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CASE REPORT

Para-anal lipoma as a rare consequence to perineal trauma. Case-report and review of the literature

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ABSTRACT

Introduction: Lipomas are the most common benign mesenchymal tumors which can be found in any part of the body. Nevertheless, their etiology and pathogenesis remain unknown. It is hypothesized that some of these lesions could result from an acute or chronic trauma.

Patients and methods: We report a case of a 54-year-old man presenting a perineal lipoma which volume grew rapidly after he fell on his buttock, in the context of inaugural epileptic seizure. Pelvic MRI showed a voluminous fatty mass, measuring $6.6 \times 5 \times 9$ cm without any signs of local invasion. Furthermore, we review the latest research on lipomas originating from traumatic lesion.

Results: The mass was completely excised in one block under general anesthaesia, using an elliptical incision and a deep dissection. We did not close the skin incision in view of the cutaneous defect. Post-operative recovery was uneventful and the patient was discharged from hospital two days after the operation. Histopathology indicated a reorganised lipoma with no evidence of malignancy.

Conclusion: Perineal lipomas are extremely rare, pathological examination of imaging guided biopsies are needed to exclude malignancy especially a well-differentiated liposarcoma. MRI remains the first option and radical surgical excision is the gold standard treatment.

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KEYWORDS
Perineal lipoma; traumatic lipoma; lipoma etiology; lipoma surgery

Introduction

Lipomas are the most common mesenchymal tumors in a human body. They are benign soft tumors, well enclosed by their fibrous capsule, and also mobile under the skin when it comes to subcutaneous lipomas. Their etiology and pathogenesis remain unknown. However, lipomas frequently show translocations involving HMGA2, resulting in over expression of the fusion protein [1–3].

These tumors are generally asymptomatic but can sometimes cause discomfort or even aesthetic concerns. Rarely these lesions grow and thus compress neurovascular structures and as consequence may be painful [4,5].

Although, they usually occur in adults between 40 and 70 years old, some perineal lipoma's cases were already described in pediatric literature but it proved to be extremely rare [6]. Most of these perineal masses are congenital; and therefore are detected and treated in early age. Although perineal masses are unusual abnormalities, 72% of these are associated with an accessory scrotum or other anorectal or urogenital malformations [7]. This frequent association is probably due to a common embryologic origin [8–10].

Generally, these tumors keep a steady volume in the time because they are growing very slowly. Nevertheless, after a localized trauma, for reasons currently still under investigation, the mass volume may brutally increase. For several years now, more and more studies and case reports have shown that several types of mesenchymal tumors, that is, desmoid tumors, lymphoid tumors and lipomas are sometimes directly resulting from an acute or chronic trauma [11].

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Fatty tumors arising at the site of previous trauma are known as traumatic pseudo-lipomas, and tend to develop with an incidence about 1% [12]. We present this perineal lipoma which pathogenic mechanism could be the patient's fall during his first epileptic seizure. In addition, given that these tumors in perineal regions are extremely rare, here we are going to put emphasis on a full preoperative evaluation and its importance in order to assess the mass depth, the risk of malignancy and its extension in neighboring structures such as sphincters.

Case report

A 53-year-old man was referred to our Colorectal Surgery Unit for the treatment of an anal swelling that has increased size since two months. The mass appears to be a consequence of a fall when he lost consciousness due to a first epileptic seizure. After this fall, the patient complained about perineal pain and an anal swelling becoming bigger day after day.

When the patient was counselled at our outpatient clinic, two months after the initial trauma, the mass had become painless but its size was still increasing rapidly and was causing discomfort when sitting in the same position for a long time. Besides, the patient presented no intestinal complaints and no blood per ano.

In his surgical history, we can notice a grade I planum sphenoidale meningioma that caused his first epileptic seizure and was completely excised in our institution. The patients' sole treatment was Levetiracetam 500 mg daily, recently prescribed by his neurosurgeon.

