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Craniofacial profile in Southern Chinese with hypodontia

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SUMMARY The association between craniofacial morphology and congenitally missing teeth is at present unclear. The aims of this study were to investigate whether hypodontia is associated with changes in the sagittal skeletal profile and to identify putative relationships between the skeletal profile and the severity of hypodontia.

In a cross-sectional analytical study, the craniofacial structure and profile based on two-dimensional lateral cephalograms of Southern Chinese hypodontia patients ($n = 49$, 24 males, 25 females, mean age 16.4 years) and a comparison group without hypodontia ($n = 41$, 15 males, 26 females, mean age 16.7 years) were compared. The hypodontia patients were divided into three subgroups according to the severity of hypodontia (mild: ≤5, moderate: 6–9, and severe: ≥10 congenitally missing permanent teeth).

All hypodontia patients had a significantly reduced mandibular plane, ANB, and face height compared with the control group ($P < 0.05$). A significant increase in chin thickness was also observed in the hypodontia patients ($P < 0.05$). As the severity of hypodontia increased from moderate to severe, a tendency to develop a retrognathic maxilla and a Class III skeletal relationship was noted in addition to the above features, making the already thick chin even more prominent. Statistically significant correlations (Pearson's correlation coefficient) were found between the number of missing teeth and SNA, NAFH, and ANB angles, the mandibular plane, chin thickness, and face height.

In Southern Chinese subjects, hypodontia was associated with a shorter face, a flatter mandibular plane, a more pronounced chin, and a Class III skeletal profile. In severe hypodontia subjects, the maxilla was more retrognathic with a greater predilection to a Class III skeletal relationship.

Introduction

Hypodontia is a common developmental anomaly characterized by the absence of one or more primary or secondary teeth, other than the third molars (Larmour *et al.*, 2005). The prevalence of hypodontia in different populations throughout the world varies considerably and has been reported to be 3.5–8 per cent (Pilo *et al.*, 1987). A meta-analysis (Polder *et al.*, 2004) of the prevalence of dental agenesis of permanent teeth suggested that agenesis differed according to geographic location and gender and was higher in Europe (males 4.6 per cent; females 6.3 per cent) and Australia (males 5.5 per cent; females 7.6 per cent). In general, the prevalence of dental agenesis has been reported to be 1.4 times higher in females than in males.

Hypodontia may present as an independent anomaly or may be associated with a craniofacial syndrome. There are more than 160 syndromes that may include hypodontia [Hallonsten and Koch, 1996, Online Mendelian Inheritance in Man (OMIM)]. Often, hypodontia is the most easily detectable part of the syndrome and thus may be an important factor in diagnosis. A genetic component has been demonstrated in non-syndromic hypodontia (Vastardis *et al.*, 1996; Stockton *et al.*, 2000). The *PAX9* gene has a key role in odontogenesis and is linked to familial and sporadic tooth agenesis (Pan *et al.*, 2008). The *MSX1* and *AXIN2* genes that affect early tooth development are also associated with tooth agenesis (Bailleul-Forestier *et al.*, 2008).

The association between craniofacial morphology and congenitally missing teeth is presently not well established. It appears that hypodontia has little effect on the general growth pattern of the jaws (Sarnat *et al.*, 1953; Roald *et al.*, 1982) and there is little correlation between missing teeth and changes in cephalometric measurements when compared with normative values (Dermaut *et al.*, 1986; Chung *et al.*, 2000). However, some authors have noted that, in subjects with hypodontia, the maxilla is more retrognathic (Wisth *et al.*, 1974; Sarnäs and Rune, 1983; Woodworth *et al.*, 1985). Tavajohi-Kermani *et al.* (2002) proposed that a decreased maxillary jaw size and maxillary tooth agenesis were generally related. They stated that most age groups had significant decreases in maxillary jaw size associated with tooth agenesis as revealed by two-dimensional (2D) area measurement. There is also a view that hypodontia influences skeletal growth of the maxilla and mandible but that the impact is insignificant unless the hypodontia is severe (Bondarets and McDonald, 2000; Bondarets *et al.*, 2002). Bondarets and McDonald (2000) found that hypodontia subjects with more than six missing teeth had a reduced anterior face height and mandibular inclination. A more prognathic mandible and reduced mandibular plane inclination were also observed in patients with more than 12 missing teeth (Nodal *et al.*, 1994).

