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Case Report

Recurrent Epithelioid Hemangioma of the Bony Pelvis Responding to Propranolol

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Keywords
Epithelioid hemangioma · Vascular neoplasm · Propranolol · Bone tumor

Abstract
Epithelioid hemangioma is an uncommon benign vascular neoplasm which can arise in bone. Resection is generally curative, but occasionally lesions recur and recurrence after surgery can be morbid and destructive. Recent case reports have described the effective use of oral propranolol to control recurrent epithelioid hemangioma of the orbit. We report the case of a 26 year old man with recurrent aggressive osseous epithelioid hemangioma in the pelvis of which has been controlled for over a year with outpatient propranolol monotherapy.

Background

Epithelioid hemangioma (EH) is an uncommon benign vascular neoplasm that generally affects the skin and subcutis. The skeleton is the second most common location for these lesions with a predilection for vertebrae and long bones of the lower extremity. Osseous EH commonly presents with localized pain, and in a minority of cases may be detected incidentally on radiographs as mixed lytic and sclerotic lesions [1–3]. Standard therapy is curettage or en bloc excision and typically carries a good prognosis. However, treatment may be complicated by multifocal lesions in an estimated 18% of patients and, less commonly, local

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recurrence. Although no standard systemic therapy has been established for refractory EH of bone, two recent case reports describe response and clinical benefit from oral propranolol in treatment of orbital epithelial hemangioma after unsuccessful surgical management [4, 5]. Here we report an impressive outcome in the case of an aggressive, multifocal and recurrent osseous epithelial hemangioma that has been controlled by outpatient propranolol monotherapy.

The patient described in this case gave his written consent that his experience be shared in this publication. None of the authors have relevant financial interests to declare.

Case Report

A 26-year-old man presented in May 2017 with one month of left hip pain. He began physical therapy but the following month sustained an intertrochanteric femur fracture while getting out of bed. This was treated with intramedullary fixation. Follow-up imaging revealed a progressive lytic lesion of the proximal femur with cortical erosion and soft tissue mass. Biopsy found this to be an epithelioid hemangioma with atypical features and extension into extraosseous soft tissue. He underwent radical resection of the proximal femur and total hip arthroplasty in October 2017. Follow-up imaging from 2/2018 showed a subtle lesion in the ischium which was not recognized at the time (Fig. 1).

X-ray imaging in May 2018 showed stable instrumentation but increasing lysis of the left ischium (Fig. 2). This was followed up two weeks later with an MRI of the pelvis and CT angiogram, which revealed a 7.4 cm lobulated expansile mass involving the ischium, inferior pubic ramus, and posterior acetabular column. This was thought to be recurrence of his epithelioid hemangioma, and he was scheduled for embolization of the ischial lesion.

He was referred to us for consideration of systemic treatment and we saw him in August 2018. By this time, he was experiencing worsening pain in his left buttock with sitting and activity. With prior case reports of successful use of propranolol in treatment of benign hemangiomas, including two recent examples of benefit in epithelioid hemangioma, we initiated monotherapy of oral propranolol 40 mg twice daily with close follow-up. He tolerated this well. After 4 weeks of treatment, the patient reported that his pain had stabilized and the lesion appeared unchanged on CT. Because of his clinical improvement and radiographic stability, he did not undergo embolization. By 5 months of therapy on January 2019 he had resolution of his pain and imaging again demonstrated no progression of the mass. He continues on propranolol to date (4/2019); imaging after 10 months on treatment shows increasing sclerosis of the mass (Fig. 3).

