Preoperative assessment of dominant occipital sinus in patients with Chiari malformation type I: anatomical variations and implications for preventing potentially life-threatening surgical complications

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OBJECTIVE The surgical treatment of Chiari malformation type I (CM-I) frequently involves dural incision at the posterior cranial fossa. In cases of persistent patent occipital sinus (OS), the sinus is usually obliterated and divided. However, there are some patients whose OS is prominent and requires crucial modification of the operative planning to avoid potentially life-threatening massive hemorrhage and disturbance of cerebral venous circulation. In the present study, the anatomical variations of the dominant OS in patients with CM-I were analyzed and the authors attempted to develop treatment recommendations for patients with CM-I with dominant OS.

METHODS The study included 213 patients with CM-I who underwent MR venography (MRV) prior to surgical treatment. OS dominance was assessed using 2D time-of-flight MRV or 3D phase-contrast MRV. Particular attention was paid to the pattern of venous outflow channels. The characteristics of the patients with dominant OS and the surgical outcomes were retrospectively reviewed.

RESULTS Dominant OS was identified in 7 patients (3.3%). The age in those with dominant OS was significantly younger than in those without (p = 0.0202). The incidence of concurrent scoliosis in the patients with dominant OS was significantly higher than in those without (p = 0.0366). All the dominant OSs were found to be of the oblique type. Unilateral oblique OS (OOS) with normal ipsilateral transverse sinus (TS) and hypoplastic contralateral TS was found in 2 patients (0.9%). The authors found 1 patient each (0.5%) who had unilateral OOS with hypoplastic ipsilateral TS and normal contralateral TS, unilateral OOS with bilateral hypoplastic TSs, and bilateral OOSs with bilateral normal TSs. Bilateral OOSs with bilateral hypoplastic TSs were found in 2 patients (0.9%). All these patients had syringomyelia. Instead of performing Y-shaped dural incision and duraplasty, surgical procedures were modified depending on the types of the OOSs to preserve their venous drainage routes. Although massive bleeding from the dominant OS during dural incision occurred in 1 patient, none suffered neurological deterioration. The syrinx volume decreased in all but 1 of the patients postoperatively.

CONCLUSIONS Assessment of the venous drainage pattern using MRV is indispensable for safe surgical treatment in patients with CM-I. The surgical procedure should be modified based on the type of dominant OS to minimize the surgical risks.

https://thejns.org/doi/abs/10.3171/2022.5.JNS212973

KEYWORDS occipital sinus; Chiari malformation type I; foramen magnum decompression; duraplasty
Chiari malformation type I (CM-I) is an anomaly due to a cranioccephalic disproportion in the posterior fossa resulting from occipital bone hypoplasia. The compression at the craniovertebral junction causes numerous symptoms that can be divided into those related to CSF circulation and those related to brainstem compression or distension. The most common symptoms are cough- or strain-related headaches. Importantly, CM-I could also affect the spinal subarachnoid space (SAS) pressure wave that is largely created by arterial pulsations in the brain. CSF flows into the spinal cord from the spinal SAS through perivascular spaces and causes the syrinx formation, and that is probably influenced by the relative timing of the arterial pulse and pressure waves in the spinal SAS. These waves can be altered significantly due to narrowing of the CSF spaces around the foramen magnum. This alteration is currently considered to cause the syrinx formation in the spinal cord.

Among various surgical methods applied for the treatment of CM-I, foramen magnum decompression with duraplasty is thought to be the most effective procedure. Dural incision of the posterior fossa is inevitable for sufficiently relaxed duraplasty, so knowing the type and course of occipital sinus (OS) to be dealt with prior to surgery is very important for avoiding devastating complications such as massive hemorrhage and disturbance of the venous outflow of the brain. In anatomical studies, some OSs were classified as dominant and draining toward the sigmoid sinus. Usually, the marginal sinus is a draining pathway from the OS toward the basilar venous plexus and the hypoglossal canal venous space. In 2008, Tubbs et al. reported an unusually large OS that instead of running along midline was turning laterally and draining into the sigmoid sinus. Hypoplasia evaluation of the TS was based on a similar way, the relationship of surgical management were investigated too.

