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“Bumpy” ride for the female cyclist: A rare case of perineal nodular induration, the ischial hygroma

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ABSTRACT

INTRODUCTION: Cycling is known to cause groin pain, typically adductor/hamstring strains and “saddle sores” (skin abrasion, ulceration and/or folliculitis). Rarely does it result in pathological mesenchymal lesions.

PRESENTATION OF CASE: We report a case of a 52-year-old female avid cyclist with chronic groin pain. Activity alteration, bike fit, and saddle modification did not ameliorate her symptoms. MRI revealed no hamstring or bony abnormality but demonstrated low T1/high T2 signal in the right perineum inferior to the ischial tuberosity.

DISCUSSION: Perineal nodular induration, or “cyclist’s nodule,” is a reactive fibroblastic and myofibroblastic pseudotumor almost exclusively reported in male cyclists. PNI can become so nodular that it has even been referred to as an accessory testicle or “the cyclist’s third testicle.”

CONCLUSION: We report an unusual case of PNI in a female cyclist and review the differential diagnosis of non-infectious soft tissue perineal pain in cyclists. Clinicians should be aware of this rare condition to avoid confusion with other mesenchymal lesions.

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1. Introduction

Cycling is a growing sport in North America, with approximately 70,000 USA Cycling members in 2013 [1] and over 89,000 USA Triathlon members in 2016 [2]. Perineal nodular induration (PNI) is a benign pseudotumor associated with saddle sports [3–6], comprised of mixed fibroblastic and myofibroblastic proliferations [6,7]. There is a paucity of sports medicine research on PNI, and it is primarily reported in uropathological literature [5,8] in association with male cycling [7,8]. This benign lesion must be included in the differential diagnosis of an elite or amateur enthusiastic cyclist presenting with groin pain, as PNI is poorly recognized and likely underdiagnosed [4,7] and must be differentiated from neoplasms and mesenchymal lesions. We present a rare case of PNI in an avid female cyclist treated at an academic medical center. This case is reported in line with surgical case report (SCARE) guidelines [7].

2. Presentation of case

A 52-year-old female presented with chronic right “sit bone” pain starting in July 2018. She initially saw an orthopedic surgeon

in December 2018 whereby she had negative x-rays, and was told it was a groin strain that would resolve. She is an avid cyclist averaging 5 h/week of cycling (60 miles/week) prior to injury. She tried bike fit modification, saddle adjustment and activity modification (including 6 months rest from cycling), but upon return to sport the symptoms recurred initially and consistently worsened 20 miles into each ride. She had no prior history of groin muscle strain/tear or saddle sores, no dermatological groin conditions or orthopedic hip conditions, and no history of perineal lesions. Family history was negative for perineal/pararectal lesions.

Physical exam revealed intact perineal skin without erythema or warmth. She had a focal area of tenderness in the right perineum near the ischial tuberosity, with a sub-centimeter nodular indurated area. She had no tenderness to the pubic symphysis or proximal bilateral hamstrings. Hip range of motion was normal with a negative FABER and Scour test. There was no lymphadenopathy.

X-rays of the right hip were unremarkable. However, due to the chronic nature of her symptoms, an MRI was obtained (Fig. 1) showing a 3.5 × 1.9 × 1.8 cm hypointense T1 and hyperintense T2 altered signal in the right perineum inferior to the ischial tuberosity consistent with PNI. There was no invasion of adductor/hamstring musculature, no groin tear, and no bony abnormality such as a stress reaction.

The diagnosis of PNI was made based on the salient points of the history, physical examination, and supportive evidence by

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Fig. 1. A. T2-weighted axial MRI showing bright T2 signal (yellow arrow) isolated to perineum without involvement of neighboring thigh musculature. B. Coronal image with blue arrow denoting symptomatic area.

MRI. Further workup including biopsy was discussed; however, the patient elected not to proceed. At 2-week follow-up, the patient admitted she did not undergo as much activity modification as she previously mentioned and decided to undergo formal pressure mapping measurements for saddle fit, as well as a proper bike fit analysis. In light of this information, the patient and team participated in shared decision making to hold off on biopsy until she had made the appropriate changes. At 1-month follow-up she had changed both the saddle and saddle nose angle with the guidance of pressure mapping, adjusted geometric bike fit, and had modified her training regimen. She was able to resume riding including hill climbs. At 6-month follow-up (2 years since symptom onset), she reported complete resolution of symptoms. She was able to fully resume cycling without pain and is now averaging more time and mileage per week (88 miles/week, 6–7 hours). She also consulted with dermatology who agreed with PNI diagnosis and that biopsy was not warranted. Again, she was offered biopsy but she declined, stating resolution of symptoms with mechanical adjustment.

