# Comparison of Facial Morphology in Two Populations With Complete Unilateral Cleft Lip and Palate From Two Different Centers

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***Objective:*** **To identify differences in craniofacial morphology of two populations with a history of complete unilateral cleft lip and palate (UCLP) treated under different protocols.**

***Design:*** **Retrospective longitudinal cohort study.**

***Setting:*** **Cleft Center of the University of Nijmegen, The Netherlands, and the Cleft Lip and Palate Program, The Hospital for Sick Children, Toronto, Canada.**

***Subjects:*** **Nineteen patients (16 male, 3 female) from Nijmegen and 19 patients (16 male, 3 female) from Toronto. Each patient was matched for sex and age with a patient from the other group. The mean ages at which lateral cephalometric radiographs were available for the Nijmegen group were 5.5, 9.9, and 18.3 years, while for the Toronto group these were available at mean ages of 5.3, 10.1, and 18.3 years, respectively. Eighteen patients from the Nijmegen group received an alveolar bone graft at a mean age of 9.5 years (range 8.2 to 13.5 years). None of the patients from Toronto received bone grafts.**

***Main Outcome Measures:*** **Eighteen cephalometric variables per radiograph were calculated at each time registration, using Dentofacial Planner cephalometric software. Statistical evaluation was performed with repeated-measures analysis of variance.**

***Results:*** **No differences were seen in the maxillary measurements. The patients in the Toronto group had significantly larger mandibles at all three time registrations.**

***Conclusions:*** **The Nijmegen and Toronto protocols resulted in similar maxillary projections in patients with UCLP. Comparison of data from other studies supports the contention that the larger profile convexity of the Nijmegen group is a reflection of a genetically determined smaller mandibular size in the Dutch population.**

KEY WORDS *cephalometry, craniofacial morphology, facial growth, orthodontics*

Patients with a history of unilateral cleft lip and palate (UCLP) commonly exhibit a degree of maxillary hypoplasia as the result of the growth-restraining effect of scar tissue from the lip and palate correction at infancy (Ross, 1987a; Kuijpers-Jagtman and Long, 2000). Consequently, these patients typically present with a Class III malocclusion, often with an anterior crossbite that may be exacerbated during the pubertal growth spurt through the maxillomandibular growth differential. The correction of this discrepancy, if severe, necessitates further surgical intervention at skeletal maturity in the form of orthognathic advancement of the maxilla, either by means of a conventional Le Fort I osteotomy (Vig and Turvey, 1985; Butow, 1986; Posnick and Tompson, 1992) or more recently through maxillary distraction osteogenesis (Hierl et al., 2003; Figueroa and Polley, 1999; Ko et al., 2000).

Several different surgical treatment protocols have been advocated for the management of children with UCLP. Various institutions around the world generally continue to operate under their traditionally established protocol, with only occasional exceptions. The Eurocleft Study showed that among the 201 European centers that are listed in the final report, 194 different treatment protocols existed for unilateral clefts (Shaw et

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Presented at the 59th Annual Meeting of the American Cleft Palate/Craniofacial Association, Seattle, Washington, May 2, 2002; and at the Seventh Panhellenic Orthodontic Conference, Athens, Greece, September 29, 2002.

Submitted May 2005; Accepted August 2005.

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**TABLE 1 Sample description**

| | *Toronto* | *Nijmegen* |
|---|---|---|
| Sample size | N = 19 (16 m, 3 f) | N = 19 (16 m, 3 f) |
| Alveolar bone graft (ABG) | None had ABG | 18 had ABG at a mean age of 9.5 yrs (range: 8.2 – 13.5 yrs) |
| Lateral cephalometric radiographs available at mean ages: | T1 = 5.3 yrs (5.0 – 6.9 yrs)<br>T2 = 10.1 yrs (8.1 – 13.0 yrs)<br>T3 = 18.3 yrs (16.0 – 20.9 yrs) | T1 = 5.5 yrs (4.0 – 6.5 yrs)<br>T2 = 9.9 yrs (8.2 – 12.3 yrs)<br>T3 = 18.3 yrs (15.4 – 22.9 yrs) |

al., 2000, 2001). Evaluation and comparison of treatment outcomes with all these different protocols is of vital importance to the field and is only possible with intercenter collaborative studies (Ross, 1987a, 1987b; Friede et al., 1991; Shaw et al., 2005).

