ENAMEL HYPOPLASIA IN CEREBRAL PALSYED CHILDREN

By

Stanley C. Herman

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INTRODUCTION
INTRODUCTION

This study is concerned with enamel hypoplasia of systemic origin and observed in cerebral palsied children. Data concerning incidence, location and type of enamel hypoplasia as observed will be tabulated and an attempt made to correlate the defects with the type of cerebral palsy present, all available relevant past medical history and any other possible etiologic factors.

A strong correlation between enamel hypoplasia and central nervous system disorders would suggest that the condition or conditions which caused the cerebral palsy are the same as those which affected the metabolism of the ameloblasts during the formative stage of growth in the developing teeth.
REVIEW OF LITERATURE
REVIEW OF LITERATURE

Orban, Sicher and Weinmann reported that amelogenesis occurs in two phases; the first being the enamel matrix formation and the second being enamel maturation. The formative stage of amelogenesis is initiated after a thin layer of dentin is formed. The ameloblasts are lengthened at this time. Next is observed a simultaneous formation of Tomes' processes and terminal bar apparatus. When the Tomes' process attains a certain length, the dentinal end undergoes a transformation into the pre-enamel matrix. When the pre-enamel matrix attains a thickness of 16 to 22 microns, it is transformed into young enamel matrix which is more homogeneous than the previous pre-enamel matrix. The enamel matrix undergoes its final transformation in the formative stage when the young enamel matrix is about 16 to 30 microns in thickness.

It has been found that the pre-enamel matrix does not contain calcium salts and that the calcification process begins in the young enamel matrix and at completion of the formative phase. The inorganic content of the matrix is about 35 per cent and organic content and water about 65 per cent. The addition of more inorganic salts and subsequent loss of water and organic material takes place in the enamel maturation stage of amelogenesis.

Diamond and Weinmann have stated that the maturation stage of amelogenesis follows a completely different pattern than the formative stage. The maturation stage begins at the incisal edge...
of the tooth or at the tips of the cusps and proceeds in planes perpendicular to the long axis of the tooth. When the maturation stage is completed, the enamel is fully developed and calcified.

Magitot' (1881) made the first report in relation to enamel hypoplasia. He reported a detailed study of the correlation between the diseases of early infancy and the distribution of enamel hypoplasia ("erosion") in the permanent teeth. His study was concerned with the findings in 40 patients, all of whom gave a clinical history of convulsions and eclamptic attacks during the first 2 years of life.

Zsigmondy' (1893) wrote a classic paper and introduced the term "hypoplasia". Until this time, the terms "atrophy" and "erosion" had been used to designate enamel defects. Zsigmondy also stated that enamel hypoplasia occurs in the primary teeth although less frequently than in the permanent teeth. He also gave a very lucid description of the lines of distribution of the defect in the enamel and dentin of hypoplastic teeth.

Berten' (1895) indicated that hypoplasia of enamel and dentin resulted from nutritional disturbances and occurred along the incremental lines which constitute the formative pattern of the tooth.

Gottlieb7 (1920) theorized that enamel hypoplasia took place when calcification of the enamel matrix did not follow immediately the formation of the organic matrix. When calcification is delayed, the organic matrix is unable to maintain its shape and it folds and collapses. These irregularities in the matrix when once calcified, form the irregular grooves and depressions re-
ferred to as hypoplastic defects.

Another explanation of hypoplasia was suggested by Klein. Klein's theory dealt primarily with degenerative changes in the ameloblasts.

Boyle (1931) reported ameloblastic degeneration with subsequent discontinuation of enamel formation in a case of vitamin A deficiency in an infant, 3½ months of age.

Mellanby (1934) showed that a deficiency of vitamin D during the period of tooth formation gave rise to deficient tooth formation which manifested itself as varying degrees of enamel hypoplasia depending on the severity of the deficiency.

Stein (1936) reported that the primary teeth of prematurely born children had a comparatively high incidence of enamel hypoplasia and suggested that premature termination of gestation and difficult adjustment to postnatal nutrition might be responsible for the enamel defects. He emphasized the fact that the distance of the enamel defects from the incisal edge was of significance in determining the time at which the hypoplasia developed. However, because of the absence of a definite landmark in the tooth, he was not able to determine with certainty the chronologic significance of the hypoplasia.

