Eruption of the central incisor, the intermaxillary suture, and maxillary growth in patients with a single median maxillary central incisor

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The occurrence of a single median maxillary central incisor (SMMCI) is a very rare condition and might be a sign of a more severe midline defect, which could be a mild degree of holoprosencephaly. Absence of the internasal and partial absence of the intermaxillary suture has been observed in a fetus with holoprosencephaly. The purpose of this study was to evaluate the intermaxillary suture, the eruption pattern of the single central incisor in the SMMCI condition, and the growth of the maxilla in a group of patients with SMMCI. A similar study was not found in the scientific literature. The material included orthopantomographs, dental radiographs, and lateral cephalometric radiographs from 11 patients with an SMMCI. The orthopantomographs and dental radiographs showed that the intermaxillary suture was abnormal anterior to the incisive foramen; however, the SMMCI erupted within the expected time interval. Superimposition on stable structures on lateral cephalometric radiographs from two untreated patients, in which growth analysis was possible, showed that the horizontal and vertical growth of the maxilla was normal. Due to the sutural midline defect it is suggested that a transversal growth analysis is included in all treatment planning of SMMCI patients. □ *Eruption; maxillary growth; single median maxillary central incisor*

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Agenesis of an upper central incisor is a very rare condition. In the literature this developmental disorder is defined as the solitary median maxillary central incisor (SMMCI) syndrome, and the condition is estimated to occur once in every 50,000 live births (1).

To most geneticists a patient with an SMMCI raises the possibility of an association with the serious developmental anomaly holoprosencephaly. It is suggested that an SMMCI may be considered an indicator of potential holoprosencephaly in the next generation (2, 3). Holoprosencephaly is a developmental defect affecting the forebrain and the face. An impaired midline formation of the embryogenic face and forebrain is the basic defect, and it shows a wide phenotypic variation (4, 5).

The chromosome abnormalities thus far described in the literature associated with SMMCI are those involving chromosomes 7, 13, 18, and 22 (1).

Many medical and dental clinicians have discussed the significance of this dental midline defect in patients with an SMMCI. Scott (6) first reported the presence of an SMMCI, with normal crown dimensions situated precisely in the midline. Since then several developmental disorders has been described. The condition has been associated with holoprosencephaly (3, 7, 8), short stature, pituitary and endocrinological deficiencies (1, 9), brain malformation and mental retardation (1, 10), hypotelorism, congenital nasal pyriform aperture stenosis (CNPAS), choanal atresia, a midpalatal ridge (1, 3, 11), and VACTERL association (vertebral anomalies, anal atresia, tracheo-oesophageal fistula, renal defects, and limb dysplasia) (12).

The face, palate, dentition, nasal cavity, and craniofacial morphology in children with SMMCI have been studied by Kjær et al. (10). The subjects described by them (10) had a characteristic external nose, an arch-shaped upper lip, and an indistinct philtrum, and intraorally all patients lacked the fraenulum of the upper lip and the incisive papilla. In addition, 9 of 10 subjects had a characteristic midaxial ridge in the palate. The analysis of the craniofacial profile in these children showed that all had a short anterior cranial base, and most had a retrognathic maxilla (10).

In a recent study it was shown in a human fetus with holoprosencephaly and an SMMCI that the internasal suture and the intermaxillary suture in the region anterior to the central incisor were absent (13). Furthermore, malformations of the nasal septum and crista galli were described (13).

It has not, to our knowledge, been ascertained whether SMMCI patients have the same midline suture deviations. It is of interest to investigate this, since such a midline defect could affect both eruption of the SMMCI and growth of the maxilla. The purpose of the present study was therefore to evaluate the extension of the intermaxillary suture in SMMCI patients and, furthermore, to