On clinical examination, we found a voluminous exophytic elongated, left para-anal mass of soft consistency, measuring 4×8 cm, located dorsally to the anus (Figures 1 and 2). Valsalva maneuver caused no modification and no bowel sounds were audible by auscultation of the mass. In addition, there was no sign of inflammation and digital rectal examination was normal. Patient showed no signs of genital abnormality. His BMI was 23.7 kg/m^2. Rectosigmoidoscopy was normal but anorectal endoscopic ultrasound confirmed the presence of a para-anal, hypo-echogenic, left posterior mass with regular contours and well delimited. The mass did not invade the anal sphincters, but caused a local compression (Figure 3). The pelvic magnetic resonance imaging (MRI) showed a pretty voluminous fatty mass, measuring $6.6 \times 5 \times 9$ cm without any signs of local invasion (Figure 4). In deep

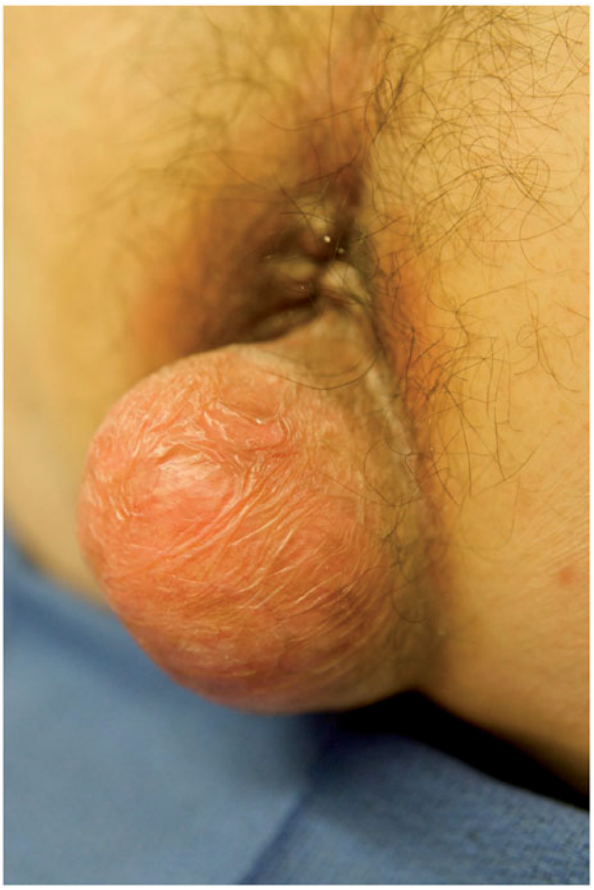

Figure 1. Soft consistency, voluminous perineal mass, measuring 4×8 cm.

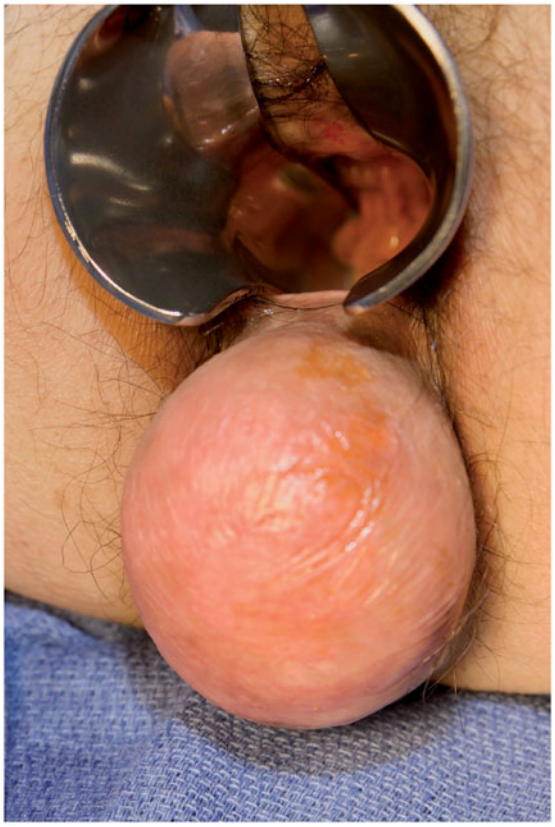

Figure 2. Lesion located posterior to the anus. There is no sign of inflammation, the digital rectal exam is normal. The picture shows the Pratt rectal speculum in the retracting the anus and allows to identify the absence of invasion of the anal canal.