Kronfeld and Schour (1939) stated in an article on neonatal hypoplasia, that hypoplastic defects were a permanent record of nutritional disturbances and of diseases that occurred during the formative period of the teeth. Neonatal dental hypoplasia designated a disturbance in the formation rather than the calcification of enamel and dentin that originated during the
In its mildest form, a neonatal disturbance is reflected as an accentuated neonatal ring in the deciduous teeth. In the severe type of neonatal disturbance, the enamel formation was arrested at birth or during the neonatal period. Postnatal amelogenesis was confined to the portion of the crown which was located cervically from the enamel area present at birth. The dentin was less responsive to metabolic and nutritional disturbances than was the enamel, and formation was arrested for only a relatively short period of time and recovery usually occurred.

Kronfeld and Schour also have stated that prenatal hypoplasia is extremely rare. Their observations supported the contention that the fetus was well protected in utero. They agreed with statements of Hess and his associates that the prenatal mineral metabolism of the mother is, as compared to the child's postnatal nutrition, of little significance to the child's teeth.

Sarnat and Schour (1941) reported that the enamel and dentin yielded an accurate and permanent record of normal fluctuations and pathological accentuations of mineral and general metabolism. They reported a clinical study of 60 patients chosen at random whose teeth showed enamel hypoplasia. Sixty-five per cent of the hypoplasia occurred in the infancy period; 33 per cent was found in the area of the teeth formed during the early childhood period, and 2 per cent was related to the late childhood period. They reported no instances of hypoplastic defects of enamel formed during the prenatal or neonatal periods.

Sarnat and Schour (1942) in a continuation of the previous study stated that the term "chronologic enamel aplasia" is pref-
erable to "enamel hypoplasia" because the former denotes both time and lack of formation. No specific etiology was found in their study group consisting of 60 children. The exanthematous diseases were not so frequently a cause as was previously thought. Enamel hypoplasia was most often found where the possible etiologic agents were rickets, hypoparathyroidism and fluorosis, but hypoplasia could not be predicted with any reliability even in the most severe forms of these diseases.

Sheldon, Bibby and Bales 15 (1945) studied the relationship between microscopic enamel defects and infantile debilities. They examined ground sections of 95 teeth from 34 patients with detailed medical histories. In more than 70 per cent of the cases there was a positive correlation between the time of formation of a band of defective enamel and the existence of some systemic disability. In 23 per cent, there were definite defects in the enamel of patients who had no histories of systemic conditions which might have produced enamel defects. In 6 per cent, there were no enamel changes in patients who had histories of disabilities which had produced enamel changes in other patients. Deficiencies of vitamins A, C, and D and also calcium and phosphorus were the most common causes of defective enamel formation.

Stein 16 (1947) found that 8 of 16 premature children had enamel hypoplasia. The position of the defect on the teeth corresponded in the time of development with the time of their births.
Miller (1955) found 13 cases of hemolytic disease associated with neurological signs of kernicterus. Each of these cases demonstrated neonatal enamel hypoplasia. He also stated that he had observed similar enamel defects in the teeth of premature children suffering from kernicterus. Miller postulated that since a clinical physiologic jaundice is present in 75 per cent of newborn children, biliverdin may be associated with the "physiologic" birth line.

Kreshover in two of his human studies, found that it was possible to correlate in many instances the occurrence of enamel hypoplasia with abnormalities of gestation or parturition. However, regardless of the disease process that may have been present, it was not possible to prognosticate from such data which babies, either prenatally, neonatally or postnatally, would manifest abnormalities in odontogenesis. He postulated, therefore, that abnormalities in odontogenesis may be considered non-specific in nature and related to a variety of causative factors.

Kreshover has shown as a result of additional studies, that enamel hypoplasia can be produced in rodents with artificially induced fever, alloxan diabetes, injections of tubercle bacilli and the viruses of vaccinia and lymphocytic choriomeningitis. The enamel hypoplasia produced was grossly and microscopically similar to that seen in humans.

