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We hope that making available the relevant information on Pachyonychia Congenita will be a means of furthering research to find effective therapies and a cure for PC.
Premature eruption in the primary dentition – a clinical and radiological study.

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SUMMARY
A longitudinal study has been made of prematurely erupted primary teeth—dentitio praecox, here defined as eruption previous to 3 months of age. The study comprises 144 children with altogether 232 prerupted teeth. The observation period varied between about 1 and 10 years, being more than 5 years in about two-thirds of the cases. An investigation has been made of incidence, hereditary factors and of complications. Statistical calculations have been made in respect of age at loss of teeth for comparison between the teeth which erupted before 30 days of age and those erupted between 31 and 90 days of age, and for comparison between these groups and a normal material. The clinical and radiological picture is described and recommendations are given for therapy.

INTRODUCTION
Premature eruption, dentitio praecox, is considered according to the older literature to exist in the primary dentition if the first tooth erupts before 4—5 months of age. In an investigation of a Swedish population Lysell, Magnusson & Thilander (1962) found that 95,5 % of children got their first tooth after 3,9 months and before 12,2 months of age. It therefore appears more adequate to define dentitio praecox in the first dentition as eruption prior to 3 months of age.

The most extreme cases of dentitio praecox are eruption previous to birth—dentitio connatalis—and during the neonatal period (first month of life)—dentitio neonatalis.

These extreme forms of dentitio praecox have attracted attention since ancient times. The literature is also rich in descriptions of individual cases. No long-term studies, however, have been made on larger series and little attention has been devoted to teeth which erupt between the neonatal period and 3 months of age.

Reports of the incidence of connatal and neonatal teeth vary. One reason may be that the investigations reported concern different populations. However, the variations may perhaps as much be ascribed to incomplete surveys. Allwright (1958) reported two cases of connatal teeth in the 6817 Chinese children born
in 1953 at the largest maternity hospital in Hong Kong. The number of cases of neonatal teeth known to him from the same series was 10. Owing to the low follow-up rate he did not consider that he could give a reliable figure for the incidence of the latter. In a Danish material based on reports from midwives and district nurses Bodenhoff (1959) estimated the figure to be at least 1:3000, but considered there was reason to presume the actual number to be greater. The table she compiled of incidences reported by different authors included figures which varied between 1:1000 (Mimura, Tokyo, 1930–33) and 1:30.000 (Puech, 1876). The connotally and neonatally erupted teeth usually belong to the regular primary dentition. In the absolute majority of cases they are central mandibular incisors.

Many different causes of dentition praecox have been proposed, both general and local, but no conclusive explanation has appeared.

Among complications which may arise through dentition praecox are hypermobility with risk of the tooth becoming loose, mineralization disturbances and traumatization of soft tissues.

In view of the many obscure factors relating to dentition praecox it was considered important to collect a material for further study.

MATERIAL AND METHOD

The present investigation was started in 1959. It was decided that it should relate to teeth erupted before 3 months of age. A request was addressed to all five maternity departments in Stockholm that newborn children with erupted teeth should be reported to the Eastman Institute. Positive replies were received from all, but the Sabbatsberg Hospital is the only institution from which patients have been referred with great regularity during the entire period of the investigation.

A request was also sent to the Child Health Centers in Stockholm that children whose teeth had erupted prior to 3 months of age should be referred to the Eastman Institute. To stimulate the mothers to visit the Institute, systematic dental treatment of their children was promised during their preschool age.

Up to 1971 records existed of 144 children with altogether 232 prematurely erupted teeth. With a few exceptions the primary investigation and subsequent examinations were made by the author. A general case history was recorded, including particulars on pregnancy, delivery, term of birth of child, birth weight, feeding and hereditary factors. The clinical status was supplemented, whenever possible, by radiographs and colour photographs. Even if the teeth were visible as mucosa-covered elevations on the crista, they were not counted as erupted until some part of the enamel was really exposed in the oral cavity.

