

RESEARCH ARTICLE

An Unusual Accessory Mandible and a Submucosal Cleft Palate—A Case Report and Review of the Literature

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An accessory mandible is a rare congenital anomaly that requires multidisciplinary management. This case report describes a female patient with an unusual accessory mandible, a dysplastic overgrowth of bone, containing teeth that extended from the midsymphyseal region. A submucosal cleft palate and cleft of the lower lip were also present. Her treatment plan took a staged approach with initial surgical resection of the accessory bone and teeth. The second stage, still in the planning phase, will correct the secondary deformity of an anterior open bite and will restore the missing lower anterior teeth. The original deformity and subsequent growth are discussed with the relevant literature.

KEY WORDS: *accessory mandible, mandibular duplication, submucosal cleft palate*

Craniofacial duplication varies in presentation, from a localized anomaly (i.e., duplication of nose, eyes, tongue, ears, maxilla, mandible, or jaw parts [accessories]; Table 1), to complete craniofacial duplication (diprosopus) (Fearon and Mulliken, 1987; Biasibetti et al., 2011). Duplication of the mandible is rare (Table 2); one of the first documented reports was by Meijer in 1883 (Wittkamp and van Limborgh, 1984). The present case report describes a girl with an accessory mandible and other associated medical conditions, including a submucosal cleft palate. The dento-skeletal development of the patient along with the relevant literature review is described.

CASE REPORT

Social and Previous Medical History

The patient was born to an African American mother who reported a family history of cleft palate but no family history of accessory jaws or facial overgrowth.

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The patient was born prematurely, at 36 weeks, with a birth weight of 2.46 kg. At the age of 1 month the patient underwent an operation for resection of an oropharyngeal mass without any complications. She underwent another operation at 3 months old where she had a transpalatal resection of a nasal choanal mass, with repair of the tongue defect. The oral mass removed at 1 month of age had fragments of salivary gland tissue, and the choanal mass was a hairy polyp. Her initial presentation at Children's Hospital Los Angeles (CHLA) was unusual in that her mother brought her to see the craniofacial orthodontist for an evaluation at age 2 at CHLA, without a primary physician referral. The patient had not yet seen a craniofacial surgeon or pediatrician. At her initial visit, she was introduced to the craniofacial surgeon and nurse to enroll her in the craniofacial team at CHLA.

Physical and Radiological Examination

On physical examination, an oral cavity lesion was identified, extending from the base of the tongue to the midline of the lower lip, measuring about $3 \times 2 \times 2 \text{ cm}^3$ (Figs. 1 and 2). The patient's profile mimicked a Class III skeletal pattern, with a prognathic mandible (Fig. 1). Intraoral examination revealed a primary dentition, the absence of anterior mandibular deciduous teeth, an anterior open bite, and some mandibular gingival overgrowth (Fig. 2). The computerized axial tomography scans with contrast (Fig. 3) and the MRI were done to evaluate the oral mass. The MRI revealed a well-circumscribed homogeneous mass, without deep extension into the tongue (Fig. 4).

At the age of 2½ years, the patient underwent surgical resection of accessory mandible and oral reconstruction.

TABLE 1 Different Types of Facial Duplication

<i>Facial Duplication</i>	<i>Study or Case Report</i>
Nose	Ghosh et al., 1971; Obwegeser et al., 1978; Barr, 1982; Kotrikova et al., 2007; Maruotti et al., 2009
Eyes	Kotrikova et al., 2007; Maruotti et al., 2009
Ears	Gadre et al., 1987; Gore et al., 2006; Pan et al., 2010
Maxilla	Fish, 1965; Avery and Hayward, 1969; Robertson 1970; Gupta, 1975; Chandra, 1978; Barr, 1982; Fearon and Mulliken, 1987; Borzabadi-Farahani et al., 2011
Mandible	McLaughlin, 1948; Davies et al., 1973; Maisels, 1981; Wittkampf and van Limborgh, 1984; Shaikh et al., 2008; Pengiran Suhaili et al., 2011
Tongue	Bell, 1971; Verdi et al., 1991



FIGURE 1 Preoperative lateral view of the accessory mandible.

Figure 5 shows the outline of osteotomy lines before resection. The operation involved excising a central wedge of malformed clefted lower lip, total lower lip reconstruction, excision of the bony mass, and resection of redundant tissue at the floor of the mouth. Following reduction of the mandible, there was a marked redundancy of soft tissue on the floor of the mouth. Using sharp dissection, a wedge-shaped tissue at midline, with the apex at the tongue, was excised to allow primary closure of the floor of the mouth. The floor of the mouth was repositioned to help creating a gingivolingual sulcus. The central aspect of the malformed lower lip was excised full thickness, and hemostasis was obtained with electrocautery. The orbicularis oris muscle was reconstructed and the skin sutured in layers. Due to concerns over swelling of the floor of the mouth and possibly airway compromise, the patient was transferred to the intensive care unit

postoperatively. Her postoperative recovery was uneventful. On the morning of her first postoperative day, the nasal trumpet was removed. Her oral intake increased over the next 2 days, and she was discharged after 2 days.

