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Painful Encysted Adiponecrosis on the Buttock: Case Report and Etiopathogenic Discussion

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1. Abstract

1.1. Purpose Encysted adiponecrosis is a benign subcutaneous lesion characterized histologically by encapsulated fat necrosis. Our purpose was to present and discuss an atypical case of adiponecrosis revealed by a chronic hard mass of soft tissue becoming painful, associated with an erythematous skin nodule of undetermined origin.

1.2. Case A 66-year-old woman had an indurated and erythematous papule on the right buttock for many years that started to become painful. Ischial bursitis related to horseback riding during youth was first evoked. On MRI, the mass extended from the skin towards the ischium as a predominantly amorphous calcified lobulated lesion surrounded by slight inflammation of skin and subcutaneous fat. Rest was advised, but the lesion became increasingly more painful and was removed surgically. Histology found old calcified encysted adiponecrosis. The role of repetitive micro-trauma or potential unrecognized equestrian cold panniculitis was discussed.

1.3. Conclusion The final diagnosis could be a mixed form between encysted fat necrosis with fistulization to the skin and old equestrian cold panniculitis, one having led to the other subsequent to possible trauma or repetitive micro-trauma. Surgical excision was followed by clinical remission.

2. Introduction

Chronic soft tissue masses are frequent findings. Causes are multiple and can be inflammatory, infectious, tumoral or traumatic. The buttock is a common site of dermatological, rheumatological, orthopedic, digestive or even gynecological pathologies.

Encysted adiponecrosis is a benign, usually asymptomatic, subcutaneous lesion characterized histologically by encapsulated fat necrosis. It occurs in areas where the adipose panniculus is thickened by nodules or subcutaneous plaques. The apparent mechanism of onset is unclear. It seems to be related to trauma that would be responsible for vascular insufficiency with secondary ischemic necrosis of a hypodermal adipose lobule then encystment of the site of necrosis [1,2].

Here, we describe the diagnostic and therapeutic approach for a patient with an atypical presentation of a chronic hard mass on the buttock that became painful.

3. Case Report

Our patient was a 66-year-old woman with a past history of breast cancer in remission after conservative surgery (lumpectomy) and hypercholesterolemia treated by diet. She had no history of smoking or excessive alcohol consumption. In her youth, she had practiced horseback riding intensively, several times a week, all year round, with exposure to cold and humidity. She stopped riding in 1980.

She first consulted in our department in 2013 for excessive passage of gas per anus. At examination, she had minor flatus incontinence and weak anal tonus, apparently related to past constipation (no history of proctologic surgery or pregnancy). There was no visible lesion of the buttock and palpation did not revealed any mass. Laxative treatment and perineal rehabilitation were advised. She again consulted in 2018 for the same reason and physical examination revealed a 5-cm diameter hard solid but mobile mass of the right buttock. The mass was painless and there was no skin anomaly. The patient reported that this nodule had been present and stable for several years. She had never had any other skin lesions on the thighs or hips. A perineal MRI was performed (Figure 1), showing an amorphous calcified lobulated lesion surrounded by slight inflammation of skin and subcutaneous fat. The mass measured 7x4 cm, extending from the skin towards the right ischium. Contra laterally, slight abnormalities of the subcutaneous fat were noticed without skin involvement. A CT-scan was also performed (Figure 2) and confirmed the existence of multiple well-delimited and calcified nodules located in the fat of the right buttock, from the skin to the ischium, and reaching about 7cm in the longest axis.

Because of the asymptomatic nature of the mass, and the absence of any criteria of radiological malignancy, a biopsy was not performed. A wait-and-see attitude was proposed and the patient was advised to rest.

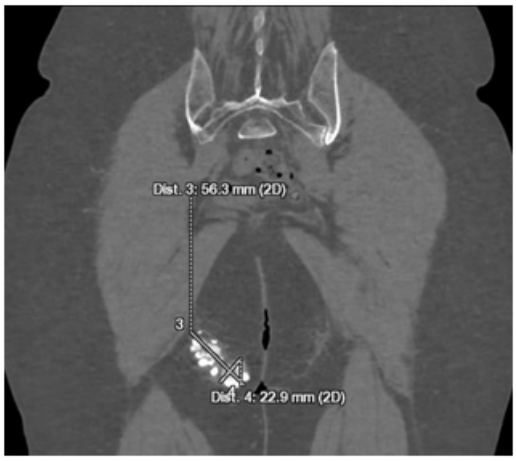
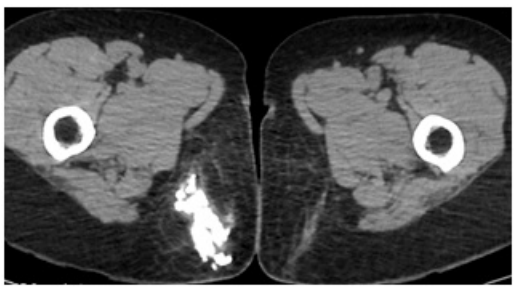


Figure 1: Pelvic MRI showing multinodular structures of the right buttock, which extended between the skin surface and deep tissue.



