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## CASE REPORTS

# Subglossopalatal Synechia in Association With Cardiac and Digital Anomalies

Albert K. Oh, M.D., Joseph Thakuria, M.D., Virginia Kimonis, M.D., John B. Mulliken, M.D.

**A 1-day-old boy in respiratory distress had a midline soft tissue band between the floor of the mouth and the posterior edge of the hard palate. There was also a soft palatal cleft, cardiac anomalies, and a hypoplastic right fifth finger and toe. Although his airway initially improved following urgent excision of the subglossopalatal band, he continued to have episodic desaturations. A tongue–lip adhesion opened his airway, and he subsequently underwent resection of juxtaductal aortic coarctation and ligation of patent ductus arteriosus and left superior vena cava. Congenital oral synechiae are uncommon. Affected infants often require prompt intervention secondary to respiratory distress and feeding difficulty. Review of the literature indicates that midline subglossopalatal synechia with cardiac and digital anomalies may be in the oromandibular–limb hypogenesis spectrum.**

KEY WORDS: *intraoral band, subglossopalatal synechiae, cardiac and digital anomalies, oromandibular–limb hypogenesis spectrum*

Congenital oral synechiae are rare, typically occurring between the upper and lower alveolar ridges or between the tongue and margins of the palate or maxilla (Valnicek and Clark, 1993; Tanrikulu et al., 2005). Neonatal respiratory distress and feeding difficulty are the most common presenting signs of an oral synechia, often requiring prompt intervention (Zalzal et al., 1986; Bagatin and Boric, 1990; Gartlan et al., 1993). There may be residual limitation of mandibular excursion following division of the band (Gartlan et al., 1993; Dalal and Davison, 2002; Denion et al., 2002; Kalu and Moss, 2004; Murphy et al., 2004; Tanrikulu et al., 2005).

Disorders associated with intraoral bands include congenital alveolar synechiae syndrome, van der Woude syndrome, popliteal pterygium syndrome, and oromandibular–limb hypogenesis spectrum (Chandra et al., 1974; Pillai et al.,

1990; Grippaudo and Kennedy, 1998; Dalal and Davison, 2002; Denion et al., 2002; Puvabanditsin et al., 2003; Sahin et al., 2005). Other recognized associations include hydrocephalus, cleft of the secondary palate, bifid tongue, patent ductus arteriosus, hypospadias, and bifid scrotum (Nakajima et al., 1979; Zalzal et al., 1986; Gartlan et al., 1993).

The authors report the case of a child with a subglossopalatal band in association with cardiac and digital anomalies and a sibling with similar digital abnormalities. Histological findings and embryologic considerations are discussed, and candidate genes are reviewed.

### CASE REPORT

A male infant, the product of in vitro fertilization, was scheduled for delivery by elective caesarian section at 37 6/7 weeks gestation to a 44-year-old G1P0 mother. There was no prenatal history of maternal infection, gestational diabetes, or exposure to alcohol, tobacco, drugs, medications, or toxins. Amniocentesis documented a normal 46XY karyotype. Prenatal ultrasonographic study demonstrated a two-vessel cord and left ventricular wall hypertrophy; however, a subsequent study did not confirm the latter finding. The 2560-g newborn was at the 50th percentile for weight and 3rd percentile for both length and head circumference. Although his Apgar scores were 8 and 9, the child was soon transferred to the neonatal intensive care unit due to respiratory distress.

Physical examination demonstrated a midline subglossopalatal band of soft tissue extending from the anterior floor

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**FIGURE 1** Soft tissue synechia extending from anterior floor of mouth to posterior hard palate. Note how the tongue is held back by the abnormal band of tissue.

of the mouth to the posterior edge of the hard palate (Fig. 1). There was also a cleft of the soft palate. Additional anomalies included microstomia, microglossia, an 8-mm lower alveolar cyst, minor hypoplasia of the auricular helices, a 2-mm left areolar tag, hypoplastic nails of all digits, and hypoplastic right fifth finger and toe (Figs. 2 and 3). Postnatal echocardiogram revealed bicommissural aortic valve, aortic coarctation, patent ductus arteriosus, patent foramen ovale, and left superior vena cava.



**FIGURE 2** Photograph of right hand, demonstrating decreased length of small finger.



**FIGURE 3** Photograph of right foot. Note marked hypoplasia of the fifth digit.

Abdominal ultrasonography demonstrated bilateral hydro-nephrosis associated with vesicoureteric reflux.

The family history was significant for a paternal half brother with a hypoplastic right fifth toe. Two maternal first cousins had Down syndrome, although their mother was of advanced age.

On the first day of life, the patient underwent urgent resection of the intraoral band under local anesthesia; the tissue was sent for pathologic examination. The patient's respiratory status improved, but over the next few days, he continued to have episodic desaturations associated with obstructive apnea. On day 5 of life, he was taken to the operating room where rigid bronchoscopy and direct laryngoscopy revealed prolapse of the epiglottis over the larynx, obstructing the airway. Release of the genioglossus and a tongue–lip adhesion opened the airway and improved the epiglottic position. Soon thereafter, he had resection of a juxtaductal aortic coarctation, ligation of patent ductus arteriosus, and ligation of left superior vena cava.

