Factors Associated with Linear Hypoplasia of Human Deciduous Incisors

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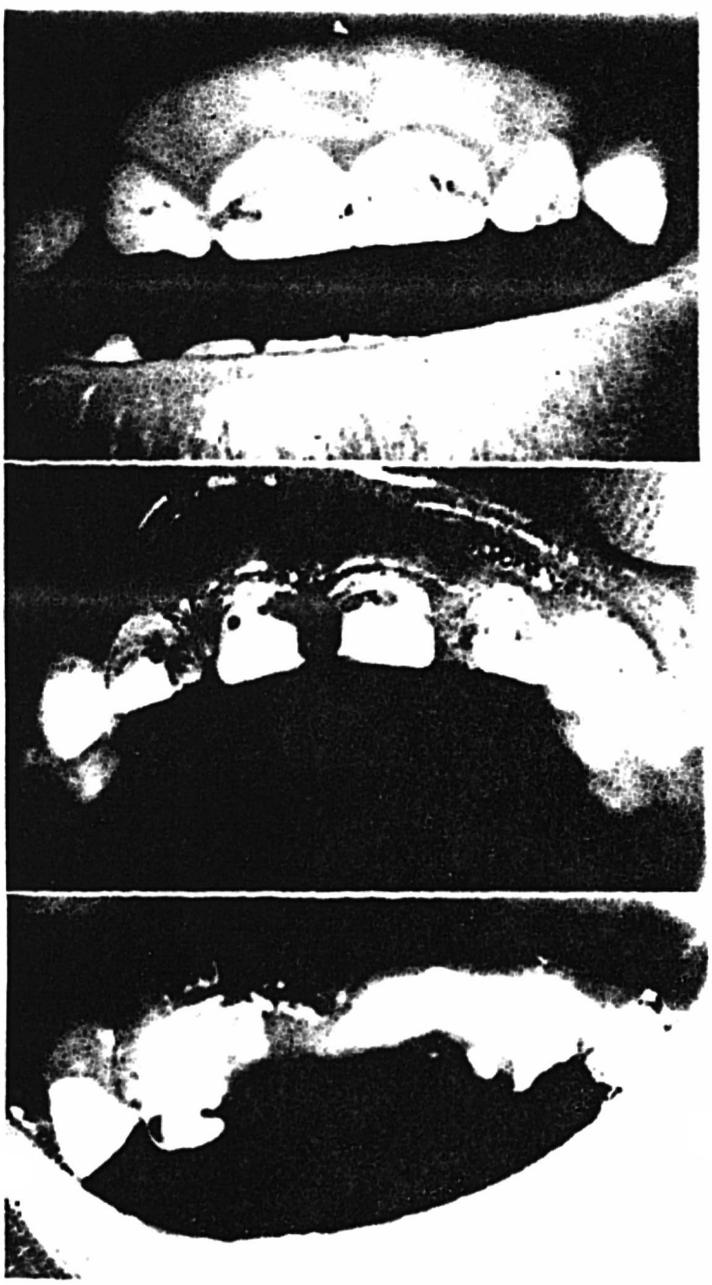
Linear hypoplasia of deciduous maxillary incisors was present in 31 of 73 Guatemalan children 2 to 3 years of age, whose medical histories were followed since birth. The presence of the line seemed to correlate with infections during the first month of life. All children were born with low serum-vitamin A levels.

In 1966 Sweeney and Guzmán[1] reported that children from three Guatemalan villages had a high percentage of caries in their deciduous teeth. A major component of this prevalence was the high caries attack in the four maxillary deciduous incisors along a visibly hypoplastic line that corresponded closely in position to the neonatal line (Illustration). Russell[2] had also commented on the paradoxically high decay prevalence of deciduous teeth, compared with permanent teeth, found in many surveys of the Interdepartmental Committee on Nutrition for National Defence (ICNND), which were conducted during the past decade in many developing nations. This pattern of decay with the associated hypoplasia has been reported sporadically in underprivileged populations of the world since 1936 but has still to be recognized by most developed countries as constituting a major disease entity. In attempting to ascertain factors associated with this lesion, we examined certain epidemiologic criteria that seemed likely to bear a relation to the presence of hypoplasia.

Materials and Methods

The population surveyed consisted of 73 children available from 95 consecutive births recorded in a longitudinal study in one village (Santa Maria Cauqué) from February 1964, through January 1966.[3] The children were all between the ages of 1.5 and 2.5 years at the time of the examination. The



Three illustrations of severe linear hypoplasia.

