Schwannomas are the most common tumors of the peripheral nerves. Intraosseous schwannoma is a rare entity; approximately 200 cases are described in the world literature, with only 1 involving the ileopubic ramus. To the authors’ knowledge, this article is the first to describe a giant intraosseous schwannoma of the ileopubic ramus, its clinical presentation and radiographic aspect, its histological findings, its biological behavior, and the differential diagnosis. It is also the first to describe surgical treatment of this entity.

A 63-year-old woman presented with paroxysmal pain at the right groin. Radiographs showed a multilocular lytic lesion of the right ileopubic ramus, extending from the acetabulum to the pubic symphysis. Magnetic resonance imaging revealed extraosseous extension in the pelvis and in the anteromedial region of the right thigh. An incisional biopsy revealed fibroconnective tissue with sparse spindle cells in a myxoid stroma; diffuse expression of S100 protein was observed by immunohistochemistry. Intraosseous neurofibroma was the first histopathologic diagnosis. The 12×7.5×4 cm lesion was entirely removed via a Letournel ileoinguinal approach. Reconstruction of the ileopubic ramus was performed with an autologous bone graft taken from the omolateral iliac crest and fixed by a pelvic reconstruction plate. Histopathological examination revealed Antoni A and B patterns with Verocay bodies, no mitosis or cellular atypia, and diffuse expression of S100 protein by immunohistochemistry, all features indicative of a benign schwannoma. The patient returned to activities of daily living with no limitations. No recurrence had occurred 24 months postoperatively.
Schwannomas are benign neoplasms arising from the myelinating Schwann cells of the nerves; they are the most common tumors of the peripheral nerves.\textsuperscript{1-4} Intraosseous schwannoma is a rare entity. Approximately 200 cases are described in the world literature, mostly involving the mandible.\textsuperscript{5-8} The pelvic ring is rarely afflicted. The sacrum is mostly involving the mandible.

The current article describes a giant intraosseous schwannoma of the ileopubic ramus, its clinical presentation and radiographic aspect, its histological findings, its biological behavior, and the differential diagnosis. To the authors’ knowledge, only 1 case of localization in the ileopubic ramus is present in the world literature.\textsuperscript{9}

The case report describes a giant intraosseous schwannoma of the ileopubic ramus, its clinical presentation and radiographic aspect, its histological findings, its biological behavior, and the differential diagnosis. To the authors’ knowledge, this is the first article to describe surgical treatment of this entity.

**Case Report**

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

A 63-year-old woman presented with a several-year history of paroxysmal pain of the right thigh. Twelve years previously, radiographs had documented a 6-cm radiolucent area localized in the right ileopubic ramus. At that time, the patient had little pain and refused further diagnostic examinations, included surgical biopsy of the lesion. Radiographic evaluations, performed almost every year, showed slow growth.

When the patient was 61 years old, she was diagnosed with colon adenocarcinoma with liver secondarism. She underwent partial resection of the colon, thermoablation of the liver lesions, and chemotherapy with oxaliplatin and 5-fluorouracil; at the time of presentation to the authors, she was disease free. In the same year, the patient sustained a traumatic fracture of the inferior wall of the ileopubic lesion, which was treated conservatively with partial weight bearing for 2 months. However, the pain became more intense, and the mass in the right groin was uncomfortable, so the patient sought medical treatment.

Physical examination showed a 7×4-cm firm, round, hard mass in the right groin; the overlying skin was intact, and no fever, warmth, or swelling was present. The hip had a full range of motion, and the patient walked without limping. No evidence existed of lymphadenopathy or neurovascular involvement. Pain was present in the right groin and thigh and was exacerbated by walking and by internal rotation of the hip.

Radiographic investigation of the pelvis showed a 7×5-cm wide lytic lesion with a well-defined margin extending from the acetabulum to the pubic symphysis. It was multilocular, with no inner calcifications; the upper cortical was disrupted without periosteal reaction.

Magnetic resonance imaging with contrast enhancement revealed cortical disruption and extraosseous extension of the lesion in the anteromedial region of the thigh and in the obturator region; inside the pelvis, the bladder was dislocated, without infiltration of the soft tissue structures. The previous fracture had not healed. The tumor measured 12×7.5×4 cm; it appeared hypointense on T1-weighted images and hyperintense on T2-weighted images and had heterogeneous enhancement after gadolinium infusion (Figure 1). Positron emission tomography showed a heterogeneous uptake in the right iliac fossa. Laboratory data were within the normal ranges.

After preoperative planning, an incisional biopsy was performed. Histologic examination revealed fibroconnective tissue composed of sparse spindle-shaped cells with wavy nuclei, arranged side by side in a loose myxoid stroma. No mitosis, hemorrhage, or necrotic areas were seen. Immunostaining for the S100 antigen was diffusely positive. The first histopathologic diagnosis was indicative of intraosseous neurofibroma. The decision was made to remove the lesion and reconstruct the ileopubic ramus with autologous bone graft from the iliac wing.

