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Orchitis the Strange, the Rare and the Unusual: Case Report and Review of the Literature

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Abstract

Orchitis are classified as granulomatous and non-granulomatous orchitis. Several agents are responsible for inflammatory and infective diseases of the testis, causing orchitis. The most common clinical features are scrotal swelling, mass and/or pain. The diagnosis is mainly histological because clinical and sonographic aspects may be not conclusive. Bacterial orchitis are characterized by diffuse enlargement and hyperemia usually without discrete mass.

We report the case of a histological proven bilateral granulomatous orchitis, with two testicular mass, diagnosed in different times. The serological markers were normal in both cases. The ultrasonographic study demonstrated an increased volume of the testis, with a hypoechoic area, low grade internal flow was noted upon Doppler evaluation. The patient underwent an inguinal exploration and a succeeding orchifunicolectomy, of the left and, some years after, of the right testis. The knowledge of all kind of orchitis that may be encountered in the routinely clinical practice is of paramount importance for a correct diagnosis and to avoid an unnecessary orchiectomy.

Keywords

Granulomatous orchitis, Infections, Testicular mass

Introduction

Testis may be affected by an unsuspected variety of orchitis, several agents and causal factors are responsible for inflammatory and infective diseases of the testis. Orchitis are roughly divided in granulomatous and non-granulomatous ones.

Granulomatous orchitis is a rare disease, first described by Grunberg in 1926 [1]. The most common clinical features that characterize granulomatous orchitis are scrotal swelling, mass and/or pain. The diagnosis is mainly histological because clinical and sonographic aspects may be not conclusive. Testis is defined as a 'sanctuary organ' since there is a blood-testis-barrier or more correctly a 'Sertoli cell barrier', which, on one hand is a mechanism of protection of the sperm against any autoimmune reaction and on the other hand inhibits the passage of several cytotoxic agents and drugs into the testicular tissue. The blood-testis-barrier can be damaged by traumas, infections, torsion etc. One hypothesized causal factor of granulomatous orchitis is the extravasation of sperm within the testis as a consequence of discontinuation of the blood-testis-barrier resulting in a secondary granulomatous reaction due to activation of the autoimmune response.

Several Orchitis Show Granulomatous Features

Testicular sarcoidosis is characterized by granulomatous lesions [2]. Sarcoidosis is a systemic disease, showing non-necrotizing granulomatous inflammation. It mainly affects lungs, eyes and skin. The causative agent remain unknown at present, suspected causes are infectious (mycobacteria, tubercolous, nontubercolous, bacteria, corynebacterium, propionibacterium, tropheryma whippleii, fungi, cryptococcus, viruses, cytomegalovirus, Epstain-barr virus, Herpes simplex virus), and non infectious ones (dusts, clay, pine, pollen, talc, metals, aluminum, beryllium, zirconium) [3]. In few cases the testis is involved, about 4%-4.5%, but only 0.5% of these patients have clinical symptoms [4]. Sarcoid most commonly affects epidydimis, solitary testicular involvement is uncommon. Testis presents with a mass, which may be painful. Testicular biopsy from the inguinal approach rather than orchiectomy may be considered mainly in bilateral lesions to exclude a testicular neoplasia [5-7].

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Malakoplakia is a rare chronic inflammatory disease, which is characterized by papule, plaque and ulceration; commonly affected sites are bladder, less frequently kidneys, ureters, testis, prostate and colon. It is described by the presence of large cells with eosinophilic cytoplasm (von Hansemann cells), with typical cytoplasmatic inclusions, so called Michaelis Gutmann (MG) bodies.

It is thought to be the result of a deficient activity of macrophages so that the partial digested bacteria lead to deposition of calcium with the formation of ulcer, plaque and papule. This is due to the strict association of malakoplakia with immunodeficiency. Often patients affected are diagnosed with lymphoma, diabetes, or are on a long term therapy with corticosteroids. Thirty six cases are reported in the literature involving testis and epididymis. A correct diagnosis requires the use of special stains to demonstrate the presence of MG bodies to help in differentiating malakoplakia from other cases of granulomatous orchitis [8].

Furthermore, testis may be affected by infections, viral infections are mainly represented by mumps orchitis. The incidence has declined since the introduction of the childhood vaccination program. 10%-30% are bilateral mumps orchitis [9]. Orchitis usually occurs 1-2 weeks after parotitis. Impairment of fertility is present in 13% of patients, while 30%-87% with bilateral orchitis experience infertility [10]. The kinetics of the clinical signs and the suggestive pathological features with hemorrhage and neutrophilic infiltrate are important in the differential diagnosis.

Primary orchitis without associated epididymitis is rare but may be caused by the human immunodeficiency virus. Pathologic changes of testis included azoospermia, hyalinization of the seminiferous tubules, and lymphocitic infiltration of the interstitium [11].

Also syphilis causes a testicular infection with mononuclear inflammatory infiltrate, vascular abnormalities and a positive immunoreactions against spirochetes. Only 11 cases are reported in literature [12].

Moreover, granulomatous epididymo-orchitis may result from a specific infectious etiology: tubercolosis, leprosy, brucellosis, actinomycosis, with a typical inflammatory infiltrate and a positive microbial agent assessment.

28 cases of brucellar epididymo-orchitis are reported in literature, mainly monolateral, showing an increase in inflammatory features: C reactive protein (CRP), leucocytosis and erythrocyte sedimentation rate, high agglutination titers ≥ 1/160. It must be kept in mind in endemic regions as patients may be successfully treated with antibiotics avoiding an unnecessary orchiectomy [13].

