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Integrating tractography in pelvic surgery: a proof of concept

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ABSTRACT

Objective: To show the interest of tractography in pelvic surgery by a demonstrative case of neurofibroma resected by robotic assisted laparoscopy.
Summary background data: Although diffusion tensor magnetic resonance imaging, along with tractography algorithms, is increasingly included in image guided neurosurgery methods, it is less frequently used for the peripheral nervous system such as the pelvic sacral plexus. We report one observation demonstrating the interest of such image data for pelvic surgery.
Methods: A 12-years-old girl with neurofibromatosis presented with a growing left pelvic neurofibroma on repeated pelvic MRI with an increased SUV (2.83) on PET scan. A diffusion tensor pelvic MRI was performed before and after robotically assisted tumoral resection, and a tractography algorithm was applied on both images.
Results: The pre-operative tractogram showed the nervous nature of the tumor in close contact with the left sacral plexus. Section of a nerve trunk encased in the neurofibroma was mandatory for the resection and well documented by the 3D enhanced vision provided by the robot. Post-operatively, the patient showed a slight paresthesia and dysesthesia of the left leg calf and of the plantar vault (left S2 territory), without any motor deficit. The post-operative tractogram showed a thinning of the left S2 sacral root and the disappearance of right aberrant nervous tracts.
Conclusions: This clinical case validates the ability of pelvic tractography to deliver a proper imaging of the sacral nervous network and emphasizes the potential usefulness of this approach in pelvic surgery management, with perspectives of image-guided surgery.

1. Introduction

The pelvis region is characterized by the richness of the vascular and nervous network dedicated to the urinary, genital and digestive tracts. Innervation of these structures is regulated by the somatic and autonomous sympathetic and parasympathetic systems. Pelvic surgery entails a risk to injure this nervous network which is poorly evaluated on classical imaging (CT scan or MRI). Diffusion tensor MRI is currently the only non-invasive way to visualize nervous fibers and has been used mainly for central nervous system (CNS) imaging and image guidance since many years [1]. This tool, along with associated tractography algorithms, was also developed for peripheral nervous system, such as the pelvic sacral plexus [2] and neurogenic tumors [3]. We report the case of a 12 years girl with Neurofibromatosis type 1 (NF1) presenting a presacral neurofibroma where a prophylactic surgery was proposed to avoid the risk of transformation to a malignant peripheral nerve sheath tumor (MPNST). Pre-operative pelvic tractography combined to robotic surgery allowed for a precise definition of the neurofibroma origin and anticipation of potential complications. Post-operative tractography confirmed the validity of this imaging technique and underlines its potential benefit in pelvic surgery.