As the collection of the material has been proceeding for over 11 years, it has been possible to keep continuous record of clinical and radiological data in many cases up to the school age. All children, except two who lost their teeth at a very early stage, were followed up for at least one year. In the majority of cases the follow-up was considerably longer. Thus, it was possible to follow up about 100 cases more than 5 years, some 40 one more than 7 years and a few cases up to 10 years. Some in up has been unavilably in the view children.

OBSERVATIONS

The children cons boys. There was reference as regards

With the except the erupted teeth central incisors. T of the lip and situated close to case a lower late in addition to the

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Among the 10 there were 5 pairs of sil were close relative siblings who were connatal teeth, a siblings the moth maternal aunt ha 3 months of age. I were definite rep in a close rela positio can acc

In the great ma and delivery had plications. Of 137 weight, 7 weighed was accordingly
10 years. Some irregularity in the follow-up has been unavoidable, however, especially in the view of the young age of the children.

OBSERVATIONS

The children consisted of 74 girls and 70 boys. There was accordingly no sex difference as regards premature eruption.

With the exception of 3 of the 144 cases the erupted teeth consisted of one or both central incisors. Two had congenital clefts of the lip and palate, the teeth being situated close to the cleft. In the third case a lower lateral incisor had erupted in addition to the two central incisors.

In no case was dentitio praecox associated with any known syndrome. Apart from the three patients with congenital clefts of the lip and palate there were no other developmental disorders. In one of the patients, more exactly one of the cleft cases, there was a supernumerary tooth, otherwise all teeth belonged to the regular dentition.

Among the children examined there were 5 pairs of siblings and one pair who were close relatives. In one of the pairs of siblings who were twins, the father had connatal teeth, and in another pair of siblings the mother, grandmother and a maternal aunt had erupted teeth before 3 months of age. In another 16 cases there were definite reports of dentitio praecox in a close relative. Some hereditary disposition can accordingly not be ruled out.

In the great majority of cases pregnancy and delivery had been free from complications. Of 137 cases with known birth weight, 7 weighed less than 2500 g. There was accordingly no manifest correlation with prematurity in these respects, nor was there any case of apparent postmaturity.

As already mentioned, it is only Sabbatsberg Maternity Department that has fairly regularly reported cases of connatal or neonatal teeth during the whole period of the investigation. During the period 1960—1970 the number of cases was 17. In addition there were also 20 children born at Sabbatsberg during the same period whose teeth erupted before 3 months of age. The number of births during the 11-year period was 25,915. The incidence of children with connatal or neonatal teeth is then 1:1524 and with eruption before 3 months of age 1:700. These figures, which are undoubtedly minimum figures, confirm Bodenhoff's (1959) assumption that her findings were too low.

Fig. 1. shows the breakdown of the material over four eruption periods, 0—14, 15—30, 31—60 and 61—90 days. It was found that the majority of the teeth which erupted during the first month of life had been observed during the first two weeks and that the majority of the teeth erupted after the first month had been observed during the third month of life.

Of the early observed teeth 72 were connatal; in 23 cases there was a bilateral connatal eruption.

Times for loss of teeth

A study of the known times for loss of prematurely erupted teeth showed that the mean age for loss of teeth erupted before one month of age was as low as 32.85 ± 3.86 months. The corresponding time for teeth erupted during the second and third months of life was 63.07
± 1.94 months. The mean difference, 30.22 months \( (t=6.09) \), is highly significant.

In a normal material (Sundvall-Hagland, 1955) the time for loss of the primary lower central incisors may be estimated at 73.51 ± 7.05 months. The difference in relation to those erupting in the second and third months of life is 10.74 months, this too a highly significant difference.

Fig. 2. is an attempt graphically to demonstrate the times for loss of individual teeth. As in many cases the teeth still remained at the last inspection, a probability calculation has been made as regards some sections of the material. The curve for teeth erupted before one month of age rises sharply towards one year of age, thereafter slowly up to 5 years. The curve for teeth erupted during the next two months of life, on the other hand, runs almost horizontal from 0 to 4 years, thereafter rising sharply. This is due to the fact that 38 % of the teeth in the first group were lost before one year of age. The corresponding figure for the second group is 2 %. There is a highly significant difference between the groups up to 5 years of age.

