European Spine Journal (2025) 34:643–649
https://doi.org/10.1007/s00586-024-08625-8

CASE REPORT

Minimal-invasive, image guided, 360-degree resection of ilio-lumbo-sacral ostechondroma, planned on the 3D model in a child with hereditary multiple ostechondroma (HMO)

Lukas Bobinski[1,2] · **Evangelos Tachtaras**[3] · **Erik Hedström**[1,2]

Received: 7 June 2024 / Revised: 9 December 2024 / Accepted: 17 December 2024 / Published online: 30 December 2024

Abstract

Background Hereditary Multiple Osteochondromas (HMO), previously known as Multiple Hereditary Exostoses (MHE), is a genetic disorder characterized by the formation of multiple, benign, exostoses (osteochondromas) growing from the metaphyseal region of long bones as well as from the axial skeleton. Lesions originating from the lumbar spine region are rare, and are most common growing from the posterior element of the vertebrae. HMO associated osteochondromas are difficult to treat due to continuous and incontrollable growth of these lesions and a lifetime risk for malignant transformation.

Case report We describe a case of a 16-year old patient with known HMO who developed a giant ilio-lumbo-sacral osteochondroma. The tumor protruded into the L4-S1 intraspinal foramina with exophytic expansion to the right psoas muscle and lumbar plexus with compression of the right common iliac vein. To plan and execute the resection and minimize the risk of complications, we used a 3D printed model of the lesion with intraabdominal vessels. The patient was operated during a two-stage procedure - first by mini-open, transabdominal, navigated resection of the lesion, followed by delayed posterior, mini-invasive, navigated resection. The outcome was uneventful and there were no signs of regrowth or malignant transformation during 4 years of follow-up.

Conclusion We describe a 360-degree surgical resection with application of a 3D printed model, navigation, and mini-invasive techniques. Our report may be useful and inspire spine surgeons to apply similar techniques to treat complex spine lesions.

Keywords Hereditary multiple osteochondroma · Exostoses · 3D printed model

Introduction

Hereditary Multiple Osteochondromas (HMO), previously known as Multiple Hereditary Exostoses (MHE), is a dominant, autosomal, hereditary musculoskeletal disorder mainly caused by mutations in exostosin-1 (EXT1) and exostosin-2 (EXT2) genes [1]. HMO presents by the formation of multiple, benign, exostoses (osteochondromas). These lesions are considered to be developmental hamartomas rather than true neoplasm [2]. However, in patients with HMO there is an estimated 3–5% lifetime risk of malignant transformation [3]. HMO is also considered the most common skeletal dysplasia with a prevalence of 1:50,000 [4].

Patients with HMO often develop six or more osteochondromas that vary in location but arise most commonly from the metaphyseal region of long bones [5]. Apart from extremities, osteochondromas can also develop from the axial skeleton including the pelvis, scapula, and spine. Osteochondromas growing in the lumbar region are rare and are described in the literature as case series or case reports that include spontaneous exostoses and HMO [6, 7].

✉ Lukas Bobinski
lukas.bobinski@umu.se

[1] Department of Orthopedics, Umeå University Hospital, Umeå, Sweden

[2] Department of Diagnostics and Intervention, Orthopedics, Umeå University, Umeå, Sweden

[3] Alpha Surgical Care Private Clinic, Umeå, Sweden

Case report

This report involved a 16-year-old girl with a known history of HMO. Her father and two younger siblings had also been diagnosed with HMO. The patient had undergone previous surgeries for osteochondromas originating around the ankle and knees. She had a previous loss of function corresponding to the right peroneal nerve. The patient's main complaint was persistent, dull pain, seated deeply within the right abdominal fossa. On clinical examination there was significant impairment of hip flexion and internal rotation. Radiological examination with computer tomography with angiography (CT-angio) revealed a large ilio-lumbo-sacral osteochondroma that protruded medially with ingrowth into the L4-S1 intraspinal foramina and exophytic expansion toward the right common iliac vein (See image 1). Magnetic Resonance Imaging (MRI) demonstrated intraforaminal impingement of several nerve roots and elevation of the psoas muscle with the lumbo-sacral plexus. Investigation with electroneurography (ENG) confirmed both proximal (lumbar nerve roots) and distal (knee) nerve impingement.

