REMOVAL OF GIANT PRESACRAL NEUROGENIC TUMORS WITH APPLICATION OF THE CUSTOMIZED 3D-PRINTED MODELS: CASE SERIES ANALYSIS AND LITERATURE REVIEW

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The characteristics and principles of surgical treatment of giant presacral neurogenic tumors are analyzed on the example of authors' own series of cases and based on the literature review. Three clinical cases of surgical treatment of patients with giant presacral neuromas are presented. Besides routine preoperative clinical examination, 3D-printed models of pelvic bones, tumors and blood vessels were made based on CT-angiography data. In two cases, giant L5 root neuromas with presacral extension were removed through retroperitoneal approach, and in one case, two-stage removal of the giant presacral neurofibroma through the median laparotomic and dorsal approaches was performed. Giant presacral neurogenic tumors are the rare pathology in the practice of spinal surgeons. Surgical treatment should be carried out by a multidisciplinary surgical team. It should be borne in mind that life-threatening complications are common in the early postoperative period. Application of customized 3D-printed models is an additional useful tool of perioperative planning.

Key Words: schwannoma, neurofibroma, retroperitoneal tumor, presacral tumor, spine, 3D model

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According to various data [1–4], giant presacral neurogenic tumors are rare. They account for 0.3–5.0 % of nerve sheath tumors of all locations: 7 % are spinal tumors, 0.4–15.0 % – retroperitoneal tumors, and last ones may cause 1 out of 40,000 hospital admissions. The giant presacral neurogenic tumors are usually considered to be nerve sheath lesions with a maximum dimension of more than 5 cm or covering 2 or more spinal segments and spreading to the presacral area more than 2.5 cm [5].

The academic papers are mainly report about selected cases. A meta-analysis of surgical treatment of benign presacral schwannomas by Pennington et al. [6] showed 123 published cases, which fulfill the above-mentioned criteria. Surgeons of various areas (neurosurgeons, abdominal surgeons, urologists, etc.) or multidisciplinary teams perform the procedures for these lesions. Different surgical approaches are described, including posterior, anterior and combined approaches as well as the use of endoscopy and robot-assisted surgery [7–9].

Considering the incidence of pathology, it seems topical to publish three additional cases. In the first case, when removing similar lesions, individual 3D models of the tumor with adjacent bone structures and blood vessels were used for preoperative planning.

The objective of the study is to show the clinical features and principles of surgical treatment of giant neurogenic tumors in the presacral space by our own series of cases and literature review.

Material and Methods

Three clinical cases of surgical treatment of patients with giant presacral neurogenic tumors are presented. Besides routine preoperative clinical examination, 3D-printed models of pelvic bones, tumors and blood vessels were made based on CT-angiography data (Fig. 1–3). The data segmentation was conducted in the MIMICS Materialise software (Belgium). The primary model was processed in a 3D graphics editor Blender 2.80. As for individual models, they were printed using the fused deposition modeling (FDM) via Z-Bolt X 4-extruder printer with the Tool Change technology for changing printheads. Each extruder had its own printing program code set in the Cura 4.6.0 slicer. A filament with a diameter of 1.75 mm was utilized as the material. The hard plastic (PLA) – for printing bone structures and tumors, and the flexible material (Flex) was for vessels.

The models were applied both in preoperative planning and for identifying surgical techniques during the operation. The multidisciplinary surgical team conducted all the procedures.

Results

Clinical case 1. A 52-year-old female patient G. was admitted to the hospital. She complained of an intermittent lower back pain with irradiation and weakness in her left leg. The symptoms bothered the patient for several years. Five months before the procedure, a space-occupying lesion in the presacral space was diagnosed by pelvic ultrasound per-

formed for a renal colic. MRI data: cystic and solid tumor up to 7 cm in diameter at the level of L5–S2 to the left of the spinal column, with the extension into the left intervertebral foramen at the level of L5–S1 (Fig. 4). During the neurological assessment, a hypesthesia in the dermatome of L5 was found in the left as well as the paresis of the left foot dorsiflexion – 4 points. Considering the lateralized attitude of the lesion at the level of the lumbosacral junction, it was decided to perform a left-sided retroperitoneal approach using Rob's technique. Following the surgical release of the iliac vessels and *m. iliopsoas* a tumor sac was lanced, the cyst was evacuated with the contents about 5 ml of a transparent liquid. An intracapsular removal was followed by intense arterial and venous bleeding from the tumor vessels. In this case it is nessessary to perform clamping of the iliac arteries at various surgery stages. The hemostasis was performed by mono– and bipolar coagulation, tamponade with a hemostatic sponge, and retroclusion. The total blood loss was about 3500 ml. It was partially compensated by reinfusion of blood via cell saver and donor-specific transfusion. The tumor had of a soft consistency. Its most part was removed by an ultrasound disintegrator.

