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## LETTER TO THE EDITOR

Operational Andrology

# Successful management of an asymptomatic bilateral synchronous testicular carcinoid tumor with a testicular-sparing surgery

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Dear Editor,

Carcinoid tumors have been described in a large range of organs, but they most commonly involve the gastrointestinal and bronchopulmonary systems.<sup>1</sup> These tumors of the testis are rare entity comprising <1% of all testis tumors and can arise as a metastasis from an extratesticular primary as a part of a teratoma or as primary tumor.<sup>2</sup> Patients affected by carcinoid tumors present symptoms of carcinoid syndrome, such as diarrhea, flushings, and bronchospasm.<sup>3</sup> We report a first case described in scientific literature of primary bilateral synchronous testicular carcinoid tumor that does not associated with carcinoid syndrome. From this publication, at the time of writing, PubMed research conducted employing keywords (“testis,” “carcinoid tumors,” and “bilateral”) revealed one case report of primary bilateral testicular carcinoid tumor associated with carcinoid syndrome and treated with bilateral orchiectomy.<sup>4</sup> In October 2014, we managed a similar clinical condition in a 52-year-old Caucasian patient presented at our Department of Urology for ultrasound check of two bilateral epididymal cysts that for about 2 months gave mild pain and discomfort. There was no evidence of fever, hematuria, and dysuria or symptoms from the lower urinary tract. His medical history was unremarkable and he denied ever being infected with a sexually transmitted disease, trauma, or systemic symptoms. On physical examination, we found a tender mass in both epididymis; it adhered to the testis with regular surface was palpated. The overlying skin was completely intact with no erythema. Examination revealed no signs of lymphadenopathy in the groin region and a mild degree of varicocele on the left side. The patient did not demonstrate any laboratory signs of inflammation (white blood cells, C-reactive protein). Laboratory tests (complete blood count, including platelets, prothrombin, partial thromboplastin levels, urinalysis) were within normal. Plasma levels of  $\beta$ -subunit human chorionic gonadotropin ( $\beta$ -HCG),  $\alpha$ -fetoprotein (AFP), and lactic dehydrogenase (LDH) were within normal ranges.

A scrotal ultrasound, using a multi-frequency linear probe (GE Logiq 7 Healthcare, Milwaukee, WI, USA), revealed a cystic anechoic lesion lying on the border between epididymal head and upper pole of both testes with dimensions of 25 mm  $\times$  17 mm and 16 mm  $\times$  12 mm for right and left epididymis, respectively. The testicular

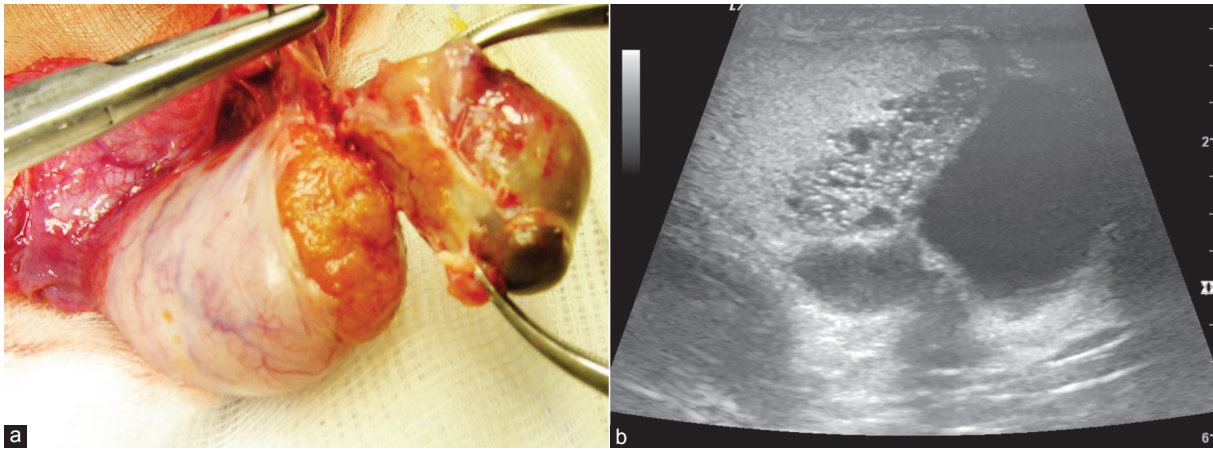
parenchyma immediately adjacent to the cysts showed decreased echogenicity compared with the parenchyma elsewhere with multiple microcysts communicating with solid lesions of mixed echogenicity, highly suspicious for malignancy. Computed tomography (CT) of the chest, abdomen, and pelvis showed no abnormal findings. In agreement with the patient, we decided to perform a bilateral testicular inguinal exploration. When a nodular area was found to be different from the surrounding tissue and to correspond in dimensions, site, and aspect to the one indicated by ultrasound, it was excised and sent for frozen section examination (FSE). A bilateral testicular-sparing surgery was performed (**Figure 1**). Resection margins were free. The lesions were well-circumscribed, mainly solid with an ill-defined cystic area filled with straw-colored fluid, measuring 16 mm  $\times$  10 mm (right testis) and 11 mm  $\times$  8 mm (left testis). Histologically tumors were composed of pleomorphic cells in a pseudoglandular and nested pattern. The tumor cell nuclei had granular chromatin, surrounded by eosinophilic cytoplasm. No teratomatous elements or other germ cell were detected. Immunohistochemical expression of CD56 and chromogranin A were highly positive in tumor cells. The patient was discharged in the second postoperative day. At 1-year follow-up, scrotal ultrasound showed normal testicles and CT showed no signs of recurrence.