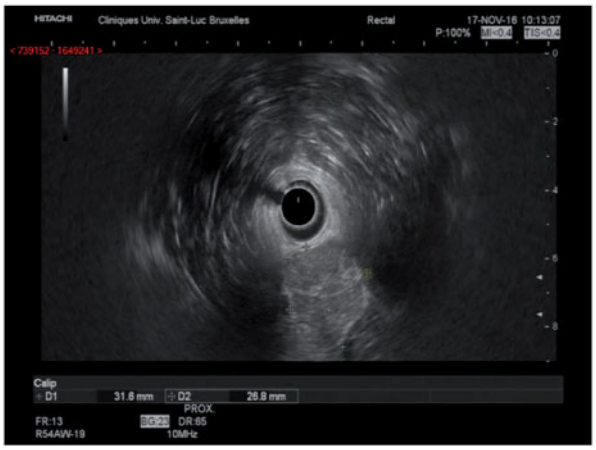

Figure 3. Eus: hypo-echogenic structure, well delimited; with regular contours.

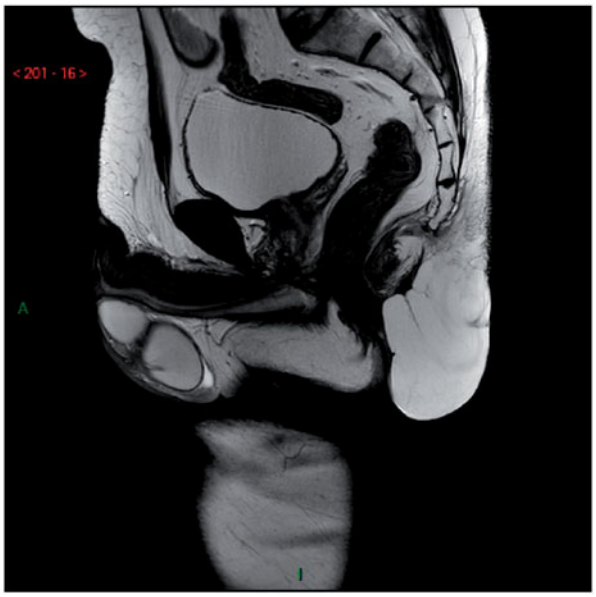

Figure 4. MRI sagittal T1: mass without any signs of local invasion.

immediate contact with the external anal sphincter, it is pushing it laterally. The MRI is in favour of a lipoma (Figures 5 and 6) but malignancy cannot be ruled out, considering a discreet 5 mm nodular portion with hypo-signal on both T1 and T2 sequence (Figure 5).

CT-scan guided biopsies were performed and microscopic examination revealed mature adipose tissues showing mainly some slightly hyperchromatic nuclei, discreetly increased in size. An MDM2 gene amplification allowed to clearly exclude the diagnosis of a well-differentiated liposarcoma.

The mass was completely excised under general anesthesia. It was resected using an elliptical incision and a deep dissection was performed aiming to remove the mass and the cutaneous tissue in one block. We did not close the skin incision in view of the cutaneous defect (Figures 7–9).

