Ultrasonography Features of Adenocarcinoma Mimicking Cystadenoma of Rete Testis

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ABSTRACT
Background: Adenocarcinoma of rete testis is an extremely rare intra-scrotal extra-testicular neoplasm, this tumor usually occurs in men over 60 years of age but there are a few cases of younger patients described as well, has a wide age range of 8-91 years. Case Presentation: In this case, we report A 23-year-old Asian male patient with right scrotal swelling for the last 3 months. Scrotal ultrasound revealed an anechoic multiloculated cystic lesion mass in the right scrotum with the size of 11.89 cm x 7.73 cm lesion that fills almost the entire testicle. There are no solid parts, visible thickening of the scrotum, and visible fluid echo intensity outside the mass. Color Doppler showed a normal flow of blood signals within the mass. Pathological examination is also needed as a gold standard diagnosis, the results concluded that it was adenocarcinoma-type testicular carcinoma. Conclusion: This case highlights the utility of radiological findings, supported by other examinations to determine the diagnosis of Adenocarcinoma of rete testis.

Keywords: Adenocarcinoma; Scrotal ultrasound; Adenocarcinoma of rete testis; Cystadenoma of rete testis; Rete testis

INTRODUCTION
Adenocarcinoma of rete testis is an extremely rare intra-scrotal extra-testicular neoplasm with about 72 cases reported in the English language literature since its first description by Feek and Hunter in 1945 (1,2). This tumor usually occurs in men over 60 years of age but there are a few cases of younger patients described as well, has a wide age range of 8-91 years (1,4).

Most patients with adenocarcinoma of rete testis present with scrotal pain and/or swelling and frequently tumors are masked by a hydronephrotic, hematocoele, inguinocrotal hernia, and epididymitis (1). The prognosis of metastatic disease is very poor and there is no standard treatment strategy defined (3,4).

CASE REPORT
A 23-year-old Asian male presented with a gradually enlarging nonpainful right scrotal swelling and heaviness for 3 months duration. There was no history of cryptorchidism or trauma in the inguinocrotal region. On physical examination, the swelling was tender and non-translucent. The overlying skin was indurated. There was no palpable lymphadenopathy, while the penis and right scrotum are normal. The clinician suspected an underlying right testicular mass for which the patient underwent ultrasonography (USG) of the scrotum. Scrotal ultrasound (US) was performed and revealed an anechoic multiloculated cystic lesion mass in the right scrotum with the size of 11.89 cm x 7.73 cm lesion that fills almost the entire testicle. There are no solid parts, visible thickening of the scrotum, and visible fluid echo intensity outside the mass. Color Doppler showed a normal flow of blood signals within the mass (Figure 1). Meanwhile, on the left testis, the ultrasound results showed that the size of the left testicle was normal and there were no tumors or cysts. Microcalcifications were found in the left testicle. Color Doppler showed normal flow of blood signal (Figure 2). No enlargement of the right and left inguinal lymph nodes was found. Ultrasonography revealed that the mass was suspected to be a tumor, but the pathologic examination needed to be in addition determined.

This patient also had an abdominal ultrasound, tumor markers, hormone levels, and chest x-ray as pre-operative preparation and evaluation of the possible primary site of a tumor. The results of the ultrasound and chest x-ray were normal. No metastases were found in the liver and spleen. Serum alpha-fetoprotein, Lactate dehydrogenase, and beta-human chorionic gonadotropin levels were in the normal range.

The patient underwent a right radical orchiectomy. Pathology examination revealed that macroscopically the testicular tumor tissue measuring 13x13x11cm was brownish in color, the tissue lamellae appeared lobulated, and red fluid was coming out from it. Microscopically, it appears that some of the testicular have become tumors and invaded the surrounding stroma with the appearance of large cell nuclei and lots of cytoplasm. The results of the anatomical pathology concluded that it was adenocarcinomatype testicular carcinoma. (Figure 3).