Among granulomatous infection, also the Bacillus Calmette-Guérin (BCG) induced tubercolous epididymo-orchitis has to be accounted. Several anatomic locations are reported: lungs, liver, bone marrow, bones, kidneys, eyes, vessels, prostate, as well as epididymis and testicles [14]. 13 cases affecting epididymis and testis are described in literature [15,16]. The testis becomes involved either from a direct extension from the epididymis or from hematogenous spread. Patients often manifest a scrotal enlargement, thickened scrotal skin, hydrocele, calcifications, scrotal abscess and a sonographic pattern characterized by diffuse enlargement of the testis with either a heterogeneous or homogeneous hypoecoic appearance, nodular enlargement or multiple small nodules.

Granulomatous orchitis either from a specific infectious may result also to be idiopathic. The idiopathic orchitis is characterized by a chronic inflammation in the interstitium and by the destruction of spermatogenic cells. The etiology is unknown, although an autoimmune or posttraumatic reaction has been hypothesized. Sonographic features include a diffuse hypoechoic aspect of the testis or focal hypoechoic intratesticular areas.

Lastly, vasculitis like polyangiitis, a systemic necrotizing granulomatous vasculite, may rarely (less than 1% of cases) affects the genitourinary system causing prostatis, urethritis, orchitis and renal injuries [17]. In this case, serologic evaluation of antineutrophil cytoplasmatic antibodies (ANCA) may be important in the differential diagnosis.

A rare case is the xanthogranulomatous orchitis with a scrotal ultrasound evidence of heterogeneous testicular areas [18].

Granulomatous orchitis has been described also as reaction after the end of chemotherapy in patients affected by Hodgkin's and non-Hodgkin's lymphoma [19].

On the Other Side there are the Non-Granulomatous Orchitis

Among non-granulomatous orchitis are described those associated with Crohn's disease, a systemic disease that sometimes involves the testis. Clinically, the testis are of normal volume and consistency, painless and characterized by a heterogeneous, hypoechoic and hypervascularized lesion at the ultrasound examination [20].

In cases of lymphoma of the testis, the majority of lymphocytes are usually B cell neoplasm with a diffuse tumor infiltrate [21].

Male reproductive system can be affected by chronic bilateral orchitis and male infertility may be the only clinical sign [22].

Furthermore, when facing with testicular abnormalities, the differential diagnosis is intricate. At first, traumatic damage and testicular torsion has to be excluded. The absence of a history of trauma and of symptoms sug-

gestive for intermittent torsion may lead our diagnosis. A not negligible differential diagnosis is also a burnt-out germ cell tumor, characterized by scar, intratubular germ cell neoplasia and intratubular calcification.

Inflammatory diseases of the testis are rare, often associated with an increasing consistence of the testis which may be suggestive for a testicular tumor. The ultrasonographic assessment is important but often not enough in discriminating between tumoral and non tumoral lesions. So that an inguinal exploration is always necessary to achieve a histological certain diagnosis.

Case: We Report a Case of Clinically Evident and Histologically Proven Bilateral Granulomatous Orchitis

In 2008, a 67-year-old man, with a history of diabetes mellitus type II, hypertension, presented at our urological clinic with a left testicular pain and swelling. At the physical examination there was a left testicular mass. The ultrasonographic study demonstrated an increased volume of the left testis in respect with the other side, with a diffuse hypoechoic area, low grade internal flow was noted upon doppler evaluation, a small hydrocele was present too. The values of α-fetoprotein and β-human chorionic gonadotrophin were normal.

Since the lesion was presumed to be malignant, we execute an intraoperative biopsy from the inguinal approach which diagnosed an inflammatory process. Nevertheless, an orchifunicolectomy was performed to obtain a deeper histopathologic analysis which revealed a granulamatous orchitis. As already mentioned, the differential diagnosis between testicular tumour and granulomatous orchitis is very complex except by histological findings. The later histopathology revealed spermatic cord and epidydimis without lesions, a diffuse stromal replacement by histiocytes with epithelioid features, some lymphocytes and plasmacells in the stroma. The epithelioid elements were CD1a-, CD68KP1+, CD-68PGM1+, CKMNF116-, CK7-, CK20-, alphainhibin-, HMB.45-, S100+ (just focally), vimentin+. A few iron pigments deposition (Perls +) was present but no MG bodies like. An interstitial fibrosis was discovered too, with seminiferous tubules hyalinized and atrophy in the subcapsular areas. By histochemistry using Periodic Acid-Schiff (PAS) technique we did not find any microbiological agent. CRP (amplification of the GP65 sequence) for mycobacterium spp. was negative.

The Conclusive Diagnosis was Granulomatous Orchitis with Epitheliod Features

About one year later during a urological control it has been found out another small lesion of the right testis, on the basis of the previous hystopathological result, we decided to follow the lesion periodically. It was unmodified, with normal α-fetoprotein and β-human chorionic gonadotrophin, until June 2016 when it showed an increasing volume, with a remarkable associated pain. The increased dimension casts doubt upon the origin of the lesion, and an inguinal exploration was planned again with an intraoperative biopsy. This last diagnosed an inflammatory process. Nevertheless, an orchifunicolectomy has been performed in accordance with the patient's wishes who was 73-year-old and did not want to effort the pain long after.

The later histopathology revealed a focal stromal replacement by lymphocytes and histiocytes and interstitial fibrosis, diffuse aspects of atrophy of the seminiferous tubules.

Conclusions

Not all lumps indicate the presence of testicular cancer, most are caused by benign, conditions such as injury, birth defects, infection, varicocele, hydrocele, epididymal abnormalities, testicular torsion, hernia etc. An ultrasound evaluetion should be enough to find out several of those pathologies, even if a correct final diagnosis is often reached by a surgical approach only. The knowledge of all kind of orchitis that may be encountered is of paramount importance to avoid unnecessary orchiectomy.

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