Due to concern about continuous growth of the osteochondroma with possible further neurological deterioration as well as a risk for malignant transformation, surgical intervention was discussed in detail with the patient and her parents during multidisciplinary consultation. The patient and her parents consented to surgery. To facilitate surgical planning we obtained a 3D printed reconstruction model of the osteochondroma that included the lumbo-sacral spine, pelvis and major vessels based on the CT investigation (See image 2 and 3).

Surgery 1: Image guided, mini-open, anterior retroperitoneal approach

The patient was securely placed on her left side on the Jackson table and supported with padded vacuum cushions with the right leg slightly flexed. The surgery was performed under intraoperative neuromonitoring with somatosensory evoked potentials (SSEP), motor evoked potentials (MEP), and direct nerve root stimulation.

After sterile preparation and draping, a reference navigation frame (small passive frame, Medtronic, USA) was attached percutaneously to the anterior, superior iliac spine (ASIS). 3D imaging was then performed with the O-arm in the lateral position. The 3D model was placed in a sterile transparent bag and used as an intraoperative template. The initial step of the surgery was exposure and mobilization of the right common iliac vein and artery followed by lateral mobilization of the psoas muscle. The resection was carried out using a high-speed drill (Midas Rex, Medtronic, USA) and micro curettes. Progress was frequently checked using a navigated pointer during exploration and decompression of intraspinal foramina (See image 4). There was a change in the SSEP corresponding to L3 and L4 nerve roots during excessive retraction of the psoas at the L4/L5 level which returned to baseline after adjustment of the retractor. There were no changes in neuromonitoring at the end of the surgery. The wound was rinsed with saline solution and closed with watertight suturing of the abdominal wall with intracutaneous sutures of the skin. The patient was mobilized the first postoperative day and discharged on the second. The patient reported anesthesia corresponding to the right

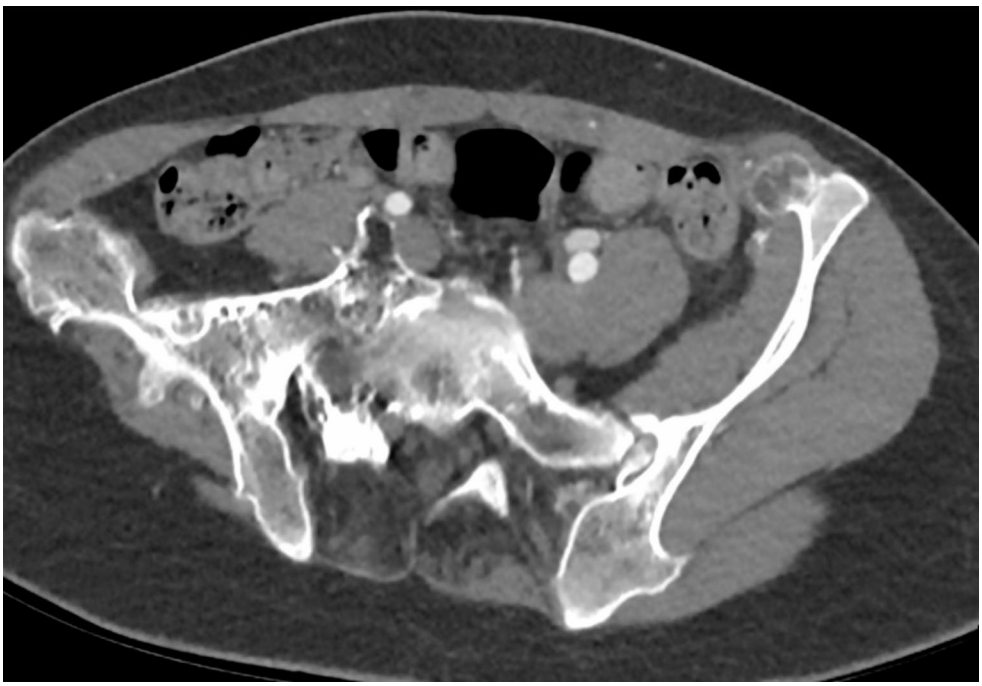

Image 1 Abdominal CT angiography demonstrates impingement of right psoas muscle and dislocation of intra-abdominal iliac vessels

