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CLINICAL reports

Solitary maxillary central incisor: clinical report

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In 1976 Rappaport et al. introduced the term *Monosuperocentroincisivodontic dwarfism* to describe patients who exhibit a solitary maxillary central incisor in both dentitions in conjunction with growth retardation.[1] The frequency of congenitally missing maxillary primary central incisors has not been reported, but the frequency of congenitally missing maxillary permanent central incisors is rare and is reported to be less than 1%.[2]

Reports describing a solitary maxillary central incisor have appeared in the literature since 1958.[3] In some patients this syndrome has been linked to known growth hormone deficiency with concomitant short stature and in other individuals it has been linked to short stature for which the precise etiology is unknown.[1,4,5] There also have been reports of a solitary maxillary central incisor in patients with normal stature and no growth hormone deficiencies.[6,7]

The purpose of this report is to present findings from a child with a solitary maxillary central incisor in both dentitions. The patient has short stature but growth hormone levels were normal.

Clinical Report

An 8-year, 2-month-old white female was referred for examination and treatment to the Department of Pedodontics at the University of North Carolina at Chapel Hill School of Dentistry. She initially was seen for an evaluation of short stature by the Division of Pediatric Endocrinology at the North Carolina Memorial Hospital. The chief dental complaint was related to intermittent discomfort associated with exfoliation of a primary maxillary incisor.

Past Medical History

The patient was a full-term product of a normal spontaneous vaginal delivery culminating a pregnancy which occurred after an unsuccessful bilateral tubal ligation. During pregnancy the mother took conjugated estrogens[a] to induce menses for approximately 6 of the first 8 weeks. Conjugated estrogens and their use during early pregnancy are known to be associated with congenital abnormalities. Diazepam[b] was taken early in the pregnancy for back-related muscle spasms. Several studies have suggested an increased risk of congenital malformations associated with minor tranquilizers taken during the first trimester of pregnancy.[8] The first trimester was complicated further by sporadic vaginal bleeding and cramping.

An extra digit on both the right hand and foot were present at birth. Both extra digits were removed by ligation shortly after birth and there were no complications related to this procedure.

Infancy and early childhood were complicated by frequent upper respiratory infections, episodes of otitis media, and mononucleosis.

Developmental milestones were reached in sequence at appropriate times. However, a review of growth charts from previous hospital records revealed a progressive descension from the normal curve between ages 3 and 7. This growth descension could not be correlated specifically with events during development; however, the mother reported that the child had a poor appetite for the first 7 years of life.

The family history was negative for evidence of endocrine, gastrointestinal, and cardiovascular disease and both parents were in good health. Both parents were short in stature; the mother was 5'1", and the father was 5'6". Two half-sibling males (same mother, different father) were of normal stature.

[a] Premarin-Ayerst Laboratories; New York, NY.
[b] Valium-Roche Laboratories; Nutley, NJ.

Dental Findings

Extraoral findings were within normal limits. Intraoral soft tissues were normal except for mild generalized marginal gingivitis secondary to inadequate oral hygiene. The mixed dentition demonstrated a Class I molar relationship bilaterally.

The most notable dental finding was the presence of a single primary maxillary central incisor, which was placed centrally between the primary lateral incisors. Radiographically, there was a single permanent successor (Fig 1).

The treatment plan consisted of oral hygiene instructions, a prophylaxis and topical fluoride therapy, occlusal sealants, and minor restorative procedures. During the course of this treatment the single primary central incisor exfoliated and a single maxillary central incisor began to emerge (Figs 2, 3). This permanent successor was located centrally, but its morphology favored that of a maxillary left central incisor.

The past dental history of the family was negative for congenitally missing teeth or other craniofacial anomalies.

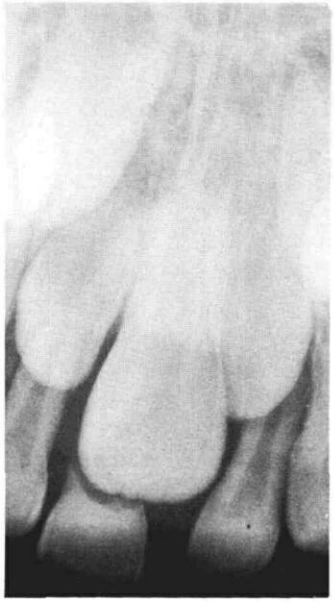


FIG 1. Periapical radiograph depicting the normally exfoliating solitary primary central incisor and a solitary permanent successor.

