Case Report: Free Latissimus Dorsi Flap in Combination With Subdural Space Reduction for the Prevention of Recurrent Hemorrhage Following Hemispherectomy

**BACKGROUND AND IMPORTANCE:** Extensive cerebral resections for the treatment of epilepsy may result in a large intracranial dead space that is prone to recurrent hemorrhage, either due to mechanical dislodgement or the development of extensive subdural membranes. Several techniques have been proposed to decrease the risk of hemorrhage by either reducing or filling the remaining intracranial dead space.

**CLINICAL PRESENTATION:** We present a case of persistent hemorrhage following functional hemispherectomy in a patient with a large porencephalic cyst. A treatment strategy involving both subdural space reduction and cranial vault filling with a vascularized free latissimus dorsi flap is discussed.

**CONCLUSION:** Subdural space reduction and cranial vault filling with a vascularized free latissimus dorsi flap is a viable treatment alternative in patients with large areas of intracranial dead space.

**KEY WORDS:** Craniocerebral Disproportion, Dead Space, Epilepsy, Free Flap, Latissimus Dorsi, Hemispherectomy, Subdural Space Reduction

Operative Neurosurgery 0:1–3, 2017

arge intracranial dead space prone to complications, including hydrocephalus, superficial hemosiderosis,<sup>1</sup> and recurrent hemorrhage, is classically described following anatomic hemispherectomy,<sup>2</sup> which has mostly been abandoned in favor of functional hemispherectomy.<sup>1</sup> However, these complications continue to be seen in any procedure that creates craniocerebral disproportion. Perhaps more relevant to modern neurosurgical practice and epilepsy surgery, we present a patient with a large porencephalic cyst who developed persistent intracranial hemorrhage following functional hemispherotomy. Explicit written consent was obtained from the patient for the preparation and publication of this report.

# **CLINICAL PRESENTATION**

At the age of 5, the here-presented male patient sustained a severe head injury secondary

ABBREVIATIONS: CSF, cerebrospinal fluid; VP, ventriculoperitoneal

to a motor vehicle collision necessitating emergent right decompressive hemicraniectomy with evacuation of an acute subdural hematoma. An enlarging porencephalic cyst and lateral ventricle required shunt insertion 2 yr later. His injuries left him with left spastic hemiplegia and hemianopia as well as impulsive behavior. Several months after the head trauma, he developed frequent staring episodes, which progressed to episodes of sensations of "tightening" of the left arm and then spreading to the leg. He would then lose awareness and demonstrate head and eye deviation to the left. Secondary generalization of these seizures could be controlled with antiepileptic medication; however, he continued to have daily focal nonmotor seizures with loss of awareness.

DOI: 10.1093/ons/opx159

At the age of 20, he underwent a functional hemispherectomy after which he became seizure free and showed improved motor function of his left arm and cognition overall. However, the procedure was complicated by cerebrospinal fluid (CSF) rhinorrhea, which was treated with surgical repair and ventriculoperitoneal (VP) shunt insertion. Over the next 17 yr, he had

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**Received,** February 21, 2017. **Accepted,** June 26, 2017.

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numerous shunt obstructions. CSF sampling consistently demonstrated xanthochromia with elevated protein. At the age of 37, he developed recurrent intracranial hemorrhages into the resection cavity (Figure). Several smaller hemorrhages were managed conservatively; however, on 2 occasions, he presented in coma and underwent frontotemporal craniotomy for evacuation after a vascular abnormality was ruled out by cerebral angiography. Despite these surgical interventions, he continued to experience shunt dysfunction secondary to recurrent hemorrhage. Three attempts to fenestrate the ventricular system to obviate the need for a shunt were made, but failed. Six weeks following his last shunt reinsertion, he developed an acute epidural hematoma. Thus, a more definitive treatment was needed. We hypothesized that the repeated hemorrhaging was related to the intracranial dead space, and thus an operation in a combined effort with neurosurgery and plastic surgery was conceived to eliminate this dead space with the shunt left in place.

The previous frontotemporal craniotomy was reopened and the multiple previous duraplasties were removed revealing a gliotic membrane lining the resection cavity and porencephalic cyst. The membrane and remaining parenchymal tissue were coagulated and lined with surgicel. A fascia lata graft was sewn to the edges of the remaining dura and laid down in the resection cavity and sutured to the falx and basal dura, thereby converting the large subdural space to extradural space. The latissimus dorsi muscle was harvested and a flap performed as described by Buncke.<sup>3</sup> Saphenous vein grafts were used to extend the flap pedicle (thoracodorsal artery and vein). The vein grafts were tunneled beneath the cheek skin anterior to the ear and into the neck where a separate incision had been made to expose the recipient vessels. An end-to-side anastomosis was then performed from the arterial pedicle to the external carotid artery. The venous limb was anastomosed end-to-end to the posterior facial vein. After placement of the bulk of the muscle into the extradural space (Figure), it was sutured to the dermis around the circumference of the defect. The bone flap was not replaced to not compromise the blood flow

to the graft. As scalp closure proved to be tight, a split thickness skin graft was performed to compensate for the scalp defect. The patient was not interested in cranioplasty to improve cosmesis. At the 7-yr follow-up, he remained stable at his neurological and cognitive baseline level without any further episodes of shunt obstruction or intracranial hemorrhage.

#### DISCUSSION

The current case report demonstrates the feasibility of a free latissimus dorsi flap in combination with subdural space reduction to fill an intracranial defect and prevent recurrent hemorrhages following resective surgery for epilepsy.

