“Bone in the penis” or fasciitis ossificans of the penis – a first time description of a pseudo-tumor at an extraordinary site

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Case Report

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Abstract

Background: Fasciitis ossicans is a rare subtype of nodular fasciitis, a benign soft tissue tumor of reactive character. It is often misdiagnosed as a malignant tumor due to its rapid growth. Most commonly fasciitis ossicans originates from the subcutis with appearance throughout the whole body, but it may arise from extraordinary sites, too.

Case presentation: We report the first case ever described of this entity deriving from the penis in a male patient presenting with a tumor on the glans penis. The tumor was resected because of a suspicion of penile cancer. Initial histopathological analysis misled to diagnosis of squamous cell carcinoma. Pathologic counseling finally led to the diagnosis of fasciitis ossicans of the penis deriving from the glans penis by showing ossification.

Conclusion: This case shows that diagnosis of fasciitis ossicans can originate from any soft tissue, even from penile soft tissue.

Background

Fasciitis ossicans is a pseudotumor of the soft tissue. It is classified as a rare subtype of nodular fasciitis – a frequent kind of reactive neoplasm of fibroblastic or myofibroblastic derivation with unknown aetiology (1). Presence of ossification in nodular fasciitis confirms fasciitis ossicans. It affects mostly adults without sex predilection but can be detected in children, too. Common locations include the extremities, head and neck, trunk and fascia or muscle of the breast. Due to its rapid growth, hypercellularity and cytologic atypia, fasciitis ossicans is often misinterpreted as malignant sarcoma (2). In some cases, it may be associated with other medical conditions such as sarcoidosis or fibrodysplasia ossificans progressiva (FOP). Clinically it presents most often as a growing asymptomatic mass. Sometimes tender, pain or bleeding appear. Excision is considered as curative; recurrence may happen after incomplete excision.

Presentation at extraordinary sites is described in case reports. However, as far as the author’s knowledge of the literature is concerned, fasciitis ossicans of the penis has never been described before.

We present the first known case of a 73-year-old man with a presentation of fasciitis ossicans of the penis.

Case Presentation

A 73-year-old man presented at the outgoing patient’s office with painful swelling of the penis for two weeks, paraphimosis and a tumor of 2x2cm arising from the glans penis narrowing the meatus urethrae externus. He took notice of the tumor of this size on this day, suffering from pain due to acute paraphimosis. Swelling of the whole penis had started already a couple of weeks before presentation at our outgoing patient’s office with onset the day after a rigid cystoscopy at a practice for urology had been
performed. At this rigid cystoscopy the patient had experienced sudden severe pain and had suffered gross haematuria afterwards. In the following days, the patient recognized an increasing swelling of the whole penis, whereas haematuria stopped the day after the traumatic cystoscopy. The urologist had only described a stenotic meatus urethrae externus without further findings. The patient had mentioned troubles with micturition for several weeks without pain or hematuria. He had sought urological consultation because of these complaints at his urological practice. About half a year prior this consultation, routine check-up was performed without any pathologies. His past medical history showed lower urinary tract symptoms due to mild benign prostate hyperplasia (BPH) and “non insuline dependent diabetes mellitus”. He did not require treatment for BPH.

At the time of first presentation at our outgoing patient's office, the paraphimosis was retracted immediately. With a suspected diagnosis of a urethral carcinoma, we performed a biopsy of the tumor. The first histopathological analysis did not reveal clear results. P-16 analysis was negative, AE 1/AE 3 stains were not detectable. Signs of ossification were already described. With the provisionally diagnosis of “papillary urethritis with metaplastic ossification with high grade squamous dysplasia – according to carcinoma in situ”, the sample was forwarded to second opinion. This analysis did not show evident results neither due to high amount of necrotic tissue. We performed MRI imaging, showing an expansive process in the distal part of the corpus spongiosum with 3.8x1.9 cm in diameter and low apparent diffusion coefficient (ADC) signal alteration with high contrast media enhancement. No signs of lymph node involvement were present. Within an informed consent with the patient, we decided to perform a partial penis amputation.