The impetus for the present study came from the observation that a number of patients with a history of UCLP from the Cleft Center at the University of Nijmegen, The Netherlands, presented in their early teens with a Class II malocclusion, the orthodontic correction of which often necessitated the assistance of extraoral traction. This was in contrast to the classic midface-deficient image of UCLP patients reported in the literature (Ross, 1987a; Friede et al., 1991; Swennen et al., 2002). A comparison study was thus undertaken to evaluate any differences in craniofacial appearance between UCLP patients treated under the Nijmegen and Toronto protocols.

## Subjects and Methods

The records of the Cleft Lip and Palate Center at the University of Nijmegen, The Netherlands, were searched for Caucasian individuals with a history of complete UCLP (CUCLP), with lateral cephalometric radiographs available roughly at 5, 10, and 18 years of age. Patients with associated anomalies or syndromes were excluded. Nineteen patients (16 male, 3 female) born between 1967 and 1991 fulfilled the above criteria. Eighteen of these patients received an alveolar bone graft at a mean age of 9.5 years (range: 8.2 to 13.5 years).

The Nijmegen sample was matched for age and sex with a sample of 19 patients with CUCLP from the Cleft Lip and Palate Program, The Hospital for Sick Children, Toronto, Canada, selected using the same criteria. These patients were born between 1954 and 1967. None of the patients from the Toronto sample received an alveolar bone graft during the period examined by the study. The majority of patients in both groups received maxillary infant orthopedic treatment (IO) and orthodontic treatment in the mixed and/or permanent dentition, as required. However, none of the patients in either group underwent facemask treatment or orthognathic surgery during the observation period. The demographic characteristics of the two samples can be seen on Table 1.

The main differences in the protocols followed in the two institutions were:

1. Maxillary IO in the Nijmegen group was carried out until the time of soft palate repair (at 12 to 18 months of age) to ensure that the maxillary segments would not collapse following repair of the lip. The method of IO used was a modification of the Hotz method, with an intraoral plate and no nasal molding. In the Toronto group, IO was discontinued after lip repair (3 to 4 months of age). The appliance used was a passive or active intraoral plate with extraoral wires for retention and no nasal molding.
2. The Millard technique was used for lip repair in both institutions, at 3 to 4 months of age in Toronto and at 16 to 18 weeks of age in Nijmegen. The surgical repair of the hard and soft palate in the Toronto group was performed in one stage through a modified von Langenbeck approach. In contrast, in the Nijmegen group, palate repair was done in two stages: the soft palate was repaired at 12 months of age through a modified von Langenbeck procedure, while the hard palate closure was delayed until patients were approximately 9 years of age.
3. In all but one subject in the Nijmegen group, the alveolar cleft was bone-grafted using a corticocancellous block from the mandibular symphysis (Freihofer et al., 1993). None of the subjects in the Toronto group received an alveolar bone graft, because mixed-dentition bone grafting was not part of the general protocol for CLP patients at The Hospital for Sick Children at that time.

The radiographs from each sample were traced and digitized by an experienced operator. A series of 18 cephalometric measurements was performed on each radiograph (see Fig. 1 and Tables 2 through 4) at each time registration (T1, T2, and T3) using the Dentofacial Planner cephalometric software (http://www.dentofacial.com/). Statistical evaluation was performed with repeated-measures analysis of variance, checking for group effect, time effect, and time-group interaction.

## Results

At T1, the only statistically significant difference was in the angle SNB, which was larger in the Toronto sample by an average of 2.6° ($p = .0204$).

At T2, the angle SNB continued to be statistically significantly larger in the Toronto group by an average of 2.8° ($p = .0126$). This was corroborated by the mandibular length measurement, which was longer by 4 mm in the Toronto sample ($p = .0432$).