In the past, several mechanisms have been suggested to explain recurrent hemorrhage in the setting of iatrogenic craniocerebral disproportion following hemispherectomy. Hereby, it is important to differentiate craniocerebral disproportion in terms of extra-axial bleeding in hemispherectomized patients from shunt-induced craniocerebral disproportion in the pediatric population. A good review on the pathophysiology and the therapeutic strategies of the latter is available from Sandler et al.<sup>4</sup>

In 1966, Oppenheimer and Griffith<sup>2</sup> published a series of 3 post-hemispherectomy patients who developed recurrent hemorrhage and delayed deterioration. At autopsy, a thick membrane resembling a chronic subdural hematoma membrane was found both below the dura and lining the remaining brain and ventricular system. Recurrent bleeding from this membrane, either spontaneous or with minor trauma, was thought to result in obstructive hydrocephalus and superficial hemosiderosis.<sup>2</sup>

Besides meticulous hemostasis to prevent membrane development,<sup>5</sup> several technical nuances to prevent this complication were suggested. In 1970, Falconer and Wilson<sup>6</sup> published a case in which they performed extensive removal of the subdural component of the membrane, resulting in improvement of the patient's symptoms for many months. These observations prompted Rasmussen<sup>7</sup> to change their practice from an anatomic complete hemispherectomy to a functional complete but anatomic subtotal hemispherectomy, which eliminated later cerebral hemosiderosis as well as the development of hydrocephalus.

Adams<sup>5</sup> proposed a modification for anatomic hemispherectomy. A reduction of the subdural space by suturing the dura down to the falx, tentorium, and the anterior and middle cranial fossa dura was advocated with the rationale to improve the splinting of the remaining brain by converting the subdural cavity to an extradural dead space. In 1985, Sorano et al<sup>8</sup> made use of a Silastic breast implant covered by a metallic mesh to fill the epidural space of a hemispherectomy patient; a 28-yr follow-up has since been published, demonstrating the integrity of the implant. In 1993, Matheson et al<sup>9</sup> published a case series of 4 patients with large resections for epilepsy undergoing preventative surgery to minimize the risk of postoperative hemorrhagic complications. Using Adams' technique, the authors and others<sup>10</sup> used omentum free flap grafts to fill the extradural space.

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Latissimus dorsi flaps are used in reconstructive plastic surgery for repair of large soft-tissue defects.<sup>11,12</sup> Levine and colleagues<sup>11</sup> presented a series of 6 patients with large intracranial dead space following postoperative infections with loss of calvarial bone. The authors used vascularized flaps of the latissimus dorsi muscle, rectus abdominis muscle or omentum to fill in the dead space and restored the vault shape with titanium meshes or Medpor implants. The vascularized tissue not only reduced intracranial dead space, but also provided a delivery system for immune cells and antibiotics to help with challenging postoperative infection.<sup>11</sup>

To the best of our knowledge, this is the first description of a free latissimus dorsi flap with Adams-type subdural space reduction to compensate for extradural space following resective surgery for epilepsy. This combined technique adds to the restorative surgical armamentarium discussed above as it proved efficient to prevent recurrent intracranial hemorrhages. Unlike in most reported cases,<sup>5,9,11-13</sup> because of our patient's preference, his cranial vault defect was not reconstructed. The "mechanical behavior of the brain and CSF" and the concept of "subdural stress" that is reduced by shunt implantation was already recognized by Hakim.<sup>13</sup>

#### Limitations

In this context and as a limitation of our case report, it should be pointed out that the lack of a closed calvarium might represent a possible confounder. The open cranial vault exposes the shunted space to atmospheric pressure, and it is a possibility that this alone may obviate recurrent hemorrhages as opposed to low-pressure compartments like in a shunted closed calvarium. However, the degree to which this effect is applicable in a situation where muscle and considerably scarred scalp fill the extradural space is unknown.

## CONCLUSION

Our case demonstrates a latissimus dorsi free flap in combination with an Adams-type subdural space reduction can safely be used to fill an extradural space to splint the remaining parenchymal and reduce intracranial dead space. Long-term follow-up has demonstrated successful prevention of both recurrent intracranial hemorrhage and shunt dysfunction.

#### Disclosure

The authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices described in this article.

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# COMMENTS

The authors present a novel technique to deal with a very complex problem. The patient suffered recurrent life-threatening intracerebral hemorrhages after a hemispherectomy for intractable epilepsy. An Adams-type subdural space reduction was followed by a latissimus free flap. This has, over a 7-year follow-up, eliminated the patient's repeated hemorrhages. This is a useful technique to have available for a very complex, albeit rare, problem.

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he authors provide a well-written, useful technical report using a latissimus free flap to obliterate the subdural space to treat superficial hemosiderosis following functional hemispherectomy. Superficial hemosiderosis is a serious complication originally associated with anatomic hemispherectomy. The repeated and chronic deposition of iron in the cerebral cortex causes neurologic and cognitive dysfunction, and may eventually lead to death. This complication led Dr Theodore Rasmussen at the Montreal Neurological Institute to refine hemispherectomy to less brain tissue resected (and less dead space created) - functional hemispherectomy - focusing on the disconnection of white matter tracts. Contemporary epilepsy surgeons may now perform functional hemispherotomy where virtually no brain tissue is resected; the key to the procedure is disruption of white matter tracts to stop the propagation of seizures. Hence, superficial hemosiderosis is an uncommon complication after functional hemi-spheretomy/spherotomy, where craniocerebral disproportion is less likely encountered. The authors' case was likely made more complicated by the patient's history of porencephaly and cerebral atrophy prior to functional hemispherectomy.

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