Methods
This study was a single-institution retrospective study of 213 patients with CM-I whose preoperative cervical MR images were available for measurement of tonsillar descent and who also underwent MR venography (MRV) prior to surgical treatment between October 2011 and March 2021. The study was approved by the ethics board of The Jikei University Hospital, and written informed consent was waived because of the retrospective design and anonymized data. The subjects consisted of 169 female and 44 male patients within an age range from 4 to 70 years (mean 30.38 years).

CM-I with dominant OS was identified using the 2D time-of-flight (TOF) MR angiography technique with an inferior saturation band to eliminate signal from arterial structures in 33 patients and 3D phase-contrast venography in 180 patients. We systematically reviewed the venograms in each case for the presence or absence of a dominant OS. When the largest caliber of the OS was more than half the diameter of the superior sagittal sinus (SSS) (measured 1 cm above the confluence of sinuses), the OS was defined as dominant. In a similar way, the relationship between the dominant OS and a hypoplastic TS was also evaluated. Hypoplasia evaluation of the TS was based on a study by Widjaja and Griffiths. The largest caliber of the TS was compared with the largest size of the SSS (measured 1 cm above the confluence of sinuses). If the caliber of the TS was less than half the diameter of the SSS, it was defined as hypoplastic.

We also assessed the following parameters on preoperative cervical MRI: tonsillar descent from McRae line, existence of syrinx, clivoaxial angle, basilar invagination, existence of scoliosis, and C1 anomaly as concurrent malformations and as charts for coexistence of genetic syndromes. Statistical analysis comparing the age and sex with the abovementioned imaging findings of those with dominant OS and those without was conducted using the Student t-test for continuous variables and Fisher exact test for categorical variables. A p value < 0.05 was used as a threshold for significance. MATLAB software (Math-Works Inc.) was used for all data analysis. The characteristics of the patients with dominant OS and appropriateness of surgical management were investigated too.

Results
As shown in Table 1, dominant OS was identified in 7 (3.3%) of the 213 patients (2 and 5 patients of 33 on 2D-TOF and 180 on 3D phase contrast, respectively). The mean ages of the patients with dominant OS and those without it were 17.29 and 30.82 years, respectively (Table 1). The difference in age was statistically significant (p = 0.0202). There was no significant difference between sexes for the incidence (p = 0.6357; Table 1).

A total of 187 patients (87.78%) with syringomyelia were observed from the entire cohort of 213 patients, including all the patients with dominant OS, who also had syrinx (Table 1). However, the incidence of syringomyelia between the patients with dominant OS and those without it was not significantly different (p = 0.6014; Table 1). The mean tonsillar descent and clivoaxial angle of the patients with dominant OS and those without it were 9.93 mm versus 11.74 mm and 141.29° versus 143.37°, respectively (Table 1). There was no significant difference in tonsillar descent (p = 0.4224) and clivoaxial angle (p = 0.6477) between the 2 groups (Table 1). Concurrent malformations of basilar invagination, scoliosis, and C1 anomaly were identified in 43 (20.2%), 25 (11.7%), and 14 (6.6%) of 213 patients, respectively (Table 1). Among these concurrent malformations, the incidence of scoliosis in the patients with dominant OS was significantly higher than in those without dominant OS (p = 0.0366; Table 1). Among the 213 patients, neurofibromatosis type 1 and Klippel-Feil syndrome were found in 1 patient each.

All the dominant OSs were found to be oblique and...
draining into the sigmoid sinus as it passes through the jugular foramen. Based on the study by Shin et al., types of OOSs were recognized, depending on the presence of single or bilateral OOSs and hypoplasia of the TS. The types of OOSs were as follows (Fig. 1): 1) unilateral OOS with normal ipsilateral TS and hypoplastic contralateral TS—type IIBb, 2 patients (0.9%); 2) unilateral OOS with hypoplastic ipsilateral TS and normal contralateral TS—type IIAa, 1 patient (0.5%); 3) unilateral OOS with bilateral hypoplastic TSs—type IIIB, 1 patient (0.5%); 4) bilateral OOSs with bilateral normal TSs—type IC, 1 patient (0.5%); and 5) bilateral OOSs with bilateral hypoplastic TSs—type IIIC, 2 patients (0.9%).

