Extraskeletal Osteosarcoma Related to Inguinal Hernia: A Rare Presentation☆

Osteosarcoma extraesquelético y hernia inguinal, una rara asociación

The incidence of neoplasms located in inguinal hernia sacs is very low and ranges between 0.07% and 0.5%.[1] Related publications usually refer to single cases. The first reported case of a tumor within a hernia sac was published by Arnaud de Ronsil[2] in 1749. Later, in 1889, Lejars[3] classified the finding of these tumors into 3 types, according to the anatomical relationship between the tumor and the hernia sac, naming them: (1) *saccular*, when the peritoneum of the hernia sac is compromised by primary or secondary malignant lesions (such as primary mesotheliomas or peritoneal metastases of prostate, colon and ovarian cancers); (2) *intrasaccular*, in cases of primary tumors of organs incarcerated in the hernia (such as cancers of the bladder, colon, appendix or neoplastic implants that compromise the omentum); and (3) *extrasaccular*, for tumors that protrude through the hernia orifice without affecting the hernia sac.

Extraskeletal osteosarcoma is a rare type of malignant tumor that occurs mainly in adults aged 50–70 years.[4,5] It accounts for less than 4% of all sarcomas and approximately 1%–2% of soft tissue sarcomas.[4,5] Their response to treatment and prognosis are worse than in primary skeletal osteosarcomas, with 5-year survival rates ranging from 10% to 46%, with a 50% recurrence rate.[4]

Extraskeletal osteosarcomas appear more frequently in the lower extremities (46.6%). Other frequent locations are the upper extremities (20.5%) or the retroperitoneum (17%).[5]

After conducting a bibliographic review, we have observed that there are currently 3 cases of extraskeletal osteosarcoma of the inguinal region published in the literature. Therefore, we consider it relevant to report this case of an unusual finding observed in our service of a rare association between the presence of an uncommon tumor like extraskeletal osteosarcoma and an indirect inguinal hernia (type L3P, according to the classification of the European Hernia Society[6]) that had been progressing for years. The inguinal mass had been the only clinical symptom of the patient.

A 74-year-old male patient, with no personal history of interest or previous surgical procedures, presented with a left inguinal hernia that had been growing for 5 years (although the growth had been more significant in recent months) and chronic incarceration. Surgery was proposed.

During the operation and dissection/isolation of the hernia sac, a polylobulated nodulation was observed in the hernia sac (which was a saccular neoplasm according to the Lejars classification), with bone-like areas that encompassed the testicular vessels, forming a block that made it impossible to preserved the testicle (Fig. 1). After informing the family members present, we conducted *en bloc* excision of the testicle with the entire hernia sac from the internal inguinal ring, which presented fat content but no visceral content (Fig. 2). Subsequently, the inguinal hernia (L3P) was repaired by inguinal hernioplasty following the modified Lichtenstein technique with placement of self-gripping lightweight polypropylene mesh.

The pathology results confirmed the presence of a well-defined nodular mass measuring 14 cm, which, when sliced, showed firm bone-like tissue with large areas of hemorrhage, which corresponded histologically with a high-grade extraskeletal osteosarcoma, predominantly osteoblastic, with necrotic foci and no involvement of the testis, invasion of the capsule or adjacent adipose tissue.

Once the pathological anatomy results were confirmed, an extension study was done with thoracoabdominal CT scan, which revealed several suspicious abdominal lymphadenopathies and images of possible pulmonary metastases. Thus, it was decided to administer adjuvant chemotherapy in the Oncology Unit.

Following the occurrence of this rare case, we conclude that in order to consider an extraskeletal osteosarcoma, the lesion must: appear in soft tissues, have no relationship with bone or periosteum, have a sarcomatous pattern and produce osteoid material or cartilaginous matrix.[4,5] Our patient presented all these diagnostic elements.

Despite its low incidence, extraskeletal osteosarcoma of the inguinal region is an entity that should be included in the differential diagnosis of inguinoscrotal masses. This is especially true in patients presenting a long-standing hernia with sudden incarceration and no signs of intestinal obstruction or local pain, since its main (and usually only) clinical manifestation is the presence of inguinoscrotal mass that increases in size.[7]

Likewise, any palpable suspicious mass in the inguinal region should be studied by ultrasound, CT or MRI[8] in order to define the preoperative diagnosis and the extension of the mass for the most appropriate therapeutic procedure in each case.

When faced with the intraoperative discovery of a mass suggestive of malignancy in the inguinal region, *en bloc* surgical resection should be performed with wide margins. In addition, it is always necessary to study by histology any hernia sacs that seem macroscopically suspicious.[8,9]

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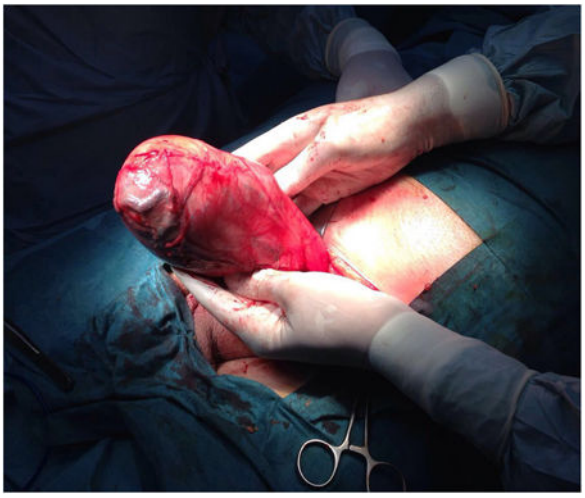

Fig. 1 – Large inguinal mass encompassing the hernia sac and testicle.

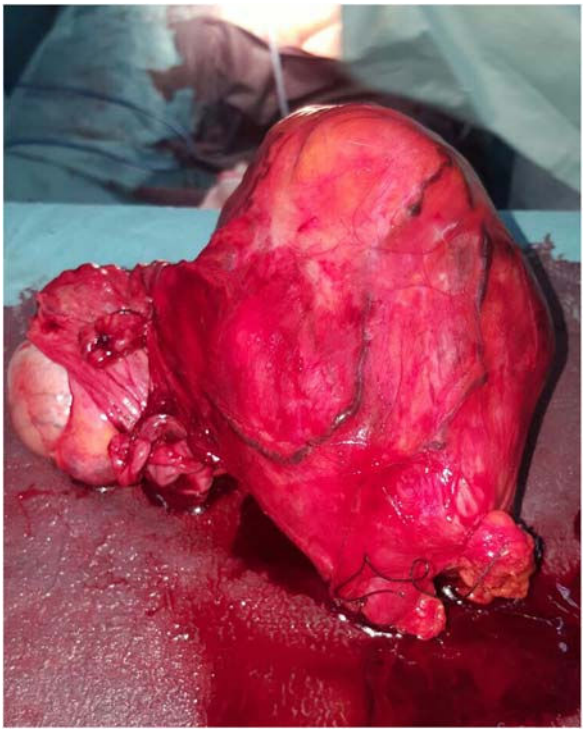

Fig. 2 – Surgical specimen: hernia sac mass and left testicle.

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