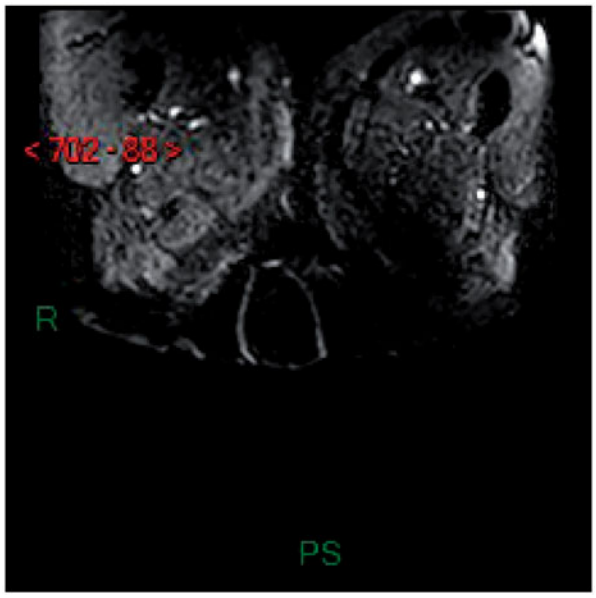

Figure 5. Fat-saturated MRI: lipoma's increased signal intensity decreases with fat-saturated sequences.

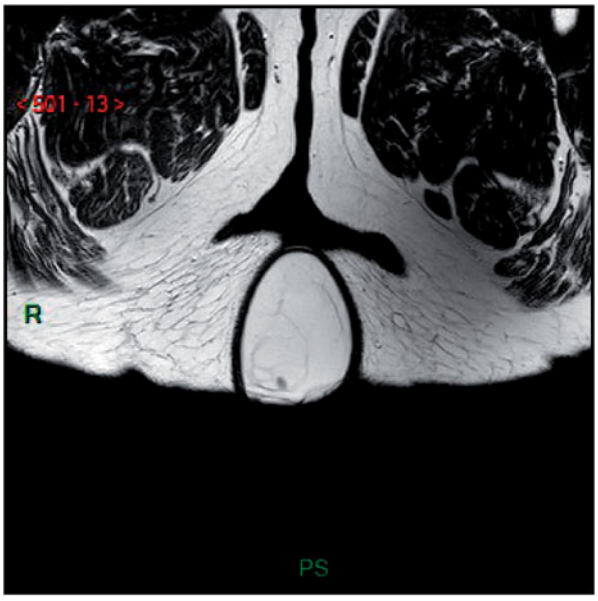

Figure 6. MRI: fatty mass, measuring 6.6 × 5 × 9. Presence of a 5 mm nodular portion in hyposignal on T1.

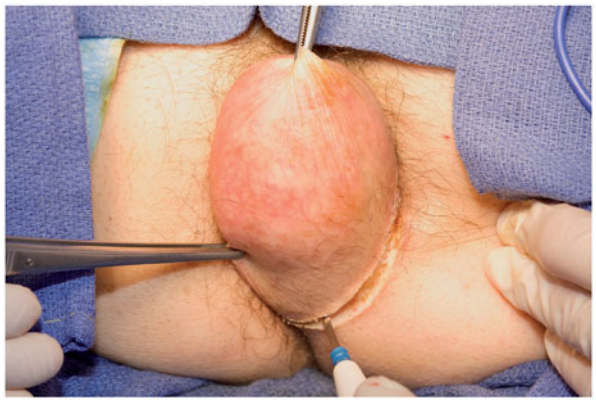

Figure 7. Elliptical incision and a dissection of the mass.

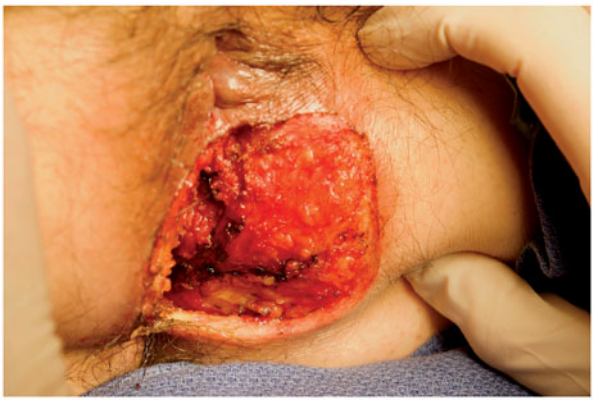

Figure 8. Radical resection of the tumor. Wound closure was not performed in view of the cutaneous defect.