Table 2 shows the clinical characteristics of the cases of CM-I with dominant OOS. All patients had syringomyelia. Surgical management was planned based on the type of OOS. For the patient with type IIAa OOS, it was decided to place a syrinx—subarachnoid shunt at the cervicothoracic junction. Two patients with unilateral OOS, normal ipsilateral TS, and hypoplastic contralateral TS (type IIBb) underwent foramen magnum decompression with unilateral duraplasty using an expanded polytetrafluoroethylene (ePTFE) sheet. Instead of the usual Y-shaped dural incision and duraplasty, the dura mater was incised in a curvilinear fashion, avoiding the unilateral OOS starting from the level of CI to the dura over the
contralateral cerebellar hemisphere. The other 4 patients ultimately required fourth ventricle–subarachnoid shunt placement. Using the limited dural opening from the level of C1 toward the cervicomedullary junction did not injure the OOS, and the shunt tube was inserted through the foramen of Magendie to the fourth ventricle. Intraoperative massive hemorrhage from OOS occurred in a patient with type IIIC OOS. The syrinx volume decreased in all but 1 of the cases at the mean follow-up of 69.4 months.

**Illustrative Cases**

**Case 1**
A 29-year-old woman presented with dysesthesia in her right upper limb. Her MRI studies showed CM-I associated with syringomyelia (Fig. 2A). Her MRV demonstrated unilateral OOS with normal ipsilateral TS and hypoplastic contralateral TS (type IIBb; Fig. 2B). Considering the venous drainage pattern, we performed foramen magnum decompression, C1 laminectomy, left-sided unilateral duraplasty with an ePTFE sheet, and tonsillar coagulation (Fig. 2C). Postoperative MRI showed significant improvement of syringomyelia (Fig. 2D).

**Case 5**
A 7-year-old boy presented with scoliosis resulting from syringomyelia. His MRI studies showed CM-I with

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**TABLE 2.** CM-I with dominant OS in 7 patients

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (yrs)/Sex</th>
<th>Type of OOS</th>
<th>Surgical Procedure</th>
<th>Surgery-Related Complications</th>
<th>Postop Syringomyelia</th>
<th>FU Period</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>29/F</td>
<td>IIBb</td>
<td>Unilat duraplasty, tonsillar coagulation</td>
<td>None</td>
<td>Decreased</td>
<td>29 mos</td>
</tr>
<tr>
<td>2</td>
<td>41/F</td>
<td>IIBb</td>
<td>Unilat duraplasty</td>
<td>None</td>
<td>Decreased</td>
<td>65 mos</td>
</tr>
<tr>
<td>3</td>
<td>15/F</td>
<td>IIAa</td>
<td>4-S shunt, Sy-Ci shunt</td>
<td>None</td>
<td>Decreased</td>
<td>104 mos</td>
</tr>
<tr>
<td>4</td>
<td>12/F</td>
<td>IIIB</td>
<td>Sy-S shunt at C7/T1</td>
<td>None</td>
<td>Decreased</td>
<td>56 mos</td>
</tr>
<tr>
<td>5</td>
<td>7/M</td>
<td>IC</td>
<td>1st op tonsillactomy; 2nd op Sy-S shunt; 3rd op 4-S shunt</td>
<td>None</td>
<td>Decreased</td>
<td>106 mos (2 mos after 3rd op)</td>
</tr>
<tr>
<td>6</td>
<td>4/F</td>
<td>IIIC</td>
<td>4-S shunt</td>
<td>Intraop massive hemorrhage</td>
<td>Decreased</td>
<td>110 mos</td>
</tr>
<tr>
<td>7</td>
<td>13/M</td>
<td>IIIC</td>
<td>4-S shunt</td>
<td>No change (re-expanded)</td>
<td>No change</td>
<td>16 mos</td>
</tr>
</tbody>
</table>

