



Benign Perineal Pseudotumor from Repetitive Microtrauma Associated with Saddle Sports: A Report and Guidelines for Management

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Abstract

Benign Perineal Pseudotumors (BPPs) can result from repetitive microtrauma in saddle sports. Also known as a 'cyclist's nodule', 'perineal nodular induration' or 'third testicle', this fibrous soft tissue mass may be referred to a number of different medical specialists for investigation and management. Soft tissue sarcomas and other malignant or benign aggressive neoplasia, albeit rare, can affect the perineal region mimicking BPP and should be excluded. MRI, needle biopsy (ideally with radiological guidance), and referral to a specialist Sarcoma Unit is indicated to ensure the safe management of this condition. For confirmed cases of BPP, management may include conservative or operative options, considering the important anatomical structures in this area and the requirement for plastic reconstructive techniques in some instances.

Keywords: Cyclist's nodule; Saddle sports; Pseudotumor; Perineal nodular induration

Introduction

Benign Perineal Pseudotumors (BPP) are fibrotic lesions located medial to the ischial tuberosity that are associated with repetitive microtrauma to the perineum from saddle sports [1-6]. The condition has been described as the 'cyclist's nodule', 'perineal nodular induration' or 'third testicle'. It is usually seen in male professional and endurance cyclists although female perineal nodules [1-4,6-9] and vulval trauma associated with cycling and equestrian sports have also been reported [5,10-13].

Patients with BPP generally present to Sports Medicine specialists or General Practitioners with an enlarging and/or painful lump. An enlarging soft tissue mass is always a cause for concern and must be investigated to exclude malignancy [2,14]. Specialist referral for ultrasound assessment, MRI and tissue biopsy to confirm the histological diagnosis is essential to exclude soft tissue sarcoma or other sinister pathology. Histologically BPP has a heterogenous appearance but is characterized by fibroblast rich, hypocellular collagenous tissue with myxoid degeneration and pseudocyst formation [1,3,4,6,7].

Case Presentation

A 48-year-old active amateur road cyclist presented to his GP with a painful lump in his perineum. He initially noticed a small mass after a cycling accident 3 years previously, but due to its enlarging nature and ongoing pain he sought medical review. On referral to the local Plastic Surgery Service, and in accordance with the National Institute for Health and Clinical Excellence (NICE) guidelines for soft tissue lesions [15], an MRI scan was performed which demonstrated a soft tissue mass with edema in the perineal region of uncertain significance. An ultrasound scan showed abnormal echogenicity of the subcutaneous fat. An excision biopsy of the mass was subsequently performed.

Intraoperatively, the soft tissue mass could not be clearly differentiated from the surrounding soft tissues and was excised from the superficial ischial perineum. Initial histology demonstrated features of angiofibroma with a benign appearance. The imaging and histopathology were then referred to the regional Sarcoma Service for advice on further management, specifically advice on the requirement for wider excision and long-term surveillance.

The multidisciplinary review of the preoperative MRI demonstrated a low T1 and T2 signal

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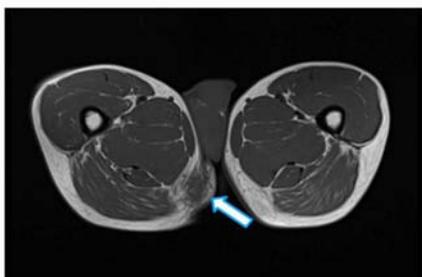


Figure 1: T1 weighted axial MRI image of the unilateral soft tissue benign perineal pseudotumor with ill defined margins and tissue edema in the right perineum.



Figure 2: T1 weighted coronal MRI image of the unilateral benign perineal pseudotumor demonstrating an inferomedial position relative to the right ischial tuberosity.

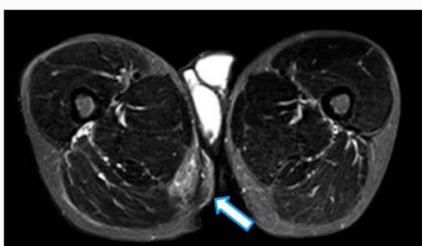


Figure 3: Fluid sequence axial MRI image enhancing the tissue edema associated with the benign perineal pseudotumor in the right perineum.

mass with ill-defined margins within the subcutaneous tissues measuring 5.7 cm × 4.5 cm × 2.5 cm with associated tissue edema of the right perineum medial to the ischial tuberosity (Figures 1-3). Histopathological review demonstrated an abundance of hypocellular hyalinized fibrous tissue interspersed with clusters of capillaries which possessed thicker than normal vessel walls. There was local chronic lymphocytic infiltrate, scattered entrapped adipocytes and linear elastotic fibers (Figure 4) and an epidermal inclusion cyst was identified at the periphery of the specimen.

The patient’s cycling history; imaging in keeping with a fibrotic lesion in a characteristic perineal location and in the absence of histological evidence of neoplasia confirmed the diagnosis of BPP. The MDT opinion was for a baseline post-surgical MRI and surveillance without the requirement for further surgery or wide excision. The patient subsequently made a full recovery and has returned to cycling.

