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Case Report

Congenital bifid tongue with lingual hamartoma: A case report and review of the literature

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ABSTRACT

A non-syndromic, congenital bifid tongue is an extremely rare malformation, the details of which are largely unknown. A female neonate was referred to our division presenting with a bifid tongue. The cleft was located in the right anterior part of the tongue. In addition, 2 tumors (10 mm and 2 mm) were located at the base of the cleft. Seven days after birth, the tumors were excised and primary closure of the cleft was performed for correction of the bifid tongue. The postoperative course was uneventful, and follow-up examination 20 months later showed no functional disturbance. Furthermore, all published cases were carefully reviewed to improve our understanding of the etiology of congenital bifid tongue. Formation of a congenital neoplasm during the early stages of tongue development could be the main etiology of a bifid tongue.

1. Introduction

Congenital bifid tongue is a rare malformation wherein the tip of the tongue is divided longitudinally [1]. It is well known that the development of the tongue takes place around the fourth week of intrauterine life. It originates from the median swelling on the floor of the pharynx and the 2 lateral lingual swellings that join the central structure [1,2]; when this fusion process is disturbed, it results in the division of the anterior part of the tongue. A congenital bifid tongue is mostly reported as one of the features associated with oral–facial–digital syndrome [3]. The occurrence of a bifid tongue in the absence of other orofacial abnormalities is extremely rare, and only 15 reports of a bifid tongue have been published in the English literature during the past 30 years [1,4–15]. Thus, the details of this condition are largely unknown. In this report, we describe the detailed clinical observations of a non-syndromic, laterally located, bifid tongue occurring concomitantly with a tongue tumor. In addition, we present a review of published cases of a congenital bifid tongue to discuss the etiology of this malformation.

2. Case report

A 26-year-old pregnant female was referred to our hospital because of abnormal fetal findings. Fetal MRI at 30 weeks gestation revealed a high signal mass on the left side of the cerebral hemisphere, along with the absence of corpus callosum (Fig. 1). Furthermore, cerebral ventricular enlargement was also observed via fetal ultrasonography.

A female neonate, weighting 3368 g was delivered by cesarean section at 36 weeks 6 days of gestation due to breech presentation. The Apgar score was 8 and 9 at 1 min and 5 min after birth, respectively. The head circumference of the neonate was 39.5 cm, which is above the 90th percentile, and no congenital cardiac anomalies were detected. MRI study investigation 1 day after birth confirmed the presence of an interhemicerebral multilocular cyst measuring 4.5 cm × 5.6 cm × 7.0 cm, along with corpus callosum agenesis and hydrocephalus. Subsequently, the patient was referred to our department due to the presence of a bifid tongue.

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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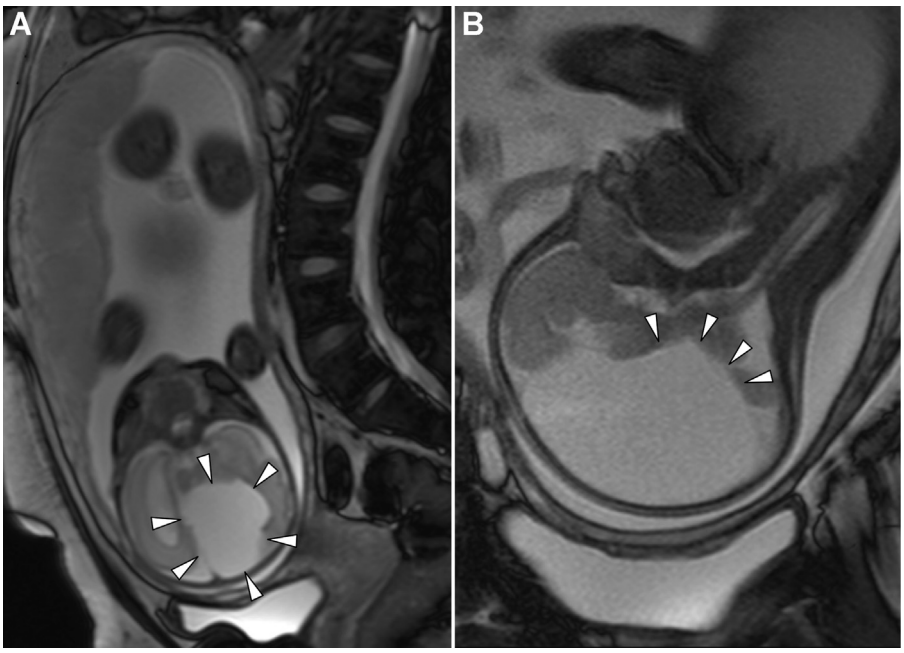