Histopathological Findings

The excised circumscribed mass had dysmorphic mucosa, soft tissues, an expanded segment of the mandible with 12 unerupted teeth, and a dentigerous cyst. The soft tissue and mucosa of the floor of the mouth specimen had regional tissue with glandular hyperplasia (i.e., hyperplastic minor salivary gland).

Craniofacial Development

Twelve years after the surgery, the patient returned for orthodontic evaluation. Her medical findings in-

TABLE 2 Examples of Mandibular Duplications/Accessories and the Associated Features

<i>Mandibular Duplication</i>	<i>Associated Features</i>	<i>Study or Case Report</i>
Two duplicated mandibular arches and alveolus (W-shaped)	Hypertelorism, duplication of pituitary gland, duplication of tongue and cervical vertebra, split notochord syndrome, cleft palate	Wittkampf and van Limborgh, 1984
Accessory unilateral mandibular body and alveolus	Duplicated mandible and patient's mandible may have common condyle, coronoid process, and part of the ramus. It may be more localized and associated with duplicated lip and macrostomia. It incorporates dentition.	Akpuaka and Nwozo, 1990; Shaikh et al., 2008
Accessory mandible and alveolus in the midline area	Bimanual dyskinesia, webbing of the epicanthal folds, scoliosis, ptosis, macroglossia, submucosal cleft palate, cleft of lower lip, dentition in the duplicated mandible	Present case report
Accessory bilateral mandible (ramus)	Klippel-Feil syndrome, fusion of cervical vertebrae, facial asymmetry, thoracic kyphosis, ocular defects, hearing loss, ventricular septal defect, cleft palate, macrostomia, pain and difficulty in chewing; may be associated with dentition in the duplicated ramus	Lawrence et al., 1985; Ball, 1986
Accessory bilateral mandible (rudimentary condyles)	Klippel-Feil syndrome, Pierre Robin sequence, occipital cleft, large foramen magnum with occipital bone clefts, submucous cleft palate	Al-Ani et al., 2009
Accessory condyle associated with coronoid enlargement. Accessory bifid or trifid condyles	It may be acquired. An asymptomatic incidental finding, temporomandibular joint sound and pain, trismus, limited mandibular opening, swelling, ankylosis, facial asymmetry	Smylski, 1952; Farmand, 1981; Thomason and Yusuf, 1986; Antoniadis et al., 1993; Artvinli and Kansu, 2003; Antoniadis et al., 2004; de Sales et al., 2004; Daniels and Iqbal, 2005; Shriki et al., 2005; Açikgöz, 2006; Tunçbilek et al., 2006; Peacock et al., 2011; Li et al., 2011
Accessory mandibular alveolus	It may be associated with duplicated lip and macrostomia.	Price and Zarem, 1979; Soneji, 2010; Pengiran Suhaili et al., 2011



FIGURE 2 Preoperative anterior view of the accessory mandible.

cluded the following: bimanual dyskinesia, webbing of the epicanthal folds, mild scoliosis, ptosis of the left eye, macroglossia, resected accessory mandible, and a submucosal cleft palate. Orthodontic examination revealed permanent dentition with a Class II malocclusion on a Class I skeletal base, tongue thrust, missing permanent lower anterior incisors, anterior open bite, bilateral posterior crossbite, and proclined upper incisors. Figure 6 shows the occlusal view of the mandibular arch taken before starting the orthodontic treatment. Figures 7 and 8 show the most recent panoramic and lateral cephalogram radiographs of the patient at the beginning of orthodontic treatment. The



FIGURE 3 A CT scan showing an axial view of the mandible. The accessory mandible containing teeth is visible.

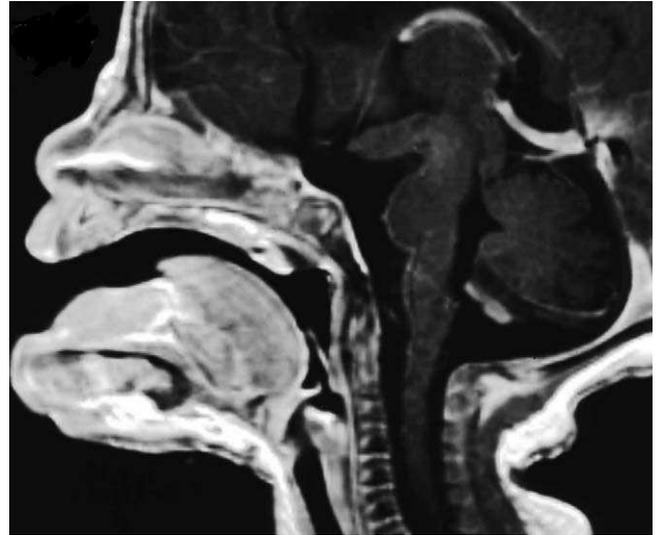


FIGURE 4 Sagittal MRI view showing the extent of the accessory mandible.