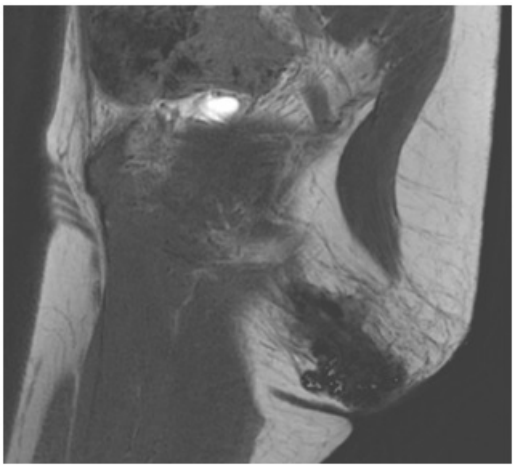
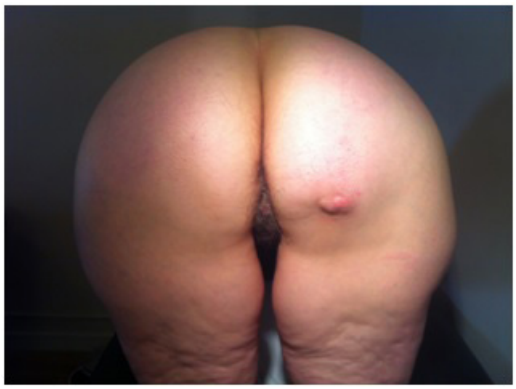


Figure 2: Pelvic CT-scan showing calcified polylobed lesion of the right buttock (approximately 7.5x4cm), surrounded by minimal contrast enhancement of deep subcutaneous fat

During the following year, the lesion became progressively symptomatic. Precisely located pain developed, becoming particularly intense when sitting. The patient thought that the mass had grown. On physical examination, the nodule appeared erythematous (Figure 3). A new CT scan was performed showing no clear modification. Due to the worsening symptoms (pain becoming daily), making the sitting position very uncomfortable, surgical removal was proposed in June 2020 and achieved via a perilesional incision, with dissection close to the lesion, extending to the ischium which was exposed. The lesion appeared in the form of a polylobate mass with thick whitish content, a presentation compatible with aseptic necrosis (Figure 4). Lymph node dissection was not performed. The wound was left open. Wound care, with disinfection, wicking and dressing, was performed once a day until complete healing.

Histological analysis of the lesion showed multiple clusters of varying sizes, composed of an amorphous, calcified eosinophilic substance with some zones of inflammatory reaction consisting mainly of resorptive multinucleated giant cells and a few lymphocytic and histiocytic cells (Figure 5). The old calcified lesions were histologically compatible with the diagnosis of encysted fat necrosis.

The wound healed completely after 3 months and the patient no longer felt bothered.



