Alagille syndrome (AGS) is a pediatric multisystem autosomal dominant disorder caused by mutations in the JAG1 and NOTCH 2 genes. The feature of the syndrome is broad-spectrum including bile duct paucity with cholestasis, characteristic facies, and anomalies in the cardiac, renal, skeletal, ocular system. Various intracranial vascular abnormalities have also been reported. We describe a case of AGS with bilateral large unruptured para-ophthalmic aneurysms, which were successfully treated with endovascular therapy.

**Subject and methods** A 14-year-old girl with genetically diagnosed AGS presented with bifrontal pressure headache. MRI of the brain showed no intracranial hemorrhage. However, MRA revealed bilateral internal carotid artery (ICA) supraclinoind segment aneurysms, which increased in size on the follow-up MRA. A diagnostic cerebral angiogram confirmed large para-ophthalmic aneurysms; 8×9×16 mm with 6 mm neck (right), 7×8×12 mm with 4 mm neck (left), with dysplastic suprachindoid ICAs and M1 segments (figure A). Therefore, a decision was made to treat the aneurysms with endovascular measures.

**Results** The treatment team was assembled with pediatric hepatobiliary and hematology services in concerning her underlying liver dysfunction, age, and anticipation of using intracranial stents or flow diversion devices with dual antiplatelet therapy (DAP). Platelet function assay, P2Y12 platelet function, and liver function tests were carefully monitored pre- and post-procedure to adjust the DAP. The right ICA para-ophthalmic aneurysm was treated with Pipeline Embolization Device and detachable coils successfully. Four weeks after, the left ICA para-ophthalmic aneurysm was also treated successfully with the same maneuver. In each treatment, the patient was discharged to her home on the following day without complication. Clopidogrel was discontinued after six months with the continuation of aspirin 81 mg daily. The bifrontal pressure sensation disappeared, and no new symptom has been reported since the treatment. One year follow up angiogram illustrates complete aneurysm dome occlusion and the flow diversion device patency (figure B).

**Conclusion** To our knowledge, this case is the first report of treated the intracranial aneurysms associated with AGS utilizing flow diversion devices. The growing body of literature describes the intracranial aneurysm and intracranial hemorrhage lead to significant morbidity and mortality in patients AGS. It would be warranted to screen with noninvasive imaging modalities, such as MRA and CTA for the patients with AGS who develop alarming neurovascular symptoms. Further, if indicated, endovascular therapy with multidisciplinary team would be considered in the setting of multisystem disorders such as AGS.

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