19-9-producing mixed gonadal germ cell tumors in testis have been reported⁹⁾. Increased levels of CA19-9 have been reported in many benign cystic lesions; for example, benign teratoma in various organs¹⁰⁾, bronchogenic cyst¹¹⁾, benign epithelial cyst of spleen¹²⁾, and liver cyst¹³⁾. In these cases, CA19-9 antigen expressing cells were identified immunohistochemically and high CA19-9 levels of the cystic fluid were often detected in them. However, we could not find CA19-9-antigen-expressing cells in our sample and strongly suspect that the scrotal mass was the cause of a high serum CA19-9 level considering the clinical course.

Muraki et al. reported a case of primary adenocarcinoma of tunica vaginalis testis expressing CA19-9 antigen¹⁴⁾.

To our knowledge, this is the first report on organized scrotal hematocoele associated with a high serum CA19-9 level. Unexpectedly, we could not find any CA19-9-positive cells in the present case. There might have been some CA19-9-producing cells in our samples, but they could not be detected immunohistochemically by our method. The fact that the CA19-9 level returned to the normal range after resection of the tumor suggests that the

continuous high serum level of CA19-9 derived from the organized scrotal hematocele. The present case supports the view that a high serum CA19-9 level may present not only in a malignant lesion, but also in a benign lesion. Attention should be paid when interpreting the significance of an abnormal level of CA19-9.

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高 CA19-9 血症を伴った陰嚢内器質化血腫の 1 例

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症例は67歳の男性で人間ドックにて、高 CA19-9 血症を指摘され、全身検索の結果、左陰嚢内の小児頭大の無痛性腫瘍との関連を疑われた。患者は15年前に左陰嚢水腫にて手術の既往があるがほどなく再腫大し放置していた。画像診断においては積極的に悪性腫瘍の存在を示唆するような所見はえられなかったが、持続して高 CA19-9 血症を認めたため高位精巣摘出術に準じて左陰嚢内腫瘍摘除術を行った。摘出腫瘍は内容暗赤色液体の単房性嚢胞であり壁の一部に圧排された左精巣を認めた。病理組織診断結果は器質化血腫であり、内容液の細胞診ともども悪性腫瘍細胞の存在は

確認されなかった。また、免疫組織化学的検索において標本中に CA19-9 陽性細胞を発見されなかったが術後よりただちに血清中 CA19-9 レベルの低下を認め、術2カ月後には正常範囲内に落ち着いた。以後、経過観察を行っているが腫瘍の再発や血清中 CA19-9 レベルの再上昇を認めていない。近年、良性疾患においても高 CA19-9 血症を伴いうる事が指摘されているが、本症例はそれを裏付ける1例であると思われる。

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