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GIANT LUMBOSACRAL SCHWANNOMA, NEUROANESTHETIC CHALLENGES INVOLVED IN SURGICAL RESECTION, MANAGEMENT AND POSTOPERATIVE EVOLUTION: A CASE REPORT

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ABSTRACT

Pelvic retroperitoneal schwannommas are rare entities that are characterized by slow growth, are oligosymptomatic, and diagnosis of the advanced stage is through compressive symptons of nearby structures, which is a challenge for surgical treatment, anesthetic management and pain control. We present the case of a 57-year-old patient with lumbosacral schwannoma, who underwent multimodal opioid-sparing anesthesia, based on intravenous lidocaine infusion, with two operative times totaling 14 hours of surgery, and its evolution and pain management after intervention. The anesthetic planning, individualizing the patient's needs, is the focus for a better outcome.

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INTRODUCTION

The retroperitoneum houses a wide variety of histological tissues and, therefore, presents a wide variety of pathologies, including benign and malignant tumors that, due to their topography, have a high potential for asymptomatic growth and evolution. They are usually pathologies diagnosed accidentally or in advanced stages (Strauss et al., 2011). In the latter case, they can be an anesthetic-surgical challenge due to the involvement of multiple vital structures, the

difficulties involved in the technical approach and their difficult postoperative analgesic management. In the present case, the multispecialized approach at the Hospital Universitário Getúlio Vargas is presented, and includes the surgical description, anesthetic management, postoperative evolution and clinical management of pain of a giant retroperitoneal schwannoma, which, in this location, is responsible for 0.7%-2.7% of all schwannomas and 0.5 %-1.2% of all retroperitoneal tumors (Kishi et al., 2002).

CASE REPORT

Female patient, 57 years old, living in a community approximately 500 km from the capital, without access to tertiary services and restricted to transport either by river or air, evolving for 5 months with protrusion on the right side of the pelvic region, which was associated with local pain radiating towards the lumbar region, and weakness in the lower limbs (worse on the right side, grade IV strength). On physical examination, a palpable (painless) mass was identified, with approximately 7.0 cm on palpation, and fibroelastic consistency in the right lower quadrant. A pelvic MRI scan was requested, which evidenced an expansive retroperitoneal lesion measuring 16.3 cm x 12.2 cm x 13.7 cm (Figure 1). Intravenous contrast allowed us to visualize the mass via a heterogeneous signal and enhancement in the topography of the right side involving the foramen and vertebral body of L5, with restriction of the diameter of the spinal canal at the level of L5-S1 and spinal infiltration at the same level.



Figure 1. Expansive retroperitoneal lesion located in right topography, compromising the vertebral body of L5 (MRI)

The patient was admitted to the Hospital Universitário Getúlio Vargas for surgical treatment, which was initiated by the oncological surgery team. Surgery was in horizontal dorsal decubitus, with a xiphopubic incision with extension of the pubic incision and right-side inguinotomy to the iliac crest, dissection of the muscular planes with isolation of the cavity and use of retractors with retroperitoneal exposure. A voluminous retroperitoneal lesion of right-hand paravertebral origin displacing the psoas muscle and iliac vessels, right gonadal vein, right ureter and vena cava to the anterior wall was identified. Retroperitoneal dissection was performed with isolation of the iliac vessels (common, external and internal), gonadal veins, ureter, inferior vena cava and abdominal aorta (Figure 2). The procedure continued using power forceps, with release of psoas, resection of the muscle lesion compromising the operative margin, dissection of the obturator fossa with isolation of the presacral space, interruption of the anterior approach due to lack of access to the spine, due to the volume of remaining lesion without anatomical margins for resection. Hemostasis of the retroperitoneum was reviewed with suture, hemostatic sponges and bone wax. No cerebrospinal fluid fistula or active bleeding was found, and the closure of the abdominal wall by planes was performed. The patient was delivered to the neurosurgery team after 8 hours and 30 min of surgery.

In the second operation, carried out by the neurosurgical team, repositioning was performed in the prone position and resection of the retroperitoneal tumor was initiated, through a posterior median

