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Case Report

IDIOPHATIC ASYNCHRONOUS BILATERAL SEGMENTAL TESTICULAR INFARCTION AND ANTICOAGULANT THERAPY: A CASE REPORT

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SUMMARY – We present an unusual case of sudden onset of pain in the left testis in a patient with a previous medical history of right orchiectomy due to hemorrhagic infarction. A partial orchiectomy was performed with complete removal of the lesion and reconstruction of the testicular parenchyma. Histopathological assessment confirmed segmental testicular infarction without the presence of malignancy. The patient subsequently received anticoagulant therapy.

Key words: *botulinum toxin type A, acute testicular pain; testicular pathologies; testicular infarction; anticoagulant therapy*

Introduction

Bilateral segmental testicular infarction is an extremely rare entity. Only a few cases have been reported previously[1-7]. The most common symptom is acute testicular pain which mimics testicular torsion, infection (epididymitis/orchitis), or testicular cancer. The pathophysiology is still not fully understood, and over 70% of reported cases are idiopathic[8]. The role of anticoagulant therapy in possible prevention and treatment is also unknown.

Case presentation

A 43-year-old man (Caucasian), father of one child, with a past medical history of right-side orchiectomy because of hemorrhagic testicular infarction nine month prior, developed acute onset of left testicular pain. The pain had increased with time. There was no history of testicular trauma, deep vein thrombosis, or pulmonary embolism. He denied prior sexually transmitted diseases, dysuria, hematuria, fevers, nausea, or vomiting. The scrotal skin was without erythema, swelling, and thickening. Physical examination revealed a left testicle with severe tenderness to palpation, but not enlarged and with absence of any intratesticular mass. No pathological lymph nodes, penile lesions, abdominal masses, or gynecomastia were detected. Complete blood count, urine analysis, urine culture, and tumor marker profile were all within the normal range. A testicular color Doppler ultrasonography illustrated absence of flow and a well-defined non-homogeneous mass in the upper pole of the left testicle. It measured approximately 25 mm. The remainder of the left testicle showed normal echogenicity and vascularization. Based on the clinical and ultrasound findings, an inguinal partial orchiectomy was performed on the same day with com-

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plete excision of the lesion. There was no evidence of recent or past torsion. Final histopathological examination revealed hemorrhagic infarction with small part of necrotic tissue, surrounded by normal parenchyma without neoplastic cells (Fig 1.). No complications occurred, and the patient fully recovered. Due to unclear etiology, the patient was transferred to the Department of Hematology for further management. Complete blood cell count, biochemical, rheumatological, and serological tests were normal. Testicular tumor markers (α-fetoprotein, β-human chorionic gonadotropin, and lactic dehydrogenase) and tests for thrombophilia were also in normal ranges. Despite the normal thrombophilia testing, we decided on anticoagulant therapy (warfarin). Transthoracic echocardiography and magnetic resonance angiography of the pelvis and lower extremities that were subsequently performed did not detect any vascular abnormalities.

For the next eight years, while taking warfarin, the patient did not have any symptoms or events.

Discussion

Segmental testicular infarction is a rare entity which can mimic a testicular torsion, infection, or a testicular cancer. Usually occurs between the second and fourth decades of the life. The pathogenesis is still not well known and is usually idiopathic[8]. Reduced or interrupted blood flow, insufficient collateral blood supply, and thrombus formation is one of proposed mechanisms[9-11]. Epididymo-orchitis is a one of the well-known underlying causes[12]. Several case reports have described anatomic variations (bell clapper deformity causing intermittent torsion), intimal fibroplasia of the spermatic artery, cholesterol embolization, trauma and following surgery, hematologic disorders such as sickle-cell disease, polycythemia, and diabetic microangiopathy, vasculitis i.e. hypersensitivity angiitis and polyarteritis nodosa, as potential causes[1,2,5-7,12-22]. Other etiological factors as well as antiphospholipid syndrome and protein S deficiency are also cited as possible causes of testicular infarction[23,24]. It would be useful to screen these patients for thrombophilia.