On the 3rd day after the surgery, the patient developed acute edema of the left leg. Dopplerography data: deep vein thrombosis of the left lower limb and pelvic veins without signs of flotation.

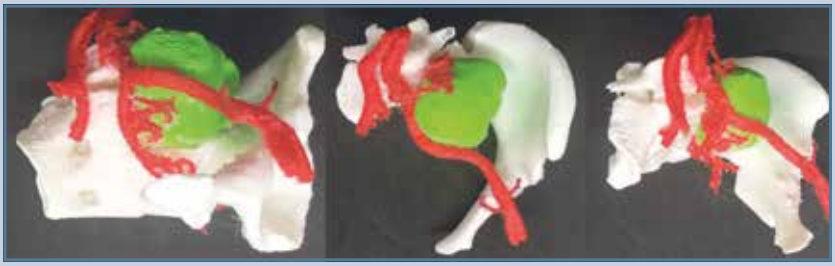

Fig. 1
3D model of a giant neurinoma of L5 root in the left with the expansion into the presacral space (case 1): L5–S2 vertebrae, left iliac bone, iliac arteries and feeding tumor vessels

The patient was prescribed anticoagulant therapy and wearing compression knitwear. On the 14th day after the surgery, the patient was discharged from the hospital. The paresis of the left foot (dorsiflexion) progressed to 2–3 points neurologically. Histological findings: plexiform neurofibroma (Fig. 5).

Clinical case 2. A 62-year-old female patient S. was admitted to the hospital. She complained of pain in the lower back with radiation on the posterior surface of the legs, more on the right, as well as weakness in the right leg. The symptoms bothered the patient for about six months. MRI data: a space-occupying lesion of the right L5 root, extending from the intervertebral foramen into the retroperitoneal space to the level of S2 (Fig. 6). CT data: lytic lesions in L5 and S1 bodies. The neurologic impairment is represented by a lower monoparesis in the L5 nerve distribution on the right up to four points.

A surgical approach was performed by Rob's technique (retroperitoneal, on the right, as in the previous case). A tumor partially covered by *m. iliopsoas* was visualized in the bifurcation area of the aorta and distally under the right common iliac artery and vein and in the bifurcation area of the iliac vessels. The tumor was a conglomeration of several nodes of a dense consistency, glomerated with the anterolateral surface of the L5 and S1 vertebrae and not susceptible to destruction by an ultra-sonic disintegrator. The removal was performed due to fragmentation by an electric knife. For surgical release of the blood vessels and eradication of tumor, *v. iliaca* interna was ligated. Also,

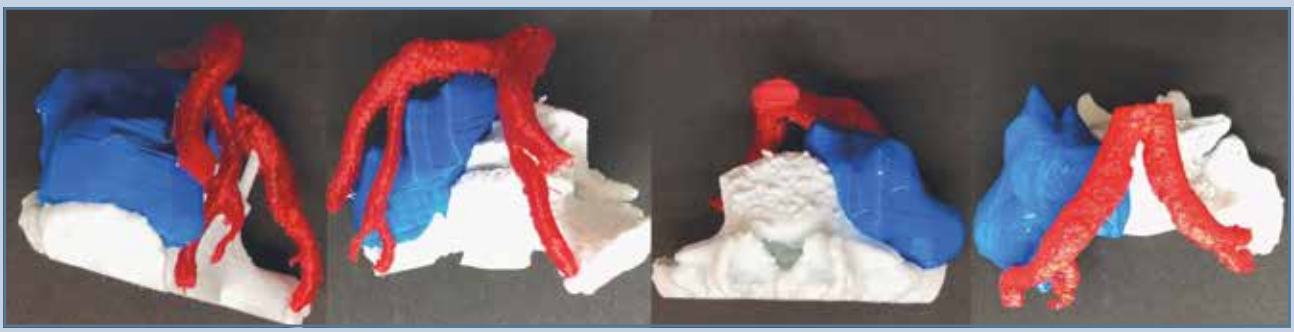

Fig. 2
3D model of a schwannoma of the right L5 root (case 2): a tumor, L5–S1 vertebrae, iliac arteries