Discussion

We submit our experience to add to the growing body of case reports describing patients with vascular neoplasms who have benefitted from treatment with propranolol. We were astounded that this drug could halt a lesion as aggressive as the one seen in our patient. Trends over the past decade show propranolol is a widely accepted therapy for infantile hemangiomas, and two recent case reports suggest it may be a viable treatment for epithelioid hemangioma of the orbit as well [4–6]. Although propranolol is most often clinically used as a beta blocker, it also has non-adrenergic actions including regulatory roles in adipogenesis, angiogenesis, and cell death [7–9]. Wagner et al recently reviewed the use of propranolol in treating

neoplastic disease [10], but it remains unclear whether adrenergic suppression or off-target effects on G-protein coupled receptors are responsible for the clinical efficacy of propranolol against neoplasms. Exploring the overlap between the range of activity of propranolol and the gamut of receptors expressed by vascular tumors would be an important next step in understanding this clinical phenomenon.

Statement of Ethics

The patient described in this case gave his written consent that his experience and images be shared in this publication.

Disclosures Statement

None of the authors have relevant financial interests to declare.

Author Contributions

All authors contributed equally to this publication.

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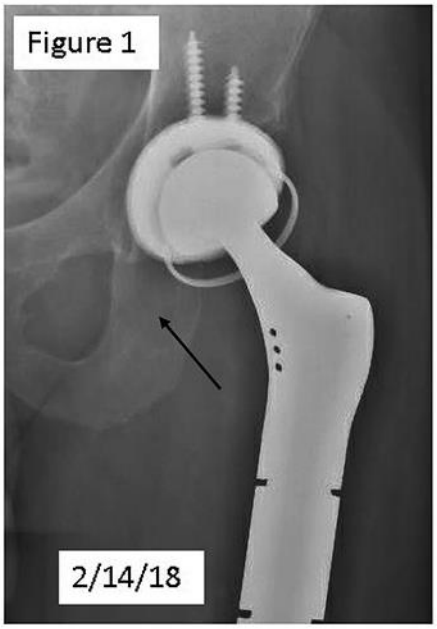

Fig. 1. Subtle lucent lesion in the ischial root.

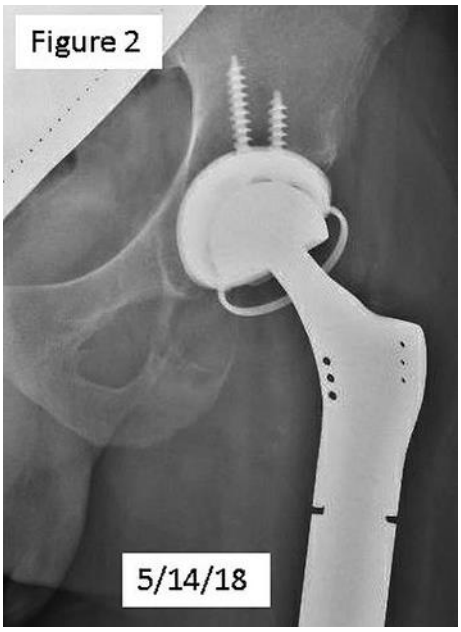

Fig. 2. Increased size of the lucency 3 months later.

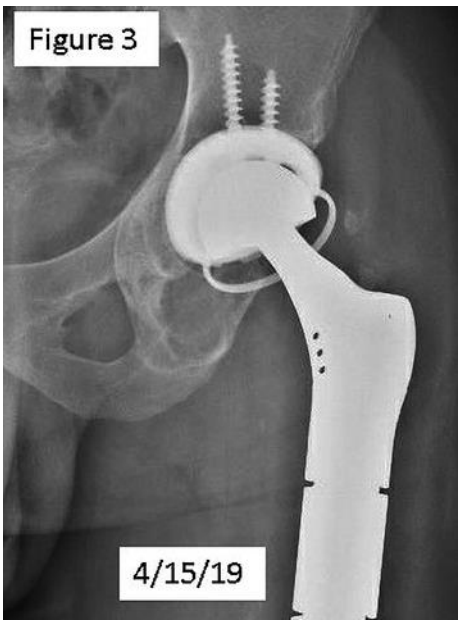

Fig. 3. Sclerosis of the lucent lesion after 1 year of propranolol.