In 1956, Perlstain and Massler reported that in a series of approximately 250 children with cerebral palsy, the incidence of dental enamel dysplasia was found to be 24 per cent, of which
approximately one-half was neonatal and the other half was of prenatal variety. They also stated that neonatal dental dysplasia occurs in about 7 per cent of the normal population, and that prenatal dental dysplasia is rarely seen in normal children. In children with kernicterus, due to the Rh factor, the incidence of prenatal dental dysplasia was 58 per cent. When the kernicterus was due to other factors, such as anoxia or prematurity, the incidence was 22 per cent. In kernicterus, not due to the Rh factor, neonatal damage to the enamel was more commonly seen than prenatal damage, the reverse being true when the Rh factor was the cause. It was further observed that cerebral palsyed children of the group of spastic paraplegics associated with prematurity, had a high incidence of prenatal dental dysplasia. In this group, the prenatal dysplasia corresponded to the age of prematurity, i.e., it represented a premature neonatal line.

Miller and Forrester (1959) reported that they examined 12 cerebral palsyed patients between the ages of 3 and 5 years. Eleven had normal teeth and one had severe enamel hypoplasia. This particular child was premature, with a birth weight of 3 lbs. 13 oz. and had a severe neonatal jaundice.

Miller and Forrester in the same study, examined 109 normal children, ages 1 to 4 years. All of these children were making routine visits to an infant welfare clinic. Mild hypoplasia was found in 4 cases, but upon examination of their histories, it was noted that 3 of the children had been born prematurely with birth weights of approximately 5 lbs. The fourth was a child of a mother who had syphilis; this condition was discovered and
treated during pregnancy and the child was otherwise normal.

Via and Churchill 27 (1959) reported enamel hypoplasia in 54 per cent of 219 cerebral palsied children; whereas, only 8 per cent of 93 normal children had enamel hypoplasia. They reported the highest incidence of enamel hypoplasia (100 per cent) in those cerebral palsied children with choreoathetosis, and in those with simple spastic diplegia (64 per cent). A definite correlation was found to exist between the time the abnormal factors occurred and the time the enamel hypoplasia was estimated to have occurred.

Pohl 28 (1950) described cerebral palsy as a group of neuromuscular disorders in which there is impairment or loss of muscular control due to brain injury. Depending upon the region of the brain affected, the muscle activity is disturbed in a particular manner. A number of clinical types of cerebral palsy are recognized. The spastic type is characterized by stiffness, results from a lesion of the cortex of the brain which normally controls voluntary movements; the athetoid type is characterized by involuntary motion, is due to a lesion of the basal ganglia which normally control automatic and associated movements and certain aspects of posture; the ataxic type, characterized by loss of equilibrium and muscle coordination, results from a lesion of the cerebellum which normally controls balance and coordination; the tremor type is characterized by vibratory movements resulting from a lesion of the basal ganglia and in this respect is related to the athetoid, although the tremor type is very rare; the rigidity type is characterized by
extreme extension of the body resulting from widespread brain damage; the mixed type is characterized by the physical phenomenon of both the spastic and the athetoid types. The extent of involvement is variable and is dependent on the extent of brain injury and whether the pathology is unilateral or bilateral. The resultant body involvement of one side is designated as hemiplegia while when only one limb is affected, the condition is referred to as monoplegia. When both lower extremities are involved the extent of the condition is referred to as paraplegia; while in quadriplegia there is involvement of upper and lower extremities on both sides of the body.
METHOD

A clinical examination of the teeth of 120 cerebral palsied children, between the ages of 2½ and 10½ years, was conducted in the Cerebral Palsy Dental Clinic at Indiana University. The examination was completed with the aid of a good light, mirror and explorer. Complete medical records were available for all of the children in the observation group.

A patient was considered to have enamel hypoplasia if a defect in the enamel was observed, which was characterized by a grooved ring or concavity of visible and palpable depth about the crown of the tooth. The hypoplastic area may have been smooth, pitted, discolored, or not discolored, and was distinct from enamel hypocalcification. The hypocalcified areas were not depressed as contrasted with the hypoplastic areas.

The developmental chart by Schour and Massler was used to determine the approximate age at which the fault in the enamel occurred.

Detailed information concerning gestation, birth and subsequent development was obtained from all available medical records and from parents whom were believed to be reliable. An example of a recorded examination can be found in Figs. 1, 2 and 3.

The control group consisted of 117 normal children between the ages of 2½ and 10½ years, selected at random from the Pedodontic Clinic of Indiana University School of Dentistry and a Public School survey. A normal child in this study was one who
was not cerebral palsied or one who has not had a chronic or serious illness early in life.