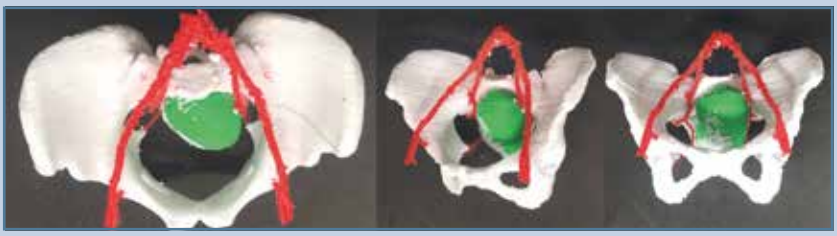

Fig. 3
3D model of a giant neurofibroma at the level of S1–S5 with pelvic bones and iliac arteries (case 3)

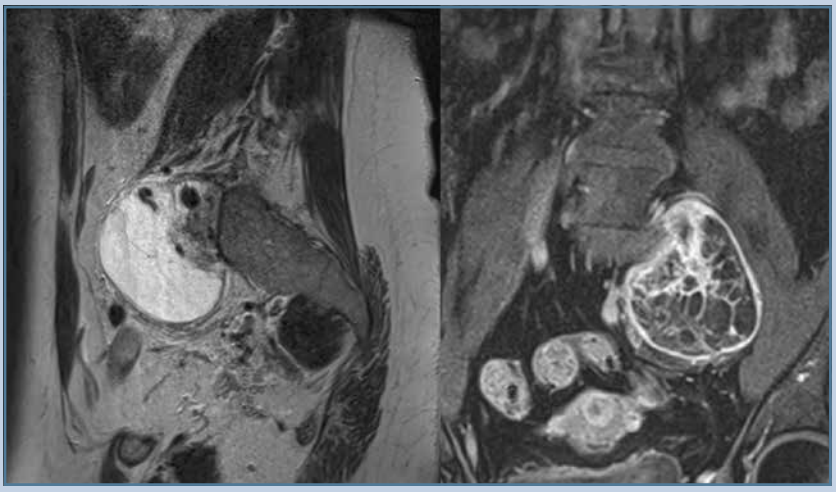

Fig. 4
MRI scan of a 52-year-old female patient G. before the surgery

the intersection of the *v. iliaca* interna was performed at the confluence with the *v. iliaca communis*. In the process of tumor isolation, an exiting root was found. A motor response was obtained from the muscles of the lower leg during stimulation. Considering the impossibility of isolating the fibers, the root was intersected. The blood loss was 400 ml. On the 1st day, the patient was verticalized. No significant increase in neurological impairment was observed. The postoperative period was unremarkable. The patient was discharged from the hospital on the 7th day. MRI data: total resection. Histological findings: schwannoma.

Clinical case 3. A 30-year-old patient S. was admitted to the hospital. She complained of an intermittent pain on the posterior surface of the left leg from the sacrum to the foot, muscle weakness in the legs, numbness of the left toes, the outer surface of the upper third of the thigh and buttocks on the left. A paresis of the left leg muscles in 4 points and the hypesthesia in the specified area were neurologically determined. Eleven years ago, according to MRI data, a space-occupying lesion of the true pelvis was diagnosed (Fig. 7). After 4 years, an abdominal delivery was performed. During this procedure, the tumor was supposed to be removed. However, it was not performed due to technical difficulties. After a while, the patient began to notice an amelioration in the pain intensity in the leg with the progression of numbness and weakness. According to the control MRI data, there was an increase in the tumor size with an expansion into the true pelvis and into the spinal canal at the level of S2–S4. Considering the significant intracanal tumor volume and a large node in the presacral space, it was decided to perform a two-stage removal through the anterior and posterior approaches. The anterior approach was performed by a midline laparotomy in the patient's supine position with her legs wide apart. The surgical release of the loops of the small intestine into the retroperitoneal space was performed. Both ureters, ovaries, fallopian tubes were visualized. A surgical release of the rectum along the mesorectal layer was performed. The en-block resection was impossible due to the tight fixation of the tumor to the sacrum. As in case 2, the tumor was of a dense consistency. An ultrasonic disintegrator could not destroy it. Finally, it was removed by fragmentation with an electric knife. In the removal of the left flank (due to intense venous bleeding from the tumor vein) – *v. iliaca interna* flow; attempts at hemostasis by electrocoagulation were unsuccessful; a defect of *v. iliaca interna* was visualized and it was sutured with a 3/0 prolene; the tumor vessel was stitched, coagulated, and transected. The tumor was separated from the sacrum with a mono-knife. The S2–S3 roots in the left were transected under neurophysiological monitoring. During the surgical release of the lower-right segment, there was an intense venous bleeding from the tumor vessel. We performed tamponade by surgicell collagen sponge. Drainage was inserted into the presacral space. The blood loss was 2000 ml.