In the histological processing of the specimen, further staining showed desmin negative, smooth muscle actin positive, antigen expression of CD10 slightly positive and CD34 marking vessels. The tumor derived from the glans penis reaching the urethra but without breaching the mucosa. Mitosis rate was rare and only in the basal cell layer without p-63 hyperexpression and Ki-67 proliferation index was low in 10%. Centrally, the tumor showed clear signs of ossification with abundant amounts of osteoid, osteoblasts and osteoclasts (see Fig. 1, 2) There were no signs for squamous cell carcinoma.

The patient could leave hospital on the second day after surgery. In the follow-up after three weeks, the patient showed unobtrusive postsurgical circumstances. He did not mention any troubles with micturition. In the follow-up after one year, patient did not mention any complaints. No signs of recurrence could be seen.

**Discussion**

In this case report, we present the case of an extraordinary location of fasciitis ossicans, which has never been described before. In this case, tumor derived from the glans penis. The description of this case should assist in diagnostic evaluation of patients with unclear tumor formations of the urethra/penis.

Initially, hyperplasia of soft tissue does not show ossification. The development of ossification is a deferred but rapid process with 2–6 weeks until clinical presentation from onset. Simultaneously,
maturation of bone tissue takes place. The tumor reaches a characteristic macroscopic size of approximately 3 cm on average, but can get bigger too in rare cases. In most cases it stays asymptomatic, but may cause tender, pain, bleeding or neuropathical sensations. Radiological examination shows soft tissue tumor with circumscribed growth without infiltrating the underlying tissue. Imaging however is nonspecific and may be difficult to differentiate from sarcoma. Spreading or progression towards malignancies have not been described so far. Since resection is a curative treatment and recurrence seem only to appear in cases of incomplete resection, surgical approach should be the aim in every case, even if resulting in functional impairment.

Fasciitis ossificans is a benign process of abnormal, extraskeletal ossification in inflamed fascial tissue. It can be reactive to trauma or surgery or, like in this case, secondary due to chronic inflammation (3). Fasciitis ossificans is composed of fibroblastic connective tissue, cartilage, bone and osteoid (4), in contrast to myositis ossificans, where no growing ossification is present. The origin of connective tissue is the main distinguishing feature. However, due to the ossification, misinterpretation as osteosarcoma can lead to inappropriate treatment. Ubiquitin-specific protease 6 (USP6)-rearrangements could be identified as a consistent marker in nodular fasciitis, confirmed by fluorescence in-situ hybridization (FISH) and might offer diagnostic adjunct in differentiation of fasciitis ossificans too (5).

Conclusion

We report for the first-time case of fasciitis ossificans of the glans penis in a male person, presenting with tumor formation of the glans mimicking penile cancer. Proof of bone formation in the specimen revealed the diagnosis. Due to the advanced stage of the disease and massive lower urinary tract symptoms, organ sparing surgery did not seem an accurate approach. Even if fasciitis ossificans of the penis is very rare, its diagnosis should be taken into account in case of unclear tumor formation.

Declarations

Ethics approval and consent to participate:

All procedures performed in this study were in accordance with the ethical standards of the institutional research committee (ethic committee St. John of God Hospital) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. In order to increase accuracy and transparency of this case report, “CARE” guidelines and “CARE” checklist were taken into account. Written informed consent for participate was obtained at the beginning of the writing procedure.

Consent of publication:

Written informed consent was obtained from the patient for publication of the case report at the beginning of the writing procedure.

Availability of data and materials:
Patient reporting data and material are stored at the clinic's patient documentation file. The dataset analysed during the current study are available from the corresponding author on reasonable request

**Competing interests:**

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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**Author’s contributions:**

SL: Project and Project/Protocol development, Data collection, manuscript writing

OK: Data presentation, images

AS: scientific support, manuscript writing

EC: Project development, manuscript writing

**References**


**Figures**
Figure 1

Top left: mineralized bone. Down right: osteoid

Figure 2
Mineralized bone beside osteoid