The aims of the present study were to investigate whether hypodontia is associated with changes in the sagittal skeletal profile in Southern Chinese in Hong Kong and to identify potential relationships between the skeletal profile and severity of hypodontia.

Subjects and methods

Study design and sample

This was a cross-sectional analytical study which included two groups of patients: a hypodontia group with a variable number of missing teeth and a comparison group with all teeth other than the third molars present.

All hypodontia patients seen in a multidisciplinary hypodontia clinic at the Prince Philip Dental Hospital, University of Hong Kong, during 2005 were considered for inclusion in the study. The hypodontia clinic is the only specialist unit of its kind in Hong Kong and receives referrals from throughout Hong Kong. From patient records, the following clinical information was recorded: age, gender, site and number of missing permanent teeth, family history, and any known syndrome. Information from the patient records of a total of 90 Southern Chinese patients (39 males and 51 females) aged 9–30 years was included in the study. The hypodontia group comprised 49 patients (24 males and 25 females) with a mean age 16.4 years [standard deviation (SD) 5.0]. The hypodontia patients were further divided into three subgroups according to the severity of hypodontia. Those with up to five missing teeth were classified as having mild hypodontia, 6–9 missing teeth as having moderate hypodontia, and 10 or more missing teeth as severe hypodontia.

The control group was formed by random selection of patients who had a normal number of teeth, excluding third molars, and a Class I skeletal profile from the patient records of the Discipline of Pediatric Dentistry and Orthodontics. The control group consisted of 41 patients (15 males and 26 females) with a mean age of 16.7 years (SD 5.5).

Inclusion and exclusion criteria

Patients with congenital craniofacial syndromes related to hypodontia such as ectodermal dysplasia, cleft lip and palate, and Williams' Syndrome were excluded. Patients with diseases and syndromes associated with a typical facial appearance, for instance Down syndrome and gigantism, were also excluded. A requirement for inclusion into either group was the availability of a pre-orthodontic treatment lateral cephalogram.

Radiographic assessment

The radiographs for all subjects were taken in the same conventional cephalostat. The distance from the focus to the median plane of the head was fixed while the distance from the median plane of the head to the film varied. The radiographs were taken with the patient in the standing position, the teeth in habitual occlusion, and the Frankfort plane parallel to the floor.

Tracing and computing techniques

Craniofacial morphology for each subject was first evaluated with a traditional cephalometric procedure. Hard and soft tissue cephalometric landmarks were marked. The radiographic images were then scanned prior to digitization. A transmissive scanner (Scanmaker 9800 XL, Microtek, Hsinchu, Taiwan) at 400 dots per inch was used to transfer the images into the computer-assisted simulation system for orthognathic surgery (CASSOS) 2001 software program (2000 SoftEnable Technology, Hong Kong). The radiographic images were then digitized using a computer. Seventy-one soft and hard tissue landmarks were recorded for each cephalograph. A customized cephalometric analysis consisting of 23 standard angular and linear measurements was used in this study.

Error analysis

One investigator (DWSC) scanned the images into the CASSOS program and digitized the hard and soft tissue landmarks. Intra-operator error was evaluated by re-digitizing 20 cephalographs 2 weeks after initial digitization. Error analysis was performed using paired *t*-tests. There was no statistically significant difference between the cephalometric analyses ($P > 0.050$, Table 1).

All data were analyzed using the Statistical Package for Social Sciences (version 11.5, SPSS Inc., Chicago, Illinois, USA).

Results

There were no statistically significant differences in age ($P = 0.777$) between the hypodontia and control groups or male to female proportions within each group (hypodontia, $P = 0.886$; control group, $P = 0.090$).

In the hypodontia group, there were a total of 352 congenitally missing teeth (mean 7.2, SD 5.1; range 1–20). Five individuals had only one tooth missing and one subject had 20 missing teeth. Fifty-one per cent had six or fewer missing teeth (Figure 1). In terms of tooth groups, the mandibular central incisors and mandibular and maxillary second premolars were the three most commonly missing teeth. The distribution of missing tooth types is shown in Figure 2.

