

Morphological Findings in Unoperated Cleft Lips and Palates

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A total of 353 children with cleft lips and palates were examined with regard to the morphology of the tissue surrounding the clefts in the lip, jaw, and palate. Photographs, radiograms (panoramic views of the maxilla), and study models were taken prior to operation. Results indicate a close relationship between the *degree and condition of the cleft* and the *extension of the vermillion*, the number of *tooth buds* adjacent to the cleft, the form and size of the alveolar borders, and the number and type of incisors.

In isolated cleft palates, the cleft edges are always symmetrical and there is a gradual transition from broad-arched, total clefts of the hard palate to the bifid uvula having no impact on articulation.

Introduction

In addition to genetic considerations, anthropometric studies are becoming more and more important in the study of developmental disturbances of the lips, jaws, and face. Better morphological understanding may result in more favorable therapeutic results.

Various types of cleft lips and palates, the most frequent malformations in this region, are operated upon in the first year of life in most parts of the world, and almost all surgeons start with labioplasty. However, the sequence of operations on the alveolus and the hard and soft palates varies. These differences are due not only to the individual preferences of surgeons and to the varieties of cleft morphology. They result also from quantitative and qualitative differences in the tissue surrounding the clefts, the anatomical and topographical characteristics of which are difficult to identify and document prior to operation. Despite the great importance of preoperative data, publications on morphological problems have been somewhat rare and have often reported findings on very small populations (Bohn, 1964; Fahrenberg, 1967; Mayer-Bohmke, 1965; Pfeifer, 1963, 1966a; Schwalbe, 1909; Teuchert, 1966; Trost, 1970). These limitations are serious when one recognizes the variability that is present.

This great variety of congenital facial malformations is easy to understand if one considers not only the morphological description but also development on the basis of normal morphogenesis.

In typical malformations of the face, the borders of developmental areas are

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determined by early embryonal principles and do not coincide with the anatomic description of the adult face. (The main border line extends over the ala nasi and along the boundaries of the premaxilla). Disturbances in this area result in the different types of clefts of the lip, alveolus, and palate (Pfeifer, 1974).

In patients with cleft lips and palates, differences in the soft and hard tissues of the nose, lip, maxilla, teeth, and the hard and soft palates are so significant that, in a classification of cleft lip and palate, morphogenetic considerations must take precedence over anatomical considerations.

This kind of grouping is included in a classification that was agreed upon at the 4th International Congress on Plastic Surgery in Rome in 1967 and was accepted by the representatives of more than 50 countries. (Report of Subcommittee on Nomenclature, 1969).

However, this international classification refers only to cleft characteristics. When considering morphological differences in the tissues surrounding the cleft, it is important to establish teratological orders. These are based on embryonal characteristics ranging from microforms to extreme forms of clefts. There are three different teratological categories: cleft of the lip and primary palate; cleft of the lip, alveolus, and palate; and isolated cleft palate. All of them reveal a gradation in characteristics which follow a certain pattern.

The purpose of this work was to study pathological findings in the region of the cleft alveolus. It was thought that these developmental abnormalities might manifest themselves in the form of a teratological order according to E. Schwalbe (1909).

This investigation was performed as a continuation of studies by Pfeifer (1963, 1966a), Teuchert (1966), and Trost (1970).

Material and methods

353 children, in-patients of Nordwestdeutsche Kieferklinik, University of Hamburg, West Germany, were examined in the years 1968 to 1970. The 253 patients with cleft lips (with or without cleft palates) were, on the average, six months old. The 100 patients with isolated cleft palates had a mean age of 4½ years (see Table 1).

Patients with bilateral cleft lips and palates include those with bilateral clefts of the hard palate even if the lip on one side did not show signs of clefting. Out of the 353 patients, 101 (29%) had clefts of the lip and alveolus; 152 (43%) had clefts of the lip, alveolus, and palate; and 100 (28%) had isolated cleft palates. These percentages do not reflect the absolute frequency of cleft types in Hamburg for the years encompassed by the investigation. Not all available subjects were studied, so subject numbers differ from one evaluation to another.

The morphological study was performed by evaluating:

1. maxillary panoramic radiograms
2. extra- and intraoral photographs
3. study models of the upper jaw

The radiograms, photographs, and study models were all taken prior to the first operation.

