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Case Report

ADENOMATOID TUMOUR OF THE EPIDIDYMIS AND REVIEW OF LITERATURE

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ABSTRACT

Adenomatoid tumour(AT) of paratesticular region is a rare benign mesothelial neoplasm and mainly involves the epididymis. A definitive preoperative diagnosis is often not possible and it is difficult to differentiate clinically and radiologically from malignancy which may end up in unnecessary orchiectomy. Histopathological examination along with immunohistochemistry is diagnostic and conclusive for the diagnosis of adenomatoid tumour.

Herein, we reported a case of seventeen-year old male who presented with intrascrotal mass suspicious of malignancy.High inguinal orchiectomy was performed. Histopathology and immunohistochemistry confirmed the diagnosis of epididymal adenomatoid tumour.

INTRODUCTION

A 17-year old boy presented with a painful, tender swelling in the right hemiscrotum since 10 days. He was initially treated at local facility with scrotal support, analgesia and antibiotics for one week before presenting at our hospital. Clinical examination revealed a firm, mildly tender intrascrotal mass. Right testis could not be separately palpated. Scrotal skin, spermatic cord, inguinal region and left testicle were normal.

Ultrasonograpgy confirmed a solid, heterogenous lesion occupying the right hemiscrotum arising from the right epididymal area and adjoining testis with lack of clear delineation as to the exact site of origin. Preoperative laboratory investigations including blood chemistry studies were within normal limits. Serum levels of tumour markers [beta HCG (beta subunit of human chorionic gonadotrophin), AFP (alfa fetoprotein) and LDH (lactate dehydrogenase) were within normal limits. Our first clinical impression was a probable germ cell neoplasm.

Subsequently the patient underwent a total right orchiectomy-epididymectomy by the inguinal approach and the specimen was sent for histopathological examination. Gross specimen revealed a well circumscribed mass (size 8 x 5 x 4.5 cm.) mainly arising from the body of epididymis. Cut section was dark brown with hemorrhagic areas without involvement of testicular parenchyma [Fig.1]. Histological examination of the mass revealed multiple, irregular spaces (vacuolated cytoplasm) lined by cuboidal and flettened epithelial cells surrounded by varying amount of fibro-connective tissue stroma [Fig. 2].

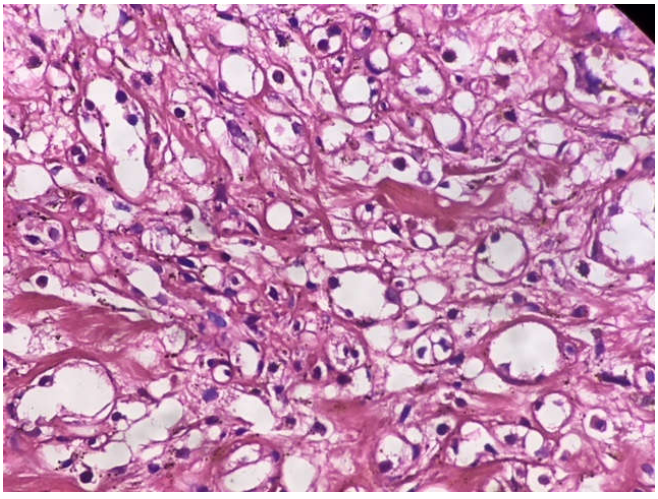

Figure 2 Typical histopathological picture showing vacuolated spaces with cuboidal and flattened cells and connective tissue stroma

Immunohistochemical evaluation was positive for tumour markers HMBE 1, pan cytokeratin, Vimentin, smooth muscle Actin and Calretitin thus documenting the diagnosis of adenomatoid tumour. Postoperative course was uneventful. During 12 months of follow up, patient remains asymptomatic and without any sign of recurrence.