Further Medical Work Up

To investigate further the patient's short stature, additional medical work up was undertaken by the Division of Pediatric Endocrinology. The review of systems was negative for evidence of cardiorespiratory, gastrointestinal, genitourinary, neuromuscular, or endocrine disease. The neurological examination was within normal limits.

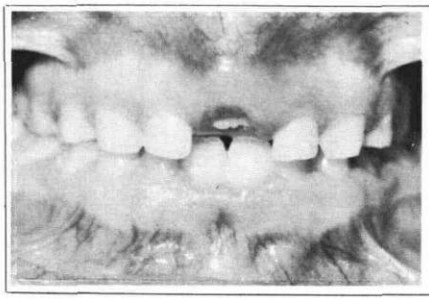
The physical examination revealed a height, weight, and head circumference with measurements in less than the third percentile. No pathology was seen on the skull radiograph. Although the chronological age predicted by the skull radiograph was 7 years, it did reveal a significant delay at 3 years, 4 months.

Laboratory tests including electrolytes, CBC, urinalysis, T_4, and somatomedin-C revealed normal values. The T_4 test is useful in determining thyroid gland function and the somatomedin-C test identifies some types of growth disorders as well as low levels of growth hormones. The growth hormone response was normal as was the karyotype.

After 6 months, the patient was examined again by the Pediatric Endocrinology Division. Height was still less than the third percentile, but the height velocity since the initial examination was calculated to be in the 90th-97th percentile. The weight at reexamination was still less than the third percentile, but the weight velocity since the initial examination was calculated to be in the 3rd-10th percentile. Based on these findings, the Pediatric Endocrinology staff recommended no treatment except for routine monitoring in their clinic.

Discussion

There are fewer than 25 reports involving a solitary primary or permanent maxillary central incisor in the literature.[1,3-7,9-12] Growth hormone deficiencies in conjunction with short stature have been found in 7 individuals.[1-5] Short stature with no associated growth hormone abnormalities have been reported for 3 individuals.[4,5] There are 3 reports in which a solitary maxillary central incisor was present and normal growth hormone levels and normal stature were re-



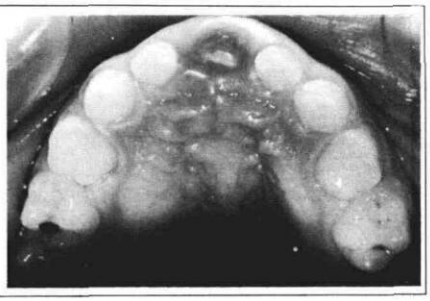


FIG 2 (*left*) and FIG 3 (*right*). Intraoral photographs following exfoliation of the solitary primary central incisor. Note that the permanent successor is beginning to erupt.

corded.[6,7] In other reports, growth hormone levels were not presented.[3,9-11]

A positive family history for the presence of a solitary maxillary central incisor was given in 2 reports suggesting a genetic determination,[9,12] however, no conclusive genetic studies were reported.

Other reports which possibly might fit the category of a solitary central incisor have been reported recently in the context of other clinical anomalies. For example, Ellisdon and Marshall reported 6 instances of "connation of maxillary incisors."[13] Two of these 6 reports easily could fall into the category of a solitary maxillary central incisor. A recent report by Bazan of fusion of maxillary incisors across the midline also presents a condition closely resembling a solitary maxillary central incisor.[14]

It is apparent that the diagnosis of this dental anomaly may be difficult. Because of the wide variation in clinical data in the reported cases, it is clear that monosuperocentroincisivodontic dwarfism, as described by Rappaport et al., is not the proper terminology for all patients with a solitary maxillary central incisor. Still, all patients with a solitary maxillary central incisor should have a thorough medical examination to verify the status of growth hormone levels.

The etiology of this condition, with or without growth hormone changes, is elusive. The role of genetics is not supported in most of the reports of solitary teeth. In this report, the mother took drugs known to be associated with teratogenic effects and the child had congenital abnormalities in the form of extra digits. This history prompts speculation that the developing tooth buds also might have been affected by the teratogenic drugs.

Although the solitary maxillary incisor condition may or may not be associated with growth hormone deficiency, affected patients should receive a definitive medical examination with endocrinology studies because growth hormone problems can be managed therapeutically if detected at an early age.

To further understand the etiology of this condition, it is important for clinicians to obtain detailed prenatal histories from the mothers of affected children.

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