TABLE 1. Classification of 353 patients with cleft lips and palates according to three morphogenetic classes (patients with a bilateral cleft of the hard and/or soft palate in combination with a unilateral cleft lip were entered in this table as "bilateral".)

main class	location of cleft	number of patients				age (arithmetic mean)
		uni-lateral	bi-lateral	median	total	
I	lip, alveolus	89	12	—	101 (29%)	6 months
II	lip, alveolus, palate	100	52	—	152 (43%)	6 months
III	palate	—	—	100	100 (28%)	4 ½ years
Total		189	64	100	353	

Results

MORPHOLOGICAL CHARACTERISTICS OF CLEFT LIPS. The following was found regarding extension of the vermilion in 312 clefts studied in 253 patients. (A bilateral cleft in one patient was counted as two clefts.)

1. In 85 per cent of the 194 unilateral cleft lips, the vermilion reached to the cleft. In only 15 per cent of the cases did the vermilion fall short of the cleft. In 127 patients, the cleft was on the left side, while in 67 cases, it was on the right.
2. For 118 bilateral cleft lips (59 patients studied), the findings were very similar. As in the unilaterals, a direct relationship seemed to exist between the extent of the cleft and the extent of the vermilion. In narrow, partial clefts, the vermilion usually ended *before* the end of the cleft, while in broad, total clefts it continued to the end of the cleft, i.e. to the orifice of the nose.

MORPHOLOGICAL CHARACTERISTICS OF THE TISSUE SURROUNDING ALVEOLAR CLEFTS. In the study of the morphological characteristics of the tissue surrounding the alveolar clefts, we distinguished among (1) variations in the number of teeth, (2) positional anomalies, (3) anomalies in the shape of tooth buds adjacent to the cleft.

Variations in the Number of Teeth. According to the radiograms, the number of teeth varied only with regard to the lateral deciduous incisors. We distinguished between missing, single, and double anlagen (see Figures 1, 2, and 3).

Disregarding the cleft palate and grouping the patients on the basis of intact, partially cleft, and totally cleft alveolar ridges, it became obvious that the incidence of missing anlagen was insignificant in cases with intact ridges (only 1%). In cases with partially cleft ridges, there was a seven-per-cent incidence of missing anlagen while, in partial cleft lips and alveolus, double anlagen were observed in 58 per cent of the cases. A single anlage was found in 35 per cent of the cases. With regard to the canines, we never found the anlagen to be duplicated or missing.

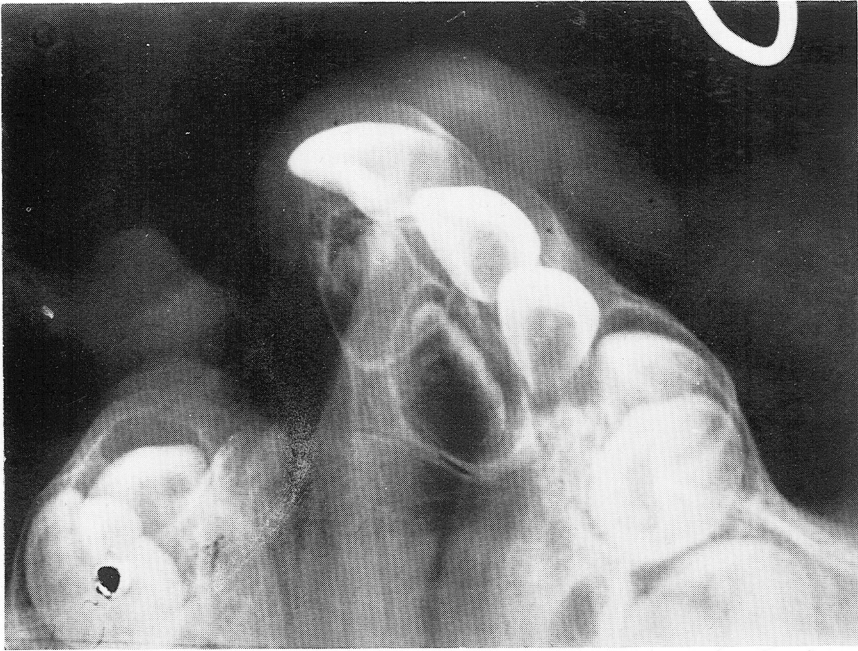


FIGURE 1. Missing anlage of the lateral deciduous incisor (bud).



FIGURE 2. Isolated anlage of the lateral deciduous incisor (bud).



FIGURE 3. Double anlage of the lateral deciduous incisor (bud).