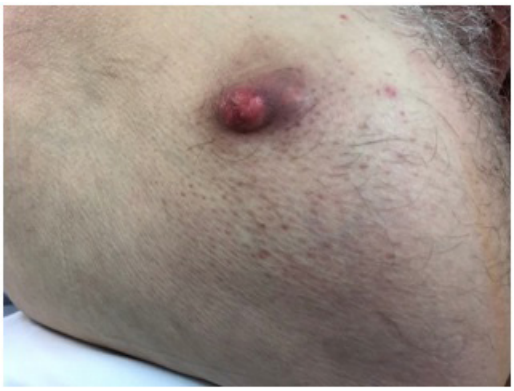


Figure 3: Clinical view of the painful erythematous nodule

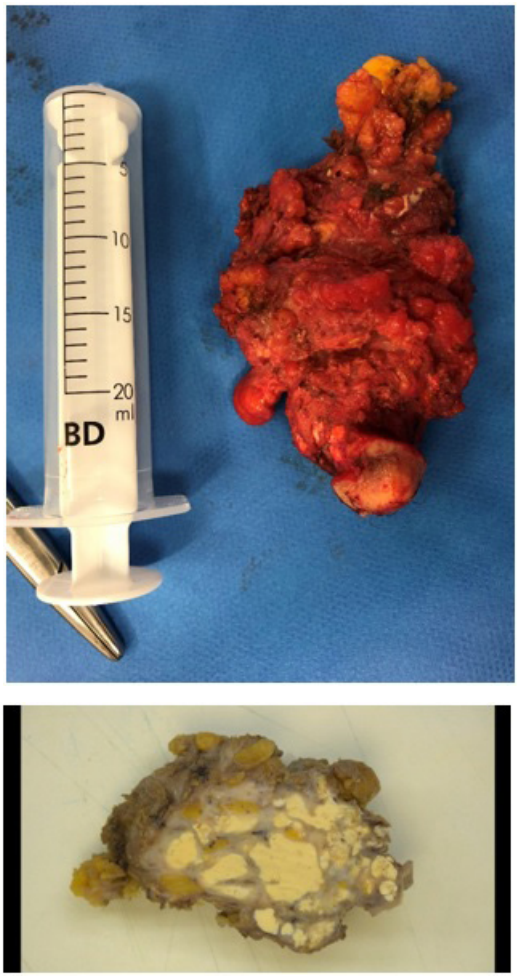
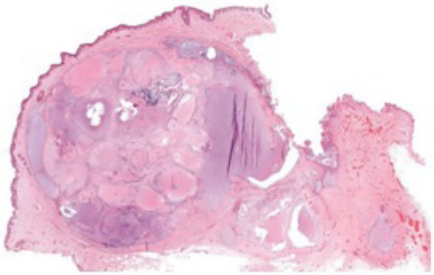


Figure 4: Postoperative view of the resected lesion before and after formalin



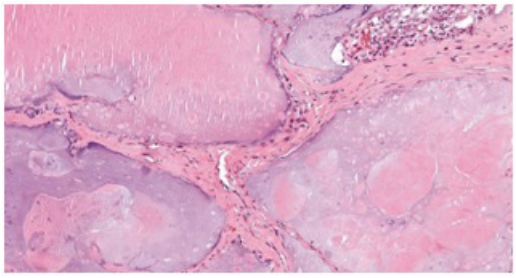


Figure 5: Old calcified encysted adiponecrosis lesions. Multiple clusters of varying size, composed of an amorphous, calcified eosinophilic substance with minimal inflammatory reaction.

4. Discussion

In this case of chronic soft tissue mass, before imaging the first diagnosis suggested by the clinical symptoms (pain and some minimal inflammation), together with the perception of deep contact with the ischial tuberosity, was ischial bursitis. The bursa, located between tendon insertions over the ischium and the muscle gluteus magnus, is an inconstant synovial pocket facilitating sliding that can bleed, inflame or become infected. Ischial bursitis follows prolonged sitting and repetitive micro-injuries. It is manifested by sharp, one-sided pain of the buttock. A swelling is sometimes palpable over the ischium [3].

Clinically evoked, this diagnosis requires cross-sectional imaging (ultrasound, CT scan or MRI) for confirmation and elimination of differential diagnoses. Although there are no specific imaging signs giving the diagnosis of ischial bursitis, what is most commonly found is an irregular cortex of the ischial tuberosity or calcification over or near the tuberosity. By insisting on the typical localization of ischial bursitis, KH Cho's study specifies the importance of differentiating ischial bursitis from peri-rectal abscess, hematoma, epidermoid cyst, dermoid cyst or hydatid cyst when the lesion presents a cystic appearance: the bursa usually widens towards the ischiorectal fossa inferomedially, the common hamstring tendon, located laterally, acting as a barrier [4]. In our case, MRI was not in favor of this diagnosis: here the lesion was located in the fat, on the medial side of the ischium, while no bursa was visible. MRI also allowed us to rule out the diagnosis of an infectious or malignant lesion affecting bone, skin, or perianal or rectal tissue. Contrast MRI is needed to visualize soft tissue masses, especially if they exceed 5 cm or show a suspicious character on the ultrasound or CT scan. Moreover, biopsy material or a surgical specimen are needed to confirm the diagnosis of tumor growth, and can also help rule out an infectious process.

Essentially, the diagnosis of ischial bursitis is clinical and radiological, but there have been a few histological descriptions in the literature reporting cartilage alterations, walls lacking lining cells consisting of fibrous connective tissue, and unspecific chronic inflammatory cells [5-7]. To our knowledge, adiponecrosis lesions have not been described. Adiponecrosis is a reversible pathology

that heals with rest. In our case, as radiological study and histological analysis did not show any element in favor of bursopathy, and the adiponecrosis was massive, the most likely differential diagnosis was nodular cystic fat necrosis of subcutaneous tissue.