4-S = fourth ventricle–subarachnoid; FU = follow-up; Sy-Ci = syrinx–cisternal; Sy-S = syrinx–subarachnoid.
caudal descent of the brainstem (Chiari 1.5 malformation) associated with syringomyelia and basilar impression (Fig. 3A). His MRV demonstrated bilateral OOS with bilateral normal TS (type IC; Fig. 3B). Due to the significant amount of venous drainage to the OOSs, the patient underwent tonsillectomy; however, the volume of syrinx after the surgery did not change. Therefore, a syrinx–subarachnoid shunt was placed at a second operation. Although the syrinx was slightly shrunken afterward, we suppose that there was no efficient CSF flow across the craniocervical junction. That required a fourth ventricle–subarachnoid shunt, which proved to be efficient, and the syrinx size decreased (Fig. 3C–E).

**Case 6**

A 4-year-old girl who was diagnosed with neurofibromatosis type 1 presented with scoliosis. Her MRI studies demonstrated CM-I associated with progressive syringomyelia (Fig. 4A). Her MRV demonstrated bilateral OOS with bilateral hypoplastic TS (type IIC; Fig. 4B). She underwent fourth ventricle–subarachnoid shunt instead of Y-shaped duraplasty because the OOS was thought to be an important alternative drainage route (Fig. 4C). During the surgical procedure, massive hemorrhage occurred when the dura was opened and the OOS was partially injured. Hemostasis was obtained with hemoclips; however, the obliteration of the OOS caused significant cerebellar swelling. Therefore, the hemoclips were removed and sinusplasty to preserve the venous drainage was performed without neurological sequelae. Postoperative MRI showed decreased volume of the syrinx (Fig. 4D).

**Discussion**

The goal of surgical treatment for CM-I is to restore adequate circulation of CSF by decompressing the cervicomедullary junction. According to a recent systematic review, bony decompression alone often cannot achieve favorable improvement in clinical and radiological outcomes. Various surgical techniques have been proposed in combination with bony decompression, such as duraplasty, removal of the outer layer of the dura mater, arachnoid opening, resection or shrinkage of the tonsils, shunt placement, and obex area (foramen of Magendie area) exploration. Considering the duraplasty for CM-I, the variation of OS should be considered prior to the procedure because it involves obliteration and division of the OS. If the OS is prominent and functioning as a main drainer, unpredictable massive hemorrhage and disturbance of brain perfusion might occur. These complications would lead to devastating consequences. Other techniques will also require dural manipulations to some extent, so special care should be taken when the OS is dominant.

There are several anatomical studies of the OS in patients without CM-I. Two cadaver studies showed the presence of the OS in 64% and 93% of cases, respectively. In the retrograde jugular venography study of Dora and...
FIG. 3. Case 5. A: Sagittal T1-weighted MR image of the posterior fossa and cervical spine revealing a CM-I with caudal descent of the brainstem (Chiari 1.5 malformation) and basilar impression with syringomyelia. B: MR venogram revealing bilateral OOS (arrows) with bilateral normal TS (type IC). C: Intraoperative photograph showing placement of fourth ventricle–subarachnoid shunt. D: Postoperative sagittal CT image showing the shunt tube placed into the fourth ventricle (arrowhead). E: Postoperative sagittal T2-weighted MR image showing resolution of the Chiari malformation and decrease of the syrinx cavity. Figure is available in color online only.

FIG. 4. Case 6. A: Sagittal T1-weighted MR image of the posterior fossa and cervical spine revealing a CM-I with syringomyelia. B: MR venogram revealing bilateral OOS (arrows) with bilateral hypoplastic TS (type IIIC). C: Postoperative sagittal CT image showing the shunt tube placed into the fourth ventricle (arrowhead). D: Postoperative sagittal T1-weighted MR image showing resolution of the Chiari malformation and decrease of the syrinx cavity.
Zileli, the OS was seen in 92% of the patients. On the contrary, imaging studies using 2D-TOF MRV showed relatively lower incidence rates of OS, at 10% and 13%. The different incidence rates of OS may result from the inability to demonstrate flow within the OS, whose inner diameter is 3.27 mm on average. MRV may be inadequate for depicting the cases of small sinus; however, such a small OS is thought to be nonsignificant in many cases because of its very limited contribution to the venous outflow. Other than the presented 7 cases of the dominant OS, we sometimes encounter some amount of venous bleeding during Y-shaped dural incision; however, no patients experienced brain swelling resulting from venous congestion.