cephalometric assessment revealed a shortened anterior cranial base, a Class I skeletal base, an increased lower facial height, and very proclined maxillary incisors. The patient is undergoing orthodontic treatment to correct her dento-skeletal deformity with orthognathic surgery after adolescent facial growth is complete.

DISCUSSION

Certain medical conditions have been associated with facial duplication: facial clefts (Cameron et al., 1993; Tharanon et al., 1998; Celebiler et al., 2007; Woods et al., 2008; Hou et al., 2011), cleft lip and/or palate (Avery and Hayward, 1969; Robertson, 1970; Gupta, 1975; Fearon and Mulliken, 1987, Soneji, 2010), Klippel-Feil syndrome (Chandra, 1978; Lawrence et al., 1985; Ball, 1986; Al-Ani et al., 2009), or the Pierre Robin sequence (Al-Ani et al., 2009). This case report also includes a distinct association of different anomalies: accessory mandible, bimanual dyskinesia, webbing of the epicanthal folds, mild scoliosis, ptosis of the left eye, macroglossia, and a submucosal cleft palate.

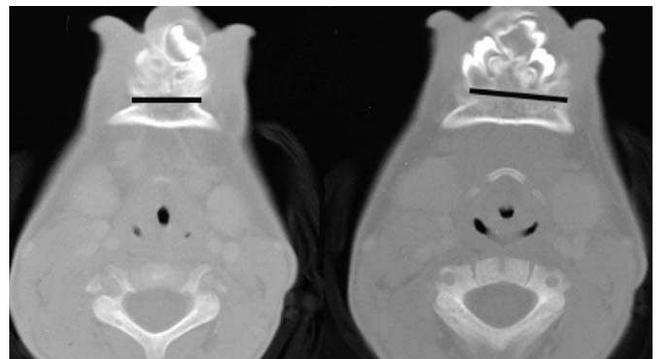


FIGURE 5 Osteotomy lines prior to surgery at the age of 2½ years.

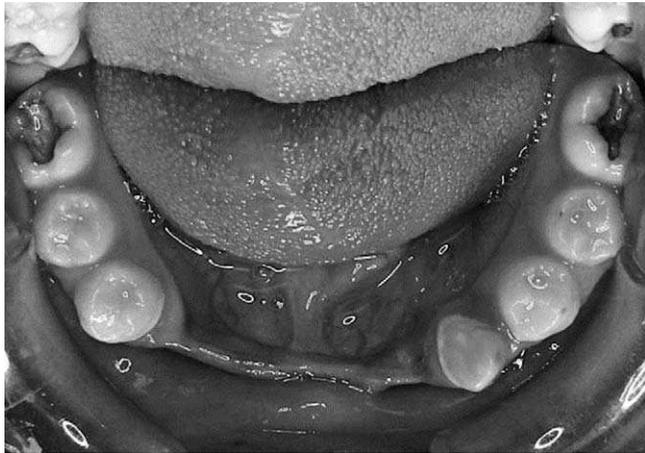


FIGURE 6 The occlusal view of the mandibular arch taken before starting the orthodontic treatment.

To our knowledge, this is a unique presentation of an accessory mandible because it contains multiple teeth and a dentigerous cyst (please see the “Differential Diagnosis” section). The teeth lacked symmetry and developed in a disorganized manner. Given that the accessory mandible was attached to the midsymphyseal midline, this structure may have resulted from dysmorphogenesis in fusion of the right and left mandibular processes of the first branchial arch. It is possible that fusion and patterning of the mandibular process were not properly regulated at the midline and resulted in uncontrolled and continuous growth (D’Souza et al., 2010). The proposed developmental mechanism that may be responsible for similar dysmorphogenesis includes mutation of the *Dlx* homeobox genes, expression of neural crest cells, and contribution to the patterning of subdivisions of the first pharyngeal arch skeleton and musculature (Depew et al., 2002; Heude et al., 2010).



FIGURE 7 The panoramic radiographic view of the patient at age 14.



FIGURE 8 The lateral cephalogram radiograph of the patient at age 14.