Thus, it became clear that there is a developmental connection between the cleft width and the number of teeth anlagen in the tissue surrounding the cleft. In clefts up to 5 mm wide, double anlagen prevail. Although anlagen are found to be missing in cleft alveoli, 4–6 mm wide, their incidence is especially frequent in clefts measuring 11 mm or more in width (see Table 2).

In summary of this portion of the paper, it may be said that the number of double anlagen decreases with increasing width of the alveolar cleft, while the frequency of missing anlagen increases.

The striking frequency of double anlagen in narrow clefts and the increasing frequency of missing anlagen associated with wide clefts was also observed when approaching the problem from another point of view. In computing the widths of cleft alveoli in cases of double, single, and missing anlagen, it was found that the average cleft width for cases with double anlagen was 7.5 mm; for cases with single anlagen, 10.6 mm; and for cases with missing anlagen, 14.1 mm.

Positional Anomalies. In unilateral clefts, it was found that the wider the cleft, the more severe were the anomalies in the position of the anterior tooth buds, e.g. tilted, rotated, and/or dislocated.

A tooth was described as *rotated* if it was turned around its longitudinal axis in mesial or distal direction but was normally positioned in the dental arch. It was *tilted* when it was in its proper position within the dental arch but was inclined in a palatal, labial, mesial, or distal direction around its horizontal axis. By tilting it in the opposite direction, it could be brought back into its normal position. A tooth was *dislocated* if it was positioned outside the dental arch. All three positional anomalies may occur singly or in combination (Mayer-Bohmke, 1965). (See Table 3).

With an intact alveolar ridge, central deciduous incisor buds showed positional anomalies in 42 per cent of the subjects studied. In partial clefts of the alveolus, 13 out of 19 (68%) exhibited positional anomalies while, in complete clefts, 104 out of 118 (88%) were affected. Seventy-three per cent (140) of the study sample of 191 demonstrated these 248 positional anomalies. Of these 248 findings, 134 (54%) were tilted; 112 (45%) were rotated; and two (1%) were dislocated.

The teeth were most frequently tilted (134 out of 140 affected teeth). Buds tilted in a distal direction were found most often. Rotated tooth buds were also frequently observed (112 buds out of 140), while dislocations were rarely observed (2 tooth buds).

Positional anomalies in the lateral deciduous incisor buds next to the canines were observed in 87 per cent of the subjects studied. Tilted buds were again most frequent and accounted for 45 per cent of the positional anomalies. They were followed by rotated tooth buds (38%) and dislocated buds (17%). (See Table 4.) Positional anomalies in the lateral deciduous incisor buds located in the premaxilla were not studied.

Size and Shape of Tooth Buds. Size and shape of the tooth buds are only to a small extent related to the width of the cleft alveolus. In cases of double anlagen

TABLE 2. Absolute and relative frequency of double, single, and missing anlagen in relation to the width of the cleft alveolus.

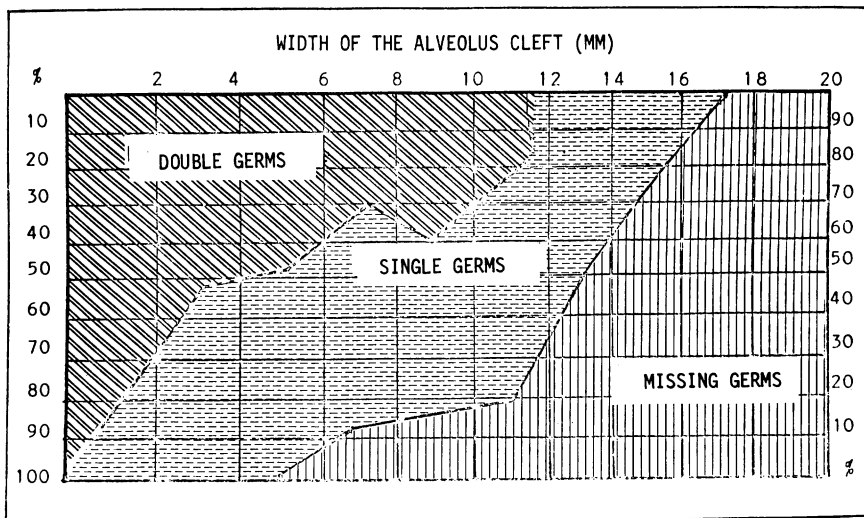


TABLE 3. Frequency of positional anomalies (tilted, rotated, dislocated) of deciduous central incisor buds on cleft side in unilateral cases (two patients with a cleft soft palate in combination with a unilateral cleft lip were not entered in this table. They were considered "bilateral".)