The majority of patients (34, 69.4 per cent) had hypodontia in both jaws. Three (6.1 per cent) had only maxillary tooth agenesis and 12 (24.5 per cent) had mandibular tooth agenesis. Bilateral missing teeth were far more common than unilateral (83.7 per cent versus 16.3 per cent).

Angular and linear cephalometric measurements for the hypodontia and control groups are shown in Table 2. Five of 23 measurements showed a statistically significant difference between the hypodontia and comparison groups ($P < 0.05$).

The mandibular plane was found to be flatter in the hypodontia group compared with the control group as indicated by the decrease in MPFH and MPSN values (P = 0.009 and P = 0.003, respectively). MPFH refers to the angle between the mandibular plane (the line tangent to menton and gonion) and the Frankfort Horizontal plane (from porion to orbitale). Similarly, MPSN refers to the angle between the mandibular plane and the sella-nasion line.

The y-axis, an assessment of the facial growth axis, was also smaller in the hypodontia patients (P = 0.004). ANB, indicating the sagittal relationship between the maxillary and mandibular alveolar bases, was also found to be significantly smaller in the hypodontia group (P = 0.022). Furthermore, the thickness of the chin button, as measured by the distance from pogonion to the Na-B line, was significantly longer in the hypodontia patients than in the control group, indicating that the hypodontia patients on the whole appeared to have a thicker chin button.

The relationship between the severity of hypodontia and craniofacial morphology was further investigated by dividing the hypodontia patients into three subgroups. There were 21 patients (43 per cent) in the mild subgroup (≤5 missing teeth), 15 patients (31 per cent) in the moderate subgroup

Table 1 Error analysis: validation of cephalometric analysis.

	First tracing		Second tracing		P value
	Mean	SD	Mean	SD	
SNA (°)	80.96	1.90	80.21	2.83	0.247
NAFH (°)	88.48	2.58	87.49	3.56	0.128
FA (°)	87.61	3.59	87.64	3.45	0.942
SNB (°)	79.10	2.32	78.49	3.29	0.591
ACB (mm)	68.31	7.18	72.29	10.20	0.225
GoM:SN (%)	104.49	6.95	100.34	24.83	0.540
MPFH (°)	25.23	5.92	26.04	6.48	0.278
MPSN (°)	32.76	4.40	32.85	4.82	0.907
ANB (°)	1.86	2.39	0.86	1.38	0.076
PPMP (°)	24.99	6.97	25.35	6.25	0.677
YFH (°)	60.83	4.56	60.87	4.24	0.895
Percentage nose (%)	49.02	4.40	47.03	3.40	0.322
NA:AGn (%)	106.7	14.53	106.88	9.81	0.952
Pog to NB (mm)	1.97	1.91	2.06	1.76	0.607
UD-SN (°)	104.50	4.93	105.82	6.39	0.459
U1-SN (°)	103.26	5.17	103.14	5.79	0.902
U1PP (°)	111.05	4.64	110.62	5.27	0.726
U1-L1 (°)	135.77	9.65	136.51	10.66	0.666
L1-MP (°)	88.21	5.57	87.51	5.43	0.559
OP-FH (°)	7.81	5.49	7.57	3.98	0.787
UOP-FH (°)	10.31	4.70	10.74	3.85	0.533
Overjet (mm)	3.82	2.00	3.69	1.88	0.484
Overbite (mm)	3.33	2.39	3.37	2.25	0.908

Paired t-tests; SD, standard deviation.

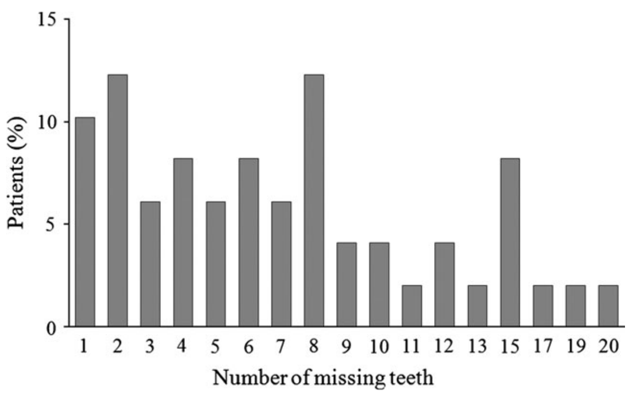

Figure 1 Distribution of missing teeth (n = 352) among patients in the hypodontia group (n = 49).