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TABLE 1

MICROORGANISMS INVESTIGATED IN SERIAL FECAL EXAMINATIONS

Intestinal Protozoan Parasites:
(1) *Entamoeba histolytica*
(2) *Entamoeba coli*
(3) *Entamoeba hartmanni*
(4) *Endolimax nana*
(5) *Iodamoeba butschlii*
(6) *Dientamoeba fragilis*
(7) *Chilomastix mesnili*
(8) *Giardia intestinalis*
Intestinal Microbial Parasites:
(1) *Candida albicans*
(2) *Shigella* species
(3) *Salmonella* species
(4) *Escherichia coli*
(5) Micrococci and Staphylococci
Intestinal Viruses:
(1) Enteroviruses
(2) Adenoviruses

73 children were examined independently in natural light and graded according to the following criteria: When no evidence could be seen on the maxillary incisors of a slight groove, stained or unstained, the lesion was considered to be negative; if a groove or depression could be seen that was slight and definite but unstained or slightly stained, it was rated as very mild; if grooving was deep and stained brown-black, it was rated as severe; and, in addition, if it was carious, it was rated very severe. If one examiner rated the subject negative for hypoplasia, and the other rated the child as very mild, the child was listed as questionable and considered as negative for prevalence calculations. According to this rating, some children with the very mild category of lesion might have been rated as negative or questionable in the previous survey.[1]

Nearly complete medical histories and cord-blood serums of these children were available to ascertain the following information: cord-blood-serum vitamin A levels, birth weight, estimate of gestation length, birth order, family size, ages of and types of infectious disease during the first 35 days postpartum, and ages of intestinal infection, or colonization, by protozoan parasites, *Candida* yeasts, enteric pathogenic bacteria, and enteroviruses and adenoviruses (Table 1) during the first four weeks of life.

Results

The data from examination of the teeth of the children are shown in Table 2. For 16 of the children, the examiners did not agree whether the lesion was present or absent because of the mildness of the hypoplasia; hence these children were rated questionable and considered negative. Thirty-one of the 73 children demonstrated the lesion, for a prevalence of 42.5% for all those examined

Table 2 also summarizes other data only for those who definitely had the lesion and those who definitely were without it. The questionable category was omitted. It is seen that no statistically significant differences existed between birth weight, percentage of normal weight at 6 months of age according to the Iowa standards, and vitamin A or carotene concentrations of the umbilical cord-blood serum, although those children with the lesion had a lower average percentage of normal weight at age 6 months. Family size indicated that more people were resident in the homes of those not showing the lesion than those with the lesion; however, the increase, which averaged less than one member per family, is explained by the data on the birth-rank order and is not statistically significant.

The rank order of births of these chil-

TABLE 2

PRESENCE OF HYPOPLASIA, VITAL STATISTICS, AND CORD-BLOOD-SERUM VITAMIN A AND CAROTENE CONCENTRATIONS OF CHILDREN FROM SANTA MARIA CAUQUE

Group	No. of Children	Birth Weight (gm)	Weight at 6 Months of Age (% Normal for Iowa Standards)	No. of People in Family	Cord-Blood Serum Vitamin A (μg %)	Cord-Blood Serum Carotene (μg %)
With the lesion	31	2596	66.8	6.2	14.8	31.3
Without the lesion	26	2600	69.8	6.8	14.6	29.0
Questionable lesion	16	...	...	...	...	...

dren (Table 3) suggested that those without the lesion had more older siblings than those with the lesion, thus helping to explain the larger family unit found for those without the lesion.

Data concerning the numbers and kinds of infectious diseases contracted during the first month of life are presented in Table 4 for children for whom these kinds of data were available. Children who demonstrated the lesion had nearly twice the prevalence of infectious diseases during the first 35 days as those who did not, (66.7% and 37.5%, respectively). These differences are statistically significant at the 5% level by the chi square test. Considering only those diseases of a severe nature, ie, diarrhea, thrush, and stomatitis, the totals were 8 and 2, respectively, for those with (27) and those without (24) the lesion.

In evaluating the age and prevalence of the first infection or colonization with the organisms listed in Table 1, only one category differed in either number or age of colonization between the two groups, the number of virus isolations obtained from the feces at ages less than 28 days. Seven isolations were found from those with the hypoplasia and two from the group without it. This difference is not statistically significant.

No individual with or without the lesion was represented by more than one infectious disease during the five weeks, and in each group only one child from whom viruses were isolated had not had an episode of infectious disease during the first 35 days of life.

