

Painful erections secondary to rare epithelioid hemangioma of the penis

Evan Barber, MD,* Trustin Domes, MD, MEd, FRCSC†

*College of Medicine, University of Saskatchewan, Saskatoon, SK; †Saskatoon Health Region, Saskatoon, SK

Cite as: *Can Urol Assoc J* 2014;8(9-10):e647-9. <http://dx.doi.org/10.5489/cuaj.1833>
Published online September 9, 2014.

Abstract

Epithelioid hemangioma is a rare benign vascular tumour that atypically involves the penis and usually presents as a painful nodule. A 35-year-old man presented with a 5-month history of painful erections without a clinically apparent lesion or deformity. Magnetic resonance imaging (MRI) with pharmacologically induced erection demonstrated a 1.3-cm nodular lesion deep to the skin at the base of his penis. Following local excision of the lesion, which was diagnosed as an epithelioid hemangioma, the patient was symptom free. This case demonstrates a rare etiology of painful erections with a unique presentation. To the authors' knowledge, it is also the first report of MRI with intracavernosal injection of trimix to assess for a specific cause of painful erections.

Introduction

Epithelioid hemangioma is an uncommon benign vascular tumour that commonly affects the head and neck and typically presents as a superficial painful nodule.^{1,2} First described in 1969 as superficial angiolymphoid hyperplasia with eosinophilia, the lesion is microscopically typified by well-formed capillary-sized vessels, epithelial-like endothelial cells with large nucleoli and prominent nucleoli, absent nuclear atypia, and often secondary inflammatory infiltrate.³⁻⁵ The pathogenesis has been subject to debate with suggestions that many cases are reactive in nature, secondary to vascular damage and repair, rather than neoplastic.⁴ Involvement of the penis is rare, with fewer than 30 reported cases in the English literature.⁶ Of these cases, there is a tendency for the lesion to present as a clinically evident painful nodule on the dorsal aspect of the shaft of the penis.⁷

Case report

A 35-year-old man presented with a 5-month history of atypical penile pain occurring with erections. His pain was more marked on the right and was severe, such that nocturnal erections would rouse him from sleep leading to insomnia. He denied a history of penile trauma and no deformity or curvature was related to his erections. His medical and surgical history were unremarkable and he had no risk factors for priapism or Peyronie's disease. On examination, his external genitalia were unremarkable with no palpable penile lesions or plaques. The patient underwent Doppler ultrasound of his penis with intracavernosal injection of trimix to induce erection, which showed no vascular or structural abnormalities, but did reproduce his pain. Magnetic resonance imaging (MRI) was then performed with pharmacologically induced erection and revealed a non-specific 1.3-cm nodular lesion 2.8-cm deep to the skin at the base of the penis, right of the deep dorsal vein (Fig. 1). The mass was excised without complication and a pathological analysis of the mass demonstrated an epithelioid hemangioma (Fig. 2, Fig. 3). Given its uniqueness, an external pathologist specializing in soft tissue lesions confirmed the diagnosis. Since local excision and pathological analysis were definitive in this case, elaborate immunohistochemical analysis of the lesion was not performed. At his most recent follow-up, the patient was asymptomatic with no erectile dysfunction and no further pain with erections.

Discussion

Painful erections are commonly due to clinically evident entities, such as Peyronie's disease and priapism. When erectile pain is atypical, more rare etiologies should be considered, such as penile cancer, soft tissue lesions, or parasomnia if symptoms are primarily nocturnal. This case of



Fig. 1. T2-weighted magnetic resonance, sagittal section, demonstrating a 1.3-cm nodular lesion at base of penis deep to skin intimately associated with the dorsal neurovascular bundle.

benign epithelioid hemangioma of the penis demonstrates a rare lesion with the unusual presentation of painful erections with normal external genitalia. In prior case reports, this tumour has most commonly presented as a solitary painful nodule on the dorsum of the shaft of the penis, occasionally with induration or multiple lesions.^{1,5-9} There is one previous case of associated painful erections, and a second with reported dyspareunia; however, these were associated with a palpable lesion.^{5,8} Ours is the first reported case of an epithelioid hemangioma that was neither visible nor palpable on the penis that required imaging to discover.