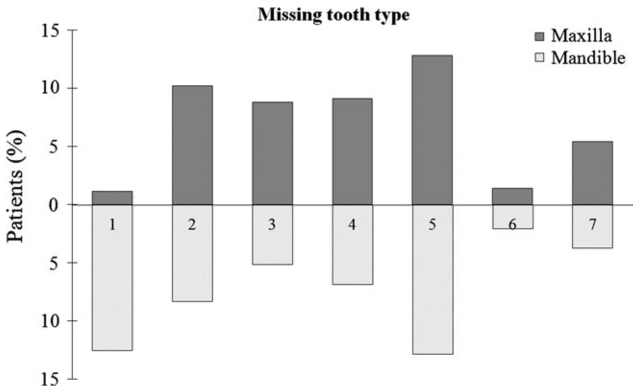

Figure 2 Distribution of missing tooth type in the hypodontia group (n = 49).

Table 2 Cephalometric measurements in the hypodontia and control groups.

	Hypodontia (n = 49)		Control (n = 41)		P value
	Mean	SD	Mean	SD	
SNA (°)	80.75	3.28	81.58	3.00	0.216
NAFH (°)	88.00	4.02	89.30	3.22	0.093
FA (°)	87.80	3.62	86.61	2.77	0.089
SNB (°)	79.74	3.95	78.90	3.29	0.282
ACB (mm)	68.68	6.51	69.67	3.41	0.386
GoM:SN (%)	105.80	7.38	105.07	7.28	0.636
MPFH (°)	24.89	6.23	28.36	6.07	0.009*
MPSN (°)	32.10	5.48	36.01	6.66	0.003*
ANB (mm)	1.04	4.02	2.67	2.14	0.022*
PPMP (°)	23.41	6.26	25.95	6.43	0.062
YFH (°)	60.45	3.69	62.67	3.37	0.004*
Percentage nose (%)	46.07	4.64	46.74	6.82	0.595
NA:AGn (%)	105.36	11.12	103.91	7.96	0.473
Pog to NB (mm)	1.65	2.24	−0.07	1.55	0.000*
UD-SN (°)	107.40	7.58	107.58	5.78	0.901
U1-SN (°)	102.56	18.02	105.86	6.34	0.268
U1PP (°)	111.03	19.02	115.77	6.55	0.131
U1-L1 (°)	128.00	26.59	125.80	10.14	0.617
L1-MP (°)	91.21	8.51	90.20	14.70	0.703
OP-FH (°)	10.81	5.95	9.70	4.23	0.337
UOP-FH (°)	12.65	6.64	11.30	4.18	0.269
Overjet (mm)	2.78	4.79	3.41	1.95	0.438
Overbite (mm)	2.46	3.26	2.32	1.83	0.824

Independent t-test; SD, standard deviation.
*P < 0.05.

(6–9 missing teeth), and 13 patients (27 per cent) in the severe subgroup (≥10 missing teeth). There were statistically significant differences in SNA and NAFH angles in addition to the five previously mentioned cephalometric parameters between the hypodontia subgroups (Table 3). SNA and NAFH (angle between the NA line and Frankfort Horizontal plane) indicates the position of the maxilla in relation to the cranial base. These values were significantly reduced in the severe hypodontia subgroup (analysis of variance, P = 0.045 and P = 0.006, respectively, Table 3). Statistical testing (Student–Newman–Keuls test) showed that SNA and NAFH values were significantly smaller in the severe subgroup compared with the mild and moderate subgroups.

Correlations between the number of missing teeth and cephalometric measurements were confirmed for SNA, NAFH, MPSN, ANB, the *y*-axis, Pog-NB, and U1-SN (the angle between the maxillary central incisor and the SN plane; Pearson's correlation test, $P < 0.05$). None of the correlations was strong since the correlation coefficient ranged from 0.22 to 0.37 (Table 4).

