Anomalies of the Lateral Incisor in Cases of Harelip and Cleft Palate.

By

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The object of the work presented here is to attempt to ascertain what anomalies can be found in the lateral incisor of patients with harelip and cleft palate, and how these anomalies appear in the temporary and the permanent dentitions.

The investigation has been made during the week 4/26—5/4 1948, at *Statens Institut for Talelidende* (The State Institute for Patients with Defects of Speech), Hellerup, Copenhagen. This institute directs the centralized treatment of the Danish patients with harelip and cleft palate, and has therefore at its disposal the material necessary for such an investigation.

Through the great courtesy of Mr. H. Bering Liisberg the Chief of the Institute, and also the organizer of the centralization plan, I obtained permission to examine those cases of interest. The dental clinic of the institute was placed at my disposal, and I was given all the necessary information concerning the patients besides the assistance of a nurse. For all of this I am greatly indebted to Mr. H. Bering Liisberg.

It is a well-known fact that patients born with a harelip, or with a harelip and a cleft palate, show irregularities of their teeth. This observation has proved to be of interest also beyond the sphere of odontology, i. e. to embryology and genetics. However, according to literature it is apparent that previous investigations of the teeth are in many ways inaccurate and imperfect. This is mainly due to the fact that it has been too difficult to collect sufficient material with good case histories to eliminate the possibility of errors.

It is primarily at the lateral incisor in the cleft area that the irregularities prevail: supernumerary tooth, congenital absence, hypo- and hyperplastic variations and malformations, and displacements. At times some of the same deviations from normal are observed in the adjacent central incisor and the canine. Generally the bite shows malocclusion. Beyond these facts the foundation for further conclusions has often been imperfect. This is illustrated in the three most comprehensive works which I have seen on the subject, written by odontologists.

PREISWERK (1908) and BILLING (1912) found, with 32 and 46 patients respectively, of different ages and without x-ray examination, that absence of the lateral incisor was the most frequent anomaly.

Contrary to this MILLHON and STAFNE (1941), with 81 patients of different ages, observed that reduplication of the lateral was more frequent than absence. They also asserted that the tendency to form supernumerary teeth is in direct proportion to the tendency towards delayed or improper fusion of the embryonic facial processes in question.

To all three works, however, it applies that one does not always know what influence earlier treatment — operation, extraction — may have had. With older patients one does not know anything about the teeth of the first dentition.

Concerning the temporary teeth in harelip and cleft palate patients there are, as a whole, only a few observations to be found published. The better part of these is rendered by a surgeon, VEAU (1934). From his vast experience he has stated that the foetus has nearly always one lateral on each side of the cleft. The medial of these is always insufficiently fixed in the jaw bone and will as a consequence often fall out and disappear at an early stage of the child's life. The precanine tooth at the back of the cleft has, however, a better hold and a better nutrition and will often remain. About the permanent lateral VEAU says: "La regression s'accentue encore: il m'a semblé que l'incisive médiane est plus souvent absente que la dent précanine."

Irregular form of the lateral incisor, mainly hypoplasia, has often been noticed in former investigations.

The discussions about the interpretation of the various findings have chiefly taken two directions.

First it has for many years been a matter of disagreement how it could be understood that in one case two lateral teeth

could be found, one on each side of the cleft, and in another case absence of same. Furthermore, how the fact was to be explained that when there was one single lateral in the cleft area, it sometimes appeared at the medial side of the cleft, and sometimes at the distal side of it. According to some recent works on the development of the premaxilla (INOUYE 1912, PETER 1921) it has been maintained that as a rule the cleft strikes the germ of the lateral and splits it in two parts. One or both may be developed or destroyed. This was considered a satisfactory explanation for all the findings. However, this way of reasoning has been attacked from odontological quarters. MATHIS (1935) stated that "the splitting of the lateral's germ" presupposed an earlier presence of the entire germ, what was out of the question with the conception of the morphogenesis of the cleft formation which the embryologists alleged. Instead of this he asserted that the border of the cleft meant an extension of the oral mucous membrane in the precanine section, which at the same time involved a possibility of the development of an extra tooth at the back of the cleft: the lateral here meant an additional contribution from the elongated dental lamina. But even MATHIS's hypothesis will not directly cover the findings which have formed the basis for the second question under debate, and of which also a brief account will be given.