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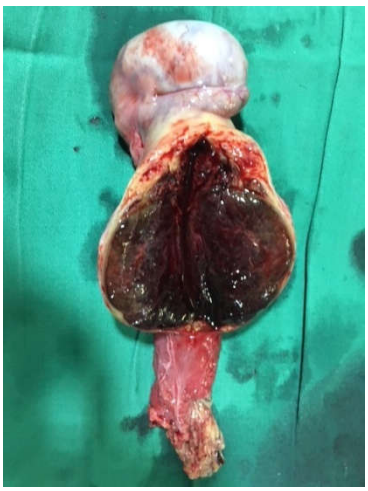

Figure 1 Cut section showing large mass arising from epididymal body

DISCUSSION

Adenomatoid tumours of the paratesticular region are rare benign tumours of mesenchymal origin. Epididymal epithelial tumours are a rare subvariant of paratesticular tumours with adenomatoid tumours being the most common followed by the papillary cystadenoma and the leiomyoma [1]. Most of these usually arisein the epididymis while only 14% arise from testicular tunica[2]. Adenomatoid tumours can also be located in the spermatic cord, prostate, ejaculatory ducts and in female genital tracts such as fallopian tube, uterus and ovarian area.In a study by Beccia *et. al*. [3], 75% of epididymal tumours are benign. 73% of benign tumours are adenomatoid tumours, 11% leiomyoma and 9% papillary cystadenoma. Other benign entities like angioma, lipoma, dermoid cysts, fibroma, hamartoma, teratoma and cholesteatoma comprise the rest 7%.

Adenomatoid tumours usually present either as an incidental finding or a slow growing scrotal mass in patients between 30 and 50 years of age [4] and usually less than 2 cm. in size. It is usually slow growing, painless, intrascrotal mass for several years and rarely may have pain.It is uncommon in children and rarely more than 3 cm.[5].Usually there is no correlation between the tumour and any previous scrotal inflammation or trauma. The naked eye appearance is that of a grey, yellow or white tumour with a glistening surface[6]. Typically, an adenomatoid tumour is represented by cuboidal cells with vacuolated cytoplasm and with large gaping spaces (pseudotubular spaces) [angiomatoid pattern] surrouned by collagenous stroma and muscle fibres. Three basic patterns are: tubules, cords and nests [7]. Peculiar thread like bridging strands (TBS) crossing the pseudotubular spaces (ultrastructurally they are formed by apposition of attenuated cytoplasm of two adjacent mesothelial cells) are a consistent morphologic feature of adenomatoid tumours [8,9]. Lymphoid aggregates (particularly towards the periphery) are found in almost all male adenomatoid tumours. Signet ring cells and lipoblast like cells are not uncommon and can mimic malignancy [8,10]. Manuel Nistal *et. al*. [11] described AT as a peculiar variety of benign mesothelioma. He highlighted the characteristics of tubules and the cells lining those tubules as : (a) acid mucopolysaccharides in the tubular lumen (reinforcing the hypothesis of mesothelial origin), (b) presence of abundant microvilli in the tubular lumen, junctional complexes, enlargement of intercellular spaces and bundles of microfilaments in the cytoplasm, (c)epithelial cells with well-developed Golgi complex and secretory vesicles and (d) existence of microvilli in the intracytoplasmic spaces in the epithelial spaces (an interesting feature).

Immunohistochemically, Adenomatoid tumour is positive for markers such as Calretinin, CK (AE1/AE3), EMA, Cam 5.2, CK 5/6, CK7, Vimentin, WT1 and HBME 1. Calretinin (a calcium binding protein of S-100 protein family expressed both in cytoplasm and nucleus) also shows its mesothelial origin. Presence of calretinin confirms adenomatoid tumour but does not exclude malignancy [7]. Absence of AFP, LDH, CEA, beta-HCG and p53 can reasonably exclude malignancy. This type of immunohistochemical profile of paratesticular tumour is strongly supportive of a mesothelial cell origin [12].