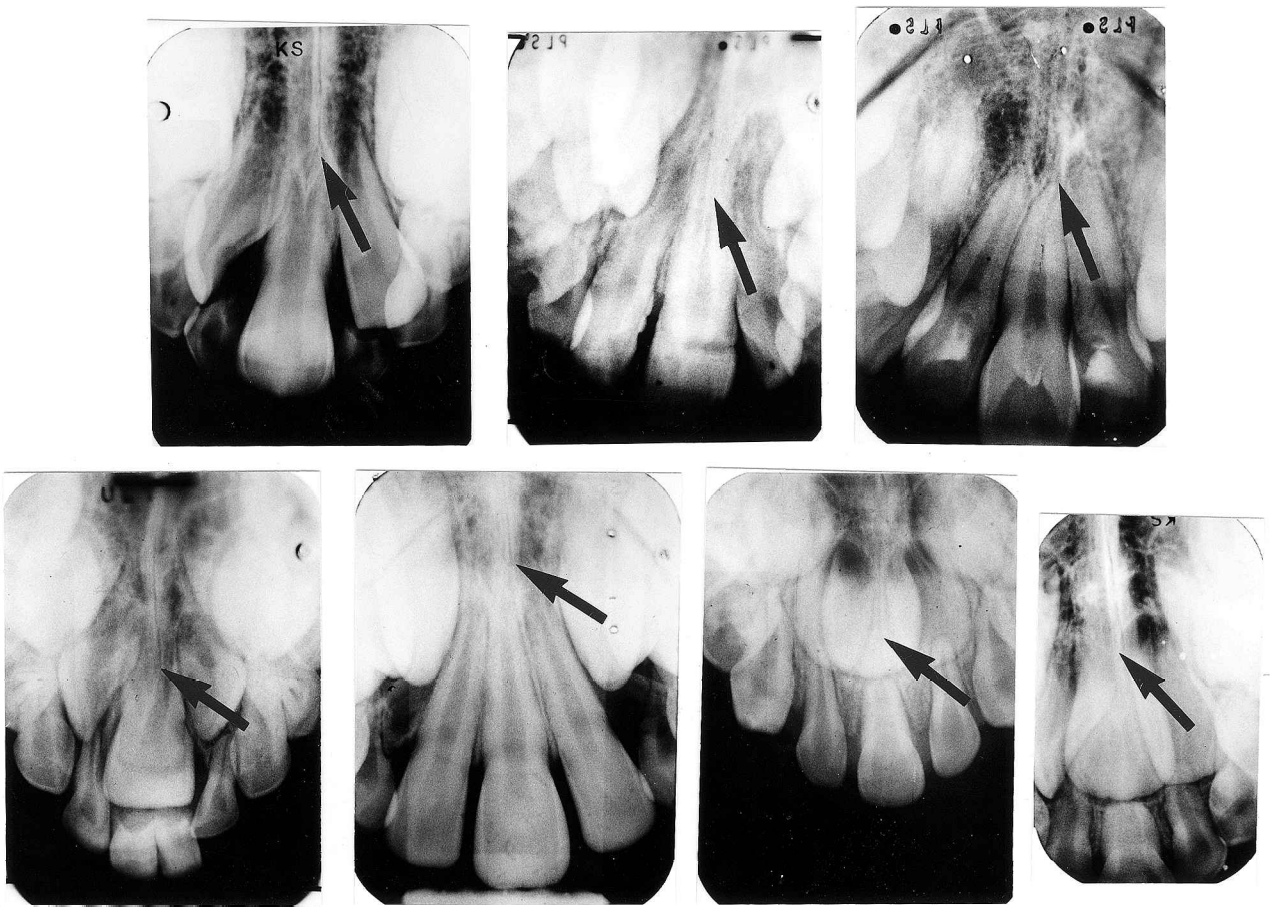

Fig. 1. Morphology of the interincisal part of the intermaxillary suture in seven of the subjects with a single median maxillary central incisor. The arrows indicate the location of the caudal end of the suture. In all subjects the suture appeared malformed and not visible in the incisal alveolar bone.

elucidate the eruption pattern of the single central maxillary incisor and the growth of the maxillary bone.

Subjects and methods

Subjects

The sample included 11 patients (10 females, 1 male) with an age range of 8–17 years. Nine of the patients were from the community care system in Denmark, one patient from a private practice in Germany, and one patient from a public University Hospital in Germany. The 11 subjects all had an SMMCI.

The material comprised orthopantomographs and dental radiographs. From two orthodontically untreated female patients two lateral cephalometric films taken at various time periods were available for studying the maxillary growth pattern.

Method

The morphology of the intermaxillary sutures was studied on orthopantomographs and dental radiographs, and the eruption pattern of the SMMCI was recorded from the same radiographs. From 2 of the 11 patients lateral cephalometric radiographs existed, taken at different ages before orthodontic treatment. The earliest and the latest lateral cephalometric radiographs from the two untreated subjects were superimposed on anatomically stable structures in accordance with the method described by Björk (14). For detection of errors of superimposition, a control tracing was prepared for each set of lateral radiographs. This is a procedure recommended by Björk & Skieller (15) for routine clinical purposes.