<i>alveolar ridge</i>	<i>patients</i>			<i>positional anomalies found</i>			
	<i>total</i>	<i>positional anomalies</i>		<i>total</i>	<i>tilted</i>	<i>rotated</i>	<i>dislocated</i>
		<i>without</i>	<i>with</i>				
No cleft	54	31 (58%)	23 (42%)	32	22	10	—
Partial cleft	19	6 (32%)	13 (68%)	19	11	7	1
Total cleft	118	14 (12%)	104 (88%)	197	101	95	1
Total	191 (100%)	51 (27%)	140 (73%)	248 (100%)	134 (54%)	112 (45%)	2 (1%)

TABLE 4. Frequency of positional anomalies (tilted, rotated, dislocated) of deciduous lateral incisor buds next to canines in unilateral cleft alveoli.

<i>alveolar ridge</i>	<i>patients</i>			<i>positional anomalies found</i>			
	<i>total</i>	<i>positional anomalies</i>		<i>total</i>	<i>tilted</i>	<i>rotated</i>	<i>dislocated</i>
		<i>without</i>	<i>with</i>				
No cleft	52	17 (33%)	35 (67%)	43	23	12	8
Partial cleft	7	1 (14%)	6 (86%)	12	5	5	2
Total cleft	76	—	76 (100%)	169	73	67	29
Total	135 (100%)	18 (13%)	117 (87%)	224 (100%)	101 (45%)	84 (38%)	39 (17%)

of lateral deciduous incisor buds, the medial anlage is generally larger than the lateral. In 51 per cent of the cases, single anlagen of lateral incisors on the cleft side are as large as the contralateral buds. In 33 per cent of the cases, they are larger than the contralateral anlagen.

FINDINGS IN THE PALATAL REGION. In the palatal region, we distinguished among morphological characteristics of clefts involving the lip, alveolus, and palate and of isolated clefts of the palate.

1. Patients with clefts of the lip, palate, and alveolus always exhibited a considerable number of anomalies in their anterior teeth. Anomalies in position and shape of the lateral deciduous incisor buds were almost the

- rule. The posterior width of the palatal cleft (distance between the maxillary tuberosities) was, in the majority of cases, greater than the anterior width (line connecting the anterior parts of the palatal shelves). In more than half of the cases (60%), the anterior width of the palatal cleft was greater than the width of the alveolar cleft. The nasal septum was always fully developed. However, in clefts of the hard palate, the septum did not reach the height of the palatal vault. In bilateral clefts, the angle between the edges of the palatal shelves was smaller than in isolated cleft palates, i.e. the anterior width was almost as great as the posterior width.
2. In isolated cleft palates, anomalies of anterior teeth were rare. Positional anomalies were found considerably less often than in combined clefts. The edges of the clefts were almost always symmetrical. In clefts including the hard palate (47 of 100 cases), the nasal septum was never fully developed.