It has long been stated that patients with different degrees of harelip, but without a trace of cleft in the jaw, may show deviations similar to those which have been mentioned above. Where such an aberration on a front tooth appears, without a sign of simultaneous harelip, some geneticists give good reasons for assuming that this may represent a so-called "microform" of harelip and cleft palate, provided the patient has a relative with a manifest cleft. It is then presumed that the growth-disturbing effects of the hereditary disposition mostly have been overcome at an early stage of the embryonic life; only traces on a tooth, most likely on a lateral incisor, remain. (MENGELE 1939).

P. FOGH-ANDERSEN (1942) says about the same subject: "— — — there can probably be no doubt that dental anomalies in relatives of harelip patients may be due to the same disposition as that which induces the cleft formation, but the cases are difficult to distinguish from the rather frequently occurring dental anomalies that are hardly due to this disposition."

Concerning the existence of microforms an agreement seems prevalent. But the term apparently needs a further revision and a sharper limitation before an estimate of the microforms and their significance becomes possible.

A clarification of the problems mentioned in this introduction may very likely have several consequences, i. e. as to how we must understand the morphogenesis of certain anomalies in the section of the lateral incisor, whether the processus alveolaris has a cleft or not, and also for future works on the heredity of these malformations. In order to achieve this, we must presume that further investigations into the teeth of patients with harelip and cleft palate are of importance.

The Investigation.

Owing to the short time at my disposal, only patients between 3 and 7 years of age were selected for examination. Moreover this age was regarded as the most expedient for the following reasons:

Through clinical examination and x-rays one could get a survey of the conditions in the temporary as well as the permanent set of teeth.

One must be allowed to assume that such young patients had either never been treated by a dentist before, or that fairly definite information about extractions of front teeth was available.

71 children attended, accompanied by one or both parents. As regards to 10 patients the examination was impossible or the x-rays unsuccessful. These cases are excluded from the investigation. There are x-rays of the anterior teeth of the maxilla of the remaining 61 cases. In 52 of these, impressions were also taken and models poured. One patient with isolated cleft palate was unoperated, the rest had been operated on at the same surgical department, Diakonissestiftelsens Hospital, Copenhagen, by Surgeon-in-Chief V. FOGH-ANDERSEN. Labioplasty had been performed at the age of 2 months, with simultaneous palato-vomerplasty when necessary. The cleft palates had been closed at the age of 2 years. Records with the diagnoses, photographs taken before and after the operations and other information were examined, and specific information from parents were noted.

Besides this material 2 cases of the same group of age, with isolated harelip (operated), have been included. Thanks to Miss INGEBORG BROCHMANN, Chief of the School Dental Service in

Drammen, x-rays, models and definite information about these children are at hand.

In most cases a pronounced displacement of certain teeth in the cleft area was found, and also malocclusion of a characteristic form where the alveloar process was cleft. A more detailed mention of these conditions is considered unnecessary, as it would not add anything to the observations of E. HARVOLD, recently presented in his work on the development of the upper jaw by harelip and cleft palate (1947).

According to parents' information it was noted in one case that a tooth had been lost before the normal shedding time, "a tooth in the palate on the left side" (case No. 54). With regard to the poor retention in the jaw offered especially to the lateral incisor on the foremost side of the cleft, it must be taken into account that this is almost certainly not the only case of too early lost deciduous lateral. On the other hand, every anlage to permanent lateral incisors must have been registered on the x-rays.

The material has been divided into 3 groups as follows:

Group A: Isolated prealveolar harelip. Neither by clinical examination, nor by x-rays, is there a traceable cleft in the processus alveolaris. 15 cases, of which 1 is bilateral, in all 16 cleft sides.

Group B: Harelip and alveolar cleft (with or without associated cleft palate). 33 cases, of which 7 are bilateral, in all 40 cleft sides.

Group C: Isolated cleft palate. 15 cases.

Group A.