In a neonate with accessory craniofacial parts, computerized tomography (CT), MRI, and magnetic resonance angiography are often necessary for evaluating the degree of duplication and planning the required reconstructive and cosmetic procedures (Hähnel et al., 2003). Treatment consisted of removing the duplicated/accessory mucosa and periosteum and preventing the development of a retention cyst or further bone formation (Davies et al., 1973; Maisels, 1981; Pengiran Suhaili et al., 2011). Although successful in removing the accessory mandibular deformity, the initial treatment created an edentulous mandibular ridge and an anterior open bite. Future orthognathic surgery and dental implants are needed to replace the missing lower anterior teeth and to correct the dento-skeletal deformity. These should be should be

deferred until after the completion of growth (Borzabadi-Farahani, 2011). Early resection of the accessory mandible carries the risk of damage to the permanent tooth germs; therefore, surgical resection is deferred until the age of 3 or later to allow better differentiation between usable and unusable tooth germs (Woods et al., 2008; Pengiran Suhaili et al., 2011).

Differential Diagnosis

The differential diagnosis for this mandibular anomaly includes cystic teratoma, multiple supernumerary teeth (MST), Tessier type 30 cleft, and duplication of facial structures. A teratoma is a tumor consisting of multiple tissues, not indigenous to their site of origin, and proliferates in a disorganized manner (Weaver et al., 1976; Byard et al., 1990). In contrast, duplications grow in an orderly fashion, resembling the local tissue architecture (Fearon and Mulliken, 1987). The present patient displayed a well-circumscribed homogeneous mass, which was not consistent with teratoma. Although the accessory jaw contained multiple teeth, it did not present like syndromic MST, which is associated with medical conditions such as cleft lip and palate, cleidocranial dysplasia, and Gardner syndrome (Vahid-Dastjerdi et al., 2011). This patient did not present like the nonsyndromic form of MST either, which tends to occur in a bilateral manner in the mandibular premolar region within the normal alveolar bone structure (Yusof, 1990; Batra et al., 2005; Kalra et al., 2005; Orhan et al., 2006; Inchingolo et al., 2010; Alvira-González and Gay-Escoda, 2011). This patient has a median cleft of the lower lip that was classified by Tessier as a type 30 craniofacial cleft (Tessier, 1976). Various forms of median mandibular clefts exist, ranging from minor clefts of the lower lip (Arshad, 1995) to complete clefts of the mandible, with the absence of the hyoid bone, thyroid cartilages, and manubrium (Halli et al., 2011). However, this patient had only a lower lip cleft that did not extend into mandibular bone.

Accessory mandibles have been considered a variant of craniofacial duplication that can range from complete to partial duplication. Complete craniofacial duplication (diprosopus) is thought to be a form of conjoined twinning (Machin, 1993; Wu et al., 2003) and occurs in 1 per 2800 to 1 per 200,000 births (Machin, 1993; Hähnel et al., 2003). The rarest type of all conjoined twins is the diprosopus (0.4%), with two faces, one head, and one body (Machin, 1993; Hähnel et al., 2003). Most neonates with diprosopus are stillborn (Hähnel et al., 2003). Facial duplication occurs in symmetrical and asymmetrical forms (Fearon and Mulliken, 1987), and patients with asymmetrical facial duplication may develop an accessory mouth on a side of the face (Maisels, 1981). The partial duplications of facial

structures have been reported for nose, eyes, ears, maxilla, mandible, and tongue (Table 1). The accessory jaws can present as duplicated jaw portions, with or without teeth, such as a duplicated condyle, ascending ramus, palate, or alveolar arch. Accessory mandibles are rarer than accessory maxillas (Shaikh et al., 2008; Borzabadi-Farahani et al., 2011) (Table 2).

Although mandibular duplication is extremely rare, duplicated parts of the mandible such as the condylar head has been reported in the literature (Table 2). Accessory bifid or trifid condyles were first described on dried skulls and cadavers (Hrdlicka, 1941; Honee and Bloem, 1969; Szenpetery et al., 1990). They were first described by Schier (1948) and can be asymptomatic with a postnatal etiology of infection or trauma to condyles (Thomason and Yusuf, 1986; Antoniadis et al., 1993; Artvinli and Kansu, 2003; Antoniadis et al., 2004; Daniels and Iqbal, 2005; Li et al., 2011) or due to other etiological factors (Blackwood, 1957; Gundlach, 1983; Quayle and Adams, 1986; Zohar and Laurian, 1987). Proposed mechanisms for mandibular duplication during development are the split notochord syndrome (Avery and Hayward, 1969), developmental anomalies arising from sequestered totipotent cells (Davies et al., 1973), first branchial arch duplication (Maisels, 1981), and duplication of the mandibular growth centered around the margins of the stomatodeal plate (Barr, 1982).

In summary, this is a rare presentation of an accessory mandible that contained supernumerary teeth and was associated with submucosal cleft palate and cleft of the lower lip.

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