By T3, the average SNB angles in the two groups differed by 4.7° ($p < .0001$), and the mandibular length of the Toronto

**FIGURE 1** Reference landmarks on the lateral cephalometric tracing. ***Hard tissue landmarks:*** A-point = the deepest point on the anterior contour of the maxillary alveolar process; ANS = anterior nasal spine, the tip of the osseous anterior nasal spine; B-point = the deepest point on the anterior contour of the mandibular alveolar process; Ba = basion, the most anteroinferior point on the margin of the foramen magnum; Co = condylion, the most posterosuperior point on the head of the mandibular condyle; Gn = gnathion, the most anteroinferior point on the outline of the chin; Go = gonion, the most posteroinferior point on the angle of the mandible; LIA = lower incisor apex, the apex of the most labially placed mandibular central incisor; LIT = lower incisor tip, the incisal tip of the most labially placed mandibular central incisor; Me = menton, the most inferior point on the outline of the chin; N = nasion, the most anterior point of the frontonasal suture; Pg = pogonion, the most anterior point on the outline of the chin; S = sella, the geometric center of sella turcica; UIA = upper incisor apex, the apex of the most labially placed maxillary central incisor; UIT = upper incisor tip, the incisal tip of the most labially placed maxillary central incisor. ***Soft tissue landmarks:*** CT = columella tangent, the point of intersection with the nasal outline of a tangent to the columella from Sn; G′ = soft tissue glabella, the most prominent point on the soft tissue drape of the forehead; LS = labrale superius, the most prominent point on the vermilion border of the upper lip; Pg′ = soft tissue pogonion, the most anterior point on the soft tissue outline of the chin; Sn = subnasale, the point where the base of the columella of the nose meets the upper lip.

**TABLE 2 Means and standard deviations (SD) for cephalometric values of the two samples at T1 (NS = not significant; U1 = UIA-UIT; L1 = LIA-LIT).**

| | *Toronto (SD)* | *Nijmegen (SD)* | *Diff.* | *p* |
|---|---|---|---|---|
| SNA (°) | 81.0 (3.9) | 79.4 (2.8) | 1.6 | NS |
| SNB (°) | 75.1 (2.0) | 72.5 (2.9) | 2.6 | **0.0204** |
| ANB (°) | 5.9 (3.8) | 7.0 (2.4) | 1.0 | NS |
| Ba-N-ANS (°) | 67.2 (4.3) | 65.7 (2.4) | 1.6 | NS |
| Ba-N-Pg (°) | 56.7 (1.9) | 54.9 (2.4) | 1.8 | NS |
| ANS-N-Pg (°) | 10.5 (4.3) | 10.8 (2.9) | 0.2 | NS |
| Ba-N (mm) | 96.3 (3.7) | 97.0 (4.2) | 0.7 | NS |
| Ba-ANS (mm) | 88.9 (4.6) | 88.4 (3.7) | 0.5 | NS |
| Md length (Co-Gn) (mm) | 94.0 (4.6) | 91.8 (4.4) | 2.2 | NS |
| SN-MP (SN-GoGn) (°) | 35.8 (3.9) | 37.6 (4.5) | 1.8 | NS |
| Md axis angle (BaN-CoGn) (°) | 74.1 (3.1) | 76.2 (4.1) | 2.1 | NS |
| AUFH (N-ANS) (mm) | 41.5 (2.0) | 41.6 (3.0) | 0.1 | NS |
| ALFH (ANS-Me) (mm) | 60.5 (4.3) | 60.1 (4.1) | 0.4 | NS |
| ANS-Me/N-ME (%) | 62.1 (2.2) | 61.1 (2.3) | 1.0 | NS |
| U1-SN (°) | 75.8 (6.2) | 71.1 (8.1) | 4.7 | NS |
| L1-MP (L1-GoGn) (°) | 84.4 (5.7) | 84.7 (6.1) | 0.3 | NS |
| Soft tissue convexity (G′Sn-G′Pg′) (°) | 10.9 (5.6) | 12.7 (5.8) | 1.9 | NS |
| Nasolabial angle (CT-Sn-LS) (°) | 120.1 (11.1) | 117.9 (13.5) | 2.2 | NS |

