Three-dimensional printing and neuroendovascular simulation for the treatment of a pediatric intracranial aneurysm: case report

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The use of simulators has been described in a variety of fields as a training tool to gain technical skills through repeating and rehearsing procedures in a safe environment. In cerebrovascular surgery, simulation of skull base approaches has been used for decades. The use of simulation in neurointervention to acquire and enhance skills before treating a patient is a newer concept, but its utilization has been limited due to the lack of good models and deficient haptics. The advent of 3D printing technology and the development of new training models has changed this landscape. The prevalence of aneurysms in the pediatric population is much lower than in adults, and concepts and tools sometimes have to be adapted from one population to another. Neuroendovascular rehearsal is a valid strategy for the treatment of complex aneurysms, especially for the pediatric population. The authors present the case of an 8-year-old boy with a fusiform intracranial aneurysm and documented progressive growth, who was successfully treated after the authors rehearsed the placement of a flow diverter using a patient-specific 3D-printed replicator system model.

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FOR many years simulation has been used in a variety of fields, from aviation to general surgery, as a training tool to gain technical skills through repetition while also rehearsing procedures in a safe environment.^{12,17} The use of simulation in neurointervention to acquire and enhance skills before treating a patient is more recent, but the lack of good models and deficient haptics have limited its utilization. The advent of 3D printing technology and the development of new training models have modified this landscape. The use of simulation in neuroendovascular training has been shown to result in shorter fluoroscopy times, better acquisition of roadmaps, and useful angiographic projections.¹¹

The prevalence of aneurysms in the pediatric population is much lower than in adults, and concepts and tools sometimes have to be adapted from one population to another. Simulation technologies allow neurointerventionalists to practice a procedure in a safe environment before treating a more vulnerable pediatric patient, especially in the setting of a complex vascular lesion.⁴ Additionally, the

simulation allows the operator to better select tools for the procedure, possibly reducing the radiation dose (which is even more critical in pediatric patients) as well as detecting pitfalls for the selected devices in a risk-free environment. In this paper we present the case of a pediatric patient with a dissecting intracranial aneurysm (IA) who was treated after we rehearsed the placement of a flow diverter using a patient-specific 3D-printed replicator system model.

Case Report

History and Examination

An 8-year-old boy, with a history of treatment for a dissecting posterior wall aneurysm in the right internal carotid artery (ICA) using coil embolization and flow diversion, over 3 years of follow-up developed a fusiform aneurysm of the supraclinoid segment of the left ICA with a saccular component and documented growth on serial imaging. The patient was initially going to be treated with off-label use of a Pipeline embolization device (ev3, Covidien), but

ABBREVIATIONS IA = intracranial aneurysm; ICA = internal carotid artery; MCA = middle cerebral artery.

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FIG. 1. A: Patient-specific replicator system model of the patient's cerebral vasculature set up in the angiography suite. **B and C:** Angiographic oblique images of the silicone vessels after injecting contrast, depicting the fusiform aneurysm with a saccular component in the ICA. **D and E:** A Leo+ stent was deployed from the left MCA to the proximal segment of the ICA, and later, a SILK flow diverter was telescoped. The final construct did not properly expand (arrowhead) due to mismatch of the MCA-ICA and the sharp curvature of the MCA. **F:** 3D-printed silicone vessel demonstrating the sidewall aneurysm of the ICA (arrow) and the final construct kinking in the proximal segment of the MCA (arrowhead). Figure is available in color online only.

the procedure was aborted due to the increase in size of the parent vessel, measuring 6 mm at the largest diameter. After considering numerous treatment options, it was decided that the parent vessel was too large for any flow diverter available in the US, and that clip placement was not ideal due to the likely dissecting nature of the lesion. Therefore, we believed the patient would benefit from the SILK flow diverter device (Balt Extrusion). We received US FDA and local IRB approvals for a compassionate use exemption for the off-label use of the SILK flow diverter.