longitudinal incision, followed by plane dissection with exposure of the spinous processes, laminae and joints of L3-S1, with difficulty, due to the volume of the tumor mass and infiltration at the medullary level. This was followed by laminectomy in L5-S1, bilateral pediclefixation L4-S1 and unilateral fixation L5, associated with foraminotomy with exeresis of the expansive lesion in the neural foramen of L5 and S1. This took 5 hours and 30 min, which brought the whole surgical procedure to a total of 14 hours. Cytomorphological and immunohistochemical examinations confirmed the diagnosis of schwannoma. The anatomopathological examination suggested the diagnosis of undifferentiated pleomorphic sarcoma and the histological examination identified spindle cells of a storiform pattern, with eosinophilic cytoplasm with little distinction of borders, pleomorphic and multinucleated nuclei with coarse granular chromatin and other atypical cells. Immunohistochemistry exams revealed S-100 tumor cells, though no CD117 (KIT) or EMA (MUC1) cells were identified. Anesthetic induction was performed with dexmedetomidine (0.5 ug/kg in 10 min), ketamine (1 mg/kg), fentanyl (3 ug/kg), magnesium sulfate (30 mg/kg), propofol titrated according to clinical response (total dose of 1 mg/kg) and rocuronium (0.6 mg/kg) and maintenance with sevoflurane, lidocaine (1 mg/kg/h), dexmedetomidine (0.5 ug/kg/h), ketamine (18 ug/kg/min), remifentanil (0.1-0.3 ug/kg/min) plus intermittent boluses of rocuronium (0.1-0.2 mg/kg) as needed. To maintain hemodynamic parameters during the perioperative period, hydration with 8 mL/kg/h of crystalloids was performed, and the use of norepinephrine was necessary with a gradual increase in dose (final total dose 0.15 ug/kg/min), as well as a transfusion of a unit of concentrated red blood cells. Patient was extubated in less than 24 hours after admission to the ICU. Analgesia was promoted with 100 mg of tramadol every 8 hours and rescue with morphine (0.1 mg/kg) every 6 hours (two doses in total) with pain scores via visual analog scale on mobilization and at rest respectively: 2 and 0 at 24 hours, 2 and 0 at 48 hours and 1 and 0 at 72 hours when discharged to the ward. After 2 months of hospital discharge, the patient reported paresthesia and neuropathic pain in the lower right limb, which was diagnosed via a Dn4 questionnaire. Pregabalin was then started with dose progression to control with 225 mg/day after 4 months of use. Thirty sessions of radiotherapy were indicated for the treatment of a lesion measuring 5.7 x 7.0 cm (Figure 3) that one year after the procedure was not intraoperatively resectable.



Figure 2. Dissection and isolation of iliac vessels (common, internal and external), right gonadal vein with ipsilateral ureter, inferior cava and abdominal aorta



Figure 3. Expansive lesion in the body of L5, with extension to the foramen, free lymph nodes without other lesions identified in the control

DISCUSSION

Surgical Discussion: Anatomically. the we understand retroperitoneum as the space bounded anteriorly by the peritoneal cavity, posteriorly by the paraspinal musculature, superiorly by the diaphragm, and inferiorly by the levator ani muscles, forming natural extension with the pelvis (pelvic diaphragm) (Liles et al., 2009). The iliac vessels, inferior vena cava and its tributaries, the aorta and its main branches, duodenum, pancreas, portions of the colon-ascending and descending, kidneys, perirenal fat and adrenal glands are structures that make up the retroperitoneum (Liles et al., 2009). Retroperitoneal lesions, when neoplastic, can comprise up to 80% of cases that are characterized as malignant. The diagnosis of sarcoma is more prevalent in most patients with mass in the retroperitoneum is this primary, extra visceral or unifocal soft tissue, and these lesions usually produce few symptoms until they reach sufficient dimensions to generate extrinsic compression to adjacent structures. Nonetheless, most patients arrive at specialized medical care with a diagnosis of abdominal mass discovered at random (Stoeckle et al., 2001). Due to the clinical, anatomical location of the lesion, epidemiology and complementary examinations, and soft tissue sarcoma with infiltration into the vertebral body of L5, which was presented as the main diagnostic suspicion in this case, it is worth emphasizing the need for differential diagnosis with other benign or malignant tumors such as paraganglioma in the anatomical region of the midline adjacent to the aorta or vena cava.

Schwannomas are primarily benign tumors that arise from the myelin sheath of nerves, composed of Schwann cells (Colecchia et al., 2020; Handa et al., 2019), and are slow-growing, non-aggressive, have a low risk of recurrence (Dau et al., 2022), and commonly affect the thoracic, cervical and lumbar regions, in contrast to the rarely affected sacral region. As they are primarily benign and locally invasive tumors, they are usually detected when the tumor mass grows large enough to cause pressure on adjacent structures and cause nonspecific symptoms (Strauss et al., 2011; Colecchia et al., 2020; Handa et al., 2019). When originating from spinal nerves, they can present neurological symptoms such as neuropathic pain or pain syndromes (Dau et al., 2022). Pelvic and sacral schwannomas are rare and originate from sacral nerves or the hypogastric plexus (Dau et al., 2022). Their diagnosis is usually late due to the space provided by the pelvis, since they only compress neighboring structures after significant growth (Handa et al., 2019).Histologically, they can present two different types of cell organization patterns: Antoni Type A and Antoni type B. Antoni Type A is characterized by an organized pattern with elongated cells and the presence of Verocay bodies, while Antoni Type B has a disordered and hypocellular pattern (Colecchia et al., 2020; Handa et al., 2019). In immunohistochemical evaluation, S-100 protein positivity is a marker for diagnostic confirmation. The tumor is also positive for the markers vimentin and neuron-specific enolase (NSE), and negative for smooth muscle actin (SMA) and CD117 (Colecchia et al., 2020; Dau et al., 2022). Klimo et al. (2003) propose a classification of sacral schwannomas using three categories, according to the location and extent of the tumor in the sacral region. Type 1 tumors are located in the sacrum and do not extend to another region; type 2 tumors extend from the sacrum to the

presacral region or subcutaneous space; and type 3 are located in the pelvis or retroperitoneum.