On the 2nd day after the surgery, about 1500 ml of fresh blood was simultaneously effused through the drainage during the patient's verticalization. Therefore, an emergency revision relaparotomy was performed that showed blood clots aling the drainage. The surgical exploration of the abdominal cavity showed the absence of any bleeding points. The stitches were removed from the retroperitoneal space. In the small pelvis, about 50 ml of blood was found in clots. A surgical exploration of the area of

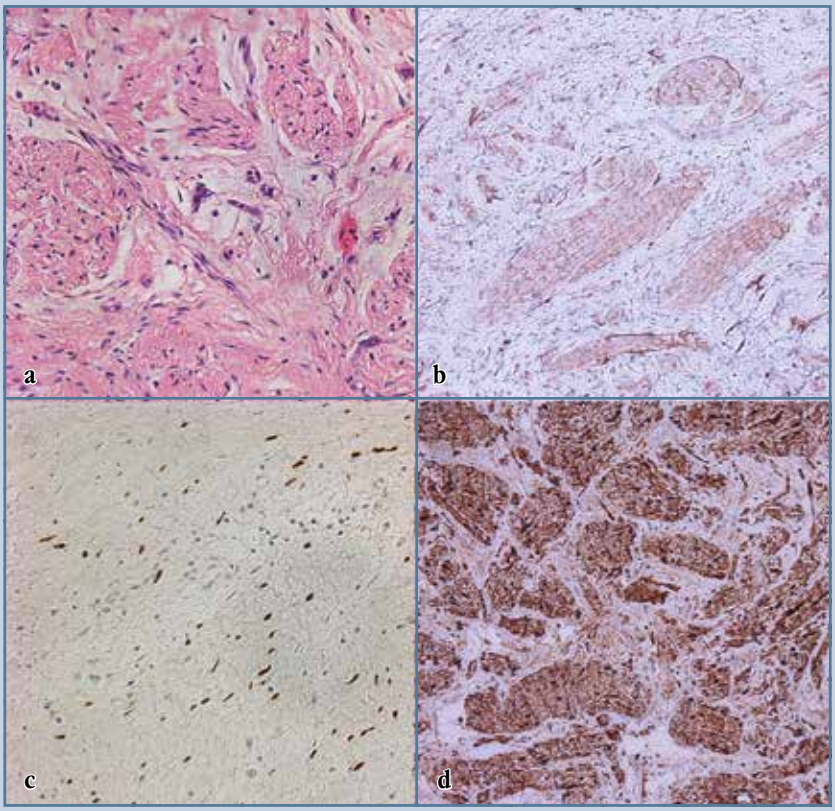

Fig. 5
A plexiform neurofibroma: **a** – the tumor is marked by elongated cells and collagen fibers with nerve fibers of classical structure; hematoxylin and eosin stain; the tumor is being expressed: **b** – GLUT1; **c** – SOX10; **d** – S100; mag. 100

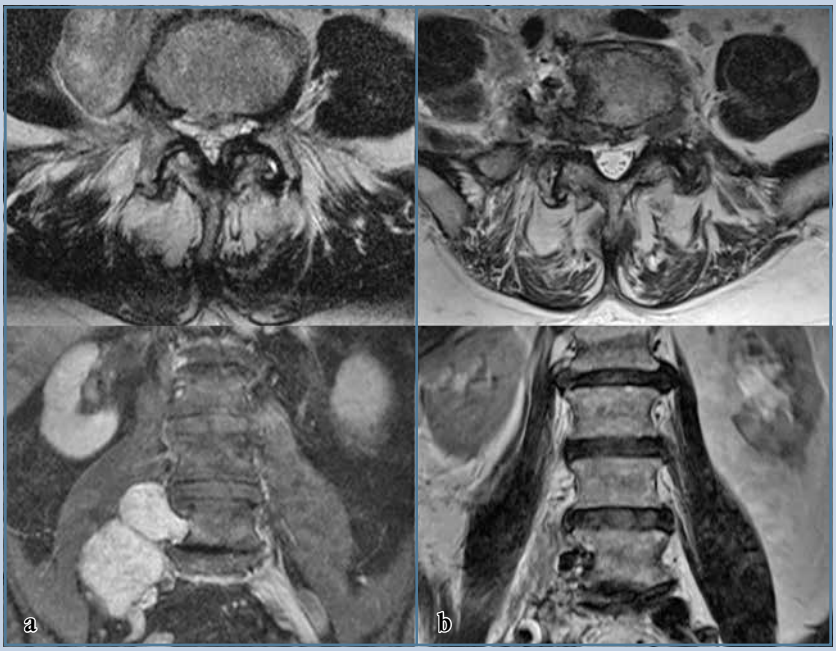

Fig. 6
MRI scan of a 62-year-old female patient S. before **(a)** and after **(b)** the surgery