3. Discussion

Perineal nodular induration, also known as ischiatic hygroma [4,9,10], is a pseudoneoplastic lesion caused by repetitive micro-trauma from friction and vibration between the saddle and ischial tuberosities with shearing of the superficial perineal fascia [3,4,10]. Although the exact pathophysiology is debated [7], chronic lymphoedema and resultant fibrous tissue is a proposed mechanism [5,11]. In men, PNI develops as a mass next to the median raphe, posterior to the scrotum [3]. Among cyclists, it has been referred to as an accessory testicle [3,9], and is dubbed the “cyclist’s nodule” or “the cyclist’s third testicle” [9] due to its characteristic singular nodularity. As such, PNI is predominantly reported in males [6]. However, PNI can occur in females [5] and is likely underdiagnosed [6,7]. Other sports with saddle emphasis, such as equestrian sports, mountain biking, and even seated lawnmowing are also considered risk factors for PNI development [12,13]. European clinicians may be more aware of this condition [4,7,14] due to the established cycling culture compared to North American countries. Furthermore, there is potential for misdiagnosis in females [5] as the vulvar region can develop a wide range of mesenchymal lesions [5,11]. PNI has been mistaken for lipoma [6,8], and has even been referred to a specialty sarcoma center for concern of a growing paratesticular mass [6] which led to serial MRIs and biopsy. With better awareness, PNI can be diagnosed with a thorough history and physical exam [3,14], avoiding a more expensive workup. In uncertain sce-

narios where the diagnosis is unclear, advanced imaging can be considered [15,16].

In most clinical situations, ultrasound has been reported to be sufficient for imaging evaluation [15] and usually shows a well-delineated, hypoechoic mass with heterogeneous echo signal and edge shadowing, no doppler flow [4]. Ultrasound has an additional advantage in that it can be used to monitor treatment response [4]. Beyond ultrasound, MRI may be helpful as it provides information regarding lesion extent, as well as PNI features of fibrosis and hypovascularity [16]. MRI shows irregular hypointense T1 and hyperintense T2 signal in the perineum, just under the ischial tuberosity [6]. With PNI, there is no invasion of surrounding muscle tissue [14]. Low intensity signal on diffusion weighted imaging (DWI), with high ADC value consistent with no restricted diffusion is also reported [16].

Although a benign growth, the reported histopathology of PNI varies if biopsy is obtained. In one study [6], biopsy demonstrated collagenous tissue with fragmented elastic fibers interspersed with fibroblast-like spindle cells. Additional studies [6,8] showed fibroblastic proliferation with spindle and epithelioid cells, fibrinoid degeneration, and numerous blood vessels with entrapped fat cells. In all cases, collagenous myxoid degeneration and pseudocysts [4,10] appear with a haphazard mixture of adipose tissue, blood vessels, and nerve fibers with spindle-shaped fibroblasts [5]. Histopathologically, PNI may mimic nerve sheath tumors, fibromatosis, angiomyoma, and fibromyxoid sarcoma [6]. Importantly, atypical features (pleomorphism, cytological atypia, necrosis or mitotic figures) are absent [6].

In unclear clinical situations, we provide an algorithm (Fig. 2) to help with decision making. Although this case is in a female, PNI must be considered in both sexes. A starting differential for similar lesions includes lymphadenopathy, hydrocoeles/varicoceles, epidermal/sebaceous cysts, aneurysms, urethral pathologies/ syringocele, and hernias [3,5,6,8,11,14]. Localized lymphoedema and vulvar hypertrophy can occur in females. Cancer must be suspected if there is an unexplained mass, with possible diagnoses including pseudosarcoma, fibroblastic/myofibroblastic tumors, paucicellular fibrous proliferations, and mammary type fibroblastoma [3,8].

Treatment of PNI involves rest and removing the offending agent, in this case uneven saddle pressure. Saddle alteration and bike fit modification to selectively load the ischial tuberosities and unload the perineum is desired. In the author’s experience flatter rather than dome-shaped saddles help create an ischial “bridge” over the saddle thereby offloading the perineum. Soft gel cushions cause the pelvis to sink deeper and may paradoxically increase per-