From the correctly superimposed tracings the growth changes in the sagittal and vertical dimension of the maxilla were assessed. The sagittal translation of the maxilla was assessed by angular measurements in accordance with the method described by Iseri & Solow (16). The nasion and sella points of the first film served as

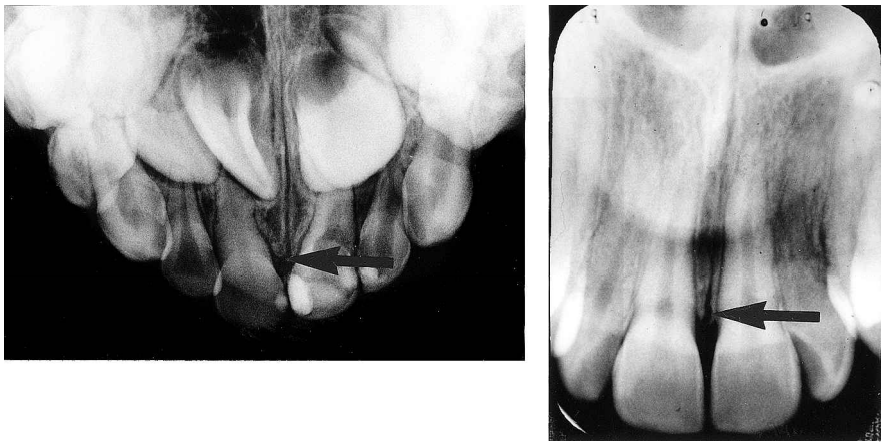

Fig. 2. Normal morphology of the interincisal part of the intermaxillary suture for comparison with Fig. 1. The arrows indicate the normal incisal extension of the suture.

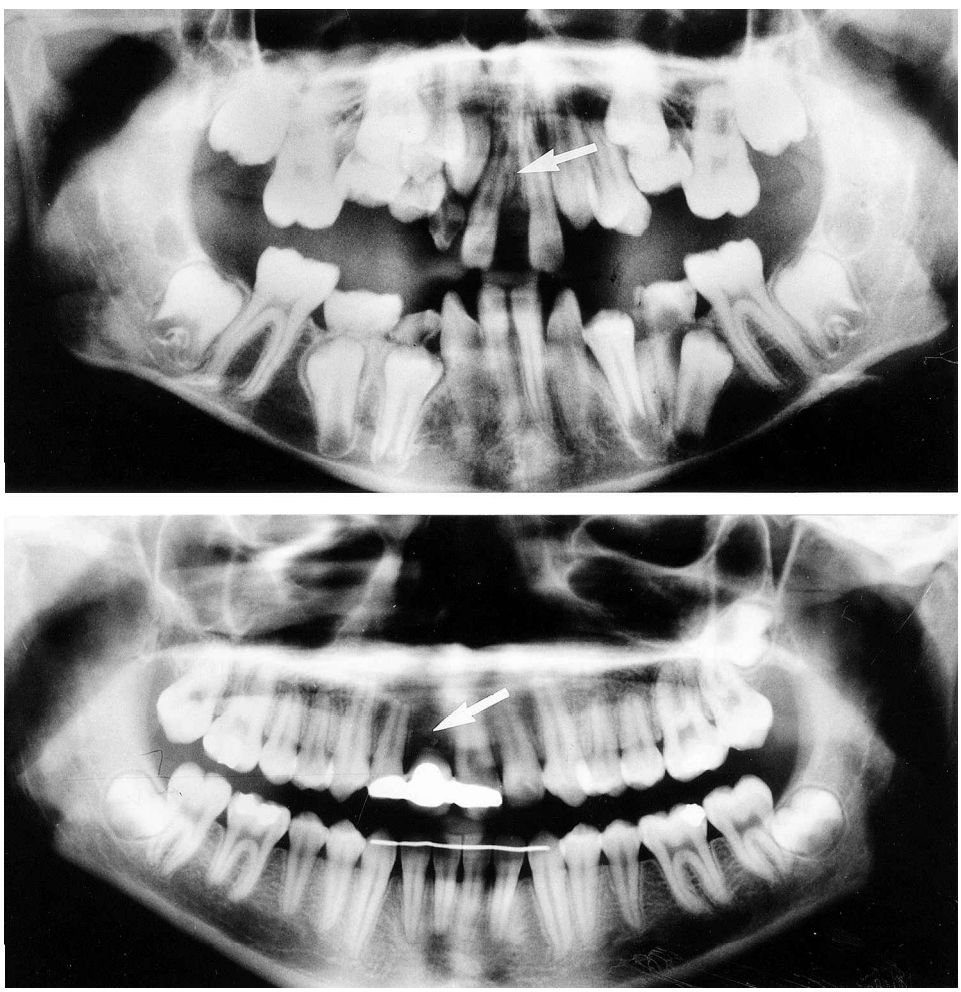

Fig. 3. Orthopantomogram of two female subjects in whom the malformation in the area of the interincisal part of the intermaxillary suture can be observed. Absence of the sutures in the regions marked by arrows.

