



## Original contribution

# Primary signet ring stromal tumor of the testis: a study of 13 cases indicating their phenotypic and genotypic analogy to pancreatic solid pseudopapillary neoplasm<sup>☆, ☆ ☆</sup>



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Received 14 April 2017; revised 11 June 2017; accepted 4 July 2017

**Keywords:**

Testis;  
Pancreas;  
Primary signet ring stromal tumor;  
Solid pseudopapillary neoplasm;  
Analogue

**Summary** Primary signet ring stromal tumor of the testis (PSRSTT) is an extremely rare tumor described only twice in the literature. Pancreatic-analogue solid pseudopapillary neoplasm (SPN) of the testis is a recently reported entity with morphological overlap with PSRSTT. We reviewed our files to find all cases of PSRSTT to better characterize this entity. We studied 13 cases of PSRSTTs using histological, immunohistochemical (IHC), and molecular genetic methods and compared the results with pancreatic SPN. Grossly, the size of PSRSTTs ranged from 0.5 to 2 cm (mean 1.1). Microscopically, PSRSTTs predominantly showed a proliferation of low-grade epithelioid cells containing characteristic cytoplasmic vacuole dislodging the nucleus (signet ring cells) separated by fibrous septa into trabeculae and nests. The immunoprofile was characterized by immunoreactivity for  $\beta$ -catenin, cyclin D1 (nuclear positivity for both antibodies), CD10,

<sup>☆</sup> Competing interests: The authors have no conflict of interest to disclose. Neither ethics approval nor informed consent was required for our study.

<sup>☆☆</sup> Funding/Support: This study was partly supported by the National Sustainability Program I (NPU I) Nr. LO1503 and by the grant SVV–2017 No. 260 391 provided by the Ministry of Education Youth and Sports of the Czech Republic, Prague, 11800, Czech Republic.

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vimentin, galectin-3, claudin 7,  $\alpha$ -1-antitrypsin, CD56, and neuron-specific enolase and negativity for chromogranin, inhibin, calretinin, SF-1, NANOG, OCT3/4, and SALL4. In some cases, the IHC panel was restricted because of a limited amount of tissue. Molecular genetic analysis revealed mutations within exon 3 of the *CTNNB1* encoding  $\beta$ -catenin in all analyzable cases. Based on histological similarities between pancreatic SPN and PSRSTT and their identical IHC and molecular genetic features, we assume that both neoplasms share the same pathogenesis, and thus, PSRSTT can be considered as a testicular analogue of pancreatic SPN.

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## 1. Introduction

Primary signet ring stromal tumor of the testis (PSRSTT) was first described in 2005 by Michal et al [1] and subsequently by Kuo et al [2] as single case reports. Microscopically, both tumors were characterized by the proliferation of low-grade epithelioid cells containing characteristic cytoplasmic vacuole dislodging the nucleus to the periphery of the cells (signet ring cells), which were separated by fibrous septa resulting in a trabecular and/or nested architecture [1]. Pancreatic solid pseudopapillary neoplasm (SPN) is a rare tumor with uncertain histogenesis traditionally encountered in the pancreas [3]. However, recent small series and several case reports have described an ovarian tumor identical to that seen in the pancreas [4-8], and most recently, a pancreatic analogue solid pseudopapillary neoplasm of the testis (PA-SPN) has been described by our group [9]. Microscopically, PA-SPN consisted of 2 distinct components: a signet ring cell component histologically identical to that seen in the PSRSTT blending with a component identical to pancreatic SPN by showing a solid (and in minor parts also pseudopapillary) component comprised by poorly cohesive low-grade cells with eosinophilic cytoplasm. The purpose of this study is to morphologically, immunohistochemically, and molecular genetically investigate 13 cases of PSRSTT and compare their features with pancreatic SPN and with 1 published case of PA-SPN [9] to establish their possible pathogenetic relationship.

## 2. Materials and methods

Cases cross-matching the keywords *testis, signet ring stromal tumor, unclassified sex cord tumor, male adnexal tumor of probable Wolffian origin, and Sertoli cell tumor, benign, NOS* were retrieved from the Plzen tumor registry; they came from the period 1993-2017. Additional cases were retrieved from the routine and consultation files of the authors. Upon re-evaluation, 13 cases of primary testicular tumors that fulfilled the diagnostic criteria for this study were selected. The clinical information was extracted from the registry records, and follow-up data were obtained from attending clinicians. In all but 2 cases, paraffin blocks or unstained reserve slides were available for the study. To compare PSRSTT with SPN, we reviewed 20 pancreatic cases from the Plzen tumor registry. For conventional microscopy, tissues were fixed in formalin, routinely processed, and stained with hematoxylin-eosin.

## 2.1. Immunohistochemistry

The immunohistochemical (IHC) analysis was performed using a Ventana BenchMark ULTRA (Ventana Medical System, Inc, Tucson, AZ). The list of antibodies and the basic technical specifications are summarized in Table 1. Because of the limited amount of tissue blocks or reserve slides, the utilization of the entire immunohistochemical panel was restricted. The following stains were performed on most cases:  $\beta$ -catenin, CD10, CD56, neuron-specific enolase (NSE), inhibin, calretinin, S100, and OSCAR. These additional stains were applied in only a few of them: vimentin, synaptophysin, chromogranin, SF-1, OCT3/4, SALL4, NANOG, cyclin D1, AE1/3, galectin-3, MIB-1, claudin 5, claudin 7, and  $\alpha$ -1-antitrypsin. Antibodies were visualized using the enzymes alkaline phosphatase or peroxidase as detecting systems (both purchased from Ventana Medical System, Inc).

## 2.2. Molecular genetic analysis of the $\beta$ -catenin gene

Mutational analysis of exon 3 of the *CTNNB1* gene was performed via polymerase chain reaction and direct sequencing as described previously [10].

## 3. Results

### 3.1. Clinical features

The clinical features are summarized in Table 2. The age of the patients at the time of diagnosis ranged from 23 to 58 years (mean 39, median 35). Follow-up was available for 5 patients. One patient died of unrelated disease (prostatic adenocarcinoma). The remaining patients were alive and well without progression, recurrence, or evidence of metastatic disease. None of the patients had a tumor in the pancreas and/or suffered from familial adenomatous polyposis.

### 3.2. Gross and microscopic findings

Grossly, the tumors were predominantly well circumscribed and encapsulated, solid, and gray in color. No cystic or necrotic foci were noted. The size of the tumors ranged from 0.5 to 2 cm (mean 1.1, median 1). Twelve of 13 tumors were located in the testis (Fig. 1A); the remaining tumor was paratesticular. All

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