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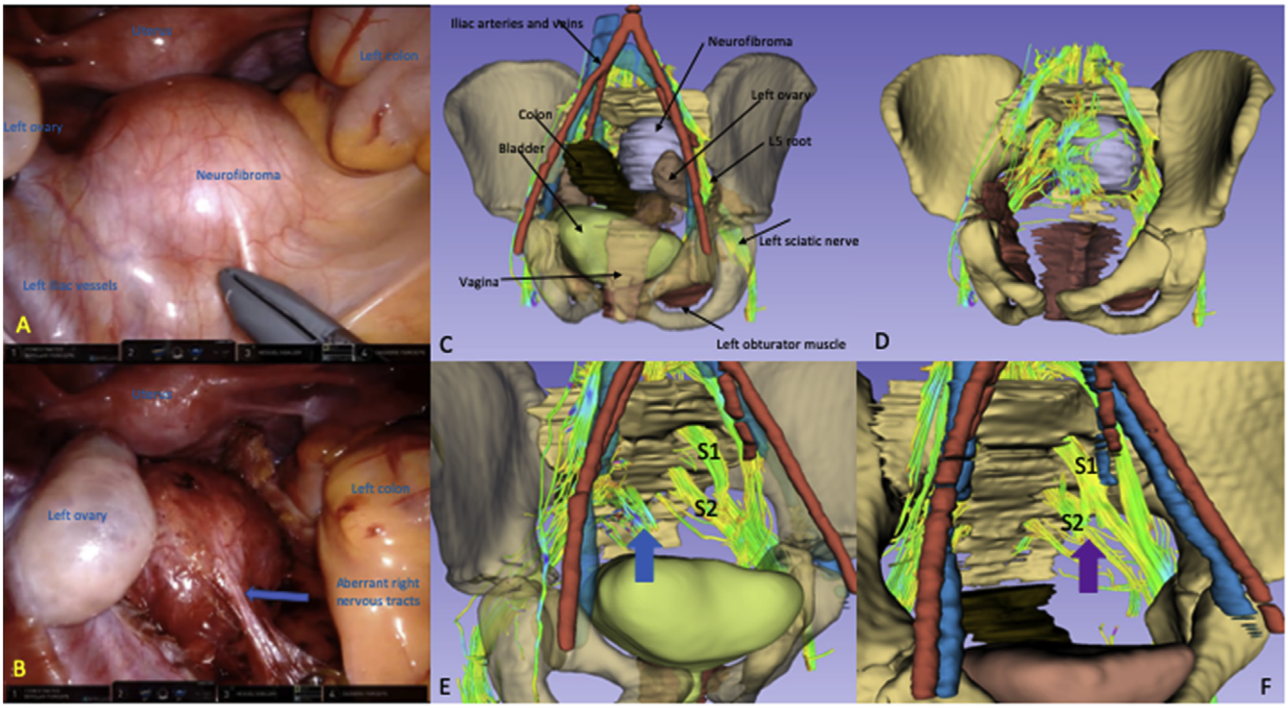

Fig. 1. Results
A = Robotic surgical view of the pelvis inspection before the dissection of the pre-sacral tumor.
B = Robotic surgical view during the dissection, showing the nervous tracts linking the sacral plexus and the tumor and that were sectioned for tumor removal (Fig. 1E).
C = 3D model of the pelvis performed on the pre-operative MRI, with manual delineation of each organ using Slicer 3D 4.6.1 Segment Editor [1] and including the sacral plexus tractogram and the pre-sacral neurofibroma (in purple).
D = Merge of both manual segmentation and tractogram of the neurofibroma, in contact with the left part of the sacral plexus.
E = Pre-operative tractogram without the representation of the neurofibroma. Note some aberrant fibers coming from the right side of the sacral roots (blue arrow) and reaching the tumor (blue arrow). S1 = sacral root S1. S2 = sacral root S2.
F= Post-operative tractogram. Note the thinning of S2 root (purple arrow), where the tumor was supposedly attached, in agreement with the post-operative clinical findings of the lesion of sensitive S2 territory. It also shows the disappearance of the aberrant nervous tracts coming from the right side of the sacral plexus. S1 = sacral root S1. S2 = sacral root S2.
[1] https://www.slicer.org.

2. Patient and method

2.1. Clinical history

A 12 years old girl was followed for NF1 when she was diagnosed with a pelvic neurofibroma. The MRI showed a slowly growing lesion of 50 × 45 × 53 mm, extending within the 2nd left sacral foramina. A PET-scan revealed a SUV max index of 2.83 with a tumor/liver ratio of 1.6. A percutaneous biopsy was performed, that showed a neurofibroma with cytological abnormalities but without any sign of malignancy, and no upraise of the mitotic index. The surgical resection of the neurofibroma was scheduled with a robotically assisted approach (Da Vinci Xi robot, Intuitive Surgical ®).