Discussion

BPPs are an uncommon condition associated with saddle sports [2]. Repetitive micro-trauma to the ischial perineal area sustained

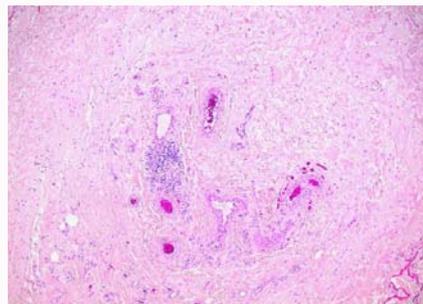


Figure 4: Biopsy slide with Hematoxylin-eosin stain (Magnification x50). The nodule is composed of hypocellular hyalinized fibrous tissue with clustered blood vessels. Focal chronic lymphocytic infiltrate, scattered entrapped adipocytes and linear elastotic fibres are also present.

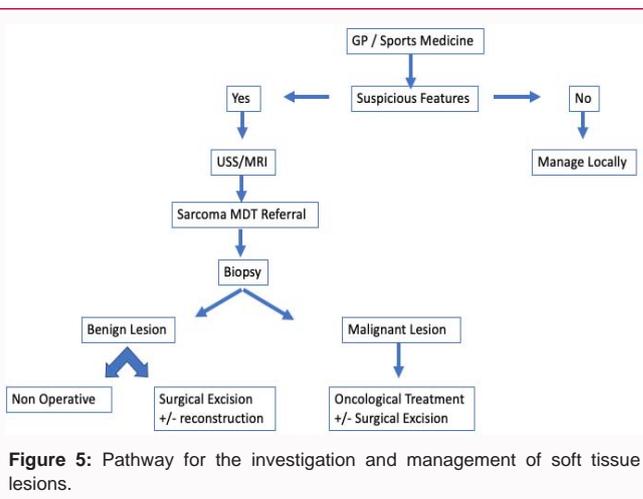


Figure 5: Pathway for the investigation and management of soft tissue lesions.

from the saddle can result in the formation of a fibrotic lump and is not infrequently reported in amateur endurance and professional cyclists, hence termed the cyclist’s nodule, although other terms exist such as perineal nodular induration [1-10,16].

BPP is more common in males although females may also develop the condition from similar saddle sport activities such as cycling [6] and equestrian sports [5,10-13]. On clinical examination the lesion may be solitary or bilateral and related to the ischial tuberosities in the posterior perineum (Figure 2), extending up to the root of the penis in males [3,4,6,7] or vulva in females [5,10,12]. The lesion is often slow growing and may be either mobile or fixed to underlying structures [2-4,7].

Chronic changes to the connective tissue occur but are not necessarily homogenous between cases which can complicate diagnosis. The lesion has previously been described as avascular in some reports [5,17], although the current case demonstrated vascularity with increased abnormal blood vessels. Generally the lesions have edema, collagen degeneration, myxoid change, fibroblastic infiltration, reactive spindle cell and cyst formation [1,3,4,6].

Soft tissue sarcomas of the perineum are very rare but have a poor outcome if there is delay to diagnosis and correct management [18]. Other neoplasia such as anal verge or ischiorectal fossa tumors can also present as a perineal soft tissue mass and mimic benign disease such as BPP [19,20]. In addition, locally aggressive benign pathology such as angiomyxomas of the vulva [21], which are difficult to

manage surgically, will recur and progress with incomplete excision [22]. All of these scenarios could be devastating in a young healthy population such as that typical for BPP. It is therefore essential to investigate all suspicious soft tissue lesions in this region appropriately within specialist centers, irrespective of a history of saddle sports. Accordingly, soft tissue lesions should be managed according to the guidelines in Figure 5. The NICE [15] and UK Sarcoma Guidelines for suspected sarcoma dictate that suspicious features which prompt referral or investigation include any lump which is enlarging [14], painful, or more than 5 cm in diameter. Imaging with ultrasound or MRI, biopsy for confirmatory tissue diagnosis and expedited referral to a specialist sarcoma service for MDT discussion is advised. Following this pathway will ensure that benign lesions such as BPPs are managed correctly and reduces the risk of inadvertent incomplete excision of tumors that may require neoadjuvant therapy.

Following diagnosis the management of symptomatic BPP is surveillance or surgery. Non-operative measures include reassurance, observation and symptom control with clothing adjuncts or improving saddle fit [12,13,23]. Intralesional injection with corticosteroids may offer symptom control [1,3,4]. Surgical options include excision alone or more extensive surgery with flap reconstruction for larger lesions [3,8]. A thorough surgical knowledge of the perineum will avoid potential damage to anus and sphincters, urogenital structures and the closely related pudendal nerve and vessels. The perineum is a well vascularized area and the excision of even small symptomatic lesions may damage the pudendal neurovascular structures and lead to blood loss or nerve injury.

Conclusion

BPPs are soft tissue inflammatory pseudotumors arising in the perineum due to repetitive microtrauma associated with endurance saddle sports. Although benign, malignant disease cannot be fully excluded without careful clinical assessment, imaging and tissue diagnosis. Referral to a specialist unit for histological opinion is recommended if there is doubt in diagnosis or management. Management is either reassurance or conservative treatment or surgical with larger symptomatic tumors. The latter requires careful marginal excision to avoid perineal structural damage and may require plastic surgery reconstruction with larger defects.

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