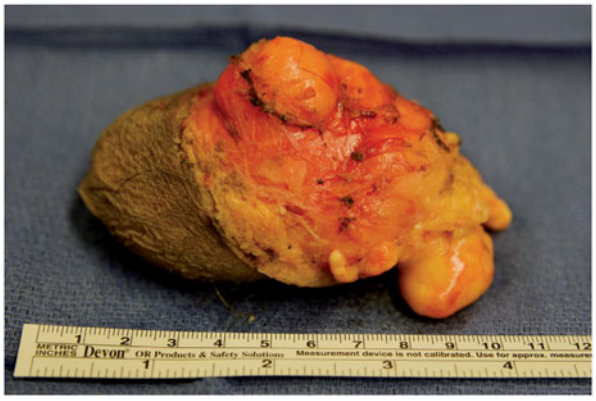

Figure 9. Macroscopic specimen of the fatty mass after complete resection.

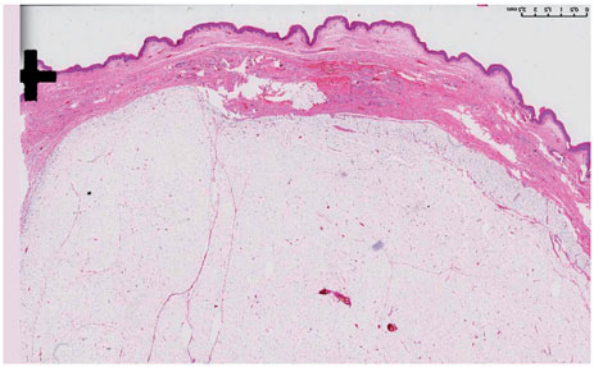

Figure 10. Histological examination revealed that the mass was composed of fatty tissue.

Post-operative recovery was uneventful. Local care and grade I and II analgesics were prescribed and the patient was discharged from hospital two days after the operation. Histolopathological examination indicated a reorganised lipoma, consisted of mature adipose cells, with no evidence of malignancy (Figures 10 and 11).

The patient came back for his out-patient counselling two weeks postoperatively. The scar evolved positively without evidence of infectious complication.

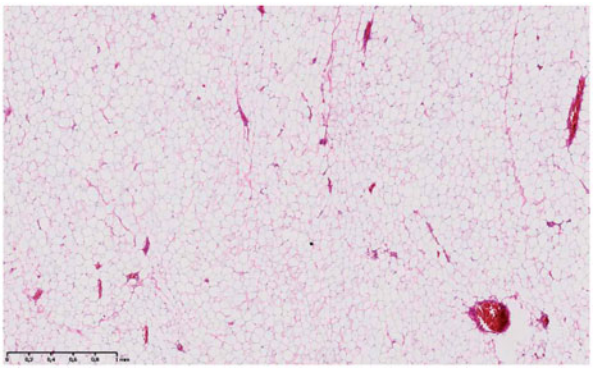

Figure 11. The mass consisted of mature adipose cells, with small periphery nuclei, pushed by a large, single vacuole.

Discussion

Lipomas localized in perineal region are extremely rare which warrants a serious and cautious management. A through imaging evaluation is mandatory, considering that treatments of perineal masses is highly depended on their differential diagnosis.

For our patient, we performed a rectosigmoidoscopy, an endoscopic ultrasound and a pelvic MRI which was followed by a CT guided biopsy. These examinations confirmed a lipomatous benign lesion. An excision was performed under general anesthesia.

The main differential diagnoses of perineal masses are: primary or secondary hernia and epidermoid or dermoid inclusion cyst. Liposarcoma and lipoblastoma are very rare and therefore not a main differential diagnosis.