The gestation age of the children as estimated by the physician in charge (Table 5) indicated that 3 of 24 children without the lesion were less than 40 weeks in utero, and had had a disease episode; whereas 7 of 27 children with the lesion were less than 40 weeks in utero, and only 2 of the 7 had not had an infectious disease. These differences were not statistically significant.

TABLE 3

RANK ORDER OF BIRTH OF CHILDREN (% OF TOTAL)

Child	With the Lesion	Without the Lesion
1st born	16.7	4.2
2nd born	16.7	12.5
3rd born	10.0	16.7
4th born	16.0	16.7
5th born	6.7	8.3
6th born	13.3	4.2
7th born	10.0	12.5
8th born	3.3	12.5
9th born	6.7	4.2
10th born	0.0	0.0
11th born	0.0	4.2
12th born	0.0	4.2

TABLE 4

NUMBERS AND TYPES OF INFECTIOUS DISEASES OF CHILDREN DURING THE FIRST 35 DAYS OF LIFE

Number of children	With the Lesion (27)	Without the Lesion (24)
Infectious diseases		
Diarrhea	3	0
Thrush	4	2
Conjunctivitis	9	2
Respiratory	1	5
Stomatitis	1	0
Total	18	9
Prevalence (%)	66.7	37.5

Discussion

In comparing the prevalence of linear hypoplasia in children 4 to 6 years of age from Santa Maria Cauqué as seen in 1963[1] with children from the same village as seen in 1967, the overall prevalence appears to have increased from 32% in 1963 to 43% four years later. However, the children examined by us in 1967 were between 1.5 and 2.5 years of age, and in no child was decay so severe as to preclude identification of the line; whereas in 1963 no child was examined who was less than 4 years of age, and many demonstrated decay or attrition of the maxillary incisors so severe that ascertaining the presence of the line was impossible. Coupled with this was the fact that most likely some children reported in this study who were classed as very mild would have been rated negative or questionable had they been seen in 1963. For these reasons, we feel that for this village

TABLE 5

NUMBER OF CHILDREN LESS THAN 40 WEEKS IN UTERO AND THEIR INFECTIOUS-DISEASE HISTORY

Group	With the Lesion	Without the Lesion
Number of children	27	24
Number of children less than 40 weeks in utero	7 (5)*	3 (3)

* Parentheses indicate the number of children who were less than 40 weeks in utero and who had a disease episode during the first 35 days postpartum.

the prevalence of 42% is a more reliable estimate of the true prevalence than the 32% previously reported for the children of ages 4 to 6 years.

Recently, Meyer and Baume[4] have studied teeth from Hong Kong and French Polynesia affected with this lesion. Histologically and microradiographically, the hypoplasia appeared to be associated with periods of ameloblast arrest during the neonatal period.

Sweeney and Guzmán[1] suggested that some degree of hypovitaminosis A during the neonatal period might partially explain the presence of this hypoplasia, since one of the more common nutritional deficiencies with a known association with hypoplasia found in most underdeveloped countries was vitamin A. In 1943, Lewis, Bodansky, and Shapiro[5] determined serum-vitamin A levels of newborns in the United States and reported that the cord-blood level closely resembled the maternal-serum level at birth but rapidly fell during the first 24 hours postpartum from about 21 μg% to 10.5 μg%, not to return to maternal level until the third or fourth day. This observation suggested to us that if the newborns from Guatemala and other regions had a much lower serum-vitamin A level, the result of the mother's pregnancy, infection, hard work, or low dietary intakes of vitamin A and precursors, the child's serum level after 24 hours might fall low enough that a deficiency bordering on the severe could be present for three or four days or longer. Such a low level might result in a long enough period of metaplasia or arrest of the ameloblast to produce the hypoplasia seen, or might predispose the child to infection.

One report suggested that such a situation prevailed, at least in Guatemala. Castañeda et al,[6] in following serum-vitamin A and carotene levels in 145 newborns, found the average natal level of vitamin A to be about 12 μg%, which fell below 9 μg%, then returned to the natal level of 12 μg% at the end of 84 hours postpartum. Interestingly, in their study the serum-vitamin A level had not risen above 15 μg% at the end of 30 days.

In our group of children for whom cord-blood-serum-vitamin A data were available, the average concentration was very similar to the average concentrations found by Castañeda et al,[6] although slightly higher. Thus, although no differences were found in the cord-blood-serum-vitamin A and carotene concentrations between those with and without the lesion, the low concentrations found in both groups indicated that these children were born with decidedly different blood levels than their counterparts in North America. That the maternal level was also low was indicated by the cord-blood concentration.

