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Case Report

A rare case of glial choristoma of the tongue associated with cleft palate

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ABSTRACT

Glial choristoma is considered a developmental malformation characterized by brain heterotopias without connection to the central nervous system. In this article, we report a rare case of glial choristoma occurring on the dorsum of the tongue with cleft palate in a 3-month-old male infant. Complete surgical excision was performed prior to palatoplasty. There were no postoperative complications or evidence of recurrence after 5 years of follow-up.

1. Introduction

Glial choristoma is a developmental malformation of heterotopic central nervous system tissue. The pathogenesis of glial choristoma is hypothesized to be due to hernia or separation of neuroglial tissue before complete closure or through arrested closure in the osseous cranium [1] or overgrowth of the developing neural lobe that may prevent closure of the cranial opening [2]. The nasal region is the most frequently affected site [3], while the oral and maxillofacial region involving the palate, tongue, oropharynx, and so on is a relatively rare site [4]. Glial choristoma of the tongue is extremely rare, with only 23 cases including the present one having been described. Only three cases, including the present case, of glial choristoma of the tongue concurrent with cleft palate have been reported in the English literature [5]. In this article, we report our experience with a rare case of glial choristoma with cleft palate.

2. Case report

A 3-month-old male infant was referred to our hospital for the examination of a tongue lesion. He was born of a full-term normal delivery and weighed 3134 g. His growth was normal, with no significant medical problems. The only abnormal finding on physical examination was a cleft of the soft palate. The family history was unremarkable. One month after birth, his mother noticed a cleft palate, and the child was examined by plastic surgeons at another hospital. For another 3 months after birth, she noticed a tongue mass and consulted another pediatric clinic. The patient was referred to our hospital for examination of the tongue. The tongue mass had findings suggestive of tumor. On clinical examination, a pedunculated and slightly elastic but hard tumor-like mass measuring approximately 22 mm × 17 mm was present in the posterior and right lateral part of the dorsal tongue. The mass was covered with normal mucosa (Fig. 1). The clinical appearance was suggestive of a benign tumor. A cleft of the soft palate was observed (Fig. 2). Breathing and feeding were unaffected by these lesions. He could feed with almost no nasal emission of milk before palatoplasty. On MR imaging, the mass extended from the surface of the tongue into the muscle layer and showed a low-intensity signal on T1-weighted imaging and a heterogeneous and slightly high-intensity signal on T2-weighted imaging (Figs. 3 and 4). A dilated vascular structure and flow void were not seen around the lesions. From the above findings, diagnoses such as myofibroma, neurofibroma, and lymphangioma were suspected. In addition, a leptomeningeal cyst was seen in the middle cranial fossa on MR images. The leptomeningeal cyst was followed without treatment. We waited until he grew up to be of sufficient weight because this tumor was not

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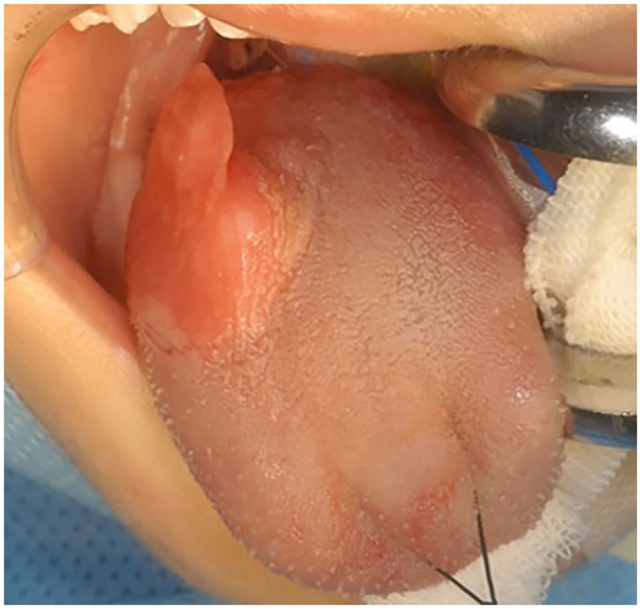

Fig. 1. Preoperative photograph of a pedunculated tumor-like mass in the posterior and lateral part of the dorsal tongue.

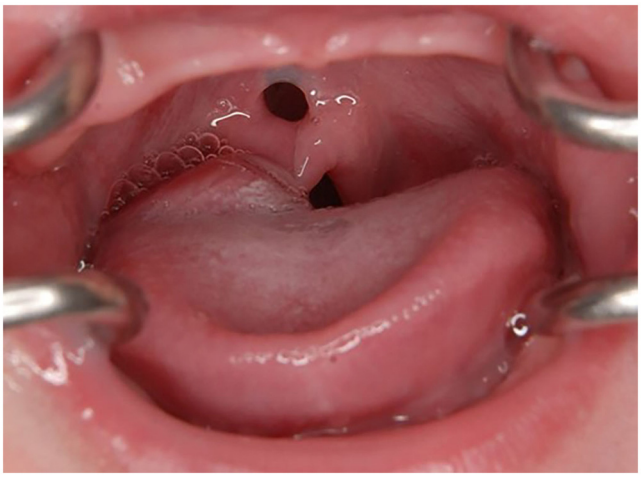

Fig. 2. Photograph of the cleft soft palate.