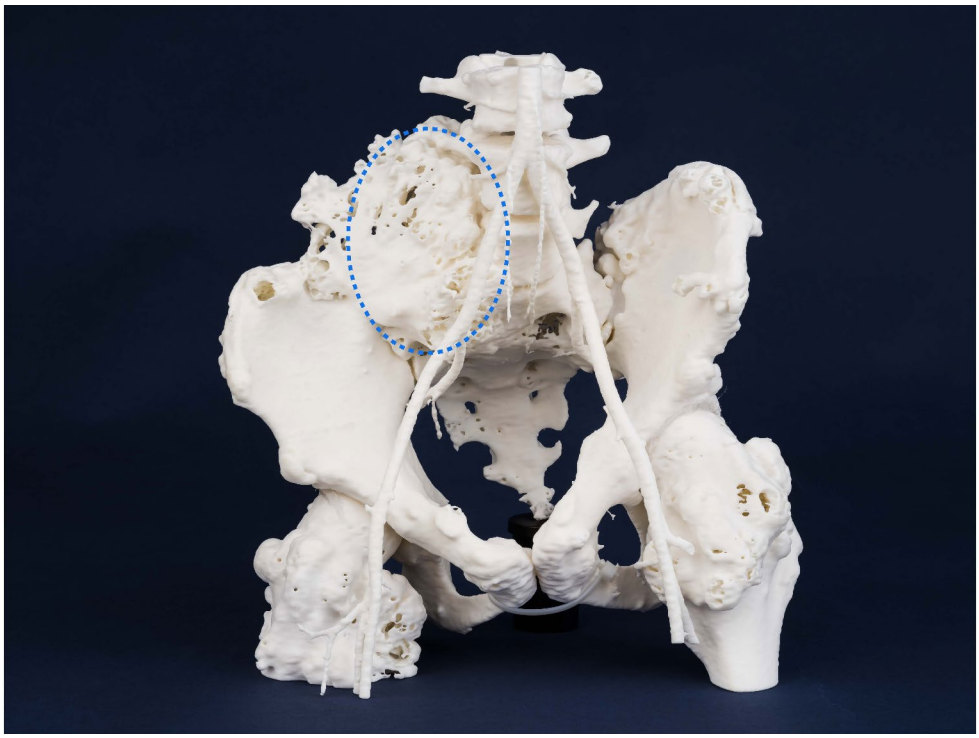

Image 2 Anterior view of the 3D model. The planned resection is marked with the blue dotted circle

Image 3 Posterior view of the 3D model

L3 dermatome and dysesthesia within the L4 dermatome, which resolved spontaneously within four weeks after the surgery. The motor neurological status was unchanged upon discharge. A control CT scan confirmed an adequate resection (See image 5).

Surgery 2: Image guided, mini-invasive, posterior approach. The second stage surgery was deliberately postponed in order to evaluate the neurological outcome and pain control after anterior resection

