CASE REPORT

Rare presentation of a testicular angiofibroma treated with testis sparing surgery

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Summary

Introduction: Testicular benign tumors are very rare (<5%). Testicular Angiofibroma (AF) is one of those, however the gold standard of treatment and follow-up is still unclear.

Case report: A 47 years-old man with only one functioning testis was referred to our clinic for a palpable right testicular mass and atrophic contralateral testis. Patient underwent testis-sparing surgery with inguinal approach and intraoperative frozen sections examination with diagnosis of AF.

Final histology confirmed AF. Post-operative follow-up was uneventful. Clinical and ultrasonographic follow-up was negative after 8 months.

Conclusion: We report a conservative surgery in a patient with AF of the solitary testis. AF is a benign para-testicular fibrous neoplasm that could be misinterpreted as malignant tumor and treated with orchietomy. Testis-sparing surgery is recommended in this case with intraoperative pathological examination. The excision of the mass is enough but in front of a possible recurrence a long follow-up is advisable.

Key words: Testicular neoplasms; Angiofibroma; Testis-sparing surgery; Small testicular tumours.

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Introduction

Angiobroma (AF) is a benign neoplasm arising mainly in vulvo-vaginal region in woman and in inguino-scrotal region in men, it is very rare in male genitalia (1). Because of the variability of clinic findings and uncertain classification, the gold standard of treatment and follow-up are still unclear. Testis-sparing surgery (TSS) with intraoperative frozen sections of small testicular lesions is recommended (2) to avoid over-treatment of benign tumors like AF despite those are uncommon.

We present a case of an AF in a young man with an only one functioning testis.

Case report

A 47 years old man was referred to our department for a palpable and painless right testicular mass found incidentally during a shower. The left testis was atrophic because of a torsion at age of 15. He had not further previous medical or surgical history. Tumor markers (alpha-
Figure 3. Intra-operative picture of the testicular mass.

Histological examination confirmed the AF. Any complication was observed in the post-operative time.
Clinic and ultrasonographic follow-up after 3 and 8 months was negative for recurrence.

**DISCUSSION**

Testicular and para-testicular benign fibromatous lesions are rare, arise in most cases from tunica vaginalis and the 15% of cases from tunica albuginea (like this one) (2, 3). The aetiology is uncertain: while some authors consider them non-neoplastic and reactive process, others consider them true neoplasms arising from the cellular tunics if inflammatory cells are not present; these lesions can arise from the testis (fibromas of gonadal stromal origin) or from testicular tunics (3).

In particular, angiomyofibroblastoma-like (AMF-like) tumors are benign mesenchymal neoplasm with extremely low incidence (4), initially described in women by Nucci et al. (5), and then in men by Laskin et al. (6). Iwasa concluded that cellular AF and AMF-like tumors are very similar but they differ in the stroma/vascular ratio in favor of AF (7). No cases of metastatic disease or recurrence are described (8).

AF arises mainly in inguino-scrotal region in men but is very rare in the testis, it originates from perivascular stem cells that differentiates into fatty and myofibroblastic cells (9); it is composed by prominent blood vessels, low cellularity with spindle cells (9) within collagenous stroma, without mitotic activity, necrosis, nuclear pleomorphisms. Testicular parenchyma is not remarkable inside the lesion and a vascular axis is described in the center.

The histological differential diagnosis includes aggressive angiomyxoma, angiomylipoblastoma, spindle cell lipoma, solitary fibrous tumor, perineurioma and leiomyoma (5).

The ultrasonography findings of these lesions are not frequently described, and there is a large variation of characteristics: fibromatous tumors are described as hyper- or iso- or hypoechoic, with or without posterior acoustic shadowing (10). In some paper are described also magnetic resonance imaging (MRI) findings: are lesions with intermediate signal intensity in all sequences, more hypointense on T2- than in T1- weighted sequences and failed to enhance after gadolinium administration (10). The differentiation of an inflammatory fibrous pseudotumor from a true neoplasm is based on clinical, radiographic, gross and microscopic features. Clinical and ultrasonography features (as the appearance of extratesticular masses) may indicate the benignity of the lesion, but the specificity isn’t sufficient to permit confident differentiation between benign and malignant process; therefore, a histological evaluation of the lesion is mandatory.

Testis-sparing surgery is indicated in selected cases (bilateral testicular tumor or monorchid patient) (11); in cases of endophytic masses intraoperative ultrasound (IU) and collaboration with the pathologist can help to detect and enucleate the entire lesion (12); in this case a wedge resection was performed without IU because tumor was mostly exophytic and margins were clearly detectable.

The major concern for TSS is the small size of lesions send for frozen sections: the low quantity of tissue and the missing of intraoperative immunochemistry can affect the diagnosis (1, 13). The definitive histologic report is the only one to comply the final diagnosis. Follow-up is still important also if the diagnosis is a benign tumor. We don’t have many data in literature about follow-up of these lesions, because are very rare and most of papers report only a case report, like our experience; in the largest case series published composed by 40 (1) and 9 (3) men were negative for recurrence. Best strategy for testicular follow-up is ultrasound, because the sensitivity for small and not palpable lesions.

**CONCLUSIONS**

Frozen sections are recommended in case of TSS that is an effective treatment for small testicular lesions to avoid over-treatment, limiting functional outcomes in cases of benign tumor, mostly in monorchid patients. Further studies and biggest series are needed to confirm the right follow-up to detect any recurrence.

**REFERENCES**


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