Nothing was wrong in with the central incisor or the canine on the cleft side in any of the patients. Table A gives a survey of the laterals found on this side, and also notes about abnormal form and/or size. Where a supernumerary is found the mesial lateral is designated with x and the distal with y. Where duplicate laterals in the temporary set are beeing succeeded by a single one in the permanent set, an (x) or (y) has been added in order to show which one of the two temporary laterals the permanent one seems to succeed, as judged by x-ray. Generally the temporary laterals maintain their usual incisor form, even with considerable variation in size. The greatest deviation in form among them shows the hyperplastic "T-form", which will later be dealt with. The permanent lateral incisors vary more decidedly in form, but as they always are unerupted it is often difficult from the x-ray only to characterize them otherwise than as "big" or "re-

Table A.

Case No.	Sex		Age yrs.	The harelip (only prealveolar cleft.)		Decidous lateral			Permanent lateral		
				Side	Description	Supernumerary x + y	Single	Absent	Supernumerary x + y	Single	Absent
1		♂	5	R	1/2 of the lip	+				+ (y) rudimentary	
				L	small notch in the prolabium.		+ T-form			+	
4	♀		4	R	1/2 of the lip	+ x T-form y reduced				+ (y) reduced	
8		♂	7	L	1/3 of the lip	+ x big y reduced				+ (x) big. with tuberculum dentis	
9		♂	7	R	1/2 of the lip	+			+ x reduced y »		
10		♂	6	L	»		+				+
13		♂	6	L	3/4 of the lip		+ big			+	

29	♀		6	R	complete		+			+ reduced	
45		♂	5	L	1/2 of the lip (+ cleft palate: soft and 1/2 of hard.)		+ T-form		+ y reduced		
58		♂	4	L	1/2 of the lip (+ cleft palate: only velum)	+ x very big y big				+ (x)	
59	♀		4	L	1/2 of the lip	+ x big					+
60		♂	7	L	»		+ with tuberculum dentis			+ rudimentary	
61	♀		3	R	»		+			+	
62		♂	3	L	»		+ big		+ y reduced		
72	♀		7	L	»		+			+ reduced	
73		♂	4	L	»	+			+ y reduced		
15	5	10		5 R 11 L		7	9		4	10	2

duced", after comparison with the corresponding lateral on the opposite side.

In the following, duplicates and/or hyperplasia will be designated as "overproduction", and absence or hypoplasia as "underproduction". From table A it is seen that the 15 cases with a total of 16 cleft sides show the following conditions for the lateral incisors:

	Deciduous teeth	Permanent teeth
supernumerary (x + y)	7	4
single (x or y)	9	10
absence	—	2
overproduction	12	5
normal production	4	4
underproduction	—	7
hyperplastic teeth	10	1
normal »	11	7
hypoplastic »	2	10

It will be observed that some of the duplicates in the temporary dentition are even hyperplastic.

Where x- and y-laterals can be classified, we notice:

	Deciduous teeth	Permanent teeth
hyperplastic x	4	1
normal »	3	4
hypoplastic »	—	1
hyperplastic y	1	—
normal »	4	—
hypoplastic »	2	6

With a view to how the permanent laterals succeed the deciduous in each patient, we arrive at the following facts:

Deciduous laterals	number of times succeeded by		
	supernumerary	single	absence
7 supernumerary	2	4	1
9 single	2	6	1
	overprod.	normal prod.	underprod.
12 overproduction	5	3	4
4 normal production	—	1	3

In one case (No. 9) there is, on the sound side, opposite the cleft, an irregularity that indicates microform. The findings on the right side are (see table A): incomplete harelip, supernumerary lateral in both dentitions. On the left side the lip is normal, but instead of 1 there are 2 deciduous laterals here

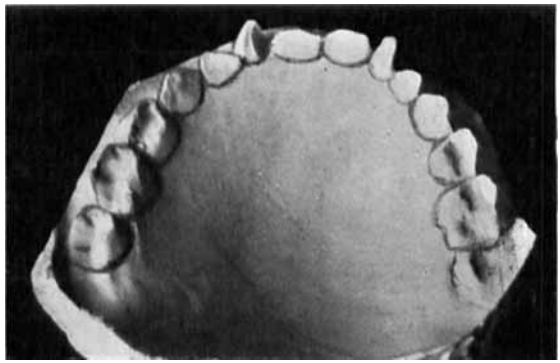

Fig. 1.

also, see fig. 1. These are, however, according to the x-ray, succeeded by only 1 permanent lateral, the crown of which is situated between the roots of both deciduous laterals and with the distal corner rotated lingually through approximately 90°.