All the children in both the observation and control groups were old enough developmentally to have a full complement of primary teeth.

Seven teeth with no evidence of enamel hypoplasia were obtained from cerebral palsied children to be studied microscopically. The teeth were selected at random from cerebral palsied children for whom extractions were indicated.

Ground sections were prepared from the extracted teeth in the following manner. First the teeth were reduced uniformly mesial-distally until they were approximately 300 microns in thickness. This primary procedure was done on a dental lathe using mounted diamond discs and stones and keeping the teeth wet at all times. The teeth were further reduced to approximately 150 microns, using Speed-Wet Durite paper under water. Next, they were polished using an Arkansas stone, levigated alumina and water. At this time, the ground sections approximated 80 to 120 microns in thickness. The sections were washed in water and then they were agitated in acetone for 45 minutes in order to dehydrate the sections. The acetone was removed by soaking the ground sections in Xylene for five minutes. A clean glass slide was then wetted with Xylene and the polished, dehydrated ground section was placed on the glass slide. Several drops of Canada Balsam were placed over and around the tooth. Next, a coverslip was placed and allowed to harden for
three days.

The ground sections were studied microscopically and the findings were related to the medical histories, and also to ground sections of teeth from normal children, as observed in histology textbooks.
Figure 1. Form used to record findings during clinical examination and review of past medical history.
I. Classification of Patient
   A. Cerebral Palsy  B. Normal

II. Types of Cerebral Palsy
   A. Athetoid  C. Ataxia
   B. Spastic  D. Tremors  E. Rigidity

III. Distribution
   A. Monoplegic  C. Quadriplegic
   B. Diplegic D. Hemiplegic  E. Paraplegic

IV. Possible Etiological Factors
   A. Birth injury  E. Convulsive seizures
   B. Congenital  F. Brain hemorrhage
   C. Anoxia  G. Encephalitis
   D. RH incompatibility  H. Meningitis

V. Hypoplasia
   A. Yes  B. No

VI. Location (chart)

VII. Type
   A. Smooth  C. Discolored  E. Secondary caries
   B. Pitted  D. Not discolored  F. Hypocalcification

VIII. Correlation with Schour and Massler chart:
   Direct correlation between difficulty at birth and hypoplasia.

IX. Distribution
   A. Prenatal  B. Neonatal  C. Post Natal

X. Brief summary of the following:
   A. Gestation
      Mother had influenza when 5 months pregnant.
   B. Birth
      3 lbs., 13 oz.  Breathed with difficulty - cyanotic.
   C. Subsequent development
      Retarded

XI. X-rays  yes——no
   Photos  yes——no
   Teeth for histological study  yes——no

XII. Additional Remarks
Figures 2 and 3. Copy of Massler and Schour chart used for recording of areas of hypoplasia during the clinical examination.

DECIDUOUS TEETH

LEFT

- Infancy ring (10 mos.)
- Neonatal ring (newborn)
- Prenatal period
- Infancy period (birth to 10 months)
- Childhood period

RIGHT

- Infancy ring (10 mos.)
- Neonatal ring (newborn)
- Prenatal period
- Infancy period (birth to 10 months)
- Childhood period
RESULTS
RESULTS

The results of the clinical examination of 120 cerebral palsied children revealed that 43 of these children had enamel hypoplasia, while in the normal group of 117 children, only 7 were affected.

The results of the cerebral palsied group were further divided to see if any relations existed between type of cerebral palsy and time of the development of enamel hypoplasia. The cerebral palsied group was divided into the following types: Athetoid, Spastic, Ataxic, Tremor, Rigidity, Mixed Spastic and Athetoid and Diagnosis Unknown. The hypoplasia was divided into three groups depending whether the enamel hypoplasia occurred during the prenatal, neonatal or infancy period. The prenatal period was also subdivided depending on whether the child was born prematurely. (Table 1).

Athetoid — The rate of enamel hypoplasia in a group of 18 athetoids was 44 per cent. Three cases of enamel hypoplasia were prenatal in origin and none of these were born prematurely. Four cases had their origin in the neonatal period. One case of hypoplasia occurred during the infancy period.