Shin et al. investigated the OOS using bone subtraction 3D CT venography in 1805 patients. They found that it was present in 41 patients (2.3%). They emphasized the necessity of a delicate approach in infratentorial craniotomy to avoid OS injury, especially in type II and type III groups, in which patients have hypoplastic or aplastic TS. This type of prominent OS was reported in several case reports, and the OS made surgery via suboccipital craniotomies problematic because of potentially fatal complications such as massive bleeding and brain swelling.

In this study we encountered 7 cases of CM-I with dominant OS out of 213 cases (3.3%). In all these cases the OS could be considered an OOS. We usually perform foramen magnum decompression with Y-shaped duraplasty using an ePTFE sheet and sutures; however, we had to modify our surgical strategy for these cases because of the potential for massive hemorrhage and devastating sequelae resulting from the division and obliteration of a dominant OS. We considered 2 factors to assign surgical risks of dominant OOS—venous congestion (importance for venous outflow) and hemorrhage. When a dominant OS has a hypoplastic ipsilateral TS, we estimate that the venous outflow through the OS has no appropriate alternative drainage pathway and its occlusion carries a high risk of venous congestion. If dominant OOSs are identified bilaterally, dural incision over the OS could cause massive hemorrhage. Bilateral OOSs have a high risk of intraoperative massive hemorrhage. Basically, we classified the type of venous drainage into 3 types depending on the combination of the risk factors that an incision of the posterior fossa dura carries, which are as follows (Fig. 1).

**Low-Risk Group**

In this group there is low importance for venous outflow and low risk of hemorrhage. The venous drainage consists of unilateral OOS with normal ipsilateral TS—types IA, IB, IIBa, and IIBb.

**Moderate-Risk Group**

In this group there is high importance for venous outflow and low risk of hemorrhage. The venous drainage consists of unilateral OS with hypoplastic ipsilateral TS—types IIa, IIb, IIIa, and IIIb; bilateral OOSs with normal TSs—type IC; or bilateral OOSs with unilateral hypoplastic TS—types IICA and IICb.

**High-Risk Group**

In this group there is high importance for venous outflow and high risk of hemorrhage. The venous drainage consists of bilateral OOSs with hypoplastic bilateral TSs—type IIIC.

**Surgical Options**

In cases of unilateral OOS with normal ipsilateral TS type, dural incision on the contralateral side is thought to be safe because the normal TS could become an alternative drainage pathway even in cases in which the OS is injured. If confirming the absence of venous engorgement after temporary occlusion of the OS, its obliteration of the OS might be possible. Therefore, unilateral duraplasty could be the first option for the low-risk group.

In the unilateral OOS with hypoplastic ipsilateral TS type, duraplasty could be dangerous because there is no ipsilateral alternative pathway when the OOS is injured. In these cases, fourth ventricle–subarachnoid shunt or unilateral duraplasty on the contralateral side could be an option. Fourth ventricle–subarachnoid shunt could be inserted through the limited dural incision, which is from the level of C1 to the caudal side of the OS instead of the usual Y-shaped dural incision. Considering the risk of injuring the OS, a meticulous dural incision should be performed. The bilateral type is also associated with danger. Even in the case of normal bilateral TS, massive hemorrhage can occur with dural incision and if there are no other drainage pathways; as in case 6, ligation and division of the OS can result in catastrophic venous infarction. Dural incision must be limited to preserve the OS and because of that, duraplasty should be avoided in the high-risk group. Posterior fossa bony decompression alone could also be an option as an initial surgical intervention.

Our surgical management was modified depending on the type of the OOS. Although intraoperative massive hemorrhage occurred in case 6, the modification could provide appropriate clinical outcomes without death. There are no universally accepted selection criteria to choose the best procedure; moreover, no single surgical procedure is best suited for all the situations we have presented. Surgeons will do better to individualize surgical management depending on the patient’s situation and based on the anatomical and venous outflow pattern. Considering the possible risks of duraplasty for CM-I, understanding the venous drainage pattern of the posterior fossa is one of the important factors. Therefore, we presented our treatment strategy, which could be considered as recommendations for their management (Fig. 5).