Penile MR imaging has been recommended for the assessment of penile prosthesis problems, priapism, penile fracture and penile fibrosis. Penile MRI can be enhanced with intracavernosal injection to produce superior imaging of the tunica albuginea and corpora cavernosa, and demonstrate deformities specific to erections.¹⁰ There are reports of this technique also aiding in the staging of penile cancers.^{11,12} In this case, trimix injection was used before the ultrasound and MRI to achieve higher quality anatomical images, and to reproduce the symptomatic state so that potential tumescence-dependent deformities could be demonstrated. To the authors' knowledge, this is the first reported case of penile MRI with intracavernosal injection to assess for a specific cause of painful erections. Though the culprit lesion would have been visible without pharmacologically induced erection, we were not aware such a lesion existed prior to the MRI and wanted to reproduce the patient's symptoms with

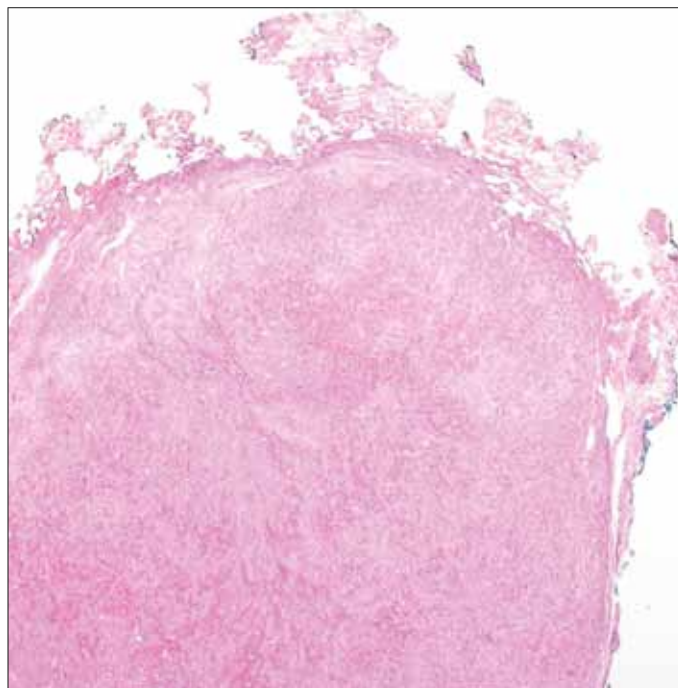


Fig. 2. Low power view of well-circumscribed nodular lesion.

the hope that the MRI would detect a soft tissue abnormality that may have only been visible during erection. This case demonstrates that MRI with intracavernosal injection may be an option to assess for non-palpable soft tissue lesions in the setting of isolated penile pain during erection of unknown etiology.

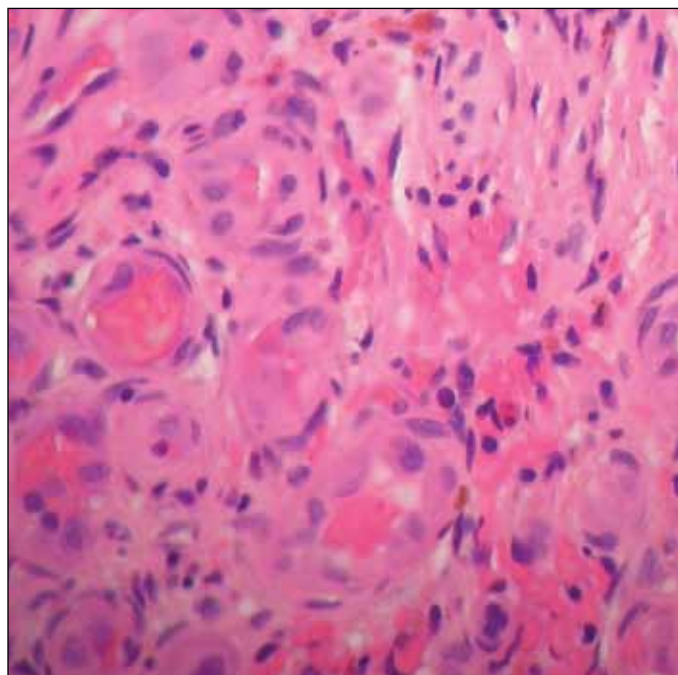


Fig. 3. Epithelioid cells with well-formed vascular lumens, consistent with an epithelioid hemangioma.