Spastic — The rate of enamel hypoplasia in a group of 80 spastics was 39 per cent. Ten of these cases of enamel hypoplasia were prenatal in origin and from this group 4 were born prematurely. Fifteen patients had enamel hypoplasia which originated during the neonatal period and 6 during the infancy period.
Ataxic — Four ataxic children were examined. Of this number, one had enamel hypoplasia which had its origin during the neonatal period.

Tremor — None of the children examined had the tremor variety of cerebral palsy.

Rigidity — None of the children examined had the rigidity type of cerebral palsy.

Mixed — Thirteen cerebral palsey children were examined who were classified as mixed spastic and athetoid. Of this group, 23 per cent had enamel hypoplasia. The 3 patients with enamel hypoplasia all had the enamel defects produced during the neonatal period.

Diagnosis Unknown — At the time of the dental examination, a medical diagnosis as to type of cerebral palsy had not been determined in 5 cases. None of these exhibited enamel hypoplasia.

In 30 of the 43 cases of enamel hypoplasia, it was shown that there was a direct correlation between the time at which the probable etiologic factor occurred which caused the cerebral palsy and the location of the enamel hypoplasia based on the time of development of that particular area of enamel. In 9 cases there was a negative history regarding the etiology of the brain damage, and in 4 cases there was no correlation between the enamel hypoplasia and the probable etiologic factor causing the brain damage. (Table 2).

The results of the microscopic study of the 7 ground
sections taken from 5 cerebral palsied patients are found in Table 3. Photomicrographs of areas of deficient or defective enamel formation can be found in Illustrations 4, 5 and 6.

**H. H.** — During the microscopic study of a ground section of the maxillary left first permanent molar, a line of deficient enamel formation was observed. This abnormal area coincided in the time of development with an acute febrile disease (Friedreich's ataxia) at 5 months of age which lasted for a period of 3 weeks. (Illus. 4).

**J. G.** — The study of the maxillary left primary cuspid revealed an area of intrinsic staining and abnormal enamel formation at the level of cervical one-third of the crown. This patient presented with encephalitis at 3 months of age. (Illus. 5).

**D. M.** — The microscopic examination of the maxillary left, and mandibular left and right primary central incisors revealed a line of abnormal enamel formation in the crowns and lines of intrinsic staining in the dentin which coincided with the fact that the child had been born after 7 months gestation with a birth weight of 2 lbs., 8 oz. (Illus. 6).

**D. S.** — The microscopic examination of the mandibular right primary incisor did not reveal an abnormal histologic appearance. In this case the mandibular right primary lateral incisor crown development was complete when the probable etiologic factor occurred.

**T. W.** — The microscopic examination of the maxillary right primary central incisor did not reveal any histologic abnormality. In this case, the etiology of the cerebral palsy was unknown.
TABLE 1.

Distribution of Enamel Hypoplasia

<table>
<thead>
<tr>
<th>Prenatal Diagnosis</th>
<th>Neonatal Infancy</th>
<th>Total No. of Patients With</th>
<th>No. of Hypoplastic Teeth</th>
<th>Frequency of Hypoplasia</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Birth to 1 month</td>
<td>1 to 12 months</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Full Term</td>
<td>Premature</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Athetoid</td>
<td>3</td>
<td>4</td>
<td>8</td>
<td>18</td>
</tr>
<tr>
<td>Spastic</td>
<td>6</td>
<td>15</td>
<td>31</td>
<td>80</td>
</tr>
<tr>
<td>Ataxic</td>
<td>1</td>
<td>1</td>
<td>4</td>
<td>25%</td>
</tr>
<tr>
<td>Tremors</td>
<td>0</td>
<td></td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>Rigidity</td>
<td>0</td>
<td></td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>Mixed</td>
<td>3</td>
<td>3</td>
<td>13</td>
<td>23%</td>
</tr>
<tr>
<td>Diagnosis Unknown</td>
<td>0</td>
<td></td>
<td>5</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>43</td>
<td>120</td>
</tr>
<tr>
<td>Normal</td>
<td>1</td>
<td>5</td>
<td>7</td>
<td>117</td>
</tr>
</tbody>
</table>