It is impossible from this restricted material to draw any conclusion about correlation between the size of the cleft in the lip and the anomalies in the teeth.

Group B.

Table B presents the conditions in the cleft area in patients where a more or less pronounced cleft also can be shown in the alveolar process. In the note columns any striking anomaly of neighbouring teeth has been marked off. The lateral on the foremost border of the cleft has been denoted with x, and the one behind the cleft with y. In addition to similar irregularities, as have been found in group A, there has in the temporary dentition been found one precanine tooth, a y-lateral, of the same size and almost the same form as the neighbouring normal canine (case No. 40), fig. 2. In the permanent laterals 2 distinct malformations could be seen (cases No. 32 and 40), and the reduced forms were often more pronounced than in the preceding group.

Table B.

Case No.	Sex	Age yrs.	Cleft formation		Deciduous lateral				Note	Permanent lateral				Note
			Side	Description	Super-numerary x + y	Single x	Single y	Ab-sent		Super-numerary x + y	Single x	Single y	Ab-sent	
2	♀	3	L	3/4 of the lip, submucous alveolar cleft.	+ y T-form							+ rudimentary		
3	♀	4	R	harelip complete, alveolar cleft incomplete. 1 cm. of proc. alv. missing				+					+	
5	♀	5	L	harelip complete, alveolar cleft incomplete. 1 1/2 cm. of proc. alv. missing				+					+	
6	♂	5	R	1/2 of the lip, alveolar cleft incomplete with a V-formed notch in proc. alv.	+ x big								+	
7	♂	3	R	harelip complete, alveolar cleft incomplete. 1 1/2 cm. of proc alv. missing			+		i_1 big and atypic, malformed				+	
11	♂	6	L	harelip complete, submucous alveolar cleft.			+					+ reduced		
12	♂	6	R	1/2 of the lip, alveolar cleft incomplete	+ x T-form y T-form				i_1 knocked out 3—4 yrs. ago				+	
31	♂	6	R	3/4 of the lip, alveolar cleft and cleft palate	+								+	

32		♂	6	L	1/2 of the lip, alveolar cleft and cleft palate	+					+ y big, malformed, tubercular shaped crown				
34		♂	7	L	harelip with a small bridge of skin. Alveolar and palatal cleft complete.				+					+	I_1 has too short root
38	♀		5	L	1/2 of the lip, alveolar and palatal cleft.		+					+ rudimentary			
39		♂	5	R	complete harelip and cleft palate	+					+ y reduced				
40		♂	6	R	broad complete harelip and cleft palate	+ y big, canine-shaped.						+ small, atypic, malformed			
41	♀		5	L	»		+					+ reduced			
43		♂	6	L	harelip with a small bridge of skin. Alveolar and palatal cleft complete.	+							+ reduced		
44	♀		5	R	1/2 of the lip, submucous alveolar cleft, palatal cleft complete			+						+	
46	♀		5	L	Broad complete harelip and cleft palate			+ rudimentary						+	
47		♂	4	R	»			+ reduced						+	

Table B. (Cont.)

Case No.	Sex	Age yrs.	Cleft formation		Deciduous lateral				Note	Permanent lateral				Note
			Side	Description	Supernumerary x + y	Single x	Single y	Absent		Supernumerary x + y	Single x	Single y	Absent	
48	♂	3	R	complete harelip and cleft palate	+								+	
50	♂	4	R	broad complete harelip and cleft palate			+					+ reduced		
52	♂	4	R	»	+ x T-form					+ y reduced				
53	♂	4	L	»				+					+	
56	♀	6	L	complete harelip and cleft palate	+ y reduced							+ reduced		
57	♀	3	L	harelip with a small bridge of skin. Alveolar and palatal cleft complete.			+						+	
64	♀	3	L	broad complete harelip and cleft palate		+					+			
67	♂	5	L	harelip with a small bridge of skin. Alveolar and palatal cleft			+					+ with tuberculum dentis		
28	♂	6	R	harelip incomplete, alveolar and palatal cleft		+							+	
			L	»		+							+	