Table 1. Age at which the single median maxillary central incisor (SMMCI) erupted

Subject no.	Age when investigated (years)	Emerged*
1	8	+
2	6	–
	8	+
3	5	–
	8	+
4	8	+
5	11	+
6	12	+
7	7	–
	9	+
8	11	+
9	7	–
10	5	–
	8	+
11	?	–
	9	+

* – Indicates that the SMMCI is not visible clinically; + indicates that the SMMCI has emerged through the gingiva.

fiducial points, s_t (sella transferred) and n_t (nasion transferred), and the angle s_t–n_t–ss_2 was used to assess the true forward angular displacement of the maxilla. The angle s_t–n_t–ss_2 is similar to the angle s–n–ss_2 but eliminates the apposition at nasion and shows the true forward angular displacement of the maxilla because its upper anterior reference point is the transferred nasion point from stage 1. The true forward displacement of the maxilla in the two females investigated was compared with a normal female reference group (16).

Results

In all 11 patients malformations of the intermaxillary suture superior to the SMMCI were observed in the primary and permanent dentition; furthermore, the suture was not visible in the incisal alveolar bone.

It was difficult to evaluate the precise extension because of the position of the central incisor in the midline (Fig. 1). The normal morphology of the intermaxillary suture in the primary and permanent dentition can be seen in Fig. 2.

In four subjects the anterior incisor was extracted in accordance with the treatment plan. In these subjects radiographs of the incisor region after extraction showed the absence of the interincisal part of the intermaxilllary suture (Fig. 3)

The eruption pattern of the SMMCI occurred within the normal age interval (Table 1).