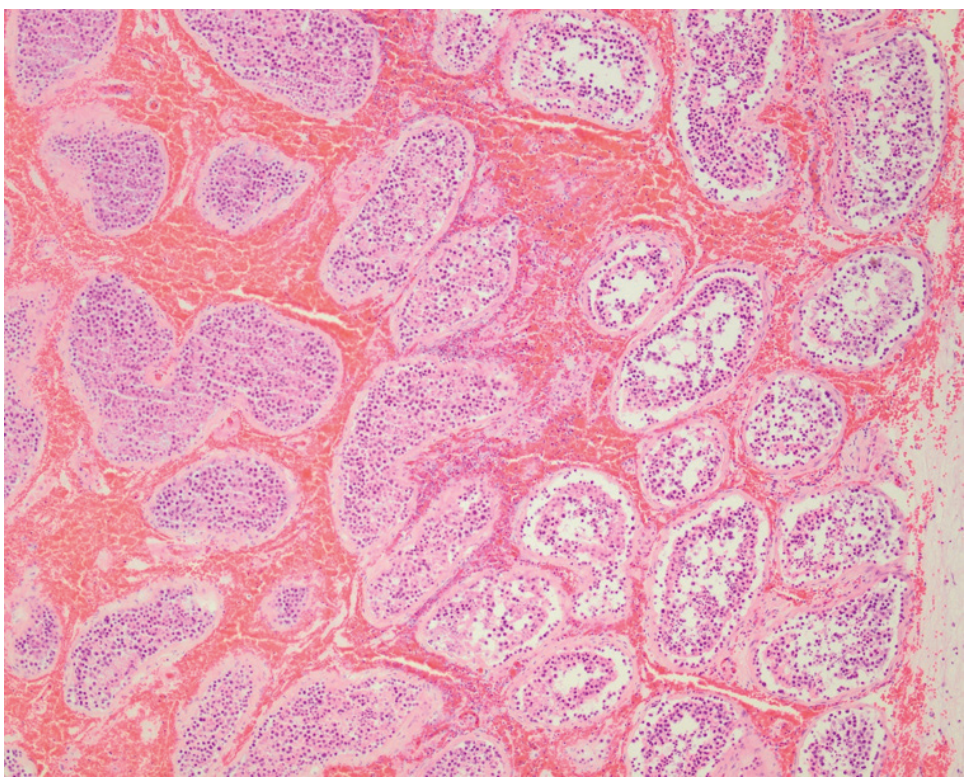

Fig.1. The testis parenchyma with some stromal hemorrhage. Seminiferous tubules in the middle are preserved with maintained spermatogenesis while those on the left and right hand side show coagulative necrosis (HE, x100), (courtesy of Assist. Prof. Tihana Regović Džombeta, Department of Pathology, KBC Sestre milosrdnice, Zagreb, Croatia).

Conservative therapy is the treatment of choice in patients with segmental testicular infarction, but surgical exploration is indicated in doubtful cases with unclear or no conclusive findings, despite advanced imaging technology. Persistent pain and extensive infarction may be an indication for partial or total orchiectomy.

Currently, there are no literature data on the use of anticoagulant therapy in these patients, but in rats, modulation of thrombosis with anticoagulants significantly reduces testicular damage after testicular torsion[25]. Therefore, in case of idiopathic testicular infarction we recommend anticoagulation to prevent further complications or recurrence of the disease.

Conclusions

Segmental testicular infarction, especially bilateral, is a rare clinical entity. To date, since the first reported case in 1909, the etiology and pathophysiology are not well understood. Additionally, there are no data in the literature on the use of anticoagulants in patients with idiopathic testicular infarction[26]. We believe that anticoagulant therapy can avoid the development of further thrombotic process and recurrent testicular infarction, which could be one of the ways to improve the treatment outcome. Moreover, we need to intensify our efforts to understand, prevent, and neutralize the mechanism of testicular infarction. Further research on the benefits of anticoagulant therapy is needed.

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Sažetak

IDIOPATSKI ASINKRONI OBOSTRANI SEGMENTALNI INFARKT TESTISA – PRIKAZ SLUČAJA

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Prikazujemo neobičan slučaj iznenadne pojave boli u lijevom testisu kod bolesnika u kojeg je prethodno učinjena desnostrana orhidektomija zbog hemoragijskog infarkta. Izvršena je djelomična orhidektomija s potpunim uklanjanjem lezije i rekonstrukcijom parenhima testisa. Histopatološkom obradom potvrđen je segmentalni infarkt testisa bez prisutnosti zloćudne bolesti. U bolesnika je nakon toga primjenjena protuzgrušavajuća terapija.

Ključne riječi: *Akutna bol testisa; Patologija testisa; Infarkt testisa; Protuzgrušavajuća terapija*