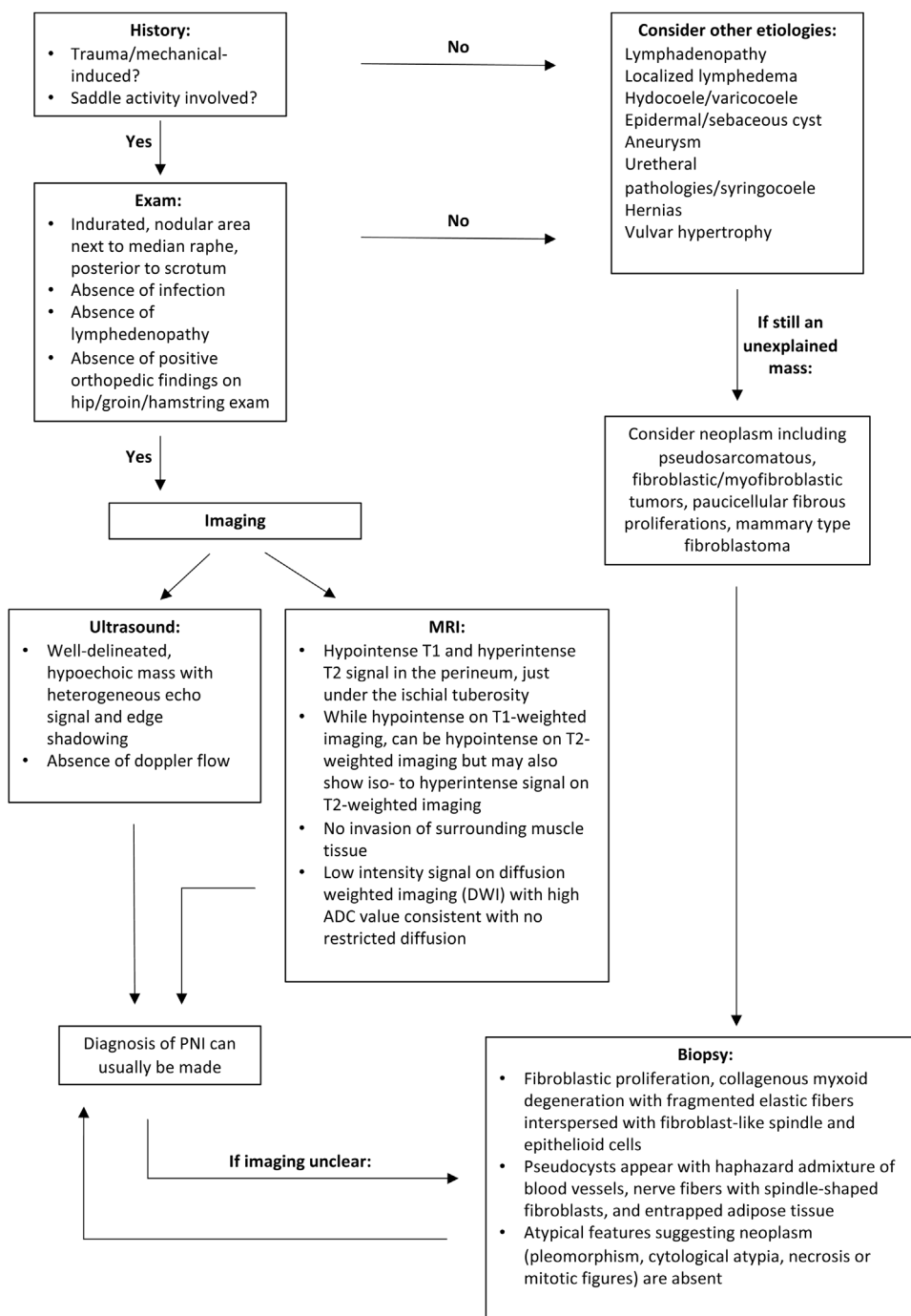


Fig. 2. Flow diagram for differential diagnosis of PNI.

ineal pressure, whereas harder saddles should relieve symptoms. A cutout or channel through the middle of the saddle may also help. With rest, saddle, and bike fit modification, most cyclists are able to return. Elite cyclists, however, may opt for medical treatments as they have already maximized bike and saddle fit. Intralesional corticosteroid injections may provide relief [6,7,14,17], but are recommended for smaller lesions due to risk of subcutaneous atrophy [4]. Hyaluronidase injections have also been utilized in some countries [3,4,14]. Finally, surgical excision and skin flap may be considered [3,14,17] for severe cases refractory to treatment. The athlete must be counseled on risk of scar tissue and recurrent symptoms after surgery. That being said, conservative management in males has been described [3,14,17].

4. Conclusion

What this case adds to the existing literature: PNI is a poorly recognized condition, and while predominantly reported in male cyclists it can occur in female cyclists or athletes engaging in other saddle sports and is likely underdiagnosed. It is poorly reported in female cyclists. With the recent growth of cycling in North America, PNI may become more prevalent and sports medicine clinicians need to be aware of this condition. It can be misdiagnosed for other masses or mesenchymal lesions, leading to expensive and invasive work-up. Thus, a thorough history and physical in the appropriate clinical context can lead to an accurate differential and the correct diagnosis, with supporting imaging findings by ultrasound and/or

MRI. Cytopathology can be utilized in unclear cases. With adequate activity modification, correct bike and saddle fit, the cyclist should be able to successfully return to sport.

Declaration of Competing Interest

There are no conflicts of interest.

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Ethical approval

This is a case report which discloses no private HIPPA information or patient identifiers. This study is exempt.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Mackenzie Norman—data analysis, paper writing.
Kenneth Vitale—study concept, paper editing.

Registration of research studies

NA.

Guarantor

Kenneth Vitale.

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