A case of atrial septal defect closure relieving refractory migraine

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Abstract

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Case report

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Authors

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Abstract

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Keywords:

atrial septal defect, migraine, patent foramen ovale

Key clinical message

Closure of an atrial septal defect dramatically improved the symptoms of refractory migraine in our case.

Introduction

Migraine is the most common disabling headache disorder, reducing the quality of life in children and adolescents, as well as in adults. Patients with an atrial septal defect (ASD) have a high prevalence of migraine.[1] Few studies have examined the association between migraine and ASD closure.[2] Here we report a case of dramatic improvement in migraine symptoms after the closure of ASD.

Case presentation

A 14-year-old Japanese girl with postural orthostatic tachycardia syndrome (POTS) was referred to our hospital because of persistent headache for five months. She had no medical history other than POTS, and her developmental milestones were normal. Her brother had a history of POTS. The headache was located at the bilateral temporal regions, was aggravated by physical activity, followed by photophobia, and was not accompanied by disturbances in vision, sensation, or speech. The pain and fatigue worsened in the morning. The frequency of pain was at least 15 to 20 days a month, and the duration of pain was 8 to 12 hours. The laboratory data showed no abnormalities, and brain magnetic resonance imaging showed no abnormal findings. The active standing test revealed that the heart rate increase was still >30 beats per minute within 10 minutes without decreasing blood pressure. We diagnosed her headaches as being of the migraine type and also associated with POTS. We initiated treatment with midodrine hydrochloride and propranolol hydrochloride. After six months of treatment, the frequency of headaches was reduced to a few times a month, and the duration improved to less than three hours.

Until eight months later, the frequency of headaches remained almost the same, and her school life was not affected by the headaches, but then the headaches gradually worsened, and she began to have more than 15 episodes of headache per month. Furthermore, she could hardly go to school. In order to reduce the burden, it was decided to transfer her to a school closer to home. Even during that period, she was not using analgesics more than nine times a month.

At 18 years old, four years after the onset of chronic headaches, ASD and right ventricular enlargement were diagnosed after an echocardiogram was performed to investigate transient tachycardia. The size of the ASD was 7.5 mm, and the Qp/Qs ratio was 1.32. She underwent transcatheter closure for ASD with right ventricular enlargement using the Amplatzer® device. On the fifth day after the transcatheter ASD closure, she remarked that she had no headache at all. After returning to school one week after transcatheter ASD closure, the headaches were limited to a few times a month, and they spontaneously resolved within a duration of 30 minutes. However, her heart rate increase was still >30 beats per minute within 10 minutes of the active standing test after the surgery. At the outpatient visit six months later, she still had headaches for about an hour at a time, once or twice a month. She was not late or absent from school.

Discussion

We presented a case of a girl with a history of POTS who had chronic migraine. ASD closure dramatically relieved her prolonged headaches. As the active standing test results were the same before and after ASD closure, we concluded that the improvement in interatrial structure contributed to relief of migraine.

A systematic review of case-control studies on migraine and patent foramen ovale (PFO) has shown an apparent bidirectional association.[1] Another prospective observational study of headache in patients with ASD has confirmed the high prevalence of headache, particularly migraine, in ASD patients.[2]Microemboli and vasoactive chemicals such as serotonin from interatrial communication and transient changes in oxygen concentration have been assumed as causes of migraine.[1, 3] Many observational studies have reported improvement in migraine headaches in 65%-91% of patients after PFO closure.[3] Although three recent randomized controlled trials failed to show the efficacy of PFO closure in reducing more than 50% of migraine headaches, subgroup analyses of migraine with aura showed statistically superior benefits.[4-6] By contrast, very few studies have examined the association between ASD closure and migraine. This may be because ASD is relatively rare (1.2–3.0 cases per 10,000 births) compared to PFO.[7]One study of 25 patients with ASD showed that there was no statistically significant difference in the frequency and duration of headaches.[2] ASD

closure currently does not take precedence over pharmacotherapy in treating migraine. However, our patient clinically responded better to ASD closure than to pharmacotherapy as migraine headache treatment. Our findings indicate that there may be a subgroup of migraine patients who benefit from ASD closure. Further systematic case-control studies will be required to clarify the association between ASD closure and migraine.

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None

Conflict of interest

None declared.

Author contributions

Y.H. and H.I. contributed to the conception and wrote the manuscript; M.K., T.M., and A.N. reviewed the manuscript and supervised the whole study process. All authors reviewed and approved the final manuscript.

Ethical approval

Written informed consent in accordance with the Declaration of Helsinki was obtained from the patient's parents, in addition to consent for publication.

Data availability statement

All data supporting this study's findings are available from the corresponding author upon reasonable request.

References

1. Schwedt TJ, Demaerschalk BM, Dodick DW. 2008. Patent foramen ovale and migraine: a quantitative systematic review. Cephalalgia 28:531-540. https://doi: 10.1111/j.1468-2982.2008.01554.

2. Riederer F, Baumgartner H, Sándor PS, Wessely P, Wöber C. 2011. Headache in 25 consecutive patients with atrial septal defects before and after percutaneous closure–a prospective case series. Headache 51:1297-1304. https://doi:10.1111/j.1526-4610.2010.01824.

3. Kumar P, Kijima Y, West BH, Tobis JM. 2019. The Connection Between Patent Foramen Ovale and Migraine. Neuroimaging Clin N Am 29:261-270. https://doi: 10.1016/j.nic.2019.01.006.

4. Tobis JM, Charles A, Silberstein SD, et al. 2017. Percutaneous Closure of Patent Foramen Ovale in Patients With Migraine: The PREMIUM Trial. J Am Coll Cardiol 70:2766-2774. https://doi: 10.1016/j.jacc.2017.09.1105.

5. Mattle HP, Evers S, Hildick-Smith D, et al. 2016. Percutaneous closure of patent foramen ovale in migraine with aura, a randomized controlled trial. Eur Heart J 37:2029-2036. https://doi: 10.1093/eurheartj/ehw027.

6. Dowson A, Mullen MJ, Peatfield R, et al. 2008. Migraine Intervention With STARFlex Technology (MIST) trial: a prospective, multicenter, double-blind, sham-controlled trial to evaluate the effectiveness of patent foramen ovale closure with STARFlex septal repair implant to resolve refractory migraine headache. Circulation 117:1397-1404. https://doi: 10.1161/CIRCULATIONAHA.107.727271.

7. Pradat P, Francannet C, Harris JA, Robert E. 2003. The epidemiology of cardiovascular defects, part I: a study based on data from three large registries of congenital malformations. Pediatr Cardiol 24:195-221. https://doi: 10.1007/s00246-002-9401-6.