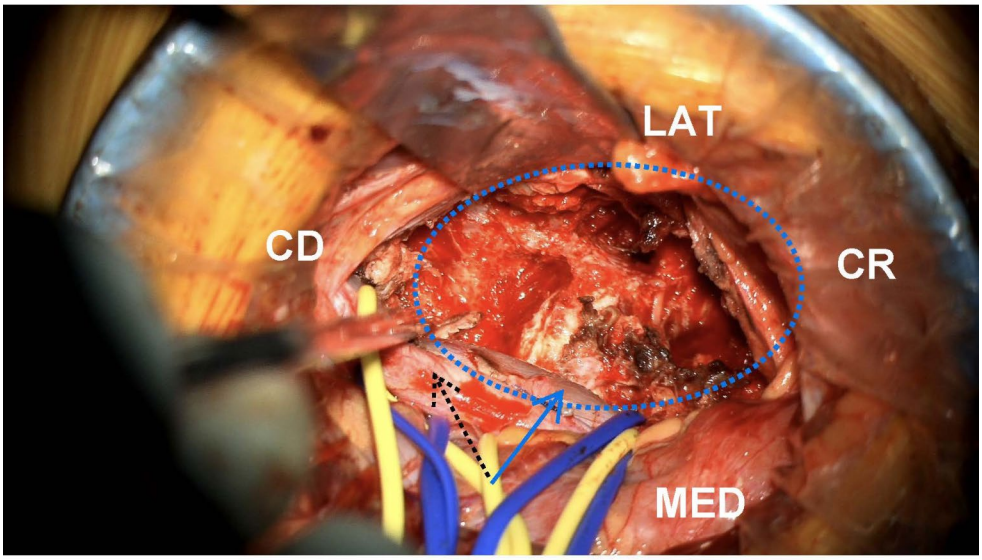

Image 4 Intraoperative view at the end of anterior surgery. Orientation: CR- cranial, CD-caudal, LAT-lateral, MED-medial. The psoas muscle is retracted laterally under the retractor blade. The blue arrow indicates the common iliac vein. The black dotted arrow indicates the common iliac artery. The blue dotted circle corresponds to the extension of anterior resection as planned on the 3D model

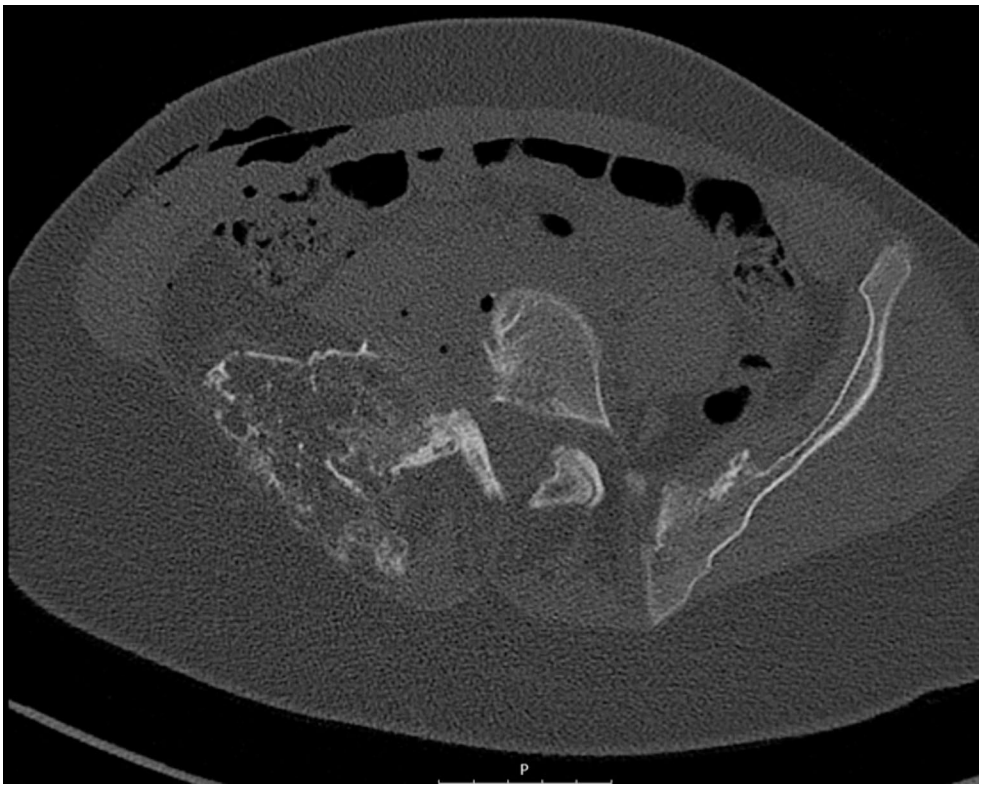

Image 5 Control CT examination demonstrates adequate resection of the lesion, including decompressed intraspinal foramina, after the anterior surgery