Of the 121 teeth which erupted before one month of age 46 were lost during the first year of life. No less than 40 of these were lost before 3 months of age. Among the teeth remaining at one year of age very few were lost before 4 years of age. But even after deducting the earliest losses and those lost through trauma, teeth which had erupted before one month of age had a greater tendency to premature loss than teeth which erupted in the second and third month of life.

Table 1 shows the causes of the losses of teeth. The main losses recorded during the observation period were due to spontaneous exfoliation or to extraction on account of imminent exfoliation. There was a fundamental difference, however, between the extremely early lost teeth and those shed or extracted later. In the first group, in which, with two exceptions, the teeth had erupted before 14 days of age, there had not yet been any root formation. In the second group the loss of teeth had been preceded by resorption of a more or less normally developed root.

Complications
Out of the total material there were 10 cases with tongue affection, all with tooth eruption before 14 days of age. Only two of these gave rise to extraction of the causative tooth. In one case a large decubitus had formed and in the other a barely walnut-sized tissue proliferation under the tongue (figs. 3, a, b). Both cases healed rapidly after extraction of the tooth. The remaining cases healed either spontaneously or after smoothing the incisal edge by cautious grinding. The number of cases with tongue affection appears small compared with the predominant view in the literature.

Breast-feeding difficulty is another of the complications which are often reported as a sequela to dentitio praecox. In the present material such affections of different degree of severity occurred in some 50 cases. Difficulty for the mother in breast-feeding, however, has not led to extraction in any case. In the case shown in brackets in table 1 the difficulty of breast-feeding merely reinforced other indications for extraction. In most cases
Premature eruption in the primary dentition

As recorded during were due to spontaneous extraction on exfoliation. There were no exceptions, however, nearly lost teeth and later. In the first 14 days of eruption period, there been any root exposure. The group showed loss of teeth due to resorption of the crown developed root.

![Graph](https://via.placeholder.com/150)

Fig. 1. The material distributed over the four eruption periods.

![Graph](https://via.placeholder.com/150)

Fig. 2. Graphic presentation of probable times for loss of teeth erupted 31-90 days (-----) and before 30 days of age (---) and for teeth in the latter group with the exception of those lost prior to 3 months of age or through trauma (-----).
Fig. 3 a. Decubitus lesion under the tongue of a 3-week-old girl with connatal tooth 81 (01-).

Fig. 3 b. Tissue proliferation under the tongue of a 2-month-old boy with connatal tooth 81 (01-).

Fig. 4 a. Acute gingival abscess at connatal 81 (01-) with severe attrition in a 15-month-old boy.

Fig. 4 b. Connatal tooth 81 (01-) with moderate attrition and discoloration in a 4 years 3 months old boy. Radiograph (Fig. 10) shows severe intraradicular resorption.

Fig. 5. Toothlike formation in region 71 (-01) following extraction at four weeks of age of connatal 71 (-01) in a 16-month-old girl.

Fig. 6 a. 81 (01-) 71 (-01) at day of life. The visible part of 1 mm high, and rested on a 5 papilla. 81 (01-) exfoliated at was removed at about 5 mor on 14th day.

Fig. 6 c. 81 (01-) erupted at years. Photo on 9th day.

Fig. 8. Extreme attrition of p71 (-01). Photo at 5 years 8 m-
Fig. 6a. 81 (01) 71 (-01) erupted on second day of life. The visible part of the crowns were 1 mm high, and rested on a 5 mm high gingival papilla. 81 (01) exfoliated at 17 days; 71 (-01) was removed at about 5 month of age. Photo on 14th day.

Fig. 6b. 71 (-01) erupted on first day of life; persisted 5 years. Photo on 16th day.

Fig. 6c. 81 (01-) erupted at birth; persisted 6 years. Photo on 9th day.