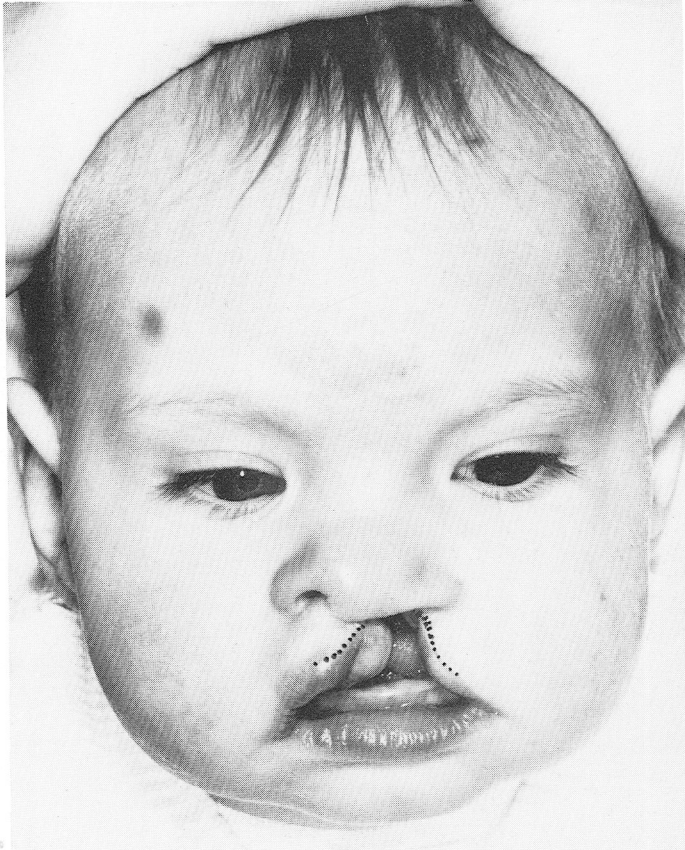


FIGURE 4. Total left cleft of lip and palate: primary cleft; vermilion (dotted) extending into the orifice of the nose (cleft end).

Discussion

In a continuation of morphological studies of clefts of the lip, alveolus, and palate undertaken by Pfeifer (1963, 1966a), Teuchert (1966), and Trost (1970), we examined 353 patients using the same criteria. So far as we know, this is the largest sample of pre-operative patients studied to date. Relationships were found between (1) the width of the alveolar cleft and the extension of the vermillion, and (2) the width of the alveolar cleft and the number, form, and position of tooth bud anomalies close to the cleft.

The extension of the vermillion permits conclusions regarding the morphogenesis of labial clefts. In a *primary* cleft lip resulting from non-junction of the embryonal tissue, the vermillion reaches up to the orifice of the nose (end of the cleft) (Pfeifer, 1963, 1966a, 1966b). (See Figure 4.) If the vermillion terminates *before* the end of the cleft (*secondary* cleft), the cleft may have

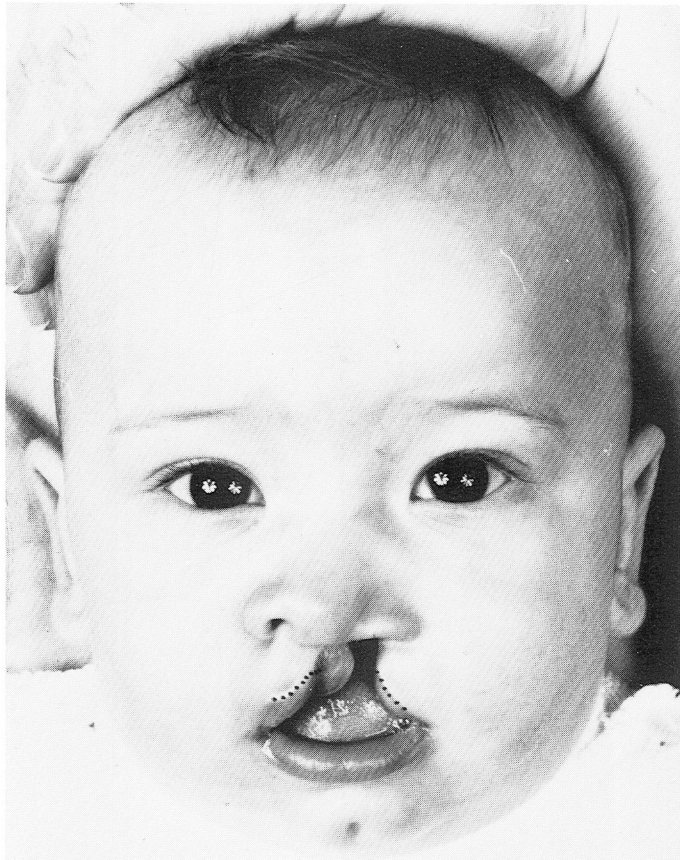


FIGURE 5. Left cleft of lip and alveolus: secondary cleft (vermillion dotted).

developed as the result of tearing of the mesenchyma (Pfeifer, 1963, 1966a, 1966b) (Figure 5). Our data confirm the results of previous investigations (Pfeifer 1963, 1966a; Teuchert, 1966; Trost, 1970) in that we also found that secondary cleft lips were seen more often with partially cleft alveolar ridges than in association with wide complete alveolar clefts. This finding "fits" modern theories, suggesting that secondary labial clefts manifesting themselves with a vermilion that does not reach to the end of the cleft itself are the result of tearing processes *after* primary physiological junction of the embryonal labial tissue.