The residual mass caused persistent discomfort in the lumbar paravertebral region, hence the patient and her parents opted for resection of the remaining mass. The resection was performed through a posterior mini-invasive approach under intraoperative neuromonitoring (SSEP, MEP) as well as direct nerve-root stimulation to minimize the risk for accidental injury to the lumbo-sacral plexus. The patient was placed in a prone position on a Jackson table. The navigation reference frame (small passive frame, Medtronic, USA) was attached percutaneously to the contralateral posterior, superior iliac spine (PSIS). A 3D image was obtained with the O-arm at the beginning of surgery to plan both skin incision and position of the mini-invasive retractor (MetrX Quadrant, Medtronic, USA) (See image 6). The resection was performed under the microscope using a navigated high-speed drill (MidasRex, Medtronic, USA). The resected material was sent for histopathological investigation. The wound was washed out with saline and closed in layers with a water-tight closure. There were no changes in neuromonitoring throughout the surgery. Postoperatively,

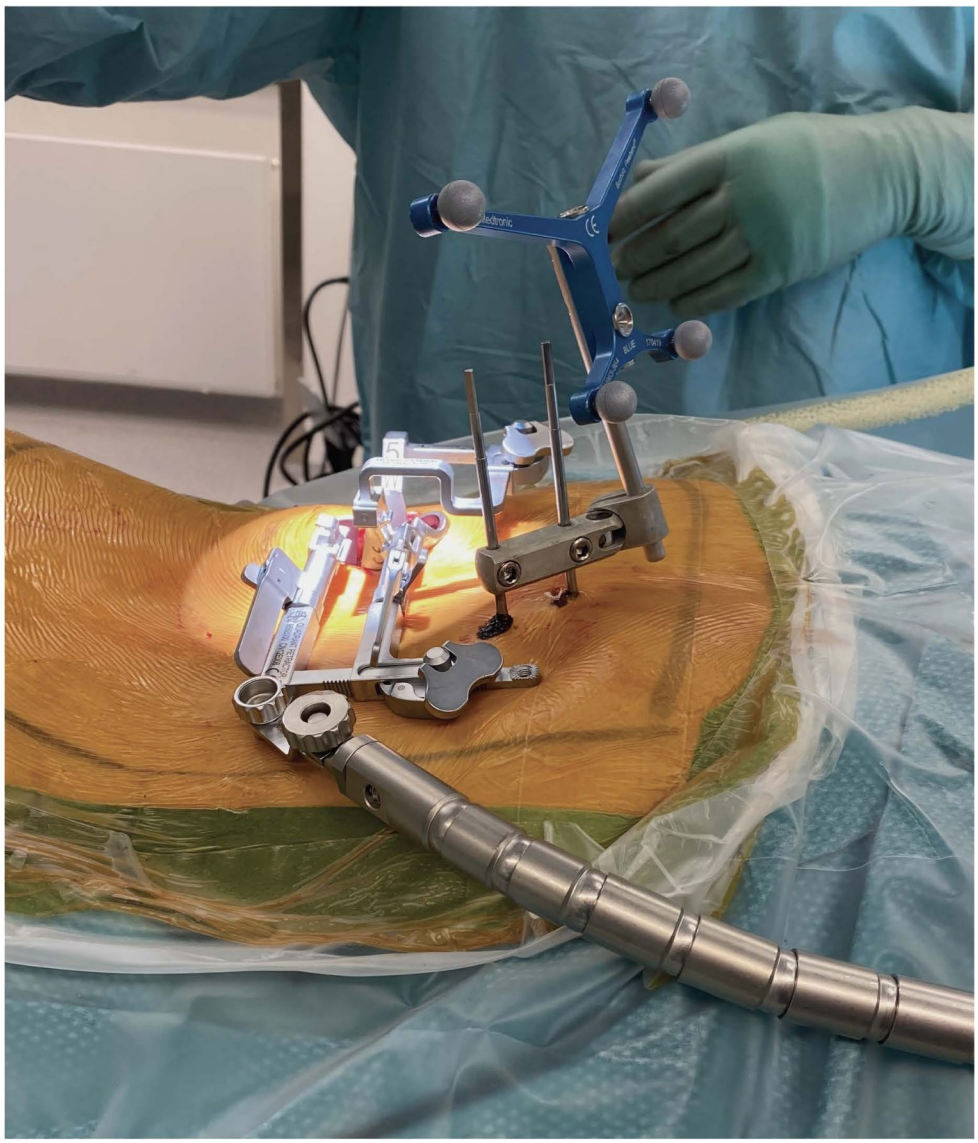

Image 6 Position of the navigation reference and the tubular retractor

the patient was mobilized on the first day and discharged on the second. The neurological status at the discharge was unchanged. A control CT scan confirmed adequate resection with only small, lateral portion left attached to the iliac crest (See image 7).

Follow-up

The patient was discussed during a multidisciplinary tumor board. Histopathologic examination confirmed osteochondroma without any signs of malignant transformation. The postoperative period was uneventful as there were no postoperative complications. The patient was followed clinically and radiologically for four years. The dull pain in the right fossa resolved completely. The neurological function and sensation in the right leg returned to the initial status before the first surgery. The patient required additional surgery of the hip, also due to osteochondroma.

Discussion

The HMO associated osteochondromas are usually densely calcified, connected to the bone marrow of the underlying bone with a thin cartilage cap on the surface of the lesion. The condition can be challenging to treat as these tend to grow in number and size, cause movement restriction, pain, lead to early onset of degenerative joint disease as well as development of spine deformities like scoliosis. In spine, they commonly develop from the posterior elements of the spine but they can extend into the spinal canal close to neighboring neurostructures, with cauliflower-like growth pattern. This can compromise the function of, the spinal