the operation was performed. It revealed no bleeding points. When the peanut displaced the obturator internus muscle, the arterial bleeding started from the muscle vessel. It was stopped by suturing. A hemostatic sponge was applied to the bleeding area. The blood loss was 200 ml.

The further postoperative period was unremarkable. On the 2nd day after the surgical exploration, the patient was verticalized. The wound was healed by primary intention. Neurological status was without negative changes. The duration of a hospital stay was 34 days. Histological findings: a plexiform neurofibroma (Fig. 8).

The posterior stage was performed in patient after 3 months. The patient was in a face-down position. The incision was made along the midline in the projection of S1–S4. The sacral canal was lanced. The dissection of the neurofibroma was performed from the neural structures. The tumor is tightly agglomerated with the anterior wall of the sacral canal. An electric knife was used to remove the lesion. Hemostasis. Blood loss was 100 ml. The patient was verticalized on the 1st day. The postoperative period was without complications.

A control MRI after the second stage of surgery treatment revealed a residual fragment of the tumor at the level of the sacral foramina S3–S4 up to 1 cm in size. In a year after the surgery, the patient retains anesthesia in the gluteal region, foot and the lower third of the lower leg. Myoparesis – up to 4 points. The radicular pain has regressed. The patient does not take analgesics, and leads a full-fledged lifestyle.

Discussion

The nerve sheath tumors are a subclass of soft tissue masses. It can be both in benign and malignant variants. The

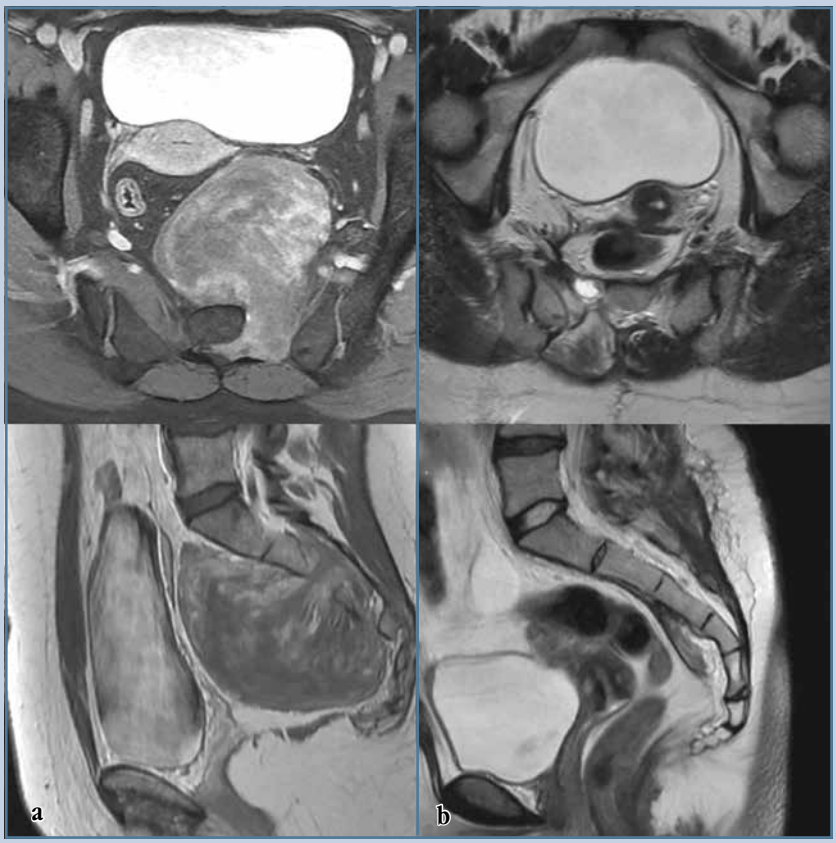

Fig. 7
MRI scan of a 30-year-old female patient S. before **(a)** and after **(b)** the surgery

nerve tumors of the lumbosacral location remain low-symptomatic for a long time and are often diagnosed as accidental findings due to mainly ventral growth and a large volume of the reserved retroperitoneal space [3]. The most part of nerve sheath tumors of this location are schwannomas. The isolated neurofibromas are less frequent than other lesions [10]. In most cases, the tumors are benign. They do not have a tendency to infestation of the adjacent structures [11]. The malignant tumors are represented by malignant schwannomas and neurofibrosarcomas [12].