30		♂	6	R	complete harelip and cleft palate	+						+ y reduced				
				L	»			+				+				
33		♂	6	R	complete harelip and alveolar cleft. No cleft palate	+									+	
				L	»			+						+ rudimentary		
35		♂	7	R	harelip incomplete, alveolar and palatal cleft complete				+						+	
				L	»				+						+	
51	♀		5	R	harelip incomplete, alveolar and palatal cleft				+						+	
				L	complete harelip and cleft palate	+									+	
54		♂	4	R	1/2 of the lip, alveolar and palatal cleft		+							+		
				L	complete harelip and cleft palate				+						+	C missing
55		♂	4	R	»	+ y reduced						y rudimentary	+			
				L	»	+ x reduced y »							+ reduced			
33	11	22		19 R 21 L		16	6	11	7			6	5	8	21	

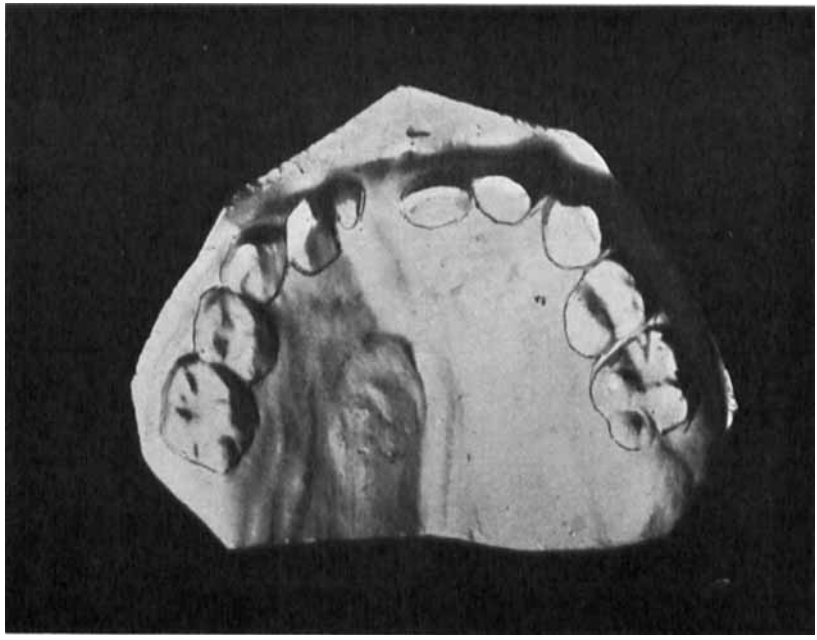

Fig. 2.

Besides, the 33 patients with a total of 40 cleft sides in group B show with regard to the lateral incisor:

	Deciduous teeth	Permanent teeth
supernumerary (x + y)	16	6
single x	6	5
» y	11	8
absence	7	21
overproduction	16	7
normal production	15	2
underproduction	9	31
hyperplastic x	3	—
normal »	18	7
hypoplastic »	1	4
hyperplastic y	3	2
normal »	19	2
hypoplastic »	5	10

Noticing how permanent laterals succeed the temporary ones with each individual patient, one arrives at the following:

Deciduous laterals	number of times succeeded by: supernumerary x + y	single x	single y	absence
16 (x + y)	5	2	3	6
6 x	—	3	1	2
11 y	1	—	4	6
7 absence	—	—	—	7
	overprod.	normal prod.		underprod.
16 overproduction	5	—		11
15 normal production	2	2		11
9 underproduction	—	—		9
	hyperpl. x	normal x	hypopl. x	absent x
3 hyperplastic x	—	1	—	2
18 normal »	—	5	3	10
1 hypoplastic »	—	—	1	—
18 absent »	—	1	—	17
	hyperpl. y	normal y	hypopl. y	absent y
3 hyperplastic y	—	—	1	2
19 normal »	2	1	7	9
5 hypoplastic »	—	—	2	3
13 absent »	—	1	—	12