Total: 180 patients with enamel hypoplasia.
<table>
<thead>
<tr>
<th>Correlation</th>
<th>No. of Cases</th>
<th>Frequency</th>
</tr>
</thead>
<tbody>
<tr>
<td>Correlation between enamel hypoplasia and probable etiologic factor</td>
<td>30</td>
<td>69.8%</td>
</tr>
<tr>
<td>causing brain damage</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Negative medical history regarding etiology of brain damage</td>
<td>9</td>
<td>20.9%</td>
</tr>
<tr>
<td>No correlation between enamel hypoplasia and probable etiologic factor</td>
<td>4</td>
<td>9.3%</td>
</tr>
<tr>
<td>causing brain damage</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Patient</td>
<td>Tooth</td>
<td>Microscopic Findings</td>
</tr>
<tr>
<td>---------</td>
<td>-------</td>
<td>----------------------</td>
</tr>
<tr>
<td>M.M.</td>
<td>Upper left 1st permanent molar enamel formation</td>
<td>Line of abnormal enamel formation</td>
</tr>
<tr>
<td>J.G.</td>
<td>Upper left primary cuspid</td>
<td>Intrinsic stain, questionable area of abnormal enamel formation</td>
</tr>
<tr>
<td>D.W.</td>
<td>Upper left, lower right and central primary incisors</td>
<td>Line of abnormal enamel formation</td>
</tr>
<tr>
<td>D.S.</td>
<td>Lower right primary lateral incisor</td>
<td>None</td>
</tr>
<tr>
<td>T.W.</td>
<td>Upper right primary central incisor</td>
<td>None</td>
</tr>
</tbody>
</table>

Results Viewed in Microscopic Sections as Compared with Medical History
Illustration 1. Example of prenatal enamel hypoplasia.

Note the line of chronologic enamel aplasia in the incisal $\frac{1}{4}$ of the mandibular primary incisors. Past medical history revealed that the patient is cerebral palseied as the result of a premature birth with a 6 month gestation and birth weight of 2 lbs., 5 oz.
Illustration 2. Example of neonatal enamel hypoplasia.

Note that only the most cervical part of the intrinsically stained areas are hypoplastic. This child experienced a severe nutritional deficiency during the first month of extra-uterine life.
Illustration 3. Example of enamel hypoplasia occurring during infancy period. Note the wide band of pitted enamel which was evident on the maxillary and mandibular permanent incisors and first permanent molars. The patient is a normal male who was severely affected with pneumonia at 6 months of age.
Illustration 4. M. M. Photomicrograph of upper left first permanent molar demonstrating a line of abnormal enamel formation marked by an arrow. The patient was cerebral palsyed with a past medical history of an acute febrile illness at 5 months of age which lasted for a 3-week duration. This condition was later diagnosed as Friedrich's ataxia.
Illustration 5. J. G. Photomicrograph of upper left primary cusp providing intrinsic stain and abnormal enamel formation in the cervical 1/3 of the crown. Patient was cerebral palsy and presented with encephalitis at 3 months of age.

A. Intrinsic Stain

B. Abnormal Enamel
Illustration 6. D. W. Photomicrograph of mandibular left primary central incisor demonstrating a line of abnormal enamel formation and lines of intrinsic staining in the dentin. Patient was cerebral palsy with a history of being born after 7 months gestation with a birth weight of 2 lbs., 8 oz.

A. Abnormal Enamel
B. Intrinsic Stain
DISCUSSION
DISCUSSION

Enamel hypoplasia was observed more frequently in cerebral palsied children than in normal children. It was found that upon careful examination of 120 cerebral palsied children, 36 per cent presented evidence of enamel hypoplasia; whereas in a group of 117 normal children, only 6 per cent were affected.

The cerebral palsied group was subdivided into six types to find if any one particular type was affected more often than the others. The types were athetoid, spastic, mixed athetoid and spastic, ataxic, tremor and rigidity. The athetoids and the spastics were affected more often than the other types. Of the athetoids, 44 per cent exhibited enamel hypoplasia; while the frequency of enamel hypoplasia in the spastic group was 39 per cent.

Another closely associated type was the mixed spastics and athetoids. The rate of enamel hypoplasia in this group was 23 per cent. One would expect the rate to be more closely related to the previous two types. At the present, there is no explanation for this observation.