Normally, the neurological manifestations are the following: lower back pain and moderately expressed radicular symptoms developing in a rather large tumors like hypeesthesia, paresis and pain in the innervation of the affected roots [13, 14]. The slow growth results in a progressive displacement of the pelvic organs, causing a disorder of their function and associated symptoms when they reach a sufficiently large size: discomfort and abdominal pain [8], constipation, [15], deep vein thrombosis [3], and dysuric manifestations.

MRI is a key diagnostic technique. The typical features are a distinct boundary, ovoid or spherical shape, growth from the intervertebral foramen [12]. Most commonly, it is not possible to adequately distinguish a benign type of a tumor from a malignant one, as well as a histological type according to X-ray examination data. [16]. According to the MRI scan, the majority of benign neurogenic tumors are shown by a homogeneous mass. About 6 % of tumors have cystic changes. The malignant tumors have heterogeneous features and contain cysts in 75 % of cases [17]. Meanwhile, degenerative changes, known in the histological literature as signs of "ancient" schwannoma, include the cyst formation, signs of hemorrhages, calcifications and fibrosis, which is not typical for neurofibromas [16]. Additionally, in most cases, the schwannoma has a capsule in the form of a thin hypodense rim along the lesion periphery [16]. The cases of misconception of the tumor as *m. iliopsoas* abscess, ovarian cyst [18, 19], aneurysmal bone cyst [20] and teratoma [21] are described. The CT angiography is an obligatory diagnostic technique before the surgery. It is aimed at determining the features of the blood supply to the tumor and its connection with the main vessels. According to CT, the tumors are dense lesions with a clear contour. The enlarged sacral foramina, bone caries, cysts and calcifications can be visualized [22].

From the macroscopic point of view, the schwannomas are well-defined nodes with a clearly defined capsule. They are usually gray-yellow or contain yellow deposits on the section. They may have areas of angiomatosis and cysts. Microscopically, Antoni type A with Verocay bodies (a palisade structures of the elongated cells) and Antoni type B of a reticular structure with loosely arranged cells were earlier considered. Today, cellular, plexiform, and melanotic schwannomas are recognized. There is a separate report about the "ancient" schwannoma with a pronounced cellular polymorphism, intracellular and cytoplasmic inclusions, which are classified as benign. The cellular schwannomas are composed of hypercellular tissue of the Antoni type A but without Verocay bodies. The melanotic schwannomas are made up of Schwann cells. However, the tumor contains a large amount of melanin, melanosomes and is reactive to melanocytic markers. Schwannomas express S100, SOX10, LEU7, and calretinin.

The neurofibromas consist of Schwann cells and fibroblasts producing collagen. Macroscopically, well-shaped nodes represent them. There are two types of plexiform neurofibroma. The first one grows from the nerve plexuses.

The second one is atypical, characterized by high cell density, monomorphic cytology, mitotic figures and/or bundle growth with cell abnormality. A localized sporadic neurofibroma of a whitish-gray color usually has a capsule, unlike a schwannoma, of a softer consistency, especially at the periphery. It can resemble a myxoma, i.e. to be jelly-like. The neurofibroma expresses S100, SOX10, GLUT1. The stromal cells can be stained with CD34 [23–25].

The necessity of performing a preoperative biopsy remains a controversial point. A number of authors concur that the amount of material obtained during aspiration and trepanopuncture is inadequate for an accurate diagnosis. The areas of cellular polymorphism in certain tumor grafts may be wrongly regarded as signs of malignancy, and the puncturation may result in the development of complications [3, 8]. An intraoperative study of frozen preparations was conducted in a number of patients [11, 26]. In our cases, all patients had typical MR signs of root tumors. The degree of malignancy, in our opinion, does not have a fundamental effect on the extent of surgery. Both in the case of benign and malignant tumors, a more favorable prognosis is associated with the degree of radical resection, and consequently it is necessary to aim for total removal [7].

As a rule, the planning of operational approach is performed considering the lesion location. The cases of laparoscopic and robot-assisted removal are described [9, 27, 28], but in most cases classical open approaches are performed.