Encysted adiponecrosis is a benign subcutaneous lesion corresponding to a particular form of traumatic panniculitis [1]. The initial lesion unnoticed, there is an encystment of adipose tissue necrosis. The histological appearance is that of a nodular lesion, consisting of necrosis surrounded by a fibrous capsule of ischemic origin. This area of adipose necrosis may have calcifications, and lipomembranous changes depending on the age of the lesion, however it has little or no significant inflammatory infiltrate [2,8]. Clinically, encysted adiponecrosis most often presents with an asymptomatic subcutaneous nodule whereas in our case, the lesion became painful secondarily and fistulized to the skin. The course finally resembles that of postoperative breast fat necrosis. Under the effect of post-operative and vascular ischemia, soft tissue may undergo fat necrosis. This occurs in areas where the adipose panniculus is important, leading to palpable and often painful subcutaneous nodules or plaques in the breast [9]. These firm, calcified lesions sometimes have episodes of inflammatory flare-ups with fistulization to the skin.

This old calcified encysted adiponecrosis, without crenulated hyaline membranes around the cystic cavities evoking lipomembranous fat necrosis but with a dermal and hypodermal perivascular inflammatory infiltration without modifications of the dermohypodermal junction, may be secondary to equestrian cold panniculitis.

First described in 1980, equestrian cold panniculitis is a panniculitis affecting the upper lateral side of the thighs and triggered by cold weather, especially in young female horse riders wearing tight, non-insulating pants [10]. This cold-associated thigh panniculitis represents a subacute vascular injury to the dermis due to prolonged and intermittent exposure to cold [11]. A Finnish study found possible risk factors: young age, female sex, tobacco, time spent riding and a high body mass index [12]. Cold may lead to an increase in blood viscosity and predisposes to panniculitis.

Our case is interesting because of the chronic form and the location of the lesion. The skin appearance also seems unusual: typically, the presentation is livid-red, infiltrated. Erythematous nodules can be observed but are infiltrated and do not exteriorize as in our case. The inflammatory infiltrate was not very pronounced in our case, as in case 3 of Ferrara's series that showed a late stage of cold-associated perniosis of the thighs with only minimal dermal and subcutaneous inflammatory infiltrate [13]. This could lead to chronic forms, which are uncommon. Usually, the diagnosis is made from the analysis of biopsy material, which shows dermal and hypodermal perivascular inflammatory infiltration without modifications of the dermohypodermal junction.

In previous studies, the lesions were usually reversible by removing the predisposing factors. The buttock area is very frequently exposed to compression by body weight, as well as friction and tension while sitting. It would therefore be less well vascularized, which could promote lesion sustainability and deep extension.

The pain reported by the patient and the atypical presentation prompted us to perform imaging explorations and even surgery. We have been unable to find any radiological description of equestrian cold panniculitis in the literature. In cases reported, masses were sometimes found deep in the lesion. For our patient, surgery appeared to be highly beneficial, providing immediate relief of the intense pain, only hours after removal of the lesion.

One other report has suggested that, in order to rule out differential diagnoses such as lupus, cryoglobulinemia or infection, additional explorations including white blood cell count, erythrocyte sedimentation rate, C-reactive protein, antinuclear antibodies and cryoglobulins, should be performed in case of an atypical localization that would have an unfavorable outcome despite correct management or persistence of the lesions in the summer [14].

Equestrian cold panniculitis is most often localized on the lateral aspect of the thighs, therefore outside the zone of compression and superficial, while in our case the lesion seems to have developed inwardly, exteriorizing to the skin. Furthermore, equestrian cold panniculitis is favored by cold which does not seem to be the case in our patient.

Our final diagnosis was encysted or encapsulated adiponecrosis. Such lesions are generally old, secondary, and sequelar, but may occasionally be arise from an old unrecognized equestrian cold panniculitis, as evidenced by the minimal inflammation, the discrete histiocytic infiltrates and the fibrosis around the pseudocysts. Indeed, the lesion we excised would correspond to old hypodermal adiponecrosis eliminated transcutaneously, which would explain the recent development of overt symptoms.

5. Conclusion

Characteristic histologic findings of a hard mass of the buttock with dermohypodermal changes and the presence of old calcified encysted fat necrosis lesions led us to describe a case of adiponecrosis possibly secondary to an old unrecognized equestrian cold panniculitis with an atypical presentation located on the medial side of the ischium in a woman who was exposed to repetitive micro-trauma due to horseback riding. CT and MRI ruled out a tumor formation, infection, and osteoarticular or other chronic inflammatory diseases. Surgical excision enabled histological examination and led to complete healing and symptom resolution.

6. Acknowledgments

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