Fig. 1. Results of T2-weighted MR images at 30 weeks of pregnancy. (A) Fetus MRI showing a cystic lesion in the interhemispheric region (arrows). (B) A sagittal image showing the absence of corpus callosum (arrows).

Oral examination revealed a 10-mm-long cleft located on the right anterior third of the tongue. A bean-shaped mass measuring 10 mm × 10 mm was located at the base of the cleft. The lingual frenum was attached on the left side of the mass. Furthermore, another small ovoid mass (2 mm × 2 mm) was observed medial to the cleft. No other oral anomalies such as cleft lip and/or palate were detected. The condition was clinically diagnosed as a congenital bifid tongue with the presence of a tongue tumor.

Seven days after birth, a cyst-peritoneal shunt was implanted by a neurosurgeon under general anesthesia. At the same time, the mass including the surrounding mucosa of the cleft was excised (Fig. 2). In the deeper layers, the mass was found to be clearly

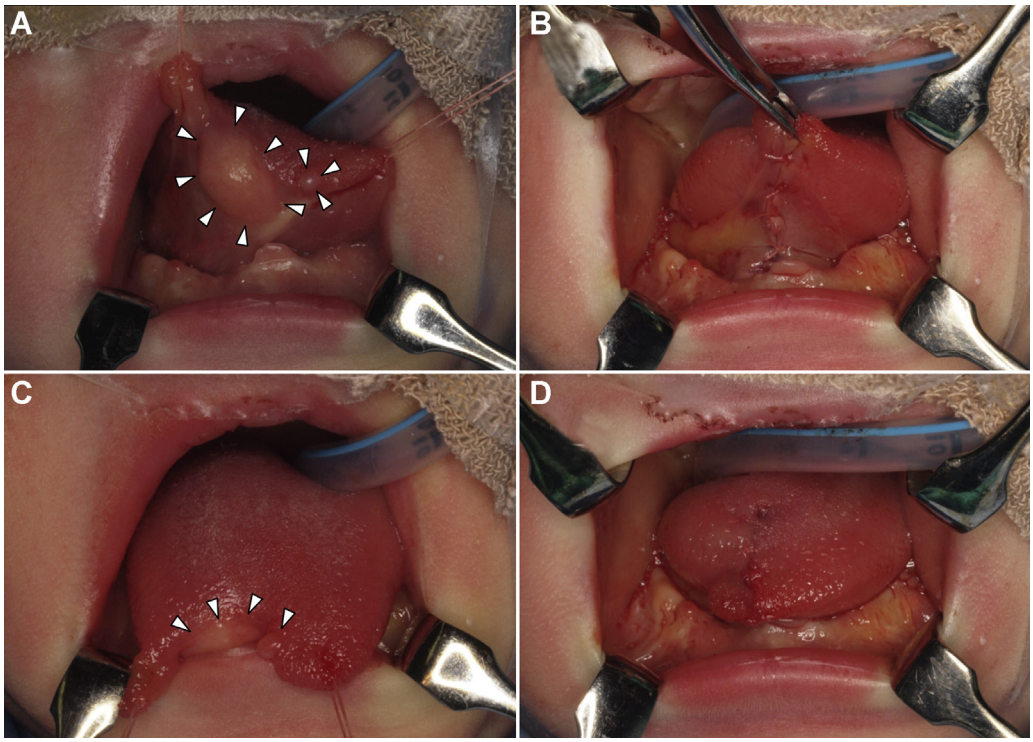

Fig. 2. Clinical appearance of the tongue of the neonate. (A, C) Preoperative intraoral view of the tongue. Two masses are seen at the base of the cleft (arrows). (B, D) Postoperative intraoral view of the tongue. The hamartomas were removed and primary closure was performed.