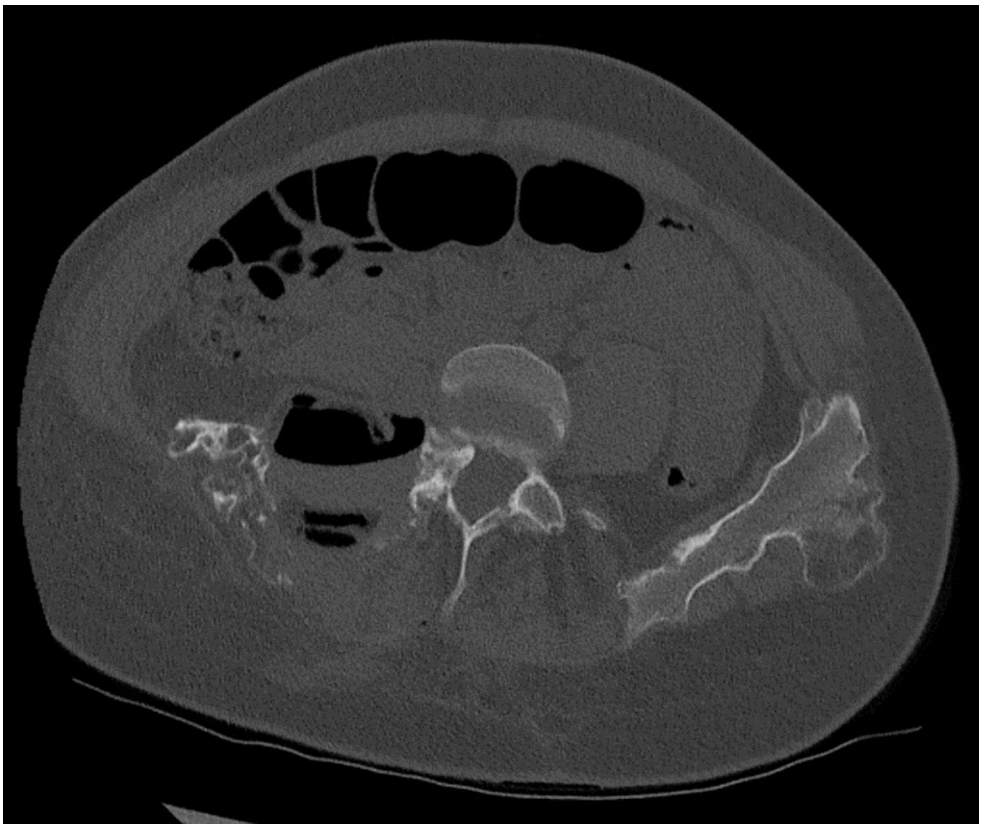

Image 7 Control CT examination demonstrates adequate resection of the lesion after the posterior surgery

cord and nerve-roots causing dysesthesia, neuropathic pain, and eventually onset of myelopathy [8, 9]. Thus, spinal osteochondromas require careful surgical treatment to prevent serious and potentially irreversible disability.

Roach et al. investigated a cohort of patients with HMO and reported that 68% of the patients developed spinal osteochondromas. Moreover, 27% of these patients presented with lesions extending into the spinal canal [7].

Moreover, there is an estimated 5% lifetime risk of osteochondromas transforming into malignant chondrosarcoma, which is both a chemo- and radiation resistant [10–12].

In our case, the on-block resection was not technically feasible because close contact and compression of neurostructures with dislocation of major intraabdominal vessels and the psoas muscle. Rymarczuk et al. described transabdominal resection of large L5 osteochondroma that caused compression of the right iliac vein but without expansion into the spinal foramina as in our case [13].

To better understand the three-dimensional anatomy and plan the extension of the resection, we used a 3D printed model based on thin sliced CT as described in previous publications [14, 15]. Hence, we decided to utilize intraoperative navigation for better control of the resection. The control of lateral extension under the psoas muscle and more importantly the depth of the intrusion into the intravertebral foramina posed the biggest challenge, which could easily lead to perioperative confusion and misjudgment within the distorted anatomy. Despite the lateral surgical position, we were able to carry out meticulous exploration and probing within the intraspinal foramina, using a navigated pointer and nerve root stimulator, enabled the resection without jeopardizing neurological function and minimize the risks of complications.

The same principles (navigation and neuromonitoring) were applied during the second surgery. Navigation enabled safe access to the foramen of L4, L5 and S1, which were obscured by the dorsal portion of the lesion. The navigated high-speed drill promoted resection with regular check-ups with the navigation to avoid accidental violation of the neurostructures. Furthermore, we were able to preserve the intraspinal facets with attached ligaments and muscles, hence there was no need for stabilization. The mini-invasive exposure enabled quick recovery and very good pain control.

Conclusions

To our knowledge, this is the first described 360-degree staged resection with a navigated, mini-open/mini-invasive approach using a 3D printed model. Our surgical strategy provided us with adequate access to the lesion and enabled

controlled resection while also minimizing risks. The neuro- and orthopedic spine surgeons may find our method useful and inspiring when treating similar lesions.

Acknowledgements The authors would like to thank Klas Sjöberg, medical photographer, who created images 2 and 3 using focus stacking technique.

Funding Open access funding provided by Umea University.

Declarations

Competing interests Dr. Bobinski received a teaching honorarium and a research grand from Medtronic unrelated to this study. Authors declare no conflict of interest related to current publications.

