

Perineal Nodular Induration in Cyclists

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To the Editor:

Perineal nodular induration in cyclists, also known as ischial hygroma, third testicle, accessory testicles of cyclists, or cyclists nodule, is an entity that is relatively well known to sports medicine specialists and to professional and amateur cyclists, but practically unknown to other doctors.^{1,2}

A 49-year-old man with no relevant history visited our department with a tender lesion in the right ischial region that had appeared a year earlier. Physical examination revealed a 4-cm nodule of rubbery consistency, covered by normal skin. Ultrasound and fine-needle aspiration studies were performed but the results were inconclusive. Magnetic resonance imaging (MRI) (Figure 1) revealed a nodular lesion with a fibrotic appearance with a maximum diameter of 3 cm, in intimate contact with the lower surface of the right ischial ramus, and changes in trabeculation and infiltration of the surrounding fat, compatible with perineal induration of the cyclist. The contralateral region revealed similar, though less marked, changes. The patient was questioned again and confirmed that he was a cyclist.

The lesion was excised and histologic study revealed a tumor lesion with poorly defined borders, consisting of a multilocular cystic formation with a fibrinous content and not surrounded by epithelial or synovial cells; there were adjacent areas of aseptic necrosis (Figure 2). The rest of the lesion showed a fibrous proliferation with fusiform cells without atypia; in addition there were areas of hemorrhage, foci of myxoid degeneration, and, occasionally, groups of mature adipocytes trapped in the fibrous proliferation. Immune staining of the fusiform cells was positive for vimentin and factor XIIIa and negative for estrogen receptors, progesterone receptors, S-100, CD34, actin, and desmin. The Ki-67 proliferation index was 0%.

Clinically, perineal induration in cyclists usually manifests as 2 nodules (1 on either side of the perineal raphe), although it occasionally presents as a single nodule (third testicle), located immediately behind the scrotum, close to the ischial tuberosity. The skin covering the nodule presents a normal appearance and the nodule usually measures between 2 and 3 cm. Palpation reveals a fibroelastic consistency and adherence to the adjacent connective tissue may be appreciated.^{1,2} Our patient consulted with a nodule located in the right ischial region, although NMRI also revealed an incipient lesion of similar characteristics on the contralateral side.

Histologically, the nodule is characterized by a central pseudocystic formation, which develops in an area of

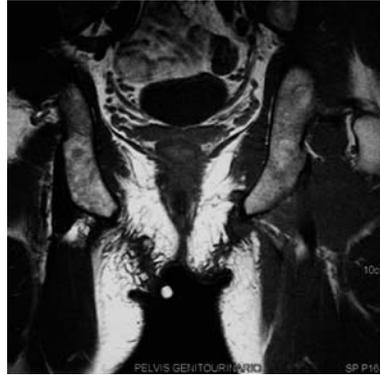


Figure 1. Magnetic resonance imaging (cutaneous marker on the studied lesion): nodule in the right perineal region, hypointense in T1 sequences.

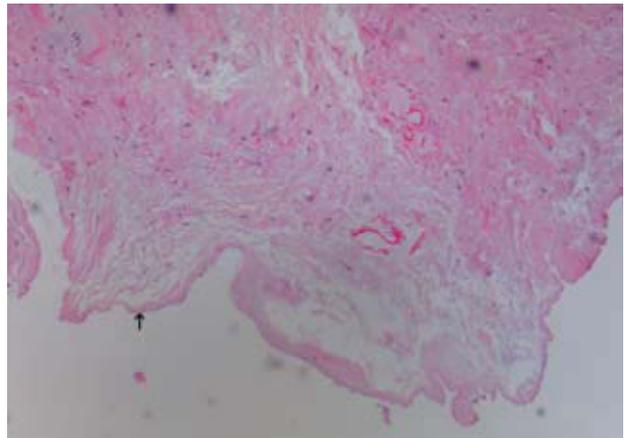


Figure 2. Cystic cavity lined by a fibrinoid material (↑) with no surrounding epithelial membrane. Below, lax conjunctive tissue with few cells and myxoid changes (hematoxylin-eosin, ×20).

aseptic necrosis that develops in the connective tissue of the surface perineal fascia. This tissue becomes homogeneous and loses its fibrous structure. Erythrocyte diapedesis and a mild reactive inflammatory infiltrate may also be observed.¹⁻³ Some of these findings are reminiscent of those of ischemic fasciitis found over bony prominences in elderly patients; this provides a clue to the etiology of this disorder.^{2,4,5} Although not universally accepted, it seems probable that the development of these lesions is linked to the compression of the soft tissue between the saddle and the ischial tuberosity, and to friction between the perineal fascia and the bony structures due to movement of the saddle; this could lead to collagen degeneration, myxoid changes, and formation of the pseudocyst.¹⁻³

Diagnosis of these nodules may be particularly difficult if a history of cycling is not detected. The differential diagnosis should be established with common minor disorders (cysts and lipomas), and with other, rarer and more important conditions such as aggressive angiomyxoma. This rare variety of myxoid tumor can infiltrate locally and is associated with a high risk of local recurrence after resection, but it has no metastatic potential. It generally affects young women and arises in the soft tissue of the pelvis and perineum. Rare cases have also been reported in men, where it develops in the scrotum and groin, presenting as masses or nodules in these areas.^{6,7}

Treatment of these perineal indurations in cyclists is generally surgical, although, if this is not possible because the patient is a professional cyclist, conservative treatment with rest and infiltrations of steroids or hyaluronidase may be tried.¹⁻³

These cyclist's nodules are a genuine handicap for professional cyclists and contraindicate cycling for amateur cyclists.¹

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Conflicts of Interest

The authors declare no conflicts of interest.

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Perianal Verrucous Papules in a Patient With Bannayan-Riley-Ruvalcaba Syndrome

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To the Editor:

Bannayan-Riley-Ruvalcaba syndrome (BRRS) is a rare autosomal dominant genetic disorder caused in approximately 60% of cases by mutations on the *PTEN* gene, a tumor suppressor gene on chromosome 10.¹ Typically, patients with this syndrome present a triad of macrocephaly, genital melanotic macules, and hamartomatous polyps in the intestine. Identical mutations of the *PTEN* gene have been reported in 80% of patients with Cowden syndrome, also known as multiple hamartoma syndrome.² Both syndromes have been reported in some families and some members may present phenotypes corresponding to both entities.³ In view of the clinical and genetic overlap of BRRS and Cowden syndrome, they are currently considered as different

phenotypic expressions of the same condition known as *PTEN* syndrome.^{1,3} This spectrum also includes Proteus and Proteus-like syndrome, in which *PTEN* mutations are found in a smaller proportion of cases.¹

There are numerous skin manifestations common to BRRS and Cowden syndromes, including oral, facial, and acral warts or verrucous papules, lipomas, trichilemmomas, and inverted follicular keratosis.¹⁻³ The presence of multiple trichilemmomas in the facial region is diagnostic of Cowden syndrome.⁴

We report the case of a 33-year-old man with no family history of interest who was diagnosed with BRRS on the basis of a range of abnormalities, the most prominent being marked macrocephaly with hypertelorism, slight mental retardation, genital lentiginos, intestinal polyps,