The patient recovered uneventfully and was discharged at 2 weeks of age. Histological examination of the excised synechia revealed central fibrous and skeletal muscular tissue surrounded by squamous mucosal lining. Uneventful takedown of the tongue–lip adhesion and repair of cleft of the soft palate was performed at 9 months of age. At 14 months of age, the patient was doing well with no obvious signs of developmental delay and head circumference had improved to the 25th percentile. After discussion, the family declined any genetic testing.

**TABLE 1 Comparison of Disorders and Cases With Congenital Intraoral Synechia**

<i>Disorder or Case</i>	<i>Intraoral Band Cleft Palate</i>	<i>Cardiac Anomaly</i>	<i>Limb Anomaly</i>	<i>Other Features</i>
Congenital alveolar synechiae syndrome	+	–	–	none
van der Woude syndrome	+	–	–	lower lip pitting
Popliteal pterygium syndrome	+	–	+	hypoplastic genitalia
Oromandibular-limb hypogenesis spectrum	+	+	+	microstomia
Nakajima et al. (1979)	+	+	+	hydrocephalus
Zalzal et al. (1986)	+	–	+	hypospadias, bifid scrotum
Oh et al. (present)	+	+	+	microcephaly

## DISCUSSION

Intraoral synechiae in association with cardiac and/or digital anomalies have been previously described. Nakajima et al. (1979) reported an infant born with a large subglossopalatal membrane, velar cleft, ankylosis of the distal interphalangeal joints of the fifth fingers, absence of the distal phalanx of a fifth toe, and patent ductus arteriosus. A male infant with a subglossopalatal membrane, soft palatal cleft, bilateral hypoplastic small fingers, third-degree hypospadias, and a bifid scrotum, has also been described (Zalzal et al., 1986). These findings are similar to those observed in our patient (Table 1). Two patients with intraoral synechia and cardiac and digital anomalies were reported by Gartlan et al. (1993). Unlike our patient, these two infants did not have anterior subglossopalatal synechiae. One infant had a posterior intraoral band and another child had syngnathia.

Respiratory distress and feeding difficulties are the major causes of morbidity in infants with intraoral synechia (Chandra et al., 1974; Nakajima et al., 1979; Zalzal et al., 1986; Bagatin and Boric, 1990; Pillai et al., 1990; Gartlan et al., 1993; Denion et al., 2002; Kalu and Moss, 2004). Airway obstruction secondary to intraoral synechiae has been reported (Nakajima et al., 1979; Zalzal et al., 1986; Denion et al., 2002) and death due to respiratory arrest, bronchopneumonia, and severe dehydration has been documented (Chandra et al., 1974; Nakajima et al., 1979; Purohit et al., 1989; Gartlan et al., 1993). The subglossopalatal synechia in our patient was between the floor of the mouth (at the base of the lingual frenulum) and the posterior border of the hard palate. We believe this caused a disruption-type cleft of the soft palate because of failure of lingual descent, interfering with soft palatal closure. Although our patient's respiratory distress improved after excision of the intraoral band, he continued to have intermittent airway obstruction that required tongue–lip adhesion.

Intraoral synechiae can involve various sites, including the alveolar ridges (Dalal and Davison, 2002; Tanrikulu et al., 2005), tongue (Kalu and Moss, 2004), the lingual base (Chandra et al., 1974), floor of the mouth (Nakajima et al., 1979; Zalzal et al., 1986), and along the midline of the secondary palate (Chandra et al., 1974; Dalal and Davison, 2002; Kalu and Moss, 2004). Gartlan et al. (1993) suggested dividing oral synechiae into two categories: (1) posterior

intraoral bands originating near the base of the tongue, representing remnants of the buccopharyngeal membrane and (2) anterior intraoral bands, which are ectopic membranes that are subclassified as subglossopalatal bands, glossopalatal ankylosis, or syngnathia. They believe that a synechia in the anterior oral cavity does not derive from the buccopharyngeal membrane. *Per contra*, Gorlin et al. (1990) underscore that intraoral vestiges of the buccopharyngeal membrane would result in an anterior synechia because the tongue develops behind the buccopharyngeal membrane. Nevertheless, skeletal muscle in the subglossopalatal band of our patient supports the position of Gartlan et al. (1993) that midline anterior intraoral synechiae are not part of the buccopharyngeal membrane which, due to lack of mesodermal penetration, would not contain muscle fibers (Moore and Persaud, 2003).