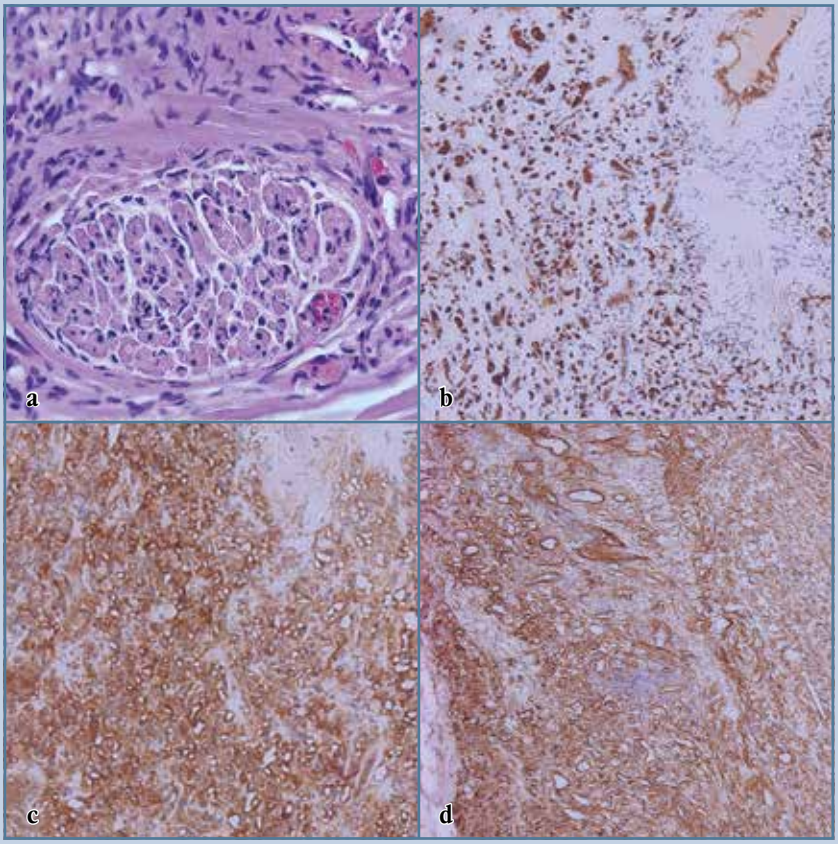

Fig. 8
A plexiform neurofibroma: **a** – with normal nerve fibers, hematoxylin and eosin stain; the tumor is being expressed: **b** – CD34; **c** – S100; **d** – Collagen IV; mag. 100

The lateral retroperitoneal approach is a priority in the lateralized tumor location. It is also possible to perform a pararectal linear or oblique transection. If the tumor has a median presacral location, a lower midline laparotomy is performed [29]. The approach should provide not only a complete visualization of the tumor, but also the possibility of surgical releases of large vessels and organs of the retroperitoneal space. If there is a significant intracanal component, it is safer to perform resection in several separate stages. Klimo et al. [10] proposed to separate sacral neurinomas into 4 types: type 1 – with tumor in the sacral canal; type 2 – with tumor exit through the sacral foramina into the retroperitoneal space; type 3 – with tumor's primary location in the sacrum, but with the spread in the ventral and dorsal directions into adjacent spaces; type 4 – with a solitary presacral location. If the tumor is located distal to the S1 vertebra, anterior approach is recommended for tumors of type 1, types 2 and 4 require isolated anterior approach, and type 3 – combined anterior and posterior approach. A number of authors have reported successful cases of radical removal of presacral schwannomas through an isolated posterior approach [26] or minimally invasive paracoccigeal one. There is a lower probability of damage to great vessels and organs of the retroperitoneal space during intracapsular removal, as well as the absence of long-term negative consequences of anterior approach (for example, erectile dysfunction). From our point of view, such an approach can be complicated to implement with a dense consistency of the tumor, the absence of a distinct boundary between the capsule and the tumor mass, and in cases of abundantly blood-supplied tumors. Additionally, it should be considered that with the development of retroperitoneal bleeding, it may be unrecognized for a certain amount of time and require an emergency operative exploration through the anterior approach, which may result in an acute blood loss. Despite the described cases of presacral schwannomas removal through the isolated posterior approach, the anterior approach, in our opinion, is a precedence.

The specialists performed lumbopelvic instrumented fixation [7, 13, 30] or the insertion of bone cement [31] in a number of cases with extensive areas of bone destruction, involving the sacroiliac joint and intervertebral joints [7, 13, 30].

