

Buerger's disease presenting as a testicular mass: A rare presentation of an uncommon disease

Jay A. Roberts, Jon-Paul Meyer

Department of Urology, Redcliffe Hospital, Queensland, Australia

Abstract

Thromboangiitis obliterans is an uncommon nonatherosclerotic segmental inflammatory disease that predominantly affects the small and medium-sized arteries and veins of the distal extremities. It was first described in 1879 and is also known as Buerger's disease. Buerger's usually begins with ischemia of small vessels producing digital infarcts and may progress to more proximal arteries and veins, producing claudication of the feet, legs, hands, or arms. Tobacco smoking is essential to the initiation and the progression of disease and it typically occurs in males under the age of 45 years. Although Buerger's most commonly affects the arms, hands, legs, and feet, it has also been reported in other vascular beds including cerebral, coronary, renal, mesenteric, and pulmonary arteries. There are also a small number of cases involving the male genitalia. To our knowledge, there has only been one English case of Buerger's involving the testis, published in 1940. Here, we present a new case of Buerger's presenting as a testicular mass in a 17-year-old cannabis smoker.

Key Words: Buerger's, testis, thromboangiitis

Address for correspondence:

Dr. Jay A. Roberts, Department of Urology, Redcliffe Hospital, Anzac Avenue, Redcliffe QLD 4021, Queensland, Australia. E-mail: jay.roberts@uqconnect.edu.au

Received: 30.10.2015, **Accepted:** 04.01.2016

INTRODUCTION

Thromboangiitis obliterans is a nonatherosclerotic segmental inflammatory disease that predominantly affects the small and medium-sized arteries and veins of the distal extremities. It was first described in 1879 and is also known as Buerger's disease after Leo Buerger who published a detailed description of the pathological findings in 1908.^[1] Tobacco smoking is essential to the initiation and the progression of disease and it typically occurs in males under the age of 45 years, though can occur in young females as well.^[1]

Buerger's is an uncommon disease with an incidence of 12.6/100,000 in the USA, though it is more common in countries with higher rates of smoking such as India.^[1] It usually begins with ischemia of small vessels producing digital infarcts and may progress to more proximal arteries and veins, producing claudication of the feet, legs, hands, or arms. There have been a few cases reported of Buerger's involving the male genitalia, particularly the spermatic cord;^[2] however, to the authors' knowledge, the only English report of involvement of the testis itself was published in 1940.^[3] Here, we report

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How to cite this article: Roberts JA, Jon-Paul M. Buerger's disease presenting as a testicular mass: A rare presentation of an uncommon disease. Urol Ann 2016;8:249-51.

Access this article online	
Quick Response Code:	Website: www.urologyannals.com
	DOI: 10.4103/0974-7796.179238

a modern case of Buerger's affecting the testis, presenting as a testicular mass.

CASE REPORT

A 17-year-old male presented with a 3 weeks history of a painless swelling in the left hemiscrotum. Examination revealed the swelling to be consistent as an epididymal cyst which was later confirmed on ultrasound examination. There was, however, a subtle mass palpable within the left testis, which had a sonographic appearance suspicious for testicular neoplasm. There had been no history of trauma or cryptorchidism. The patient had no other significant history apart from being a heavy cannabis smoker and tobacco smoker. He denied use of other illicit drugs.

Tumor markers (β -human chorionic gonadotropin, alpha-fetoprotein, and LD) were all negative. Staging chest X-ray and computed tomography scan of the abdomen did not reveal evidence of metastatic disease. Given the ultrasound appearances, malignancy was suspected and the patient underwent a left inguinal orchiectomy.

Histology revealed multiple infarcts of the testicular parenchyma with an obliterated artery adjacent to each infarct. The lesions varied in the stage of development but appeared similar in nature with evolving thrombosis and fibrous obliteration. The findings were consistent with thromboangiitis obliterans. There was no evidence of malignancy.

Further review of the patient did not reveal any further evidence of infarction though he did give a history of intermittent leg cramps and weakness, which may have been due to intermittent claudication. Unfortunately, the patient did not attend for further follow-up.

DISCUSSION

Buerger's disease is an uncommon disease of which the cause is unknown; however, tobacco smoking is central to the initiation, maintenance, and progression of the disease.^[3] It usually presents in males, though has been described in females, and almost exclusively onset in those under the age of 45 years.^[1] Diagnosis is often difficult due to lack of a unifying definition, and for some time, there was debate whether Buerger's was a separate clinical entity, though several criteria and point systems have now been proposed.^[4] Buerger's usually begins with ischemia in small distal arteries and veins, with upper limb involvement in up to 90% of patients.^[4] Frequently, multiple extremities are involved and some believe this to be essential to distinguish Buerger's from other vasculitides and atherosclerosis.^[4] Migratory superficial thrombophlebitis occurs in approximately 40% of patients

and may occur as an early manifestation of disease prior to digital ischemia.^[4,5]

As the disease progresses, it affects more proximal arteries and may present as claudication of the feet, legs, arms, or hands.^[5] Raynaud's phenomenon is present in 40% of patients. Absence of atherosclerotic risk factors other than smoking (autoimmune disease, hypercoagulable states, and diabetes mellitus) appears to be important in making a diagnosis.^[1,4] Angiography of affected limbs may reveal the typical appearance of "corkscrew collaterals."^[1]

Thromboangiitis obliterans refers to the characteristic pathological findings, which vary according to the duration of the disease. Biopsy of acute phase lesions may provide a definitive diagnosis due to the pathognomonic microscopic features in these lesions.^[4] The acute phase lesion consists of an occlusive, highly cellular, inflammatory thrombus. Polymorphonuclear neutrophils, microabscess, and multinucleated giant cells are often present.^[1,5] In the chronic phase, the pathognomonic features almost disappear;^[4] however, there remains sparing of the vessel walls and internal elastic intima, distinguishing Buerger's from atherosclerosis and other types of systemic vasculitis.^[1,5]

Central to the management is cessation of tobacco smoking, with 94% of those who abstain able to avoid amputation, whereas 43% of those who continue smoking tobacco requiring one or more amputations.^[1] Cooper *et al.*^[6] reported a 10-year risk of amputation of 38% for any amputation and 21% for major amputation.

Although Buerger's most commonly affects the arms, hands, legs, and feet, it has also been reported in other vascular beds including cerebral, coronary, renal, mesenteric, and pulmonary arteries.^[1] There are also a small number of cases involving the male genitalia, with reports of penile involvement resulting in partial or total penectomy,^[7-9] as well as a collection of reports of Buerger's isolated to the spermatic cord.^[2,10]

In 1940, Mathe^[3] reported a case of Buerger's isolated to the left testis, which to our knowledge is the only case in English other than the current report of such a presentation. Mathe reported a 53-year-old who, similarly to the present case, presented with a firm nodule in the left testis, and gave a history of leg cramps and weakness, thought to be intermittent claudication. Given the presence of a palpable mass in the testis, an orchiectomy was also performed. Interestingly, both cases were present in the left testis, and similarly all reported cases within the spermatic cord also involved the left side. No explanation for this phenomenon has yet been put forward.

Buerger's is an uncommon disease and involvement of the testis as reported here is extremely rare. It is indistinguishable from neoplasm on clinical and sonographic evaluation and therefore, orchiectomy remains mandatory. However, establishing a definitive diagnosis is important for counseling of patients, as without cessation of smoking disease progression is the likely outcome and may result in further amputation.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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