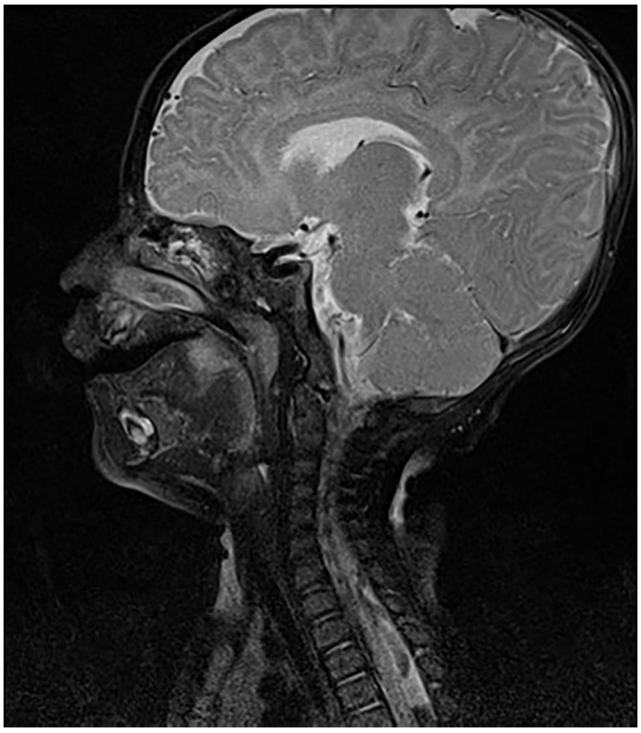

Fig. 3. T2-weighted sagittal image of MRI.

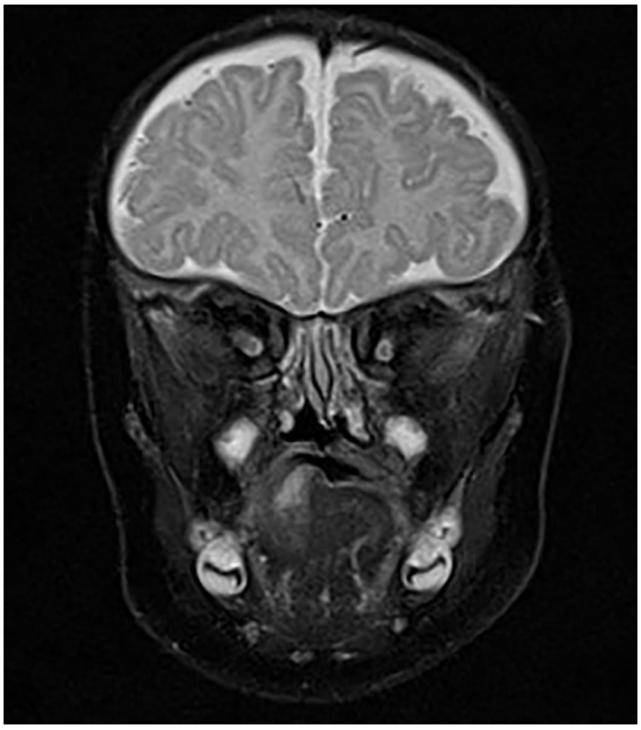

Fig. 4. T2-weighted coronal image of MRI.

suspected to be malignant. The tumor size and nature remained unchanged over the period of time before the surgery was performed. Fifteen months after birth, the tumor was excised under general anesthesia. At the time of surgery, the tumor measured 22 mm × 17 mm and was a wide-based, slightly elastic but hard mass that was immovable. The posterior part of the tumor was formed in a fold. The margin of the tumor was slightly obscure. An incision was made around the base of the tumor including covered mucosa, which was removed with a part of the lingual muscle (Fig. 5) because the border between the tumor and the muscle was indistinct. The capsule of the specimen was indistinct. The specimen was of a mottled white color and a homogeneous mass (Fig. 6). The border between the mass and muscle tissue was slightly indistinct (Fig. 7). On histological examination, considerable amounts of fibrous tissue were present under the mucosal epithelium. The marginal region between choristoma and normal surrounding tissues was indicated on H-E (Fig. 8a) and glial fibrillary acidic protein (GFAP) (Fig. 8b) staining. The border was not clear. The lesion was composed of mature glial cells mixed with abundant fibrous elements (Fig. 9). The glial cells were positive for GFAP (Fig. 10), S-100 (Fig. 11), and vimentin and negative for desmin, alpha-SMA, synaptophysin, and neurofilament. There were a few cells positive for Ki-67, a proliferative marker. The lesion of the tongue was diagnosed as a glial choristoma according to its histopathological findings. Healing of the incisional wound was good. Twenty-one months after birth, palatoplasty with a Furlow procedure was performed. The palatoplasty was slightly delayed than usual because

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