**TABLE 3 Means and standard deviations (SD) for cephalometric values of the two samples at T2 (NS = not significant; U1 = UIA-UIT; L1 = LIA-LIT).**

| | *Toronto (SD)* | *Nijmegen (SD)* | *Diff.* | *p* |
|---|---|---|---|---|
| SNA (°) | 79.1 (3.7) | 77.2 (2.7) | −1.8 | NS |
| SNB (°) | 75.4 (2.1) | 72.6 (2.7) | 2.8 | **0.0126** |
| ANB (°) | 3.7 (3.4) | 4.6 (2.6) | 0.9 | NS |
| Ba-N-ANS (°) | 65.5 (3.9) | 64.4 (3.2) | 1.1 | NS |
| Ba-N-Pg (°) | 57.2 (2.6) | 55.2 (2.3) | 2.0 | NS |
| ANS-N-Pg (°) | 8.2 (4.1) | 9.2 (3.5) | 1.0 | NS |
| Ba-N (mm) | 104.1 (4.3) | 103.7 (3.8) | −0.4 | NS |
| Ba-ANS (mm) | 94.9 (5.5) | 93.6 (3.8) | −1.3 | NS |
| Md length (Co-Gn) (mm) | 106.3 (5.4) | 102.3 (4.4) | 4.0 | **0.0432** |
| SN-MP (SN-GoGn) (°) | 35.0 (4.8) | 37.1 (3.8) | 2.1 | NS |
| Md axis angle (BaN-CoGn) (°) | 75.0 (4.3) | 76.7 (3.5) | 1.7 | NS |
| AUFH (N-ANS) (mm) | 48.3 (3.3) | 47.1 (3.1) | 1.2 | NS |
| ALFH (ANS-Me) (mm) | 65.2 (5.5) | 65.1 (4.1) | 0.1 | NS |
| ANS-Me/N-ME (%) | 60.2 (2.0) | 60.0 (2.0) | 0.2 | NS |
| U1-SN (°) | 87.4 (7.5) | 89.5 (7.4) | 2.1 | NS |
| L1-MP (L1-GoGn) (°) | 88.0 (5.6) | 85.8 (8.0) | 2.2 | NS |
| Soft tissue convexity (G′Sn-G′Pg′) (°) | 9.8 (6.9) | 10.7 (5.8) | 0.9 | NS |
| Nasolabial angle (CT-Sn-LS) (°) | 114.7 (13.6) | 115.8 (11.2) | 1.1 | NS |

sample was longer by 7.9 mm ($p$ = .0002). The SNA angle was also significantly larger in the Toronto sample by 4.4° ($p$ = .0002), resulting in no significant difference in the average ANB angle. By this time point (T3), the difference in angle Ba-N-Pog also reached statistical significance, being larger in the Toronto group by 3.7° ($p$ = .0012).

The results are presented in Tables 2 through 4, and cephalometric superimpositions of the two groups are shown on Figures 2 through 4.

**TABLE 4 Means and standard deviations (SD) for cephalometric values of the two samples at T3 (NS = not significant; U1 = UIA-UIT; L1 = LIA-LIT).**

| | *Toronto (SD)* | *Nijmegen (SD)* | *Diff.* | *p* |
|---|---|---|---|---|
| SNA (°) | 77.7 (3.2) | 73.3 (2.9) | 4.4 | **0.0002** |
| SNB (°) | 78.2 (3.8) | 73.5 (4.4) | 4.7 | **< 0.0001** |
| ANB (°) | 0.5 (3.7) | 0.2 (3.7) | 0.3 | NS |
| Ba-N-ANS (°) | 64.2 (4.0) | 62.7 (3.5) | 1.5 | NS |
| Ba-N-Pg (°) | 60.3 (3.9) | 56.5 (4.2) | 3.7 | **0.0012** |
| ANS-N-Pg (°) | 4.0 (5.2) | 6.2 (5.2) | 2.2 | NS |
| Ba-N (mm) | 114.2 (4.4) | 112.8 (4.2) | 1.4 | NS |
| Ba-ANS (mm) | 103.0 (5.8) | 100.2 (5.3) | 2.8 | NS |
| Md length (Co-Gn) (mm) | 127.2 (8.1) | 119.3 (6.4) | 7.9 | **0.0002** |
| SN-MP (SN-GoGn) (°) | 32.9 (5.5) | 35.7 (6.0) | 2.8 | NS |
| Md axis angle (BaN-CoGn) (°) | 75.6 (4.7) | 78.2 (5.6) | 2.6 | NS |
| AUFH (N-ANS) (mm) | 55.8 (4.1) | 53.8 (3.7) | 2.0 | NS |
| ALFH (ANS-Me) (mm) | 75.7 (6.1) | 75.6 (6.7) | 0.1 | NS |
| ANS-Me/N-ME (%) | 59.5 (2.1) | 60.2 (2.5) | 0.7 | NS |
| U1-SN (°) | 102.4 (7.6) | 99.2 (7.2) | 3.2 | NS |
| L1-MP (L1-GoGn) (°) | 83.4 (8.6) | 87.7 (9.0) | 4.3 | NS |
| Soft tissue convexity (G′Sn-G′Pg′) (°) | 2.5 (7.5) | 7.0 (8.5) | 4.5 | NS |
| Nasolabial angle (CT-Sn-LS) (°) | 103.8 (13.9) | 106.3 (12.4) | 2.5 | NS |