Our data on the number of duplications of anlagen of lateral deciduous incisors adjacent to alveolar clefts are consistent with those of other authors (Bøhn, 1964; Cassardelli, 1952; Pfeifer, 1963, 1966a; Teuchert, 1966; Trost, 1970). They also found a decreasing number of tooth buds with an increasing alveolar cleft width. This finding is consistent with Pfeifer's (1974) statement to the effect that the less severe a malformation is, the more often hyperplasia and duplication of the anlagen will be found. Regarding anomalies in the shape of lateral deciduous incisors, our data are generally comparable to those of Bøhn (1964). However, there is one difference. He found even more frequently than we did that the lateral deciduous incisors on the cleft side are larger than those on the contralateral side.

Frequency and distribution of the various positional anomalies of the lateral deciduous incisors differ among the various authors (Pfeifer, 1963; Teuchert, 1966; Trost, 1970). Eckstein (1958) unfortunately failed to present data on the width of the alveolar clefts in the cases in which he studied the lateral incisors.

The results of this investigation on morphological findings in cheilognathopalatoschisis representing embryonal characteristics maintained after birth indicate that they follow the pattern of a teratological order and allow conclusions regarding the mechanism of cleft formation. Another result of this study is of far-reaching importance. After measuring the cleft width, predictions can be made as to what types of dental anomalies and orthodontic problems are likely to develop in the individual patient. Thus, knowing the cleft width is not only important for the maxillofacial surgeon (from a technical point of view) and for the speech pathologist, but for the embryologist and for the orthodontist in his planning of treatment.

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References

- BØHN, A., *Dental Anomalies in Harelip and Cleft Palate*, 109 pages, Med. Diss., Oslo, 1964.
 CASSARDELLI, H., *Lippen-Kiefer-Gaumenspalten und ihre Beziehungen zur Anzahl der Zähne und zum Zwischenkiefer*, 44 pages, Med. Diss., Berlin, 1952.
 ECKSTEIN, A., *Lageanomalien oberer Milchzahnkeime bei LKG-Spalten und ihre Bedeutung für die Kieferentwicklung und den Operationstermin*, *Fortschr. Kiefer-Gesichtschir.*, 4, 140-144, Thieme, Stuttgart, 1958.

- FAHRENBERG, H. W., Vergleichende Untersuchungen bei Schneidezahnanomalien der ersten und zweiten Dentition an Röntgenbildern von Kindern mit seitlichen Kieferspaltdformen, 63 pages, Med. Diss., Hamburg, 1967.
- MAYER-BOHMKE, H., Über Zahnkeimanomalien des Milchgebisses von Kindern mit unbehandelten Lippen-Kiefer-Gaumenspaltdformen, 65 pages, Med. Diss., Hamburg, 1965.
- PFEIFER, G., Über Entstehung und Erkennung regionaler Entwicklungs- und Wachstumsstörungen bei Lippen-Kiefer-Gaumenspalten als Grundlagen der Therapie, 260 pages, Med. Habilschrift, Hamburg, 1963.
- PFEIFER, G., Morphology of the Formation of Clefts as a Basis for Treatment, In K. Schuchardt (Ed.), Treatment of Patients with Clefts of Lip, Alveolus and Palate, (2nd Hamburg International Symposium, July 6-8, 1964), p. 14-24, Thieme, Stuttgart, 1966a.
- PFEIFER, G., Die Entwicklungsgeschichte der Lippen-Kiefer-Gaumenspalten als Leitspur für die Behandlung. *M Schr. Kinderheilk.*, 114, 244-249, 1966b.
- PFEIFER, G., Systematik und Morphologie der kraniofazialen Anomalien. *Fortschr. Kiefer-Gesichtschir.*, 18, 1-14, Thieme, Stuttgart, 1974.
- Report of the Subcommittee on Nomenclature and Classification of Clefts of Lip, Alveolus, and Palate and Proposals for Further Activities. In Sanvenero-Rosselli, G. (Ed.), Transactions of the Fourth International Congress of Plastic and Reconstructive Surgery in Rome, Oct. 1967, Excerpta Medica Foundation, Amsterdam, 1969.
- SCHWALBE, E., Die Morphologie der Mißbildungen des Menschen und der Tiere, Vol. III, 1, 113-204, Die Mißbildungen des Kopfes. Fischer, Jena, 1909.
- TEUCHERT, W., Präoperative morphologische Untersuchungen der Umgebung von Lippen-Kiefer-Gaumenspalten, 32 pages, Med. Diss., Hamburg, 1966.
- TROST, R., Morphologische Merkmale bei unoperierten Lippen-Kiefer-Gaumenspaltdformen, 60 pages, Med. Diss., Hamburg, 1970.