Of particular interest in this study the finding that 69.8 per cent of the cases in which the child had both cerebral palsy and enamel hypoplasia, there was a positive correlation based on the time of the possible etiologic factors which could have caused brain damage and the position of the
enamel hypoplasia based on the developmental time of that particular area of enamel, while in 20.9 per cent of the cases, there was no known possible etiologic factors, and in 9 per cent there was no correlation between known possible etiologic factors and the temporal location of the enamel hypoplasia. It is quite possible that this correlation might have been even greater had certain parts of the medical records been more detailed and accurate with the inclusion of subtle, albeit significant, information. (See Table 2). It was interesting to note the similarity of the results in this correlation with those of Sheldon, Bibby and Bales who related microscopic enamel defects and infantile debilities. In more than 70 per cent of their cases, there was a positive correlation between the time of formation of a band of defective enamel and the existence of some systemic disability. In 23 per cent there were definite defects in the enamel of patients who had no history of systemic conditions which might have produced enamel defects. In 6 per cent there were no enamel changes in patients who had histories of disabilities which had produced changes in other patients.

In the present study no specific etiologic agents could be found which would consistently produce enamel hypoplasia. Cases were examined where the probable etiologic agents were prematurity, encephalitis, birth injury, anoxia, nutritional deficiencies, Rh incompatibility, meningitis and others.
These etiologic agents did not cause enamel hypoplasia in every case in which they were causative factors in the brain damage.

One theory proposed on the etiology of enamel hypoplasia is that the brain and the ameloblasts are simultaneously affected by the same pathological process. Another possibility is that the etiologic agent involved caused injury to the brain which in turn stimulated a severe systemic metabolic upset which affected the active ameloblasts. Whichever of the theories is true is not known, but this insult must be sufficiently severe to cause both the brain injury and the enamel hypoplasia. It should be noted that in 64 per cent of the cases of cerebral studied, there was no enamel hypoplasia visible on clinical examination suggesting that cerebral palsy is produced more readily than enamel hypoplasia. Because of the high correlation between cerebral palsy and enamel hypoplasia, and because of the apparent difficulty in the production of enamel hypoplasia, as mentioned above, the presence of enamel hypoplasia may help the clinician and the researcher to determine when the brain injury occurred in cases where the etiology is subtle or apparently unknown.

As a supplement to this clinical study, seven ground sections from five patients were prepared for microscopic study. Six of the teeth were primary and one was permanent. None of the teeth were grossly hypoplastic on clinical
examination. They were studied to determine if some hypoplasia was being overlooked on clinical examination. Upon microscopic examination the primary teeth from two patients exhibited abnormal enamel formation, while two others showed no abnormality. The one permanent first molar showed a band of abnormal enamel formation.

The medical histories of the patients from whom the teeth were extracted were studied. In four of these cases, the etiologic factors which were probably responsible for the cerebral palsy condition, were present during the development of the crowns of the teeth. (See Table 3). In one case (M.M.) there was a band of deficient enamel formation in the first permanent molar which coincided with the occurrence of an acute febrile disease for a duration of three weeks (Fig. 4). In the second case (J.G.) there was a questionable area of abnormal enamel formation which appeared darkly stained and rough which could be related to a febrile illness diagnosed as encephalitis, at three months of age. (Fig. 5). In the third case (D.W.) histologic sections of three primary anterior teeth demonstrating a marked intrinsic stain were studied. This child was born after seven months gestation and the birth weight was 2 lbs., 8 oz. On microscopic examination, the staining was evident in the dentin. Of more interest, there appeared to be two distinct neonatal lines, one being a line of abnormal enamel formation. This area could have been developing at the time of birth.
(Fig. 6). In case four (D531) the development of the crown of the teeth studied was completed before the probable etiologic factor which caused the brain damage, occurred, therefore, there was no abnormal histologic picture. In case number five (T1261) the etiology of the cerebral palsy was unknown. The crown of the tooth in question showed no microscopic abnormality.

There appeared to be some positive correlation between microscopic findings and the probable etiologic factors causing the brain damage. Abnormal enamel formation in 3 cases seemed to coincide with the time of abnormal factors which were probably the cause of the cerebral palsy. This is a very small sample from which definite conclusions cannot be determined. More study is needed in this area.
This study was concerned with enamel hypoplasia of systemic origin and observed in cerebral palsied children. Data concerning incidence, location and type of enamel hypoplasia as observed was tabulated and an attempt made to correlate the defects with the type of cerebral palsy that was present, all available relevant past medical history and any other possible etiologic factors.