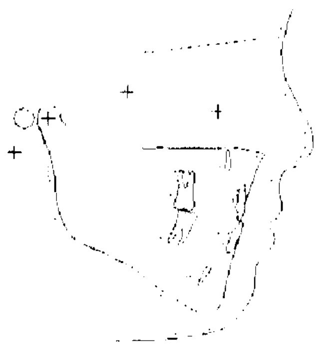

FIGURE 2 Cranial base (SN) superimposition of the average tracings at T1 (dotted = Nijmegen, solid = Toronto).

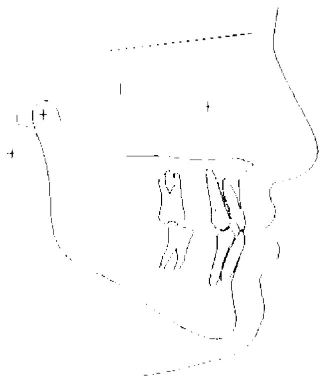

FIGURE 4 Cranial base (SN) superimposition of the average tracings at T3 (dotted = Nijmegen, solid = Toronto).

## Discussion

Cleft treatment protocols vary greatly among institutions and little evidence exists on comparison of the outcomes of each protocol. In a report issued recently, the World Health Organization (2002) recognized the absence of such data and emphasized the need for clinical studies to generate them. Intercenter comparison studies of craniofacial morphology are one of the few ways available to assess outcome of cleft treatment. Such studies, however, carry the risk of directly assuming that any morphologic differences found are a result of different techniques or appliances used. Their results need to be interpreted with caution, because the samples compared are dissimilar in more ways than one, and many sources could contribute to the differences observed.

This study presents a retrospective comparison of two populations of CUCLP, matched for age and sex, from two centers with fairly similar treatment protocols. One could hypothesize that the protocol differences in the two centers examined (e.g., duration of IO, history of alveolar bone grafting) might affect the maxillary projection or growth. Surprisingly, the maxillary projections of the two groups were similar up to the last time registration. By T3, SNA angle in the Toronto group was statistically significantly larger, although this was not corroborated by basal bone measurements (Ba-N-ANS and Ba-ANS). Since this effect did not become apparent until after the age of 10 years, it seems that IO was not implicated in this matter. It appears unlikely that the use of an intraoral passive plate for the first year of life would cause any sagittal maxillary growth inhibition. This point will hopefully be elucidated in the not too distant future by the results of the prospective clinical trial on presurgical IO that is currently under way in the Netherlands (Prahl et al., 2001). A recent publication of the dental cast analysis of the above patient sample at 6 years of age showed no significant effect of IO on sagittal or transverse dental arch dimensions (Bongaarts et al., 2004).

Because the sagittal effect on the maxilla appears to be limited to point A (which may be influenced by tooth position) rather than ANS (which is generally considered more representative of the basal bone of the maxilla), one could attribute the maxillary difference at T3 to the alveolar bone graft in the Nijmegen sample. However, the theory that the additional scar tissue from alveolar bone grafting may influence maxillary growth (Ross, 1987a, 1987b) has not been substantiated in the literature when bone grafting is performed in the mixed dentition or later (Daskalogiannakis and Ross, 1997; Semb, 1988). Moreover, the importance of mixed-dentition bone grafting in the alveolar cleft (Boyne and Sands, 1972) is recognized and appreciated. Even if unequivocal proof existed of a growth-retarding effect of mixed dentition bone grafting, it would certainly continue to be an essential part of most treatment protocols worldwide, as its short-term benefits in achieving a unified maxilla, eliminating any fistulae, and providing support

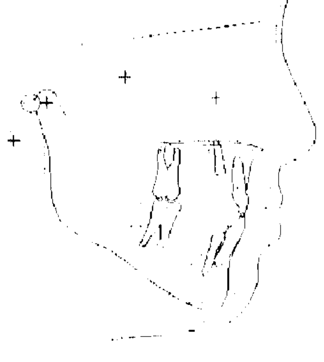

FIGURE 3 Cranial base (SN) superimposition of the average tracings at T2 (dotted = Nijmegen, solid = Toronto).