2.2. Imaging protocols

A pre-operative and a post-operative pelvic MRI were performed on a 3 T machine (General Electrics®), according to the classical protocol for children's pelvic MRI used in the radiology department. After ethical board committee agreement, two additional sequences for research purposes were added, with an overall duration of 45 min and without general anesthesia:

- Coronal T2-weighted volumic sequence: isotropic acquisition with a voxel size of 1 × 1x1 mm^3, duration = 7 min.
- Axial diffusion sequence: 25 directions, b600, quasi-isotropic resolution with a voxel size of 3.2 × 3.2 × 3.5 mm^3, duration = 5 min.

Anatomical images are segmented in order to build a 3D individual model of the patient using Slicer 3D 4.6.1 software. Results from tractography are then added to this model.

3. Results

Pre-operative imaging with tractography showed that the lesion was arising from the left S2 root. During surgery, a voluminous tumor was found, displacing both the rectum and the colon to the right (see Fig. 1A). The tumor was clearly attached to a nervous structure coming from the sacral roots, which needed to be sectioned for removal (see Fig. 1B). Tumor extraction was done within a bag after partition through the umbilical port. No blood transfusion was required. The surgery length was of 15 min. The pathology report showed a typical plexiform neurofibroma without malignancy. The patient was discharged at day 3. Post-operatively, the clinical examination showed paresthesia and dysesthesia of the left leg calf and of the plantar vault (left S1–S2 territory), without any motor deficit. These symptoms spontaneously disappeared within the following weeks. The 3D patient specific model, the analysis of the pre- and post-operative tractogram are shown in Fig. 1C, D, E and F respectively. MRI performed 8 months post-operatively did not show any tumoral residue and apparently normal sacral roots S2 and S3 while viewing standard T1 and T2-

weighted sequences. Diffusion tensor imaging with tractography however showed a thinning of S2 left root (purple arrow, Fig. 1D), where the tumor was supposedly attached, in agreement with post-operative clinical symptoms.

4. Discussion

This case report demonstrates the interest of integrating tractography in a pediatric case of pelvic neurofibroma and support its wider use in pelvic surgery with neurological concerns.

The surgery was indicated because of the high risk of transformation to a malignant peripheral nerve sheath tumor (MPNST) suggested by the results of the Pet Scan with an index above 2 and a tumor/liver ratio above 1.5 [4]. MPNST are highly aggressive sarcomas associated with a poor prognosis, for which the treatment of choice remains surgery [5,6]. The robotic assisted technique was chosen because of its advantages in pelvic surgery [7,8], and in order to avoid scar and preserve muscle in a patient whose disease is known to expose to iterative surgeries. The use of tractography integrated in a patient specific 3D model helped to identify better the connection of the tumor with the sacral plexus and to inform properly the patient and her family of the potential neurological risks. The thinning of the S2 sacral root on the post-operative sacral plexus tractogram and the disappearance of aberrant nervous tracts probably resulted from the section of these components to remove the tumor (see Fig. 1B, E and F). These results must be taken with caution due to the absence of quantitative measurements and available normal values in tractography parameters of the sacral plexus especially in children. They are however in agreement with reports about tractography and peripheral nerve sheath tumors surgery [3,9], mentioning the thinning of the involved peripheral nerve.

Tractography is a post-processing tool applied on diffusion tensor MRI images, using mathematical algorithms and work remains to be done to prove its actual accuracy, especially for peripheral nerves. The correspondence between the clinical symptoms that occurred during the post-operative course and the nerve lesion identified on post-operative imaging validates *a posteriori* the clinical relevance of tractography.

5. Conclusion

In this case report, we combined the latest surgical and imaging technologies to offer the best management for prophylactic surgery in the frame of a genetic predisposition syndrome to cancer. To our knowledge, it is the first report showing in the same 3D scene the peripheral nerve tractogram, the tumor and the surrounding organs. We are currently working to optimize and automatize both segmentation and tractography processes in order to use it in daily clinical practice.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

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Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Conflicts of interest

The following authors have no financial disclosures: C.O.M, E.M, A.V, J-B.M, Q.P, A.D, L.B, P.G, T.B, D.G, N.B, I.B, S.S.

Appendix A. Supplementary data

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