Discussion

The four most frequently missing teeth found in this study were the maxillary and mandibular premolars, mandibular central incisors, and maxillary lateral incisors. This finding is in agreement with the classic pattern of missing teeth proposed by Rasmussen (1999) but differs from other studies which suggest that the mandibular incisors are the most commonly missing teeth in Southern Chinese (Davis and Darvell, 1993; Larmour *et al.*, 2005). The majority of patients (83.7 per cent) presented with bilateral agenesis with the unilateral condition only encountered in the mild hypodontia subgroup. This contrasts with the report by Polder *et al.* (2004) that unilateral dental agenesis is more common than bilateral agenesis.

There is generally no consensus in the literature about the relationship between tooth agenesis and the sagittal position of the maxilla and mandible. In this study, when the hypodontia and control groups were compared, the sagittal positions of the maxilla and mandible were found to be within normal limits. This contrasts with the findings of Wisth *et al.* (1974), who reported a reduction in maxillary prognathism in patients with hypodontia, and Woodworth *et al.* (1985), who noted a retrusive maxilla in patients with bilateral congenital absence of the maxillary lateral incisors. However, a notable difference emerged when the hypodontia patients in the present study were divided into different severity subgroups, that is the maxilla was significantly retrognathic in those patients with severe hypodontia. This observation is in agreement with Øgaard and Krogstad (1995) who found that hypodontia patients with 10 or more missing teeth had a significant reduction in SNA, and with Nodal *et al.* (1994) who reported similar findings in patients with more than 13 missing teeth.

Table 3 Cephalometric measurements in the three hypodontia subgroups.

	Mild		Moderate		Severe		*P* value
	Mean	SD	Mean	SD	Mean	SD	
SNA (°)	81.44	3.33	81.42	3.24	78.84	2.63	0.045*
NAFH (°)	88.73	3.70	89.31	3.44	85.35	4.16	0.006*
FA (°)	87.30	3.52	87.80	3.96	88.60	3.16	0.250
SNB (°)	79.35	3.24	79.17	4.22	81.02	4.64	0.342
ACB (mm)	68.60	7.17	67.54	5.21	70.12	6.93	0.500
GoM:SN (%)	104.00	6.33	106.19	7.04	108.26	9.00	0.393
MPFH (°)	23.95	7.16	26.33	5.38	24.75	5.64	0.046*
MPSN (°)	31.21	5.71	34.21	5.56	31.12	4.66	0.009*
ANB (°)	2.07	3.63	2.25	2.92	−2.03	4.38	0.000*
PPMP (°)	23.53	6.59	25.44	6.14	20.86	5.29	0.068
YFH (°)	60.82	3.92	61.25	3.63	58.95	3.13	0.009*
Percentage nose (%)	45.48	5.73	45.77	4.36	47.39	2.59	0.742
NA:AGn (%)	104.48	9.19	101.85	9.44	110.82	14.16	0.082
Pog to NB (mm)	1.38	2.19	1.38	2.26	2.38	2.30	0.000*
UD-SN (°)	109.21	6.56	104.16	8.23	108.22	7.77	0.163
U1-SN (°)	107.73	9.11	100.44	11.16	96.63	30.41	0.077
U1PP (°)	115.47	8.97	109.22	11.51	106.02	33.18	0.119
U1-L1 (°)	125.10	12.25	138.44	12.52	117.00	55.32	0.075
L1-MP (°)	95.55	7.89	87.87	5.55	87.24	9.70	0.161
OP-FH (°)	10.25	6.21	10.58	4.71	12.73	7.66	0.548
UOP-FH (°)	11.53	6.04	13.49	5.32	13.82	9.37	0.427
Overjet (mm)	3.52	4.74	3.42	1.97	−0.400	7.46	0.062
Overbite (mm)	2.58	3.16	2.86	1.95	1.34	5.29	0.652

One-way analysis of variance; SD, standard deviation.
$^*P < 0.05$.

Table 4 Correlations between the number of missing teeth and cephalometric measurements.