FIGURE 2: Scrotal Ultrasound of the Left Testicle Shows Microcalcifications.
DISCUSSION
The rete testis is an anastomotic network of ducts that can sometimes be a site of both benign and malignant pathologic changes. Although rare, neoplasms arise and typically are both benign cystadenoma or malignant cystadenocarcinoma (5). Adenocarcinoma of the rete testis is a rare, highly aggressive tumor originating from the non-spermatogenic epithelium of the intratesticular excretory ducts (6). About 72 cases reported in the English language literature since its first description by Feek and Hunter in 1945. In general, testicular adenocarcinoma is found most often in Caucasians as much as 86%, and only 7% of cases have been reported in Asians (7). These tumors are commonly seen in middle-aged white men with a median age of 53 years, even though some literature said ages have varied from 17 to 91 years. The majority of symptoms in patients are a scrotal mass with diffuse enlargement of the testis. However, it is difficult to make a differential diagnosis with other testicular lesions, as rete testis adenocarcinoma also invariably presents with epididymitis, hydrocele, inflammatory lumps, or inguinal hernia (6,7). This patient also complained of the same thing with significant testicular enlargement within a period of 3 months. There are no specific clinical manifestations for this case so it requires supporting examinations.

Ultrasound has been shown to be a reliable and valuable tool in the diagnosis of scrotal abnormalities. This procedure is relatively cheap and noninvasive. In addition, it provides real-time imaging, reveals internal blood flow properties, causes little discomfort, and is easily repeatable, as well as being suitable for X-ray-sensitive organs as an ionizing radiation-free test. Ultrasound diagnostics are therefore recommended for confirming the presence of testicular masses (6).

Some literature mentions ultrasound images of adenocarcinoma are described as having clear fluid with no nodular or solid components and cysts of varying size. The great majority of testicular tumors are solid and predominantly hypochoic on sonographic examination and most have associated solid areas. This is supported by Tian et al, 2014 who explain that adenocarcinoma of the rete testis is typically located in the epididymis or testicular hilum, rather than the intratesticular region, as reported in the majority of the current literature. The majority of patients show other signs like hydrocele and echoic lesions on para-testicular regions. Nodular septations with cystic solid components have also been detailed as an unusual finding that varies from other pathological tumor types. In many cases, the lesions present as hypoechoic masses with poorly defined borders (5,6).

In our case, the sonographic features were predominantly the presence of multiple septations and cysts, and the absence of solid components which shows that these images are consistent with cystadenoma of rete testis (5). However, pathological examination is still the gold standard for diagnosis confirmation (6).

Pathology examination revealed that on gross examination the testicular tumor tissue measured 13x13x11cm was brownish in color, the tissue lamellae appeared lobulated, and red fluid was coming out from it. Microscopic examination revealed some of the testicular have become tumors and invaded the surrounding stroma with the appearance of large cell nuclei and lots of cytoplasm. The results of the anatomical pathology concluded that it was adenocarcinoma-type testicular carcinoma (Figure 3).
CONCLUSION

Based on those examinations we conclude that our case may meet the criteria cystadenocarcinoma of the testis, with US features and pathology examination, however, there are further examinations that need to be performed to rule out better diagnostic that is not performed on this case, such as immunohistochemical examination, we just exclude other possibilities of diagnosis based on tumor marker serum such as alpha-fetoprotein, lactate dehydrogenase and beta-human chorionic gonadotropin levels that shows normal result. US abdomen and chest x-ray also did not show evidence of metastatic disease.

Nochomovitz and Orenstein set up the exclusion standards for adenocarcinoma of the rete testis in 1984, Those are the absence of histologically similar extra-scrotal tumor that could be the primary site; tumor centered in the hilum of the testis; morphology incompatible with any other type of testicular or para-testicular tumor; and immunohistochemical exclusion of other possibilities (8). Even among strict criteria, erroneous diagnosis is common, requiring strict immunohistochemical guidelines to rule out mesotheliomas, serous adenocarcinomas of the ovarian type, and other germ cell tumors (9).

In conclusion, adenocarcinoma of the rete testis is an extremely rare tumor type with a poor prognosis. Sonography is the most promising tool for early diagnosis and increased case examples providing sonographic tumor observations must be presented to achieve an improved rate of diagnosis (6).

REFERENCES