The relationship between CM-I and the development of an OOS is not clear. The incidence of the OOS in our study might be relatively higher than that in a previous study on subjects without CM-I, because we only focused on the dominant OS and omitted small ones. Embryologically, OS derives from the plexiform occipital and marginal sinuses, which are prominent in the 4th to 5th fetal months, and have multiple venous channels on each side connecting the medial part of the TS with either the prominent marginal sinus around the foramen magnum or the superior jugular bulb. Usually these channels regress rapidly after the 6th or 7th fetal month.

There is a study in which
TS stenosis and CM-I are significantly associated and observed in 33% of the cases, and that may reflect increased intracranial pressure. The sinuses are poorly developed until after birth; however, the development of cerebellum greatly influences the change in the volume of the posterior fossa, which is most marked between the ages of 30 and 40 weeks in the fetus. Considering the pathogenesis of CM-I resulting in increased intracranial pressure in the posterior fossa and compression at the foramen magnum, the dominant OOS may be the physiological hemodynamic response as a compensatory venous pathway when the TS is hypoplastic and the foramen magnum is tight. Dominant OOS could be considered also as a persistent fetal morphology. This also could be the reason why the ages of those with dominant OS were significantly younger than the ages of those without. This speculation, however, cannot be applied for some types of OOS when there is normal ipsilateral TS. Although scoliosis could result from syringomyelia, patients with CM-I who have concurrent scoliosis have a higher incidence of dominant OOS. Therefore, these venous anatomical abnormalities might be a part of systemic anomalies of mesoderm, which are often associated with spinal congenital abnormalities.

There are limitations of this study. First, the number of patients with dominant OS is limited. Second, it is difficult to estimate an actual risk of the obliteration of an OOS before and during the surgery, especially in the case of the moderate-risk group. Intraoperative indocyanine green videoangiography could provide some useful information to estimate the venous drainage route and its dominance.
but it might be not sufficient. Therefore, it is impossible to understand the full details of the collateral circulation of the intracranial venous system, and the variants of the confluence flow pattern add additional uncertainty. Although temporary clip occlusion of the OS could be useful for evaluating the effects of obliteration on venous return, there is still a risk of delayed postoperative venous infarction resulting from retrograde venous thrombosis when the main drainage route is injured, even in asymptomatic cases immediately after the operation.\(^{21,30,37}\) Finally, digital subtraction angiography, CT venography, and contrast-enhanced MRV, which are superior to the noncontrast MRV in diagnostic accuracy, were not used for evaluation. However, the MRV techniques we used had several safety advantages: noninvasiveness, contrast agent–free imaging, and avoidance of ionizing radiation.\(^{38}\)

Considering the potentially devastating outcomes after obliterating a dominant OS resulting from Y-shaped duraplasty for CM-I, evaluating the venous drainage pattern prior to surgery is indispensable for safe surgical treatment. Even though the additional evaluation is costly and time-consuming, obtaining MRV prior to surgery is recommended, especially for the young and for patients with concurrent scoliosis.\(^{36}\)

### Conclusions

Patients with CM-I have 3.3% of incidence of dominant OS, with a higher probability in the young and in patients with concurrent scoliosis. Preoperative assessment of the venous drainage pattern is essential for surgical treatment of CM-I because a dominant OS is often the main drainage pathway and its preservation is important. The standard Y-shaped duraplasty might endanger venous outflow in such cases. All dominant OSs were of the oblique type and could be classified into 3 groups depending on the possible risks related to their obliteration. The surgical procedure should be modified based on the type of existing dominant OS pattern.

### References


Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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Conception and design: Tochigi, Isoshima, Abe. Acquisition of data: Tochigi, Isoshima, H Ohashi, Kawamura, Hatano, S Ohashi, Nagashima, Abe. Analysis and interpretation of data: Tochigi. Drafting the article: Tochigi. Critically revising the article: Isoshima, H Ohashi, Karagiozov, Murayama, Abe. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Tochigi. Statistical analysis: Tochigi. Study supervision: Murayama, Abe.

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