A strong correlation between enamel hypoplasia and central nervous system disorders suggested that the condition or conditions which caused the cerebral palsy were the same as those which affected the metabolism of the ameloblasts during the formative stage of growth in the developing tooth.

Enamel hypoplasia was observed in 36 per cent of 120 cerebral palsied children between the ages of 2½ and 10½ years as compared with 6 per cent enamel hypoplasia in a group of 117 normal children in the same age group.

In the experimental group, 44 per cent of the athetoids, 39 per cent of the spastics, 23 per cent of the mixed spastics and athetoids, and 25 per cent of the ataxics presented with enamel hypoplasia on clinical examination.

In 69.3 per cent of the cases studied in which there was both cerebral palsy and enamel hypoplasia, there was a positive correlation based on the time of the possible etiologic factors which could have caused the brain damage and
the position of the enamel hypoplasia based on the developmental time of that particular area of enamel while in
20.9 per cent of the cases, there was no known possible etiologic factors, and in 9 per cent there was no correlation
between known possible etiologic factors and the temporal location of the enamel hypoplasia.

Seven ground sections from 5 patients were studied microscopically; 6 of the teeth were primary and one was per-
manent. None of the teeth were visibly hypoplastic on clinical examination. The primary teeth from two patients exhibited abnormal enamel formation, while two others showed no abnormality. The one permanent first molar exhibited a band of abnormal enamel formation. In all the cases which exhibited enamel defects, there was a correlation with the medical history. The two teeth which did not show defects did not relate to the medical histories — one, there was completion of enamel formation before the brain damage occurred, and the other the etiology of the cerebral palsy was unknown.
CONCLUSIONS
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1. The incidence of enamel hypoplasia was observed to be 6 times greater in a group of cerebral palsied children than in a group of normal children.

2. In 69.8 per cent of the cases studied in which there was both cerebral palsy and enamel hypoplasia, there was a positive correlation based on the time of the possible etiologic factors which caused the brain damage and the position of the enamel hypoplasia based on the developmental time of that particular area of enamel.

3. No specific etiologic agents were found which would consistently produce enamel hypoplasia in the children studied.
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REFERENCES


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VITA

September 4, 1933

1947 to 1951

1951 to 1961

1958

1959

1959 to 1961

Born, Indianapolis, Indiana

Shortridge High School, Indianapolis, Indiana

Indiana University

B. S. in Dentistry

D.D.S.

Graduate Dental School, and Fellow of United Cerebral Palsy

PROFESSIONAL SOCIETIES

American Dental Association

Indiana State Dental Association

Indianapolis District Dental Society

American Society of Dentistry for Children

Indiana Society of Dentistry for Children

Alpha Omega Dental Fraternity
ABSTRACT
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A study was undertaken at the Cerebral Palsy Dental Clinic of Indiana University to study the relationships between cerebral palsy and enamel hypoplasia.

A group of 120 cerebral palsy children between the ages of 212 and 102 years were examined clinically to determine if enamel hypoplasia was present. This information was recorded with a summary of the pertinent medical history. These findings were compared with each other and with a control group of 117 normal children between the same ages.

Enamel hypoplasia was observed in 36 per cent of the cerebral palsy group as compared with 6 per cent of the normal group.

In 69.6 per cent of cases studied in which there was both cerebral palsy and enamel hypoplasia, there was a positive correlation based on the time of the possible etiologic factors which could have caused brain damage and the position of the enamel hypoplasia based on the developmental time of that particular area of enamel.

No specific etiologic agents could be found which would consistently produce enamel hypoplasia in the children studied.

It should be noted that in 64 per cent of the cases of cerebral palsy studied, there was no enamel hypoplasia visible on clinical examination, suggesting that cerebral palsy is produced more readily than enamel hypoplasia. Because of the high correlation between cerebral palsy and enamel hypoplasia in cases where both were present and because of the apparent difficulty in the production of enamel hypoplasia as mentioned above, enamel hypoplasia may help the clinician and the researcher to determine when the brain injury occurred in cases where the etiology is subtle or apparently unknown.