An unusual complication of pubic exostosis

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SUMMARY
We report the case of a left superior pubic ramus osteochondroma occurring in a 29-year-old man. The patient consulted for a multiple exostoses disease revealed by a painful induration at the base of his penis. This induration was increasing in volume and causing discomfort during sexual intercourse and when sitting for a prolonged period of time. CT-scan examination showed a pedunculated osteochondroma of the left superior pubic ramus compressing the urethra. Surgical resection of the tumor confirmed the diagnosis of osteochondroma. At a 2-year follow-up delay, all symptoms had regressed with no evidence of recurrence.

Level IV: Case report.

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Introduction
Exostoses, or osteochondromas, are bony excrescences that have developed on the bone surface, covered with a cartilaginous cap [1,2]. The two clinical forms encountered are solitary exostoses and multiple exostoses disease [2–4]. These benign tumors can be asymptomatic or manifest when a complication arises. Urogenital manifestations have been described exceptionally [5–8]. We report here a case of pubic exostosis causing urethral compression and discomfort during sexual intercourse in a man with multiple exostoses disease.

Observation
The case involved a 29-year-old male taxi driver, known to be a multiple exostoses disease carrier, who consulted for painful induration located left of the base of the penis. This swelling had recently increased in volume and had become the cause of discomfort during sexual intercourse and when sitting for a prolonged period of time.

The clinical examination revealed the presence of bony growth located on the left side of the base of the penis, painful on palpation, with no other anomaly.

The plain X-ray (Fig. 1) showed a cauliflower-like tumor measuring 6 cm along the longest axis that had developed from the left superior pubic ramus. Computed tomography (CT) demonstrated a pedunculated bony mass extending from the anterior surface of the left superior pubic ramus, exerting slight compression on the urethra. This mass was covered with a fine, regular cartilaginous cap with no calcifications in the area (Fig. 2).

The patient was operated on in the dorsal recumbent position, pulling back the bursae on the right side. The approach was transversal, centered directly on the tumor. An incision was made in the fascia of the external oblique muscle and the abdominal rectus sheath, taking care to recline the inguinal cord. To expose the inferior part of
the left superior pubic ramus, the fascia of the obturator internus was incised at its contact with the bone, along the muscle’s superomedial and inferior insertions. The muscle was then rasped at the same time as the obturator membrane at the internal part of the obturator canal, making sure not to damage the obturator neurovascular bundle penetrating the obturator canal. Once the tumor had been clearly exposed with its insertion pedicle on the superior pubic ramus, it was extracted by en bloc resection.

The pathological study of the specimen confirmed the benign status of the tumor.

At the 2-year follow-up, the patient had no symptoms and no tumor recurrence was noted (Fig. 3).

Discussion

Osteochondromas account for 40% of benign tumors and 10% of all primary bone tumors [1]. These tumors are found preferentially on the metaphyseal aspect of the fertile growth cartilages, but also on flat bones and the bones of the axial skeleton. Their frequency in the pelvis is estimated at 7% [2,3]. Osteochondromas are most often asymptomatic, but complications can arise, in particular if the tumor is voluminous or if it is located in an at-risk anatomic site.

Three types of complications occur [4]:

- extrinsic, secondary to compression or irritation of an anatomical structure neighboring the exostosis;
- intrinsic, related to a fracture of the base of the pedicle or a malignant transformation;
- mixed, related to bone deformations and interference with joint clearance, most often encountered in multiple exostoses disease.

Of the extrinsic complications, the structures compressed by the osteochondroma are frequently musculoskeletal [9], vascular, or neurological [10]. The urogenital structures are involved only exceptionally in these compressive manifestations. A literature review reports only seven cases in four publications [5–8] of urogenital manifestations secondary to an osteochondroma (Table 1). The case reported herein is the first to describe the association of urethral compression and dyspareunia secondary to exostosis.

Excision surgery is not systematic when an osteochondroma is discovered, but the indication is becoming increasingly necessary in the complex forms and in trunk and abdominal locations, where the risk for degeneration is major [2].
Table 1  Urogenital manifestations secondary to pubic osteochondroma.

<table>
<thead>
<tr>
<th>Complications</th>
<th>Number of cases</th>
<th>Authors</th>
</tr>
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<tbody>
<tr>
<td>Microscopic hematuria</td>
<td>4</td>
<td>Philips et al. [5]</td>
</tr>
<tr>
<td>Isolated dyspareunia</td>
<td>2</td>
<td>Smith and Khan [6]</td>
</tr>
<tr>
<td>Urethral compression</td>
<td>1</td>
<td>Carpintero et al. [8]</td>
</tr>
<tr>
<td>Urinary problems</td>
<td></td>
<td>Carpintero et al. [8]</td>
</tr>
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</table>

The resection of osteochondromas located in the pubic area presents certain characteristics that warrant discussion:

- the most appropriate patient installation is the dorsal recumbent position, with the lower limbs abducted on supports, which better exposes the tumor and facilitates the direct cutaneous approach;
- a transversal approach centered on the osteochondroma, which, after incision of the fasciae and elevation of the bone’s soft tissues, exposes the inferior section of the superior pubic ramus. However, one must take care not to damage the inguinal cord and the urethra and the obturator neurovascular bundle;
- the resection should entirely remove the exostosis up to the implantation base, including its entire cartilaginous cap, to prevent any tumor recurrence, a preventive measure to follow for surgery on any exostosis location.

Conclusion

Osteochondromas are frequent benign bone tumors that are usually asymptomatic. Pelvic, and particularly pubic, locations are very rare and often complex. Contrary to the usual forms, they require excision surgery to remove compression or to prevent sarcomatous degeneration.

Conflicts of interests

None.

References