It is recommended to the surgery to be performed by a multidisciplinary team [7]. If the surgery was performed through a lateral approach, then the vessel surgeon was involved in it. In case of the median approach, the operation was carried with the involvement of an abdominal surgeon. Addionally to the neurosurgical instruments, the technical equipment should include a vascular access kit, mirrors and wound retractors for wide anterior approaches, hemostatic dressings, and headlights. The surgery should be planned considering the possible profuse blood loss, with the necessary supply of donor blood and the cell-saver device. Wei et al. [7] describe the use of temporary abdominal aorta balloon occlusion in 24 cases in a series of 48 cases of sacral neurinomas. The average blood loss was at least 1600 ml (500–5000 ml). A patient died of DIC–syndrome associated with a hemorrhagic shock According to the meta-analysis, the average volume of blood loss was about 2000 ml [6]. A possible cause of blood loss reduction is preoperative embolization of tumor vessels [3, 32]. It needs to be taken into account that the tumor structure may differ significantly in density and blood supply. In the two cases described, the tumors were dense and could not be destructed by an ultrasound disintegrator. Therefore, the removal was carried out with an electric knife and scissors. In one case, the tumor was soft, cystic, but abundantly supplied with blood.

We believe that the use of an individual 3D model is a useful additional tool for perioperative planning. During the surgery it improves the understanding of the topography of the tumor and adjacent structures. Meanwhile, we do not consider this technique as having a significant impact on surgical strategy. The main disadvantage is the complexity of segmentation of the venous bed due to irregular and weak contrast during CT angiography. Previously, the advantages of models for the removal of spinal column tumors were reported [33, 34], while no data on the use of 3D models in the surgery of retroperitoneal neurogenic tumors were found. Considering the extremely low frequency of occurrence of such lesions and the variety of pathological characteristics (size, location, blood supply, etc.), it is not possible to evaluate the objective benefits of using 3D printing for specific parameters in a comparative study.

Alderete et al. [11] report a 45 % incidence of complications. They are hematoma, deep vein thrombosis and infection of the surgical site during the resection of neurogenic tumors of the presacral space. In our experience, similar complications developed in two patients. The first patient suffered from repeated bleeding, which required an urgent operative exploration revision. A deep venous thrombosis manifested the second complication. In order to detect bleeding in a timely manner, we recommend not removing the drainage with a diameter of at least one cm in the retroperitoneal space during the first day of verticalization. The most common neurological complication is the aggravation of paresis and sensation disorders [11]. Meanwhile, when removing benign neural tumors, the development of a significant neurological deficit is not typical. The authors explain this by intraoperative technique with the preservation of functionally relevant fibers. Even in our cases, when the tumor roots were detected, it was not possible to separate their fibers from the capsule. Nevertheless, we did not observe a characteristic pattern of complete lesion of the involved roots after surgery. All the patients were verticalized on the 1st or 2nd day. They walked independently, without auxiliary devices. Hypothetically, this can be explained by a prolonged growth of the tumor with a regression in the functions of the involved roots and their compensation due to intact neural lesions. If the tumor is localized in the presacral area, it is impossible to allow a bilateral intersection of the S2 root in order to avoid the pelvic organ dysfunction. Generally, in the long-term period after the removal of benign retroperitoneal neurogenic tumors, patients do not observe a significant decrease in the life quality [11]. During a meta-analysis by Pennington et al. [6], when removing benign sutures, complications developed in 25.6 % of cases. The most frequent were weakness and decreased sensitivity in the legs, urinary retention and erectile dysfunction in men [6]. The complication rate correlated with the tumor size and was higher with open anterior approach, compared with endoscopic, robot-assisted and isolated ones. Nevertheless, the groups may be heterogeneous in terms of the size and tumor location.

The chance of a tumor recurrence is primarily determined by the histologic pattern. The malignant tumors of the roots, as a rule, are susceptible to continued growth, even after total removal and a combined treatment [30]. The main recurrence predictor in benign tumors that are not associated with neurofibromatosis is the eradication degree of the tumor. A 10-year recurrence-free period was observed in 40.7 % of cases with partial removal of benign schwannomas; in 100.0 % with en-block removal, and in 95.0 % with gross total resection (the differences between the last two indicators are not statistically significant) [6]. In this case, there are also questions concerning the data comparability. This is due to the fact that with tumors of type 1–3 according to Klimo, the en-block removal involves a sacrectomy. This method is not justified for benign tumors of the membranes of the vertebral nerves.

Conclusion

The giant presacral neurogenic tumors are rare. They are not a common pathology in the spinal surgeons practice. A multidisciplinary surgical team should perform the procedure. The possibility of life-threatening complications is typical in the early postoperative period. The use of individual 3D models can be regarded as an additional useful tool for perioperative planning.

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