Through a Letournel ileoinguinal approach, the right ileopubic ramus was exposed. First, the soft tissue component of the tumor, located in Scarpa’s triangle up to the adductor canal, was entirely removed, then the bone lesion was debrided (Figure 2). The posterior cortex of the ileopubic ramus was left in situ to maintain the length of the defect, but it was too thin by itself to be a wall preventing bladder dislocation or discomfort during any Valsalva maneuver.

![Figure 1: Preoperative anterior-posterior radiograph of the pelvis showing a lytic, trabeculated lesion of the right ileopubic ramus extending from the acetabulum to the pubic symphysis (A). Preoperative T1-weighted magnetic resonance image with contrast enhancement showing a hypointense mass measuring 12×7.5×4 cm and extending into the anteromedial region of the thigh and into the obturator region, dislocating the bladder (B).](image-url)
Therefore, the decision was made to reconstruct the ileopubic ramus.

Reconstruction was performed with a 7-cm autologous bone graft obtained from the ipsilateral iliac crest from the same incision. It was fixed with a 12-hole reconstruction plate molded from the contralateral pubic tubercle to the internal iliac fossa along the iliopectineal line and 6 screws.

Macroscopic appearance of the tumor was a grey-yellow solid mass with vascular congestion and hemorrhagic spots, measuring 12×8×4 cm and weighting 400 g. Detailed histopathological examination confirmed derivation from peripheral nervous tissue. It showed a mixture of hypercellular and hypocellular areas with foci of nuclear palisading. Neither cellular atypia nor mitotic features were evident. Immunohistochemistry showed diffuse expression of S100 protein. Findings were specific for intraosseous schwannoma (Figure 3).

The patient started to walk with crutches 1 month postoperatively, and full weight bearing was allowed 1 month later. No pain or articular limitation of the right hip was reported.

Follow-up radiographs were performed every month. Complete incorporation of the graft occurred 6 months postoperatively (Figure 4). Twenty-four months postoperatively, the patient had no sign of recurrence.

**DISCUSSION**

Schwannomas are relatively common benign neoplasms that arise from the myelinating Schwann cells of the nerves (particularly the eighth cranial nerve), with a predilection for sensory nerves. However, intraosseous schwannoma is rare, accounting for less than 1% of all benign bone tumors. Most cases involve the mandible, maxilla, sacrum, and vertebrae.4 Myelinated nerves may be present intramedullary or within the nutrient canals in associations to blood vessels, so schwannomas may arise within the bone or in the canals. They should be differentiated from extraosseous schwannomas growing from nerves lying on the bony surface, which could erode the bone from outside.

The rarity of this tumor has been attributed to the paucity of sensory nerve fibers in bone.9 No agreement exists about why the mandible is the most common site of involvement; it has been attributed to the long intraosseous path of the alveolar nerve, but nerves with longer courses (like those that innervate long bones within the nutrient foramina) have a rarer involvement.7-11

Symptoms are nonspecific and related to the location and extension of the lesion. Imaging findings are nonspecific, but they suggest a benign lesion.11-15 After clinical and radiological examinations, differential diagnoses include solitary bone cyst, aneurismal bone cyst, giant cell tumor, chondroblastoma, fibrous dysplasia, and intraosseous neurofibroma.

Histological examination is mandatory for a definitive diagnosis. Schwannoma typically shows 2 types of cell arrangement: compact spindle cells (Antoni A pattern) and diffuse cells separated by loose myxoid stroma (Antoni B pattern). Encapsulation is another typical feature of schwannoma. Diffuse immunoreactivity for S100 protein is indicative of Schwann cell origin.9 Its principal histological differential diagnosis is neurofibroma, which arises from the perineural fibroblasts; it does not show an Antoni A pattern and is rarely encapsulated. Moreover, neurofibroma has a greater tendency to recur than does schwannoma.16,17

Malignant transformation for intraosseous schwannoma has not been reported, and recurrence after treatment is due to...
incomplete resection of the lesion. Curettage and bone grafting is the adequate treatment.  

To the current authors’ knowledge, this case is the second report of a giant intraosseous schwannoma of the ileopubic ramus and the first that proposed a treatment. In the current patient, the huge mass of the tumor involved bone and the adjacent soft tissues of thigh and pelvis. It required tumor excision in a difficult intra- and extraperiosteal location and bone reconstruction to allow weight bearing and activities of daily living. Further difficulties were the involvement of crucial areas such as the Scarpa triangle with the main blood vessels for the lower limb and the femoral nerve, as well as the inner pelvis with bladder dislocation.

Considering that the shape of the iliac crest could fit the pubic arch if grafted upside-down, the surgical plan included reconstruction of the excised area with a large bone graft fixed with a reconstruction plate. The graft fused uneventfully, and the patient has no discomfort or bladder dislocation during any Valsalva maneuvers.

**REFERENCES**