

Case report

Congenital parotid ectopia in accessory maxilla and facial cleft anomalies: Three cases report

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ABSTRACT

To further document the clinical features of accessory maxilla with three additional cases report. Clinical and radiological features of three cases of accessory maxilla were presented. Related literature was summarized for comparison. Ectopic parotid gland, facial cleft and accessory maxilla are three concomitant malformations in this condition. The tooth-bearing accessory maxillary duplication derives from the abnormal growth of the zygoma or zygomatic arch. Facial cleft, parotid ectopia and tooth-bearing accessory maxilla may constitute a rare congenital syndrome.

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1. Introduction

The terms of “maxillary duplication” and “accessory maxilla” have been used to describe an extremely rare clinical entity characterized by redundant tooth-bearing bony segment posterior to the maxillary tuberosity mimicking an accessory maxilla [1–3]. Up to now less than 20 cases have been documented in the English literature [1–14].

Cameron described the following clinical characteristics of this congenital anomaly: an accessory tooth-bearing maxilla arising from the inferior border of zygoma; supernumerary teeth of normal morphology in the accessory maxilla; full complement of maxillary dentition; facial cleft [8].

Ectopia of the parotid gland has not been reported in this malformation. CT imaging allows for radiological diagnosis of bone and parotid malformations [15]. We present three additional cases to illustrate the parotid malformation in this circumstance.

2. Case report

This study was approved by the Institutional Review Board of School and Hospital of Stomatology Peking University (PKUSSIRB-2012084).

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2.1. Case 1

A 13-year-old boy with facial deformity was referred to our hospital. The patient was discovered with bilateral lateral facial clefts and cleft palate at birth. Surgical repair of the facial clefts and palatoplasty were performed at 5 years old. Familial history was unremarkable.

Physical examination showed the bilateral facial scars due to the surgical repair of the lateral facial clefts (Tessier 7). Cheek prominence and shallow grooves extending from the facial scars to the bilateral zygomata were present. Hypoplasia and retrognathia of the mandible was remarkable. Cleft soft palate was found and hypernasality was prominent. Posterior to the maxillary tuberosity on each side was an abnormal segment of bone (Fig. 1). Mucosal sulcus between the accessory maxilla and the maxillary tuberosity on each side could be seen. Bilateral buccal mucosa scars and folds existed and the openings of the Stensen's ducts could not be identified. Panoramic radiography revealed two erupted and three impacted supernumerary molar-like teeth in the right accessory bone segment. One erupted tooth and another impacted molar-like supernumerary tooth were found in the left accessory bone segment (Fig. 2). Another conical supernumerary tooth was observed in the left tuberosity (Figs. 1 and 2).

CT showed that the accessory teeth-bearing bony segments extended from the inferior borders of bilateral zygomata to the maxillary tuberosities (Fig. 3A). A narrow bony gap was observed between the normal maxilla and the accessory maxilla on the left

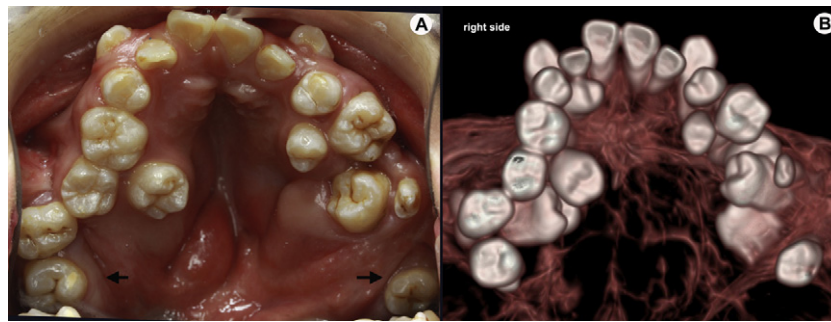


Fig. 1. Intra-oral clinical (A) and three-dimensional CT (B) views of the maxillary dentition. Posterior to the maxillary tuberosities on each side is the accessory maxilla. Two (right side) and one (left side) erupted, three (right side) and one (left side) impacted supernumerary teeth with molar shape could be observed in the accessory bone. Another erupted conical supernumerary tooth is observed on the left tuberosity. Note the narrow mucosal sulcus between the maxillary tuberosity and accessory maxilla on each side (black arrows).



Fig. 2. Panoramic radiograph shows bilateral accessory maxillae (yellow arrows) with multiple teeth posterior to the maxillary tuberosity and the full complement of the permanent maxillary dentition. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

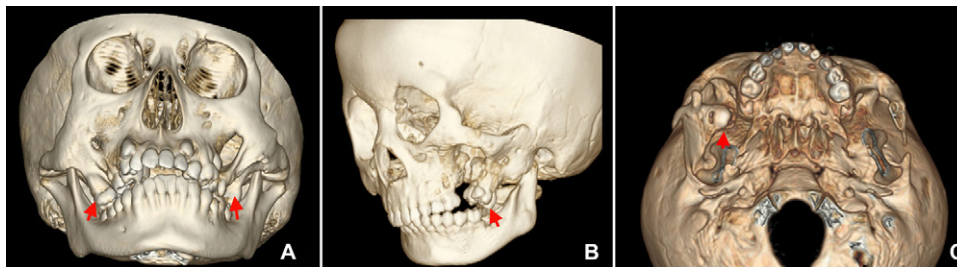


Fig. 3. Three-dimensional CT images. (A) Bilateral accessory maxillary bone segments extend from the inferior borders of bilateral zygoma to the maxillary tuberosity; (B) the accessory maxilla runs from the inferior border of the zygomatic process of the temporal bone to the maxillary tuberosity. Several supernumerary teeth are present in the accessory maxilla. Note the full complement of the primary maxillary and mandibular dentition; (C) malformations of bilateral zygomatic arches. Note the supernumerary tooth on the right zygomatic arch (red arrow) and full complement of the primary maxillary dentition. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

side. The accessory maxilla attached to the maxillary tuberosity on the right side.

Soft-tissue CT images showed that the left parotid space was composed of adipose tissue except for the vessel structures. Interestingly, a 1.4 cm × 1.5 cm × 2.5 cm soft tissue nodule with gland attenuation was found below the zygoma, suggesting the existence of an ectopic parotid gland. The right parotid gland was normal (Fig. 4A).

The accessory maxillae were not excised. Distraction osteogenesis was carried out to increase the mandible length. Orthodontic treatment was performed and functional occlusion was acquired.

2.2. Case 2

A 4-year-old girl was referred to our hospital because of facial deformity. She had undergone cheiloplasty because of congenital

left macrostomia (Tessier 7). Physical examination showed left hemifacial microsomia and cheek prominence. The left outer ear was morphological normal but a few centimeters lower compared with the right ear. Oral examination revealed that an accessory bone segment with several supernumerary molars located posterior to the normal primary dentition on the left side. Mucosa sulcus was seen between the accessory bone and the maxillary tuberosity. The opening of the left Stensen's duct could not be identified and no obvious excretion could be elicited.

Three-dimensional CT showed that the accessory maxilla extended from the zygomatic process of the temporal bone toward the maxillary tuberosity (Fig. 3B). Four supernumerary molar-like teeth were observed in the accessory maxilla. Hypoplasia of the left mandibular coronoid process was also noted. The left parotid space was composed mainly of adipose tissue (Fig. 4B). A 2.0 cm × 1.5 cm × 1.0 cm soft tissue nodule of

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