The clinician frequently faces the dilemma of differentiating the many benign scrotal conditions presenting as mass from the more serious and omnious testicular or paratesticular malignant neoplasm. Epididymal AT also creates the same problem. It is very difficult to answer few questions: Whether preoperatively epididymal AT could be diagnosed with certainty? If so, whether conservative treatment or surgical exploration? And if surgery is chosen, whether local excision or extensive surgery or orchiectomy?

In contrast to testicular tumours (where 95% of lesions are malignant and inguinal exploration with often orchiectomy is justified), most of the epididymal tumours are benign.In numerous cases, similar clinical signs and imaging studies make it difficult to distinguish adenomatoid tumours from malignant intratesticular neoplasm and often diagnosis of adenomatoid tumour results from incidental pathologic findings following orchiectomy [4,13]. Hence it is often argued that it is sufficient in most of the times to have *a scrotal exploration and local excision* to salvage the testicular endocrine and cosmetic value.

However, it is not easy to differentiate between AT or malignancy (25% of epididymal tumours are malignant and of these, 44% are sarcomas). There is high incidence of malignancy of epididymal tumours and which are often misdiagnosed [3]. Hence, there should always be a high index of suspicion and prompt multidisciplinary discussion to direct investigations and it is better to have *exploration by inguinal route and intraoperative assessment* [6,14]. Suspicion arises when it is found in younger age group or gross examination reveals suspected testicular involvement.

Altaffer *et. al*.[15] concluded that: (a) any suspected intratesticular mass should be explored via an inguinal approach, (b)all apparent inflammatory masses should be followed closely as they resolve and the patient should be explored if the mass should seem to be intratesticular at any time, and (c)patients should be informed preoperatively that there is a good chance that either the testicle will have to be removed or that they will need some other operative procedure.

An ultrasonogram may show hypo, iso or hyperechoiclesion and should be supported by clinical features. Frozen section biopsy is often advised before extensive excision or orchiectomy when diagnosis is in doubt [16,17]. Absence of severe cytologic atypia and of high mitotic rate along with lack of infiltrative pattern of growth are all features favouring a

diagnosis of AT. They opined that preoperative normal serum tumour markers combined with intraoperative frozen section histology, when indicated, can prevent unnecessary orchiectomy [18]. According to Rubenstein *et. al*. [19] when the diagnosis is uncertain but a benign process is suspected, surgery should be performed through an inguinal incision with control of the cord. Mass can be explored and can be sent for frozen section biopsy. When a benign lesion is encountered, testis sparing surgery should be performed.

But another view is that a diagnosis based on frozen section is unreliable when it has to be made in circumstances where there is less time for thought [6].Testicular ultrasound can not reliably differentiate benign from malignant intrascrotal pathology. Hence testicular preservation based on ultrasound findings should be done in extremely select cases only [20]. In our case, gross examination was highly suspicious of malignancy and we straightway went for orchiectomy.

Hence, after review of relevant literatures, we can reasonably conclude that aggressive operative approach should be undertaken in clinically suspected epididymal AT when it meets the following criteria:

1. Younger age group patients (infancy to childhood): risk of paratesticular rhabdomyosarcoma[8].
2. Large tumour>1 cm.
3. Firm, suspected inflammatory mass not resolving to antibiotics
4. Heterogenous, non-circumscribed mass on ultrasound without clear delineation from testis
5. Intraoperative appearance highly suspicious
6. Presence of signet ring cells on histopathology

Overall decision to be taken based on thorough analysis of clinical features, tumour markers, imaging studies and intraoperative findings [2].

AT of the epididymis is a distinctive clinical entity which is hard to distinguish from malignancy and diagnosis is often confirmed after total orchiectomy. A comprehensive clinical approach with clinical history, routine light microscopy and immunohistochemical markers are crucial for proper diagnosis.

***Abbreviations**:* AT- Adenomatoid tumour

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Conflicts of interest

Authors declare that there is no conflict of interest regarding the publication of this article

Consent

Patient is sufficiently anonymized in this article and our hospital guideline do not need written informed consent for anonymized case report

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