

Testicular Sex Cord Stromal Tumor with Annular Tubules in Cryptorchidism: A Case Report

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Abstract

Sex cord stromal tumors with annular tubules (SCTAT) are rare neoplasms typically found in the ovary and are often associated with Peutz-Jeghers syndrome. However, SCTAT arising in testicular tumors are exceedingly rare, only with five cases documented in literature. These tumors display unique histological characteristics. We report an unusual case seen in a 25 year old male who presented with bilateral undescended testes. Right sided orchidectomy with left sided orchidopexy was done. Histopathological examination revealed SCTAT with diffuse cytoplasmic positivity for inhibin and calretinin on immunohistochemistry. The patient exhibited no features of Peutz-Jeghers Syndrome. He is currently under surveillance. This case underscores the significance of SCTAT in testicular tumors, particularly in patients with a history of undescended testes.

Keywords: Undescended testis; SCTAT; Immunohistochemistry

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Introduction

Sex cord tumors with annular tubules (SCTAT) are a rare and distinct subset of sex cord stromal neoplasms, initially characterized by Scully in 1970. SCTATs comprise less than 1% of ovarian sex cord stromal tumors and are frequently associated with Peutz-Jeghers syndrome. While the ovarian form is better understood, similar morphology in testicular tumors is a rarity, with only 5 cases reported. [1] The paucity of literature of the SCTAT in testis can lead to missed diagnosis in undescended testis. These tumors depict characteristic morphology exhibiting ring shaped tubules containing central hyaline material and nuclei arranged in a palisading pattern. [2, 3]

Case Report

A 25 year old male presented with bilateral undescended testes. He denied any systemic symptoms or signs of hormonal imbalances. He underwent right sided orchidectomy with left sided orchidopexy. Gross examination of the right orchidectomy specimen appeared unremarkable with no discrete nodules or gray white areas identified. Microscopic analysis showed a tiny focus of neoplastic cells (0.4cm) arranged in characteristic annular tubules enclosing central eosinophilic hyaline bodies, with palisading nuclei around the periphery. The tumor cells were tall with ample amount of cytoplasm and basally located slightly elongated bland nuclei. Surrounding seminiferous tubules were atrophied containing sertoli cells attached to the thickened basement membrane and interstitium shows leydig cell hyperplasia.(Fig 1) No significant mitotic activity was noted. Immunohistochemistry showed strong cytoplasmic activity for inhibin and calretinin,(Fig 2) markers indicative of sex cord stromal differentiation. The tumor was negative for PLAP, CD117, thereby ruling out a germ cell tumor component. The patient exhibited no mucocutaneous pigmentation, gastrointestinal polyps, or family history of PJS. Genetic testing for PJS and follow up was advised.

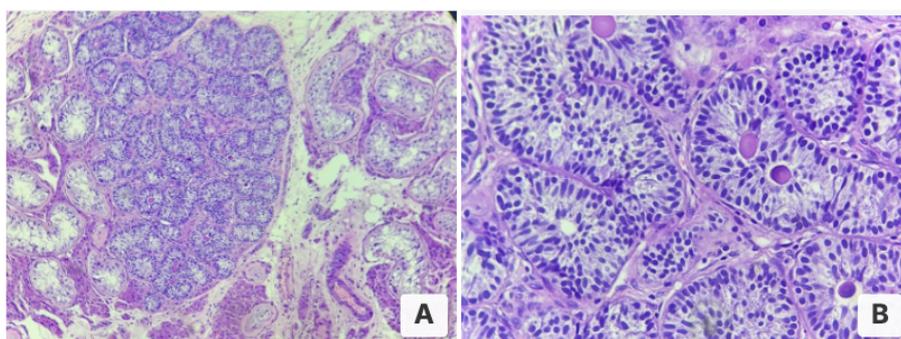


Figure 1: H&E stain (A) showing tiny focus of tumor with surrounding Leydig cell hyperplasia (100X). B: A complex annular tubular pattern consists of cells arranged around hyaline bodies (400X).

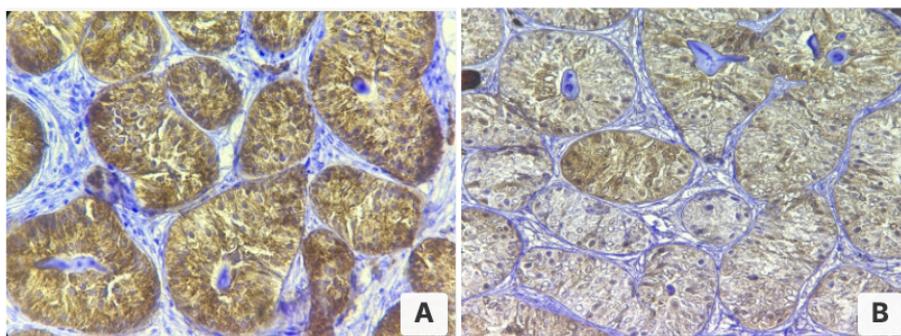


Figure 2: Immunohistochemistry. Tumor cells show intense cytoplasmic positivity for calretinin (A) and inhibin (B).

Discussion

SCTAT is a rare and histologically distinctive neoplasm that may exhibit features overlapping with other sex cord-stromal tumors, such as Sertoli cell or granulosa cell tumors. Its hallmark microscopic features include ring-like tubules surrounding hyaline material with peripheral nuclear palisading [2, 5, 6]. SCTATs are primarily found in the ovaries and are frequently associated with PJS, particularly when bilateral and multifocal. In contrast, sporadic or nonsyndromic SCTATs are usually unilateral, larger, and may show a slightly increased potential for malignant behavior [3, 9] and requires careful vigilance.

Cases of testicular SCTAT are extremely rare, with only a few published reports, including a notable case by Arora et al., which described a similar tumor in the absence of PJS [4]. Our case closely mirrors this, both in terms of morphology and immunoprofile. A critical component of diagnosis lies in differentiating SCTAT from other testicular neoplasms. The negative expression of germ cell markers such as PLAP and CD117 helped rule out seminomatous or non-seminomatous germ cell tumors [7]. Instead, the strong expression of inhibin and calretinin supported a sex cord-stromal origin.

Notably, the association of this tumor with bilateral undescended testes raises the question of a possible developmental predisposition, although no direct syndromic correlation was identified. As the long-term behavior of testicular SCTAT remains uncertain, especially in nonsyndromic contexts, close follow-up is warranted [10]. NCCN guidelines recommends 3 monthly serum markers and 6 monthly imaging studies in the initial first year. [11]

Conclusion

This case contributes to the limited body of literature on SCTAT-like tumors in the testis and is, to our knowledge, among the few reported in association with bilateral undescended testes. Recognition of its characteristic histological and immunohistochemical features is essential for accurate diagnosis and management as these subjects require follow up and regular monitoring apart from surgery. Although rare, SCTAT should be considered in the differential diagnosis of sex cord-stromal tumors of the testis, especially in patients with prior gonadal anomalies.

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