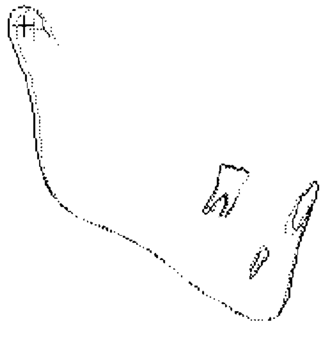

FIGURE 5 Average mandibular superimposition (on the symphysis and mandibular border) at T1 (dotted = Nijmegen, solid = Toronto).

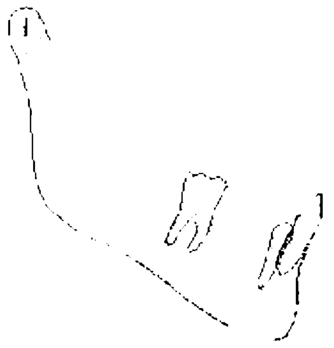

**FIGURE 6** Average mandibular superimposition (on the symphysis and mandibular border) at T2 (dotted = Nijmegen, solid = Toronto).

**FIGURE 7** Average mandibular superimposition (on the symphysis and mandibular border) at T3 (dotted = Nijmegen, solid = Toronto).

for the teeth would still far outweigh any such long-term side effects.

Surprisingly enough, the main differences found by this study were in the mandibular measurements. These differences are not likely a result of anything related to the clefts or their somewhat dissimilar treatments in the two centers. The subjects from Nijmegen had more retropositioned mandibles at T1 (at that time only reflected in angle SNB), but by T3 their mandibular length was shorter by 7.9 mm on average and their SNB angle was smaller by an average of 4.7° (Figs. 5 through 7). At first glance, one might attribute this difference to greater magnification in the Toronto cephalograms, especially when one considers that a 400-cm x-ray source-to-subject distance is used as a standard in the Nijmegen setup, in contrast to the 150-cm source-to-subject distance in Toronto. However, when taking into account the fact that the differences in other linear measurements (such as Ba-N and Ba-ANS) were statistically insignificant, one realizes that the effect of magnification in this instance is inconsequential compared to the highly significant differences in mandibular length, which were, indeed, corroborated by angular measurements. These differences in mandibular length seem to be responsible for the higher frequency of Class II malocclusions observed in the Nijmegen group in their early teenage years, compared to the patients from Toronto.

Trenouth et al. (1999) have cautioned that when comparing different populations, racial and genetic factors are likely to result in differences in the data that need to be taken into account. In their comparison of cephalometric reference values from five centers, the normative data from Nijmegen (Prahl-Andersen et al., 1979) exhibited the most pronounced Class II relationship of the five sample populations examined. It seems plausible, therefore, to assume that the greater Class II tendency in the Nijmegen compared to the Toronto sample was, to a great extent, a result of differences in the genetic make up of the general population, rather than protocol differences in the treatment of UCLP between these two centers. It appears that the maxillary inhibition from the repaired cleft in the Nijmegen sample, combined with the genetically controlled shorter mandible, resulted in a harmonious sagittal maxillomandibular relationship in this group.

## Conclusion

The protocols of the Nijmegen and Toronto centers resulted in very similar maxillary growth outcomes. The significantly smaller SNA angle in the Nijmegen group at T3 may be associated with a growth-inhibiting effect from the alveolar bone graft surgery, although this is only speculative. The Nijmegen group had significantly smaller mandibles at all three registrations, which seems to be an expression of a higher prevalence of mandibular retrognathism in the Dutch population.

*Acknowledgments.* The authors wish to thank Ms. Lynn Cornfoot for tracing and digitizing all radiographs, and Ms. Marlies Zaal for her kind help with data collection.

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