Only in group B there are patients where an operation has verged on the alveolar process, viz. 29 cleft sides where palato-vomerplasty ad modum VEAU has been performed. Considering the possibility that this may have influenced the germ for the permanent y-lateral, the investigation reveals that the 29 cases mentioned have altogether 18 y-laterals in the temporary dentition, in 10 cases of which they are being succeeded by permanent y-laterals. The remaining 11 cleft sides, where palato-vomerplasty has not been performed, have 9 deciduous laterals succeeded by 4 permanent. Consequently, we find approximately the same condition where this operation has been carried out, as where it has not been performed.

Neither for group B does the material offer any definite facts with regard to a possible connection between the size of the cleft and the character or degree of the irregularities.

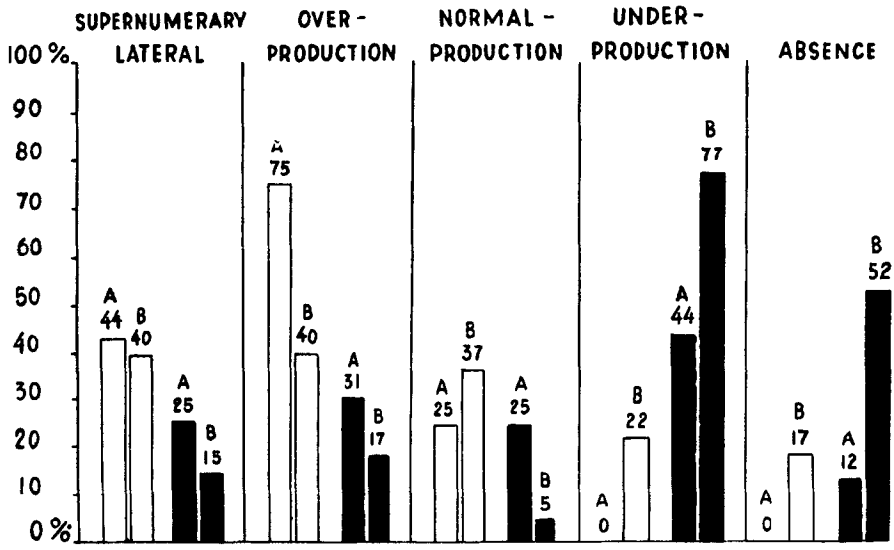

Fig. 3.

Some figures, calculated on the basis of the observations in groups A and B, are placed together in a graphic illustration, fig. 3. As will be seen, this comparison indicates several possible conclusions. But it must be emphasized that the percentages apply only to the patients here examined. For more conclusive figures much more comprehensive material is essential.

The relatively frequent occurrence of the hyperplastic temporary lateral, here called "T-form", makes further comment on this phenomenon desirable. Without having been given a specific name it has earlier been pictured and described by BOLK, COLYER,

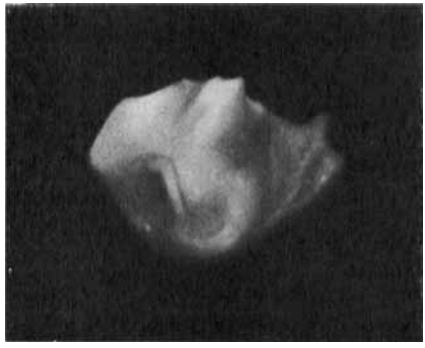

Fig. 4.

DE JONGE COHEN and EULER. It may briefly be characterized as an incisor with a pronounced tuberculum dentis, the point of which is attached to the incisal edge by a crista, a T-form being the result, see fig. 4. The labial aspect has a vertical furrow near the middle. EULER presumes that this crown form may be caused by a specific pressure in the surroundings during development, or that it is a consequence of a more comprehensive malformation, or that it is entirely of hereditary character. DE JONGE COHEN states that it belongs to the rarest teratological findings among the teeth of the premaxilla. In view of this statement it becomes particularly striking that this crown type has been found 7 times, amounting to about 10 % of the here examined temporary laterals on the cleft side in groups A and B.

Group C.