Fig. 7. Typical picture of persistent tooth; 71 (-01), erupted previous to 30 days of age. Radiograph at same age (Fig. 12). Photo at 1 year 8 months.

Fig. 8. Extreme attrition of prerupted 81 (01-), 71 (-01). Photo at 5 years 8 months.

Fig. 9. 41 (1-) in typical rotated position, not crowded. 81 (01-) erupted at birth, partially loosened and removed at 16 days of age.
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Fig. 6 c. 81 (01-) erupted at birth years. Photo on 9th day.

Fig. 8. Extreme attrition of pre-erupted 71 (01). Photo at 5 years 8 mon
Preliminary eruption in the primary dentition

Fig. 6 a. 81 (01-) 71 (-01) erupted on second day of life. The visible part of the crowns were 1 mm high, and rested on a 5 mm high gingival papilla. 81 (01-) exfoliated at 17 days; 71 (-01) was removed at about 5 months of age. Photo on 14th day.

Fig. 6 b. 71 (-01) erupted on first day of life; persisted 5 years. Photo on 16th day.

Fig. 6 c. 81 (01-) erupted at birth; persisted 6 years. Photo on 9th day.

Fig. 7. Typical picture of persistent tooth; 71 (-01), erupted previous to 30 days of age. Radiograph at same age (Fig. 12). Photo at 1 year 8 months.

Fig. 8. Extreme attrition of unerupted 81 (01-), 71 (-01). Photo at 5 years 8 months.

Fig. 9. 41 (1-) in typical rotated position, not crowded. 81 (01-) erupted at birth, partially loosened and removed at 16 days of age.
Table 1. Causes of loss of teeth with break down over the four eruption periods.

<table>
<thead>
<tr>
<th>Eruption at</th>
<th>0-14 d</th>
<th>15-30 d</th>
<th>31-60 d</th>
<th>61-90 d</th>
</tr>
</thead>
<tbody>
<tr>
<td>Extraction before examination</td>
<td>8</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Spontaneous exfoliation or extraction owing to imminent exfoliation, 0-6 m</td>
<td>31</td>
<td>1</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>Ditto, 6 m—4 y</td>
<td>4</td>
<td>3</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>Ditto, over 4 y</td>
<td>35</td>
<td>1</td>
<td>15</td>
<td>58</td>
</tr>
<tr>
<td>Tongue affections</td>
<td>2</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Breast-feeding difficulty</td>
<td>(1)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Periapical complication</td>
<td>8</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>(not caries-induced)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Trauma</td>
<td>6</td>
<td></td>
<td></td>
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</tbody>
</table>

Breast-feeding has instead stopped fairly soon. In three cases an acrylic "breast-feeding splint" extending over the alveolar process was tried with good result. The splint was made from an impression of the mandibular crista and was placed in the child's mouth during breast-feeding. Even if the experience with this splint has been satisfactory, it is questionable whether it should be used other than in special cases.

All of these complications occurred in close association with the eruption and they were of types chiefly reported in the literature. However, the literature also contains accounts of certain other complications appearing at an early stage which have not occurred in this material, for example marginal infection and osteomyelitis.

Allwright (1958) described some cases of chronic apical ositis in conjunction with congenital lower incisors which had erupted at least two years previously. He related the apical ositis to poorly developed enamel, which had worn away, after which there had been a bacterial invasion of the pulp. Allwright's description is largely similar to the 8 cases of periapical complication shown in table 1, in one of which, however, there was an acute gingival abscess. The remainder were discovered owing to fistulation or through radiographic examination. The age at onset of the complication varied between 1 year 3 months and 4 years 9 months. Caries or trauma had occurred in none of these cases. (Figs. 4 a, b, 10).

The teeth lost through trauma had all erupted before 14 days of age. This may be a coincidence, but may also be interpreted in the manner that these teeth were easily dislocated, as they were less securely attached in the jaw.

An interesting complication noticed in 8 cases was some kind of tooth formation after the early loss of a connatal tooth.
ion periods.