Three-Dimensional Printing and Simulation

Using source imaging from the most recent cerebral angiogram, the target lesion and target vessel were 3D printed using a replicator system model (Vascular Simulations) of the patient's cerebral vasculature (Fig. 1). Two strategies were rehearsed and compared using the replicator model: 1) a standalone construct with the SILK device extending from the ICA terminus to the ophthalmic segment, versus 2) a dual-device construct using the Leo+ stent (Balt Extrusion) and the SILK flow diverter. The Leo+ was deployed from the left proximal middle cerebral artery (MCA) to the ICA, and the SILK device was solely telescoped into the stented segment of the left ICA to cover the saccular component of the aneurysm. On careful inspection of the dual-device construct (Leo+ and SILK), we noticed a lack of ideal expansion of the Leo+ device in the ICA terminus due to the sharp curvature and diameter mismatch of that segment to the more proximal carotid artery (Fig. 1D–F). Therefore, the patient was treated with the strategy developed during the simulation using the SILK flow diverter only, beginning proximal to the tortuous segment.

Operative Course

The patient was previously placed on dual antiplatelet therapy (81 mg of aspirin daily and 1 mg/kg of clopidogrel

daily) and VerifyNow (Accriva Diagnostics) therapeutic levels were obtained prior to the procedure. A right femoral artery access was obtained with a 6-Fr Glidesheath Slender catheter (Terumo Interventional Systems) and the patient was given a single 50 U/kg dose of intravenous heparin intraoperatively. The initial angiogram and a 3D reconstruction demonstrated the sidewall aneurysm in the left supraclinoid ICA (Fig. 2A–D). A Neuron 0.70-inch guide catheter (Penumbra) was placed into the left ICA. Subsequently, a Vasco+ 25 microcatheter (Balt Extrusion) was advanced over a microwire to the target vessel and navigated past the left ICA aneurysm, into the distal left MCA. The microwire was removed and a SILK device (5.5 × 25 mm) was advanced into the microcatheter. Due to the kinking observed during the simulator rehearsal, we decided to deploy the SILK device extending from the ICA terminus to the ophthalmic segment. The device was deployed uneventfully and crossed the entirety of the aneurysm neck (Fig. 2E–G). Angioplasty with a Hyperform balloon (7 × 7 mm; Medtronic) was performed to improve the apposition of the SILK device. A cone-beam CT angiogram demonstrated good wall apposition of the device without parent vessel compromise (Fig. 2H and I).

Postoperative Course and Follow-Up

At the 6-month follow-up, the patient was neurologically intact but complaining of recurrent mild headaches with a modified Rankin Scale score of 1. CT angiography demonstrated patency of the left ICA and good position of the flow diverter (Fig. 2J–L). The 6-month imaging follow-up demonstrated aneurysm occlusion and patency of the left ICA (Fig. 2M). The patient will undergo a cerebral angiogram at the 1-year follow-up.

Discussion

Pediatric IAs account for less than 5% of aneurysms

FIG. 2. **A–C:** Cerebral angiogram, anteroposterior (A) and oblique views (B and C), demonstrating the sidewall aneurysm arising from the ICA. **D:** 3D reconstruction depicting the aneurysm in the left ICA and the previously coiled lesion in the contralateral side. Treatment was performed using the SILK device from the ICA terminus downward, covering the aneurysm neck. **E–G:** The SILK device partially deployed (E) and a fluoroscopic image (F) demonstrating the placement of the flow diverter (arrows). A final angiographic run demonstrated patency of the left ICA as well as its branches (G). **H and I:** A cone-beam CT scan was performed demonstrating good wall apposition of the device. **J–L:** At the 3-month follow-up, CT angiography was performed showing a stable location of the SILK device, no evidence of the aneurysm, and patency of the left ICA and its branches. **M:** At the 6-month imaging follow-up, MR angiography demonstrated complete occlusion of the left ICA aneurysm.

found in the general population.¹ These lesions differ from adult aneurysms in location and size, but overall, they tend to be more complex in morphology (giant, wide neck, dissecting).²⁰ Treatment modalities used for adult aneurysms can be applied to the pediatric population but special considerations have to be made, in particular the size of endovascular devices and longer life expectancy with a latent risk of recurrence or de novo IA formation.