The examination of patients with isolated cleft palate was carried out in the same manner as in the other groups. The largest clefts extended to the papilla incisiva, while the smaller included the velum and the uvula only.

All 15 cases had normally formed and developed deciduous, and permanent teeth. No supernumeraries were found. In one case the germs of the mandibular lateral incisors were missing the temporary as well as the permanent (No. 24). Besides this the following was found:

3 cases with completely cleft palate showed normal alignment and occlusion, No. 14, 15 and 36. The last mentioned had not been operated on.

3 cases, also with completely cleft palate, had narrow maxillary arches with crowded teeth in the front, No. 24, 26 and 37.

3 patients where the velum and part of the hard palate were cleft, had likewise crowded teeth in the premaxilla, No. 19, 22 and 23.

6 patients with more or less pronounced cleft in the soft palate showed:
1 normal conditions, No. 18,
1 malocclusion, Angle cl. III, No. 21,
1 » , » » II, div. 1, No. 20 and
3 crowded front teeth, No. 16, 17 and 26.

Summary.

In 63 harelip and cleft palate patients, from 3 to 7 years of age, the lateral incisors in the cleft area have been examined with regard to their number as well as to their form. In each case data are at hand for both the temporary and the permanent dentition.

Group A, patients with prealveolar harelip, includes 15 cases with 16 cleft sides in all. About 3/4 of them showed "overproduction", i. e. supernumerary and/or hyperplasia, of the deciduous lateral. In the permanent dentition nearly one half showed overproduction, but here "underproduction", i. e. absence or hypoplasia, was equally numerous.

Group B, patients with harelip as well as alveolar cleft, comprises 33 cases with 40 cleft sides altogether. Overproduction in the temporary lateral was less frequent than in group A. In about 1/4 of the cases even underproduction is seen. In the permanent dentition a few had overproduction, while about 3/4 showed underproduction. In both dentitions most of the laterals were found distal to the cleft. These were, however, more frequent hypoplastic than laterals medial to the cleft.

Hyperplastic deciduous teeth of a characteristic form, here called T-form, represented approximately 10 % of the laterals in groups A and B.

In group C, 15 patients with isolated cleft palate, the laterals showed no irregularities, neither in number nor in form.

Zusammenfassung.

In 63 Hasenscharte- und Gaumenspaltepatienten, im Alter von 3 bis 7 Jahren, wurden die lateralen Inzisiven im gespalteten Gebiete, ihrer Anzahl und Form nach, untersucht. In allen Fällen liegen Data for, sowohl für das temporäre als auch für das permanente Gebiss.

Gruppe A: Patienten nur mit gespalteter Lippe, umfasst 15 Fälle, in allem mit 16 gespalteten Seiten. Ungefähr 3/4 von diesen zeigten »Überproduktion«, d. h. Doppelanlagen und/oder Hyperplasien, im Bereiche des Milchlaterales. Im permanenten Gebisse zeigte annährend die Hälfte Überproduktion, hier aber war »Unterproduktion«, d. h. Agenesien oder Hypoplasien, beinahe ebenso häufig vorkommend.

Gruppe B: Patienten mit Lippe und Kiefer (eventuell auch Gaumen) gespaltet, umfasst 33 Kasus, in allem mit 40 gespalteten Seiten. Überproduktion im temporären Gebisse war weniger häufig als in Gruppe A. Etwa 1/4 von den Fällen zeigte sogar Unterproduktion. Im permanenten Gebisse hatten nur wenige Überproduktion, während ca. 3/4 Unterproduktion zeigten. Sowohl im temporären als auch im permanenten Gebisse wurde

ein lateraler Schneidezahn häufigst am distalen Rande der Spalte gefunden. Doch waren diese »distal-lateralen« Inzisiven öfters hypoplastisch als die »medial-lateralen« Inzisiven die sich in manchen Fällen am vordersten Rande der Spalte fanden.

Hyperplastische Milchlateralen mit einer karakteristischen Form, hier T-form genannt, kamen mit ca. 10 % in den Gruppen A und B vor.

In Gruppe C: 15 Patienten wo nur der Gaumen gespaltet war, zeigten die lateralen Schneidezähne keine Anomalien, weder in der Anzahl noch in der Form.

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