Epithelioid hemangioma is a benign lesion and must be distinguished by the pathologist from epithelioid hemangioendothelioma and epithelioid angiosarcoma, which are low- and high-grade malignancies, respectively.⁷

Conclusion

Given the benign nature of epithelioid hemangioma, treatment with complete local excision should be curative. This case is interesting in that local excision of the lesion was successful in completely resolving the patient's significant symptoms. Need for further treatment is not anticipated, as these lesions rarely recur; however, the patient will be followed on an as-needed basis should he develop recurrence of symptoms.

Acknowledgements: The authors thank Dr. Usharani Ganugapati, anatomic pathologist, for her figures and teaching.

Competing interests: Dr. Barber and Dr. Domes declare no competing financial or personal interests.

This paper has been peer-reviewed.

References

1. Dewan P, Francis ND, Lear JT, et al. Angiolymphoid hyperplasia with eosinophilia affecting the penis. *Br J Dermatol* 2008;159:755-7. <http://dx.doi.org/10.1111/j.1365-2133.2008.08733.x>
2. Olsen TG, Helwig EB. Angiolymphoid hyperplasia with eosinophilia. A clinicopathologic study of 116 patients. *J Am Acad Dermatol* 1985;12:781-96. [http://dx.doi.org/10.1016/S0190-9622\(85\)70098-9](http://dx.doi.org/10.1016/S0190-9622(85)70098-9)
3. Wells GC, Whimster IW. Subcutaneous angiolymphoid hyperplasia with eosinophilia. *The Br J Dermatol* 1969;81:1-14. <http://dx.doi.org/10.1111/j.1365-2133.1969.tb15914.x>
4. Fetsch JF, Weiss SW. Observations concerning the pathogenesis of epithelioid hemangioma (angiolymphoid hyperplasia). *Mod Pathol* 1991;4:449-55.
5. Ismail M, Damato S, Freeman A, et al. Epithelioid hemangioma of the penis: case report and review of literature. *J Med Case Rep* 2011;5:260. <http://dx.doi.org/10.1186/1752-1947-5-260>
6. Natali A, Nesi G, Vittori G, et al. Rare case of atypical epithelioid hemangioma of penis initially misdiagnosed as Peyronie's disease: Report with clinical, radiologic, and immunohistochemical analysis. *Urology* 2009;73:210 e7-10.
7. Fetsch JF, Sesterhenn IA, Miettinen M, et al. Epithelioid hemangioma of the penis: a clinicopathologic and immunohistochemical analysis of 19 cases, with special reference to exuberant examples often confused with epithelioid hemangioendothelioma and epithelioid angiosarcoma. *Am J Surg Pathol* 2004;28:523-33. <http://dx.doi.org/10.1097/0000478-200404000-00012>
8. Rao RN, Spurlock BO, Witherington R. Angiolymphoid hyperplasia with eosinophilia: report of a case with penile lesions. *Cancer* 1981;47:944-9. [http://dx.doi.org/10.1002/1097-0142\(19810301\)47:5<944::AID-CNCR2820470521>3.0.CO;2-B](http://dx.doi.org/10.1002/1097-0142(19810301)47:5<944::AID-CNCR2820470521>3.0.CO;2-B)
9. Srigley JR, Ayala AG, Ordonez NG, et al. Epithelioid hemangioma of the penis. A rare and distinctive vascular lesion. *Arch Pathol Lab Med* 1985;109:51-4.
10. Kirkham AP, Illing RO, Minhas S, Minhas S, Allen C. MR imaging of nonmalignant penile lesions. *Radiographics* 2008;28:837-53. <http://dx.doi.org/10.1148/rg.283075100>
11. Petralia G, Villa G, Scardino E, et al. Local staging of penile cancer using magnetic resonance imaging with pharmacologically induced penile erection. *Radiologia Med* 2008;113:517-28. <http://dx.doi.org/10.1007/s11547-008-0273-6>
12. Scardino E, Villa G, Bonomo G, et al. Magnetic resonance imaging combined with artificial erection for local staging of penile cancer. *Urology* 2004;63:1158-62. <http://dx.doi.org/10.1016/j.urolgy.2004.01.008>

Correspondence: Dr. Evan Barber, College of Medicine, University of Saskatchewan, Saskatoon, SK; evan.barber@gmail.com