Rehearsing the intervention can decrease procedural time, radiation dose, and possible complications. In our case, the use of a simulator allowed us to gain a comfort level using a novel device, on an exact replica of our patient's aneurysm, in a risk-free environment before performing the procedure. The use of the replicator also allowed us to detect performance issues with one of the devices due to an unanticipated tortuosity and size mismatch that was not appreciated on careful preoperative image analysis.

Vascular simulation has been shown to improve the skills of not only novice neurointerventionalists but also experienced operators and fellows. However, there are several types of simulators to choose from, each with its own advantages and disadvantages (Table 1). The virtual reality models allow one to rehearse a large variety of scenarios as well as the ability to upload a patient's radiographic information to create a patient-specific scenario. Neverthe-

less, they can be expensive and have been noted to lack accurate haptic feedback.⁵ Human cadavers provide accurate anatomy, and when combined with pump-driven flow models, this model provides useful simulation of the conditions of live surgery for clipping aneurysms and management of complications such as intraoperative rupture.¹⁶ Another option is to use ex-vivo models such as a human placenta, in which aneurysms can be developed, multiple surgical scenarios simulated, and complication management practiced.^{15,18} The benefits of using human placenta include low cost, ready accessibility, and most importantly, realistic haptic feedback.³ Nonetheless, ex-vivo models are not able to simulate specific patient vasculature. Pump-driven flow models with silicone vessels, like the one we used, are advantageous in that they allow direct visualization of the wire, catheter, and devices as well as the ability to use patient-specific vasculature.⁴ The drawbacks include limited haptic feedback and the fixed vascular anatomy of this model. Although surgical simulation is not novel and is greatly accepted for training residents or improving specific surgical skills, only a few studies have been reported that involved rehearsing a patient's cerebrovascular lesion operation before the actual surgery (Table 2). In fact, the majority of these studies have compared the preplanned procedure to the actual surgery for aneurysm clip-

TABLE 1. Summary of types of simulations available for cerebrovascular surgery

Simulation	Advantages	Disadvantages
Virtual reality	Simulation of replicated patient-specific lesions Durability Multiple models & metrics Minimal risks	Low to intermediate fidelity Low to intermediate haptic & force feedback Maintenance
3D printed vessel	Simulation of replicated patient-specific lesions Minimal risks Acceptable haptics for endovascular simulation	Cost (depending on material) Durability (one to a few times depending on material) Maintenance Time of fabrication Different elasticity response compared to real arteries
Human cadaver	Human tissue Approximate reality High fidelity (dissection, bypass) Minimal risks	High cost Unable to reproduce patient-specific lesions for preop simulation Maintenance Single use
Live animals	Minimal risks High fidelity (hemostasis, dissection, bypass)	High cost Unable to reproduce patient-specific lesions for preop simulation Maintenance Single use
Ex-vivo (e.g., human placenta)	Low cost Minimal risks High fidelity (hemostasis, dissection, clipping, bypass, endovascular) Multiple uses	Unable to reproduce patient-specific lesions for preop simulation

TABLE 2. Summary of studies reporting surgical outcomes following preoperative simulation for aneurysm treatment