References

1. Jennes I, Pedrini E, Zuntini M, Mordenti M, Balkassmi S, Asteggiano CG, Casey B, Bakker B, Sangiorgi L, Wuyts W (2009) Multiple osteochondromas: mutation update and description of the multiple osteochondromas mutation database (MOdb). Hum Mutat 30:1620–1627. https://doi.org/10.1002/humu.21123
2. Albrecht S, Crutchfield JS, SeGall GK (1992) On spinal osteochondromas. J Neurosurg 77:247–252. https://doi.org/10.3171/jns.1992.77.2.0247
3. Fei L, Ngoh C, Porter DE (2018) Chondrosarcoma transformation in hereditary multiple exostoses: a systematic review and clinical and cost-effectiveness of a proposed screening model. J Bone Oncol 13:114–122. https://doi.org/10.1016/j.jbo.2018.09.011
4. Schmale GA, Conrad EU, Raskind WH (1994) The natural history of hereditary multiple exostoses. J Bone Joint Surg Am 76:986–992. https://doi.org/10.2106/00004623-199407000-00005
5. Clement ND, Porter DE (2014) Can deformity of the knee and longitudinal growth of the leg be predicted in patients with hereditary multiple exostoses? A cross-sectional study. Knee 21:299–303. https://doi.org/10.1016/j.knee.2012.10.029
6. Ohtori S, Yamagata M, Hanaoka E, Suzuki H, Takahashi K, Sameda H, Moriya H (2003) Osteochondroma in the lumbar spinal canal causing sciatic pain: report of two cases. J Orthop Sci 8:112–115. https://doi.org/10.1007/s007760300019
7. Roach JW, Klatt JW, Faulkner ND (2009) Involvement of the spine in patients with multiple hereditary exostoses. J Bone Joint Surg Am 91:1942–1948. https://doi.org/10.2106/JBJS.H.00762
8. Fowler J, Takayanagi A, Fiani B, Cathel A, Sarhadi KJ, Arshad M, Lau S, Siddiqi I, Ghanchi H, Wolberg A, Hariri O (2021) Diagnosis, management, and Treatment options: a cervical spine Osteochondroma Meta-Analysis. World Neurosurg 149:215–225e216. https://doi.org/10.1016/j.wneu.2021.01.148
9. Aldea S, Bonneville F, Poirier J, Chiras J, George B, Carpentier A (2006) Acute spinal cord compression in hereditary multiple exostoses. Acta Neurochir (Wien) 148:195–198 discussion 198. https://doi.org/10.1007/s00701-005-0680-6
10. Porter DE, Lonie L, Fraser M, Dobson-Stone C, Porter JR, Monaco AP, Simpson AH (2004) Severity of disease and risk of malignant change in hereditary multiple exostoses. A genotype-phenotype study. J Bone Joint Surg Br 86:1041–1046. https://doi.org/10.1302/0301-620x.86b7.14815
11. Mesfin A, Ghermandi R, Castiello E, Donati DM, Boriani S (2013) Secondary chondrosarcoma of the lumbar spine in hereditary multiple exostoses. Spine J 13:1158–1159. https://doi.org/10.1016/j.spinee.2013.03.056
12. Strovski E, Ali R, Graeb DA, Munk PL, Chang SD (2012) Malignant degeneration of a lumbar osteochondroma into a chondrosarcoma which mimicked a large retropertioneal mass. Skeletal Radiol 41:1319–1322. https://doi.org/10.1007/s00256-012-1405-6
13. Rymarczuk GN, Dirks MS, Whittaker DR, Neal CJ (2015) Symptomatic lumbar osteochondroma treated via a multidisciplinary military surgical team: case report and review of the literature. Mil Med 180:e129–133. https://doi.org/10.7205/MILMED-D-14-00239
14. Zhang Y, Wen L, Zhang J, Yan G, Zhou Y, Huang B (2017) Three-dimensional printing and computer navigation assisted hemipelvectomy for en bloc resection of osteochondroma: a case report. Med (Baltim) 96:e6414. https://doi.org/10.1097/MD.0000000000006414
15. Fox O, Kanawati A (2021) 3D printed composite model of pelvic osteochondroma and nerve roots. 3D Print Med 7:31. https://doi.org/10.1186/s41205-021-00121-9