Number of missing teeth against	*P* value	Pearson's correlation coefficient
SNA	0.021*	−0.244
NAFH	0.015*	−0.257
FA	0.108	0.170
SNB	0.325	0.105
ACB	0.745	−0.035
MP-FH	0.113	−0.168
GOM:SN	0.418	0.086
MPSN	0.055*	−0.203
ANB	0.002*	−0.328
PPMP	0.034	−0.223
YFH	0.003*	−0.308
% Nose	0.899	0.014
NA:AGN	0.065	0.195
Pog to NB	0.00*	0.368
UD-SN	0.561	−0.062
U1-SN	0.032*	−0.227
U1PP	0.043	−0.214
U1-L1	0.543	−0.068
L1-MP	0.482	−0.078
OP-FH	0.219	0.139
UOP-FH	0.155	0.155
Overjet	0.215	−0.140
Overbite	0.904	−0.014

*$P < 0.05$.

The more retrognathic maxilla in the severe hypodontia subgroup is a probable explanation for the significantly reduced ANB in that group. It was found in the present study that tooth agenesis had no significant effect on the length and prognathism of the mandible. The increasing tendency towards a Class III skeletal relationship in severe hypodontia subjects supports the findings of Chung *et al.* (2000) who reported the linkage of more severe hypodontia with a tendency to a Class III skeletal relationship.

Face height was also found to be significantly different between the hypodontia and control groups. Previously, it has been shown that patients with anodontia develop a normal face height and in hypodontia patients a slightly increased face height (Sarnat *et al.*, 1953; Ochiai *et al.*, 1961; Mizushima *et al.*, 1962; Wisth *et al.*, 1974; Dermaut *et al.*, 1986). In the present study, it was noted that the hypodontia patients had a significantly shorter face compared with the control group. This is in agreement with the findings of Bondarets and McDonald (2000) and Woodworth *et al.* (1985) and is perhaps not surprising because it is believed that anterior face height is affected by the eruption of teeth and vertical growth of the soft tissues. The presence of teeth is a contributing factor in establishing growth of the alveolar process in both the maxilla and the mandible.

Consistent with the literature (Nodal *et al.*, 1994; Bondarets and McDonald, 2000), the mandibular plane was significantly reduced. A novel finding in this study related to chin thickness which appeared to be significantly increased. It is speculated that the flatter mandibular plane and a more prominent chin might be related to anterior rotation of the mandible.

Another area of controversy in the literature on hypodontia is the angulation of the teeth. The teeth have been reported to be more retroclined than normal (Bondarets and McDonald, 2000), showing a tendency towards protrusion in both jaws (Yüksel and Üçem, 1997) or more upright, especially in the mandible (Roald *et al.*, 1982). The latter authors suggested that their finding might be explained by the greater effect of the lip musculature on the anterior teeth in hypodontia patients. In the present study, no statistically significant relationship was found between angulation of the anterior teeth and the severity of hypodontia or between tooth angulation in the hypodontia patients compared with the controls.

It is acknowledged that the prevalence of hypodontia in Hong Kong cannot be established because recruitment into study was made from a pool of hypodontia patients attending the Prince Philip Dental Hospital. Furthermore, the study group was skewed because only those subjects who had treatment in the dental hospital and had a lateral cephalograph taken were included. If hypodontia patients, especially mild cases, do not have a significant malocclusion or an atypical facial appearance, it is probable that they would not be referred to the dental hospital. This study also did not include patients who sought dental treatment in the private sector.

A second limitation relates to patient selection. While hospital patients with a normal number of teeth and a Class I skeletal profile were considered as the control group, their cephalometric measurements may still be slightly different from the published means for normal populations. Some subjects may have a mildly retrusive maxilla with compensation by incisor angulation, thus affecting comparisons.

The fact that the maxilla and mandible continue to grow during the adolescent period should also be considered. Maxillary growth ceases on average at about 15 years of age in females and 17 years in males. Mandibular growth ceases rather later than maxillary growth, on average at about 17 years in females and 19 years in males and may continue even longer (Mitchell, 1996). Inclusion of hypodontia patients at a relatively young age may also have lead to an overestimation of the effect of tooth agenesis on craniofacial growth.

Conclusion

Based on 2D lateral cephalograms, overall, hypodontia patients tended to have a shorter face and flatter mandible compared with Class I subjects. As the severity of hypodontia increased to 10 or more missing teeth, a tendency to develop a retrognathic maxilla and a Class III skeletal relationship was noted in addition to the above features, making the already thick chin even more prominent.

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