Although the etiology of congenital oral synechiae is unknown, review of normal embryonic development provides pathophysiologic clues. The stomodeum or primitive mouth develops during the third week of intrauterine life as a transverse gap between the forebrain and the pericardium (Gartlan et al., 1993; Moore and Persaud, 2003). During the third to fourth weeks, the stomodeum and foregut are separated by the oropharyngeal or buccopharyngeal membrane, which is derived from the prechordal plate, a fusion of ectoderm and endoderm without mesodermal penetration (Moore and Persaud, 2003). This thin membrane disappears by the end of the fourth week (Gartlan et al., 1993; Moore and Persaud, 2003). The tongue, floor of the mouth, and palatal shelves develop in close contact with one another during the fifth and sixth weeks of prenatal life. By the eighth week, the tongue descends caudally and the palatal shelves become horizontal and fuse by the 12th week of development (Moore and Persaud, 2003). Remnants of embryologic tissue such as the buccopharyngeal membrane or abnormal adhesions between structures in close proximity (Gartlan et al., 1993) could give rise to intraoral bands during these phases of embryologic development (Kalu and Moss, 2004).

Although most cases of congenital intraoral bands have been sporadic, syngnathia associated with van der Woude syndrome has been reported (Denion et al., 2002; Puvabanditsin et al., 2003). Van der Woude syndrome is caused by mutations in interferon regulatory factor 6



(IRF6) (Kwang et al., 2002). Popliteal pterygium syndrome, which includes intraoral synechiae (Nauman and Schulman, 1961; Gartlan et al., 1993; Knoll et al., 2000), is also caused by mutations in IRF6 (Kwang et al., 2002). Testing for IRF6 was not done for the child in this case report, as the parents expressly refused all genetic investigations. In addition, congenital intraoral synechiae associated with IRF6 mutations represent sygnathism, although Denion and colleagues (2002) reported an infant with van der Woude syndrome and bilateral sygnathism with a subglossopalatal membrane.

Teratogenic medications have also been implicated, as several case reports have linked development of intraoral synechiae with maternal use of trimethobenzamide hydrochloride, meclizine hydrochloride, promethazine, imipramine, diazepam, chlorpromazine, and misoprostol during critical periods of facial and limb development (Nevin et al., 1980; Pauli and Greenlaw, 1982; Bokesoy et al., 1983; Pastuszak et al., 1998). Our patient's mother had no known exposure to teratogens. Others believe that mechanical factors, such as injury to the tip of the tongue or early rupture of amnion during intrauterine life, could cause intraoral adhesions (Spivack and Bennett, 1968; Torpin and Knoblich, 1969). No signs of intrauterine trauma were observed in our patient despite amniocentesis and pregnancy was uncomplicated. Finally, some have proposed a vascular etiology (David et al., 1992), with at least one report of fetal stem vessels exhibiting organized thrombi (Scott, 1991) and several cases of infants with oromandibular–limb hypogenesis spectrum associated with chorionic villus sampling (Firth et al., 1991, 1994; Scott, 1991; Hsieh et al., 1995; Chen et al., 1996).

Oromandibular–limb hypogenesis spectrum is comprised of sporadic disorders associated with intraoral synechiae, varying degrees of extremity abnormalities, cardiac anomalies, and microstomia (Grippaudo and Kennedy, 1998). Six entities that exhibit overlapping clinical features, fall into this group of syndromes, and include Moebius syndrome, hypoglossia–hypodactylia syndrome, Hanhart syndrome, glossopalatine ankylosis syndrome, limb deficiency–splenogonadal fusion syndrome, and Charlie M. syndrome (Hall, 1971; Kaplan et al., 1976). We believe that the close proximity of the face and heart in the early embryo (Jones, 2005), coupled with shared neural crest contributions to both developing regions (Kirby et al., 1983; Hutson and Kirby, 2003; Jones, 2005), may contribute to synchronous cardiac and facial malformations associated with oromandibular–limb hypogenesis spectrum, as well as other syndromes such as deletion 22q11 syndrome (velocardiofacial syndrome/DiGeorge syndrome), Noonan syndrome, fetal alcohol syndrome, and retinoid embryopathy syndrome. Patients with oromandibular–limb hypogenesis spectrum and intraoral soft-tissue bands (Purohit et al., 1989; Grippaudo and Kennedy, 1998) or cardiac anomalies have been reported (Caravella and Rogers, 1978; Bosch et al., 1984; Rarogue et al., 1988;

Deda et al., 2001; Suvarna et al., 2006). It is interesting to note that intraoral synechiae associated with oromandibular–limb hypogenesis spectrum are often subglossopalatal (Purohit et al., 1989; Grippaudo and Kennedy, 1998), as was the case in the child reported in this study.

As most cases of oromandibular–limb hypogenesis syndrome are sporadic, the paternal half-brother with a hypoplastic right fifth toe in this case report is likely aleatory. Nevertheless, it is plausible the half-sibling has decreased phenotypic expression of the same mutation possibly present in the proband. Several candidate genes, associated with oromandibular, limb, and cardiac development exist, including *Msx2* (Davidson et al., 1991; Jabs et al., 1993; Abdelwahid et al., 2001; Kwang et al., 2002; Brunelli et al., 2004), *Tbx22* (Braybrook et al., 2001, 2002; Marcano et al., 2004), sonic hedgehog (Imokawa and Yoshizato, 1998; Nanni et al., 1999; Bale, 2002), and *PVRL 1* (Suzuki et al., 2000). Nonetheless, these mutations, to date, have not been documented in patients with congenital intraoral synechiae.

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