Anesthetic Discussion

Anesthetic planning is an important strategic tool for the proper development, predictability, organization and possible decisionmaking during the anesthetic act. The reported surgical pathology presented radiological signs of spinal canal invasion in L5-S1, with destruction of the vertebral body in L5 accompanied by loss of muscle strength in the topographic region; the procedure of multispecialty approach with two surgeries in different positions; initially of dorsal decubitus followed by ventral decubitus and estimated long duration. The general anesthetic technique with continuous lidocaine infusion was chosen because it has a better hemodynamic stability when compared to the epidural technique (Weibel et al., 2018; Batko et al., 2020; Terkawi et al., 2016) if one considers the proximity and tumor compression of large vessels, in addition to spinal cord involvement being able to interfere with the distribution of the anesthetic in the neuroaxis. Continuous epidural analgesia with insertion of two catheters proximally and distally to the lesion has been described in spinal surgeries for the treatment of scoliosis with a better outcome in decreasing pain scores when compared to the use of a single catheter and the use of patient-controlled intravenous analgesia (Taenzer and Clark, 2010; Klatt et al., 2013), with the determining mechanism being the interruptions of the epidural and paraspinal spaces caused by surgical manipulation, since the use of a single catheter is insufficient (Taenzer and Clark, 2010; Klatt et al., 2013). It is worth mentioning the medical and legal implications that may arise from the use of an analgesic technique with neuraxial catheter in cases in which a postoperative neurological injury occurs (Duarte and Filho, 2021). The analgesic strategy in this case was based on continuous intravenous lidocaine infusion, mainly taking into account the first abdominal surgery, since it is known that pain of inflammatory origin is mediated by interaction with polymorphonuclear cells and by G protein coupled receptors via the muscarinic M1 receptor (Weibel et al., 2018; Hollmann et al., 2000; Hollmann and Durieux, 2000). These pathways are responsible for the intestinal effects mainly in the prevention of postoperative intestinal complications and in the development of paralytic ileus (Herroeder et al., 2007), and analgesia is obtained that is comparable to the epidural technique and with reduced postoperative opioid consumption (Koppert et al., 2004).

Due to the neurosurgical approach, the neurological condition already clinically established, the topography of the lesion and the possibility of further development of neuropathic pain, the analgesic strategy with intravenous lidocaine other classical routes of action of lidocaine would also play a role in the proposed intervention. Inhibition of the ascending stimulus transmission pathways of injured peripheral fibers and the respective dorsal root ganglia that are interconnected to them (Weibel et al., 2018; Devor et al., 1992), as well as polysynaptic reflexes of the posterior horn of the spinal cord (Woolf & Wiesenfeld-Hallin, 1985; Weibel et al., 2018), seem to be the main mechanisms proposed for its more classic analgesic effects. Associating these pathways with the effect of ketamine and magnesium sulfate on the inhibition of NMDA receptors, which prevent and inhibit the installation of the wind-up mechanism (Mion and Villevieille, 2013), the main substrates for improving pain in the postoperative outcome are obtained. Among the numerous benefits of using perioperative lidocaine in a continuous infusion, we highlight the decrease in postoperative pain scores and opioid consumption that is comparable to that of the epidural technique (Weibel et al., 2018; Dunn & Durieux., 2017; Taenzer and Clark, 2010), decreased postoperative nausea and vomiting (Weibel et al., 2018; Dunn and Durieux, 2017; Terkawi et al., 2016), decreased duration and incidence of paralytic ileus (Weibel et al., 2018; Terkawi et al., 2016), decreased length of hospitalization (Weibel et al., 2018; Herroeder et al., 2007; Batko et al., 2020; Terkawi et al., 2016) and improvement in postoperative quality of life (Batko et al., 2020, Farag et al., 2013; Terkawi et al., 2016). All these positive outcomes have been discussed in multiple studies; however, definitive clinical conducts have been difficult to define in meta-analyses due to the small size of the studies and the great heterogeneity between them (Weibel et al., 2018; Dunn and Durieux, 2017).

CONCLUSION

Despite diagnostic and therapeutic advances, oncology continues to present challenging and multifaceted cases. These challenges may be the result of institutional limitations, regional characteristics or natural history of the disease investigated, however, in the context of the patient, the individualized multimodal approach seems to be the best option, and the prolonged infusion of intravenous lidocaine is an alternative when other analgesic strategies may not be possible. Retroperitoneal schwannomas as rare entities, though they are just one example of a daily routine with advanced conditions of difficult management that health professionals face, in which, good rapport between specialized teams is adetermining factor for favorable outcomes in these presentations. Intraoperative multimodal analgesia, considering the anatomical and physiological limitations of each case, without underestimating the pain potential of the proposed surgical and neoadjuvant procedures, plays a fundamental role in this context.

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