Lipomas frequently show translocations involving HMGA2, resulting in over expression of the fusion protein. HMGA2 fusions can be detectable by immunohistochemistry, and be useful to help pathologists to distinguish between normal mature adipose tissue (HMGA2 negative) and adipose tumor such as lipoma or liposarcomas (HMGA2 usually positive). However, if the immunohistochemical detection is negative, could still be a non-HMGA2 fusion lipoma, which is the case in approximately 30%. If the immunohistochemistry is positive, then there is proof that the fusion is present and that this is a real lipoma and not pseudolipoma/pseudotumor [1–3]. We do not have this immunohistochemical detection, but it is commercially available; would need positive and negative testing on cases of established lipoma and ordinary fat.

The atypical localization of the lipoma of our patient and the uncertain etiology of it, could as well lead in the differential diagnosis to an accessory scrotum associated with a lipoma. Only three

cases in the literature have been reported describing accessory scrotum associated with a lipoma in adult, which were treated surgically [13,14].

Only a very small number of publications and case-reports have been published about perineal lipomas in adults and even less reports about perineal masses arising from trauma. Nevertheless, already in 1932, Adair described [15] for the first time, two cases of patients who had secondary lipoma due to trauma. Then several theories emerged struggling to explain the appearance of this adipose tissue, at the same site of previous acute or chronic trauma. In general, secondary lipomas appear between a period of time estimated from 5 month to 6 year after the trauma [5].

The first theory was purely mechanical, as Brooke and MacGregor [16] identified in 1969 some post-traumatic prolapse of fatty tissue through Scarpa's fascia. For these entities, the term pseudolipoma was used for non-encapsulated fatty tissue on MRI, histologically similar to lipoma, but developed in an unusual localization. Then some authors proposed other mechanical explanations such as septa ruptures, anatomical defects or herniations to explain the presence of these lipomatous masses [17,18].

Nonetheless, these mechanical theories could not explain every post-trauma lipoma. Nowadays the most recent theory tries to explain this phenomenon, involving local inflammation due to acute or blunt trauma. Hence, the traumatic impact and the microhemorrages are believed to be behind *de novo* formation of this fatty tissue. Inflammatory mediators, cytokines and growth factors released after the trauma, are reported to induce differentiation and proliferation of preadipocyte into mature adipocytes. Signorini et al. [19] are the first to suggest *de novo* formation of adipose tissue, thanks to preadipocyte stimulation. Recent genetic researches put forward the likelihood that lipoma development might result from an association of necrotico-hemorrhagic optimal mid and prolonged inflammation after an acute trauma or chronic micro-trauma [5–20].

MRI examination is undoubtedly the best way to define the tumor size, invasion depth as well as inter-sphincteric extension. It is also an excellent examination to point out urogenital or anorectal abnormalities. Moreover, on MRI, it is possible to distinguish lipoma from liposarcoma or pseudolipoma [5–12]. However, MRI cannot distinguish between lipoma and lipoma-like liposarcoma/well-differentiated liposarcoma. Pseudolipoma is a soft adipose tissue quite similar to lipoma, though there is no fibrous capsule on MRI.

The diagnostic evaluation can also be completed afterwards, with an ultrasound or a CT-scan. Even though lipoma diagnosis is essentially clinical, a voluminous 4×8 cm mass on clinical examination, which has grown quickly in size, could as well be linked to well-differentiated liposarcoma. Thus, complete imaging and, sometimes, biopsy have to be done to distinguish these two lesions.

Treatment of these masses require radical resection. It is especially the mass size, its relation with neighboring structures and the differential diagnosis that influences surgeon's decision and extent of surgery.

Conclusion

Perineal lipomas are unusual tumors which require a complete imaging evaluation in order to assess tumor extension, and eventually rule out other urogenital or anorectal anomalies. In some cases, pathological examination of imaging guided biopsies are needed to exclude malignancy especially a well-differentiated liposarcoma. MRI remains the first option and radical surgical excision is the gold standard treatment.

Disclosure statement

No potential conflict of interest was reported by the authors.

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