Fig. 10. Radiograph at 4 years 3 months of age of 81 (01-) erupted at birth (see Fig. 4 b) showing severe intraradicular resorption.

Fig. 11 a. Radiograph at about 10 months of age showing tooth formations in region 81 (01-), 71 (-01) following extraction of connatal 81 (01-), 71 (-01) during first week of life.

Fig. 11 b. Radiograph at 6 months of age of lower front of jaw showing small root formation developed after removal of connatal 81 (01-); 71 (-01) erupted at 9 days of age.

Fig. 11 c. Radiograph of the same patient at 3 years of age, showing further development of the formation.

Fig. 12. Radiograph at 4½ years of age of 71 (-01) erupted at birth (see Fig. 7).

is to poorly developed worn away, after bacterial invasion.

h’s description is cases of periapical table 1, in one of was an acute gingivitis discovered through radiograph. The age at onset of ed between 1 year and 9 months. Caries found in none of these 4).

gh trauma had all of age. This may may also be interest at these teeth were vary were less secure.

lication noticed in of tooth formation of connatal tooth.
These formations may be described chiefly as dwarfed roots. In some cases there was a rudimentary crown in which the pulp was visible as a small red spot centrally. (Figs. 5, 11, a, b, c.) In two cases the formations gave rise to periodical ostitis and in one other to irritation of the tongue. The occurrence of such structures may be explained by the fact that, on removal of the connatal tooth, all tissue with the specific ability to continue the formation of tooth had not been removed or destroyed.

A similar phenomenon was described by Ryba & Kramer (1962). In an infant girl, whose deciduous lower central incisors were erupted at birth, the one was dislodged a few days after birth and the other one was removed about three weeks later. By the age of 13 weeks two tumour-like masses had formed in the lower central incisor region. Histologic examination of these masses showed formation of both regular and irregular dentine and continued activity of part of Hertwig’s sheath.

Southam (1968) has given further examples of continued growth of dentine papillae after early loss of connatal lower central incisors.

As already noted, roughly one-third of the teeth which had erupted before 1 month had been lost before 3 months of age. Most of them had been connatal. With a few exceptions they had fallen out spontaneously or been extracted owing to a great risk of becoming loose. On examination of the clinical and radiological picture certain common features were found in these cases, which may be summarized as follows:

The tooth usually rested on a relatively high papilla-like bed—in two cases the latter was tumour-like and attached to the gingiva by a short narrow stem.

The tooth bed was more or less mobile and the tooth itself often displayed increased mobility.

The gingiva around the tooth might be reddened, and bleeding might occur at the gingival margin.

The clinical crown appeared to be long or increased quickly in length since the mucosa, which partially covered the tooth primarily, retracted within a few days.

The colour of the tooth might be pink or greyish-blue, indicating intradental bleeding. In other cases it was whitish or yellowish, but might also be normal. In some teeth the enamel displayed local defects and their consistency might appear soft as cartilage.

The teeth were often small and of abnormal shape. When occurring symmetrically they sometimes covered one another like bricks. In cases when radiographs could be taken, no root development was observed and the tooth appeared to have very little or no contact with the alveolar bone. (Fig. 6, a.)

Teeth with a longer duration among those early erupted also had a deviating pattern primarily in most cases. (Figs. 6, b, c.) The character of the deviations was the same as in the group described above, but the most extreme forms were not observed. Common to all of the connotally and neonatally erupted teeth, however, was that after a fairly short period of exposure in the mouth they assumed a yellow or yellowish-brown colour. In many the crown was relatively small even at the time of eruption. In others the crown gave the impression of uniformly dim as time went on. This plainly by general attritional layer of enamel. Furthermore, there was an in some cases very pronounced. The radiographs showed fairly short root, in most abnormal shape. The crowns usually narrow and of (Fig. 12).