Authors & Year	Study Design	Simulation Model	Type of Intervention	No. of Aneurysms	Outcomes/Comments
Duckworth et al., 2015	Case report	3D printed vessel/pump-driven flow	Flow diverter deployment	1	Use of new technology; improved technical approach for the real intervention
Ishibashi et al., 2016	Case series	3D-printed vessel	Coiling/microcatheter navigation	27	Preplanned microcatheter shaping was good in terms of accessibility, positioning, & stability; no specific data on patient outcomes
Kimura et al., 2009	Case series	3D-printed hollow elastic model	Surgical clipping	8	4 of 8 interventions performed w/ same preop strategy; residual neck in 2 patients at last follow-up
Kono et al., 2013	Case report	3D-printed vessel	Balloon-assisted coiling	1	Improvement in microcatheter navigation w/ the simulation; little discrepancy in strategy; successful treatment of the aneurysms
Marinho et al., 2014	Case series	Virtual reality	Surgical clipping	10	All aneurysms occluded; same surgical strategy in 8 of 10 aneurysms; 1 patient had postop seizures
Mashiko et al., 2015	Case series	3D-printed hollow elastic model	Surgical clipping	20	Better understanding of the vessel anatomy; 12 of 20 replicated aneurysms were clipped; 3 were endovascularly managed; 4 were not treated & 1 patient died before surgery; unexpected findings: hard wall or tissue adhesion; preplanned clipping strategy varied minimally to actual intervention
Namba et al., 2015	Case series	3D-printed vessel	Coiling/microcatheter navigation	10	Successful microcatheter navigation as the preplanned strategy; 1 complication, vessel injury
Shono et al., 2018	Case series	Virtual reality	Surgical clipping	8	5 of 8 procedures were performed as the preplanned strategy; challenges: shorter parent vessel, calcification, bleb, thrombosed lesion
Wang et al., 2017	Case series	3D-printed cranio-vascular model	Surgical clipping	6	Little discrepancy on the preplanned surgical strategy; improved understanding of the surgical view
Wang et al., 2018	Case series	3D-printed vessel	Surgical clipping	8	Similar strategy performed in the actual surgery; strategy (e.g., more clips) usually changed because of hard wall or wide-necked lesion

ping^{7,9,10,19,21,22} and endovascular management; these studies have mainly assessed microcatheter vascular access or endosaccular embolization.^{6,8,13} In general, the results of the above studies suggest that the preplanned strategy is usually performed in the actual procedure and the benefits are perceived by the operator, however, there is no consensus on how to objectively measure outcomes after simulation and there is no prediction of the long-term outcome for the patient. Also, in some instances such as aneurysm rupture, it is futile to consider performing a surgical simulation.

To date, there is no standard of treatment for pediatric aneurysms and flow diversion has been demonstrated to be a safe and effective alternative.^{2,14} However, these patients require close monitoring because they are placed on dual-antiplatelet therapy and are exposed to potential delayed events that can occur after using a flow diverter well described in the adult population.²³ In our case, the decision to treat the aneurysm was made based on progression of the lesion and the use of a flow diverter was considered the most appropriate choice, given the lack of good microsurgical and endovascular options.

Three-dimensional printing and neuroendovascular simulation should be considered as strategies for the training of new operators and the implementation of new technologies and endovascular tools. This can also be used in select complex cases in which patient-specific models can decrease procedural times, number of devices used, and potential complications. The complex and rare nature of some pediatric aneurysms gives reason to consider utilizing this strategy before undertaking such a procedure. Thus, multiple approaches and various devices can not only be considered, but also tested before having the patient treated. Admittedly, the major drawback of simulation is the cost, both from the initial investment for the simulator but also for each individual model, which must be custom-made for each patient. Development of more cost-effective 3D printing and simulation devices are needed. With a plethora of new devices and the increased number of operators decreasing the experience for endovascular treatment of IAs, the use of simulation will likely help to achieve and maintain surgical skills and possibly reduce avoidable complications. Patient-specific 3D modeling, treatment simulation, and rehearsal are valid strategies for preoperative planning in the treatment of complex aneurysms, especially in the pediatric population.

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Disclosures

Ricardo A. Hanel is a consultant for Medtronic, Cerenovus, Elum, Three Rivers Medical, Stryker, Codman, and MicroVention; owns

stock in Elum, Three Rivers, Cerebrotech, EndoStream, Synchron, Blockade, Neurvana, and InNeuroCo; and serves on the scientific advisory board of Medina Medical.

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