The growth tracings from the two subjects are shown in

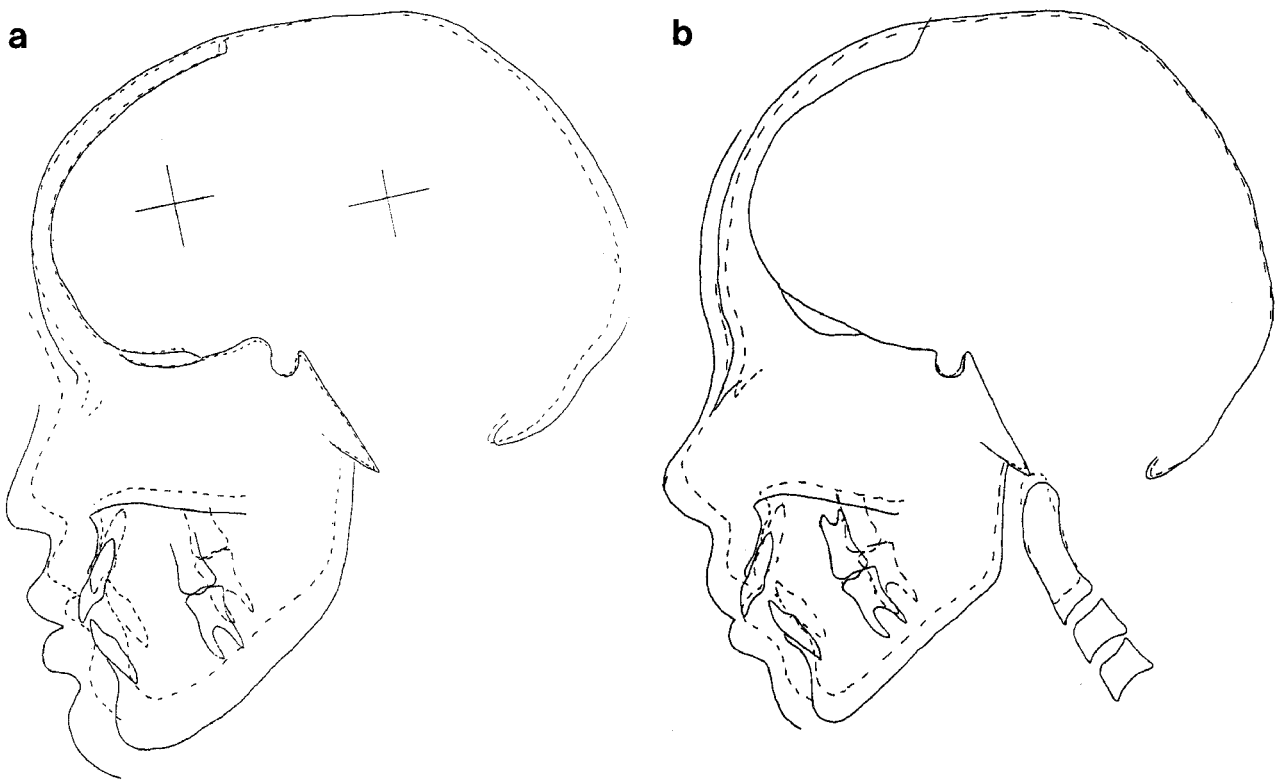

Fig. 4. Superimposition tracing of lateral cephalometric radiographs of two patients with a single median maxillary central incisor (SMMCI) before orthodontic treatment. Fig. 4A. Subject 1. Fig. 4B. Subject 2. The craniofacial growth pattern in the two female patients with an SMMCI, showing normal sagittal and vertical growth of the maxilla.

Table 2. Maxillary growth in two subjects with a single median maxillary central incisor (SMMCI). Forward translation compared with normal standard for individual subjects

Subject no.	Time period, A → B (years)	Study group: Profile 2 s_t–n_t–ss_2 and Profile 1 s–n–ss_1 (degrees)	Reference group: Expected forward translation of the maxilla (degrees)
1	8^{11}–14^{5}	3.5	4
2	10^{4}–17^{0}	2.5	3

Superscript numbers refer to months; Subjects 1 and 2 refer to Subjects 1 and 2 in Table 1. Comparison of the forward translation of the maxilla in the two female subjects compared with the forward displacement for the same time period for the reference group in accordance with Iseri & Solow (16).

Fig. 4. The true forward angular displacements of the maxilla in the study group was 2.5° for Subject 1 and 3.5° for Subject 2 (Table 2)

Discussion

Patients with an SMMCI often need orthodontic treatment. For treatment planning it is an important question whether the central incisor is erupting normally, even though a suture defect occurs in the incisor region, and the question of maxillary growth is also essential. In the present study these questions are elucidated on material in which it has been possible to study the eruption pattern, the morphology of the anterior part of the intermaxillary suture, and the growth of the maxilla in the sagittal and vertical direction.

It was shown that the eruption time of the single central incisor in the SMMCI condition was normal. The intermaxillary suture in the incisor region was abnormal. This interincisal midline defect did not influence the eruption pattern.

The craniofacial growth pattern in two patients with a SMMCI showed normal sagittal and vertical growth of the maxilla.

The craniofacial morphology of patients with an SMMCI has been described earlier (10), and the morphology of the intermaxillary and internasal suture in a fetus with holoprosencephaly has also been described (11).

Craniofacial growth analysis has not formerly been undertaken in a sample like the present one, in which a midline defect occurred in the maxilla and the dentition. In the group of 11 patients it was only possible to study the craniofacial growth in 2 subjects, since the other 9 had all received orthodontic treatment. Because the condition is estimated to occur only once in 50,000 live births (1), it is difficult to select a more homogeneous sample.

However, it is interesting to observe that the sagittal and vertical growth seemingly is normal even though it has been shown that the anterior cranial base and maxilla are both shorter than in a normal group of subjects (10). Minor growth changes of the maxillary prognathism (s–n–ss) are expected because of periosteal apposition on nasion, as previously reported by Riolo et al. (17). Therefore the s_t–n_t–ss_2 angle was used to express the true sagittal displacement of the maxilla. Our study showed that sagittal growth of the maxilla had a mean forward translation (Table 2). This can be compared with the study of normal maxillary growth by Iseri & Solow (16), performed in 14 females from the age of 8.5 to 17.5 years. It was shown in this study that the maxilla was displaced about 6° forward from 8.5 to 17.5 years (0.66°/year), corresponding to 5 mm forward displacement in relation to the anterior cranial base. The superimpositions disclosed a normal maxillary downward growth compared with the 21 subjects described by Björk & Skieller (18).

The present study showed a midline deviation in the intermaxillary suture in SMMCI patients. This defect seemingly does not affect the eruption of the central incisor. As could be expected, the sagittal and vertical growth is normal. The existing material did not include material for analysis of the transverse growth pattern. The growth that occurs midaxially in the palate would presumably be affected by the developmental midline defect.

The SMMCI condition is often associated with normal mental development, and the patients are therefore treated in normal orthodontic practices. An orthodontist would presumably meet an SMMCI patient only once in his/her professional carrier. It is therefore important to coordinate treatment experience and to collect material for scientific analysis. The goal is to achieve a better understanding of the treatment options for these patients.

The present study has shown that there is a sutural midline defect, which suggests that frontal radiographs of the cranium for frontal growth analysis and treatment control are indicated in these patients.

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