If the serum-vitamin A concentration of the mother was the sole criterion that dictated whether a child was born with a serum level conducive to creating a deficiency when the child's level fell after birth, we might have expected more of the lesions in larger families and in the children with the most older siblings because of food limitation. It seemed to work in the opposite manner, ie, those without the lesion came from the larger families and were younger members of the family. However, we have no information concerning the level of vitamin A after birth.

It is probably more valid to associate the lesion and the low serum levels of vitamin A with the prevalence of infectious disease and infection during the first month of life and with the number of premature births. Some febrile illnesses[7] cause an increased excretion of vitamin A and contribute to a dramatic decrease in serum-vitamin A levels seen during the period of illness. Although no one-to-one relationship existed between an infectious disease during the first month and the presence of hypoplasia in each child, almost twice as many infectious disease episodes were observed in the group with the hypoplasia.

The number of premature babies found in the group with hypoplasia is also of interest because Rosensweig and Sahar[8] have shown that 25% of premature babies from a well-developed country had hypoplasia resembling the hypoplasia seen by us. Whether the hypoplasia of prematurity could also be mediated through vitamin A levels is unknown, but some evidence suggests that the liver stores of vitamin A might be more unavailable by reason of the prematurity, earlier stage of development, and shorter time in which to lay down adequate stores,[9] thus perhaps rendering the premature more susceptible to infection. Certainly 80% of the children less than 40

weeks in utero had an infection during the first month, although three of the ten did not show the lesion, and each had had an infection.

In summary, then, certain findings are suggestive of a possible role of nutrition and infection in the etiology of linear hypoplasia.

Conclusion

Seventy-three Guatemalan children were grouped according to the presence or absence of linear hypoplasia of the deciduous maxillary incisors. These groups were then compared for weight at birth and at 6 months of age, number of people in the family, rank order of birth in the family, cord-blood-serum-vitamin A levels, length of gestation, and ages and types of infectious disease and gastrointestinal infections or colonizations.

The prevalence of the lesion was higher than previously reported from this village and affected 42.5% of the children. The presence of the line appeared to be correlated with infections during the first 35 days postpartum. There was a tendency for those with the lesion to have had more fecal virus isolations and to have more premature babies in their ranks than those without the lesion, but these facts were not statistically significant. All children were born with very low serum-vitamin A levels.

References

1. SWEENEY, E.A., GUZMÁN, M.: Oral Conditions in Children from Three Highland Villages in Guatemala, *Arch Oral Biol* 11:687-698, 1966.
2. RUSSELL, A.L.: *Environmental Variables in Oral Disease*, KRESHOVER, S., and MCCLURE, F. (eds), Washington, D. C.: American Association for the Advancement of Science, 1966, pp 30-32.
3. MATA, L.J.; URRUTIA, J.J.; and GARCIA, B.: Effect of Infection and Diet on Child Growth: Experience in a Guatemalan Village, *Ciba Foundation Study Group No. 31, Nutrition and Infection*, Boston: Little, Brown & Company, 1967, pp 112-126.
4. MEYER, J., and BAUME, L.J.: Pathologie de la Melandontie Infantile, de l'Odontoclasie et de la Carie Circulaire, *Rev Suisse Odontstoma* 76:48-92, 1966.
5. LEWIS, J.M.; BODANSKY, O.; and SHAPIRO, L.M.: Regulation of Vitamin A in Blood of Newborn Infants, *Amer J Dis Child* 66:503-510, 1943.
6. CASTAÑEDA, G.; MENDEZ DE LA VEGA, J.; GUZMÁN, M.A.; and AGUIRRE, F.: Estudios Nutricionales en un Grupo de Ninos Guatemaltecos: I. Del Nacimiento a un Mes de Edad, *Rev Col Med Guatemala* 6:11-21, 1955.
7. KAGAN, B. M., and KAISER, E.: Vitamin A Metabolism in Infection, *J Nutr* 57:277-286, 1955.
8. ROSENSWEIG, K.A., and SAHAR, M.: Enamel Hypoplasia and Dental Caries in the Primary Dentition of Prematuri, *Brit Dent J* 113:279-280, 1962.
9. HENLEY, T.H.; DANN, M.; and COHEN, W.R.C.: Reserves, Absorption, and Plasma Levels of Vitamin A in Premature Infants, *Amer J Dis Child* 68:257-264, 1944.