Yawning occurs in various conditions such as hypoxia, epilepsy, and sleep disorders including sleep apnea. Neural networks among the pons, hypothalamus, limbic system, and autonomic nervous system may be associated with yawning. However, intractable yawning associated with a brain tumor has rarely been reported.1

We report a rare case involving a patient who had a mature teratoma in the supramedial cerebellum and suffered from intractable yawning. After subtotal removal of the tumor, the yawning completely disappeared.

**Case Report**

**History and Examination.** A 19-year-old woman with recurrent yawning visited a local hospital, and an intracranial mass was detected on MRI. The patient visited our institution 4 weeks after symptom onset. The yawning occurred 20 times per minute and continued for approximately 60–90 minutes. Such yawning attacks occurred 2 or 3 times per week. Neurological examination demonstrated no abnormal findings. Magnetic resonance imaging depicted a high-intensity mass in the supramedial cerebellum on T1- and T2-weighted images (Fig. 1). The tumor compressed the dorsal midbrain and upper pons.

Blood gas analysis findings, including partial oxygen pressure, were within normal limits during yawning. Blood counts and biochemical data were also within normal limits. Although the patient’s score was 20/24 on the Epworth Sleepiness Scale for detection of sleep disorders (normal score < 11), polysomnography showed no abnormality. Electroencephalography also demonstrated no abnormality. After improving her daily living environment according to our advice, the patient’s Epworth Sleepi-
ness Scale score declined to 9/24. However, the yawning continued for 2 years after the decrease in the Epworth Sleepiness Scale score, and then diplopia developed. Neurological examination demonstrated a right trochlear nerve palsy. Magnetic resonance imaging showed no change in the tumor.

Operation and Postoperative Course. The patient underwent removal of the tumor through an occipital transtentorial approach performed under general anesthesia. The tumor was hard and contained yellowish fat tissue and calcified components. During surgery, we found that the tumor adhered to the dorsal side of the midbrain and upper pons and involved the right trochlear nerve and the right superior cerebellar artery. Therefore, the tumor except the part adhering to the dorsal midbrain was removed. The surgical specimen was histologically diagnosed as a mature teratoma composed of fat, muscle, and nerve cells with poor heteromorphism. The yawning completely disappeared immediately after surgery and has not recurred 7 months after surgery. Postoperative MRI showed a residual tumor at the dorsal midbrain and resolution of the brainstem compression (Fig. 2).

Discussion

Based on blood gas analysis, the Epworth Sleepiness Scale score, and findings on polysomnography and electroencephalography, intractable yawning in this patient was likely not caused by hypoxia, epilepsy, or a sleep disorder such as sleep apnea. Furthermore, the yawning disappeared immediately after surgery and did not recur after surgery. These findings suggested that the yawning was associated with a tumor in the supramedial cerebellum that compressed the dorsal midbrain and upper pons.

The parabrachial area is located on the dorsal side of the junction of the midbrain and pons and plays a role in unifying the autonomic nervous system. In the human brain, the parabrachial area is a horseshoe-shaped band of gray matter composed of the lateral parabrachial nucleus (PBN), the Kölliker-Fuse nucleus, and the medial PBN (Fig. 3 upper). These nuclei receive important afferent fibers from the cardiovascular, respiratory, and gustatory systems and project efferent fibers toward superior centers. On the other hand, the paraventricular nucleus (PVN), which is located in the medial hypothalamus, is a center of the autonomic nervous system and neuroendocrine system, and it is closely associated with yawning (Fig. 3 lower). An experimental study in rats demonstrated the occurrence of yawning upon electrical or chemical stimulation of the PVN. Furthermore, the PVN has reciprocal connections with brainstem autonomic centers including the PBN. In particular, the PVN transmits signals mainly from the lateral PBN. In the present case, any extraordinary stimulation from the lateral PBN in the brainstem to the PVN in the hypothalamus may have led to the development of pathological yawning.

In this case, MRI showed that the tumor preoperatively compressed the dorsal side of the midbrain and up-
Intractable yawning associated with mature teratoma

These findings suggested that the intractable yawning might have resulted from the tumor compressing the PBN located on the dorsal side of the junction of the midbrain and pons.

Disclosure

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

Author contributions to the study and manuscript preparation include the following. Conception and design: Saura, Beppu. Analysis and interpretation of data: Uesugi, Sasaki. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Saura. Administrative/technical/material support: Matsuura, Asahi.

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Fig. 3. Upper: The parabrachial area is composed of the lateral PBN (a), the Kölliker-Fuse nucleus (b), and the medial PBN (c). Lower: The PVN is localized in the medial hypothalamus.