Concerning teeth with the second and third variations in the clinical picture but by no means uniform (about 50%). In so far were recorded, however same kind as in the previous—with a few exceptions, especially as regards incidence of abnormality diminish the later the to

Etiology

The present investigat is able to elucidate the eruption. One might in a matter of general entire dentition. No condition of the material has respect. But at least as with the earliest eruption is clear to be so. In a tedly, the homologous during the 3-month period teeth have erupted at in some cases even regularity, Premature loss of a result in lack of space successor. In general, lack of space as a rule is not noted in t
in two cases the crown was attached to a narrow stem.
more or less mobile tooth might develop in the tooth might be pink or grayish in color. In displayed local decay might appear as long in length since the tooth covered the root by a few days.
A tooth might be pink or grayish in color. In displayed local decay might appear small and of abnormal shape. The pulpal cavity was usually narrow and of irregular shape. (Fig. 12).
Concerning teeth which had erupted in the second and third months of life deviations in the clinical picture were noticeable but by no means to the same extent (about 50%). In so far as abnormalities were recorded, however, they were of the same kind as in the preceding groups, but—with a few exceptions—less pronounced, especially as regards the root. The incidence of abnormalities seemed to diminish the later the tooth erupted.

Etiology
The present investigation has not been able to elucidate the cause of premature eruption. One might suppose that it was a matter of general early eruption of the entire dentition. No consistent examination of the material has been made in this respect. But at least as regards the group with the earliest eruption, this does not appear to be so. In a few cases, admittedly, the homologous tooth has erupted during the 3-month period, but the other teeth have erupted at the normal time, in some cases even relatively late.

Premature loss of a primary tooth may result in lack of space for the permanent successor. In general, however, no such lack of space as a result of early loss of teeth was noted in this material. When crowding was found, it was moderate. Only a few cases could be characterized as pronounced.

An observation, finally, which may be worthy of mention was that in some cases, irrespective of the time of loss of the primary tooth and irrespective of the dental arch space, the permanent tooth erupted in a rotated position. (Fig. 9).

THERAPY
On the basis of the observations made in this study certain recommendations can be given for the therapy of dentitio praecox:

Careful supervision of connatal and neonatal teeth up to 2—3 months of age in respect of tendency to loosening and the associated risk of aspiration.

Inspection of the tongue for decubital sores.

Attention to difficulties in breast-feeding.

Extraction should be done:
a. as soon as signs of loosening are observed (bleeding at gingival margin, internal discoloration, partial loosen- ing of tooth),
b. on the occurrence of severe affection of the tongue, when smoothing the incisal edge has not caused, and is not considered able to cause, any improvement,
c. in the event of difficulty in breast-feeding when conventional measures or the "breast-feeding splint" referred to above are not indicated.

Severe bleeding after extraction, concerning which a warning is issued in the literature, has not been observed in the present material.

Clinical and radiological check-up after...
$\frac{1}{2}$—1 year in respect of subsequent development of the tooth in cases when early extraction has been performed.

Continuous clinical and radiological follow-up of teeth which in any way deviate from normal, in respect of e.g. periapical complications.

In cases when subsequent tooth formations have been left intact, they should be followed up in the same way.

This investigation was started at the initiative of the Late Head of the Department of Operative Dentistry of the Eastman Institute. Dr Irene Sundvall-Hagland. The investigation was supported by a grant from Stockholms stadsfullmäktige: No 134. Bidrag till medicinsk forskning vid Stadens sjukhus och andra vårdinrättningar.

Helpful suggestions as regards the manuscript and the illustrations were given by professor Bengt Magnusson, Faculty of Odontology, University of Göteborg.

REFERENCES


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GULL BJUGGREN

En longitudinell studier — dentitio praecox 3 månaders ålder.


En särskilt intresse uppmärksammar oss att 31 av 90 tändorna hade forlorades merparten.

I en del fall förekommer spontan exfoliation av tändarten. I med något enstaka ur 14 dagar.

En annan komplikationen, som är oftast av betydelse i kroppen, är att ett fall kan återfinna sig efter en tid.

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Av dem som består av tändorna, funna man at...