In 1930, Cope<sup>5</sup> described the first case of a carcinoid tumor located in the testis which was a gastrointestinal tumor. In 1954, Simon *et al.*<sup>6</sup> reported the first case of primary testicular carcinoid. Subsequently, around 160 cases of carcinoid tumors in the testis have been reported. These cases can be divided into three groups: primary pure testis carcinoid tumors (about 70% of patients), carcinoid tumors associated with teratoma (about 20% of patients), and carcinoid metastasis to the testis (about 10% of patients). Moreover, patients with these tumors present rarely testicular tenderness, hydrocele, or epididymal cyst as our case on physical examination. In a literature review reported by Stroosma and Delaere,<sup>7</sup> 16% of the patients affected by testis carcinoid tumor had symptoms such as diarrhea, sweating, palpitations, headache, flushing, and bronchoconstriction. These symptoms occur when serotonin, produced by the tumor, is released into the systemic circulation. Serotonin is metabolized to 5-hydroxyindoleacetic acid (5-HIAA) and excreted through urine; the absence of 5-HIAA in 24 h urinary dosage prior surgery in our patient may explain the lack of carcinoid syndrome. Although most testicular carcinoids behave in an indolent manner, 10%–15% metastasize to

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**Figure 1:** The lesions were well-circumscribed, mainly solid with an ill-defined cystic area. **(a)** Carcinoid tumor testis is identified and removed. **(b)** Scrotal ultrasound of the right testis in sagittal section shows testicular parenchyma adjacent to the epididymal cyst with multiple microcysts communicating with solid lesions of mixed echogenicity.

lymph nodes, liver, skin, and skeletal system, occasionally many years after orchiectomy.<sup>1,3</sup> In literature, radical orchiectomy is the treatment of choice of carcinoid testis tumor and is curative for testis-confined.<sup>7</sup> A retroperitoneal lymphadenectomy is indicated when this tumor is a component of teratoma. Adjuvant chemo- or radio-therapy have little benefit on these tumors.<sup>2</sup> Therefore, to our knowledge, this is the first case of a synchronous bilateral primary testicular carcinoid tumor treated with a bilateral testicular-sparing surgery. The extensive use of high-frequency testicular ultrasound has led to the incidental detection of an increasing number of small, asymptomatic, nonpalpable testicular masses. Ultrasonography constitutes an excellent diagnostic method in the diagnosis of testicular masses with diameters of between 0.5 cm and 1.5 cm.<sup>8</sup>

Hopps and Goldstein,<sup>8</sup> who performed intraoperative ultrasound-guided localization and the subsequent surgical excision of small testicular tumors, proposed surgical exploration for nonpalpable, hypoechoic testicular masses. They reported on four patients treated using this approach, concluding that this type of surgery provided the opportunity to remove benign masses as well as malignant lesions with appropriate margins in selected cases. The concept of testicular-sparing surgery certainly follows a logical progression for neoplasm management that we have successfully observed in a variety of other tumors including renal and breast cancers. Until a short time ago, all testicular tumors were virtually treated with radical orchiectomy; however, testicle-sparing surgery is now advocated especially for bilateral and/or multiple lesions or in monorchid patients.<sup>9</sup> The benefits of testicle-sparing surgery include the improvement of the patient's overall quality of life, fertility, endocrine function, and the avoidance of the negative cosmetic effects of radical orchiectomy.<sup>10</sup> However, the role of the uropathologist is to establish the diagnosis of malignancy and resection margins of the tumor in FSE, taking into consideration the clinical information, tumor markers, and ultrasonographic results.

In this case report, a close collaboration between pathologist and urologist was possible conservative management without compromise oncological efficacy in the treatment of these tumors.

#### COMPETING INTERESTS

The author declares no competing interests.

#### REFERENCES

- Mazzucchelli R, Morichetti D, Lopez-Beltran A, Cheng L, Scarpelli M, *et al.* Neuroendocrine tumours of the urinary system and male genital organs: clinical significance. *BJU Int* 2009; 103: 1464–70.
- Wang WP, Guo C, Berney DM, Ulbright TM, Hansel DE, *et al.* Primary carcinoid tumors of the testis: a clinicopathologic study of 29 cases. *Am J Surg Pathol* 2010; 34: 519–24.
- Wolf M, Wunderlich H, Hindermann W, Gajda M, Schreiber G, *et al.* Case report: primary carcinoid tumor of the testicle without metastases in combination with testicular atrophy and testosterone deficiency. *Int Urol Nephrol* 2006; 38: 625–8.
- Son HY, Ra SW, Jeong JO, Koh EH, Lee HI, *et al.* Primary carcinoid tumor of the bilateral testis associated with carcinoid syndrome. *Int J Urol* 2004; 11: 1041–3.
- Cope Z. Metastasis of an argentaffin carcinoma in the testicle. *Br J Urol* 1930; 2: 268–72.
- Simon HB, McDonald JR, Clup DS. Argentaffin tumor (carcinoid) occurring in a benign cystic teratoma of the testicle. *J Urol* 1954; 72: 892–4.
- Stroosma OB, Delaere KP. Carcinoid tumours of the testis. *BJU Int* 2008; 101: 1101–5.
- Hopps CV, Goldstein M. Ultrasound guided needle localization and microsurgical exploration for incidental nonpalpable testicular tumors. *J Urol* 2002; 168: 1084–7.
- Ong TA, Yaakup NA, Sivalingam S, Razack AH. Hook wire localization for testis sparing surgery. *Urology* 2013; 81: 904–7.
- Dell'Atti L. Efficacy of ultrasound-guided testicle-sparing surgery for small testicular masses. *J Ultrasound* 2015; 19: 29–33.

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