Twee jaar spraak en taal bij schisis
Jansonius-Schultheiss, K.

Citation for published version (APA):

General rights
It is not permitted to download or to forward/distribute the text or part of it without the consent of the author(s) and/or copyright holder(s), other than for strictly personal, individual use, unless the work is under an open content license (like Creative Commons).

Disclaimer/Complaints regulations
If you believe that digital publication of certain material infringes any of your rights or (privacy) interests, please let the Library know, stating your reasons. In case of a legitimate complaint, the Library will make the material inaccessible and/or remove it from the website. Please Ask the Library: http://uba.uva.nl/en/contact, or a letter to: Library of the University of Amsterdam, Secretariat, Singel 425, 1012 WP Amsterdam, The Netherlands. You will be contacted as soon as possible.
Children with a complete cleft (a cleft in lip, jaw, hard and soft palate) or with a cleft palate only run a high risk of developing speech problems. The nasal and oral passages cannot clearly be separated and this affects the articulation of both vowels and consonants. Vowels can be hypernasal and plosive consonants, which require a build-up of pressure in the mouth, are difficult to produce. Research indicates that cleft palate children are also at risk for language problems. The interesting question is what is the relationship between the development of speech on the one hand and the language system on the other in these children and what factors influence this development. One important factor might well be hearing loss; as a group children with a cleft palate are at higher risk for middle ear infection (Otitis Media with Effusion or OME). Due to the cleft palate the Eustachian tube is less open and the ventilation of the middle ear is reduced.

Cleft lip and palate is obvious from birth and the children receive different types of treatment from an early age. Some babies are given an oral plate from birth on. In some children tubes are inserted in order to drain the middle ear. All children receive surgery to close the palate (and lip and jaw) but this is done at different ages. There is no standard programme of treatment either nationally, within the Netherlands, or internationally. It is important to know when these different treatments can best be given for optimal results in speech and language development. This development is closely related to the communicative, social, emotional, cognitive and learning development. It is therefore very important to prevent speech and language problems already at an early age.

In this study the speech and language of cleft palate children at the age of two years will be examined and compared to normal development. The influence of hearing loss and medical intervention will also be evaluated.

Cleft palate can be the result of a genetic defect but this is not necessarily the case. It is relatively frequent, around two cases per thousand births (§ 1.1). In the Netherlands the children are treated from birth, usually by a cleft palate team including an orthodontic specialist, a plastic surgeon, an ENT specialist and a speech pathologist. Some of these teams choose to insert an oral plate from birth which covers the split in the jaw and hard palate (called the
Presurgical Orthopedic Therapy or PSOT). This treatment is applied for orthodontic purposes but from research it appears also to have beneficial effects on the production of speech sounds. The operation to close the soft palate (and sometimes at the same time the hard palate as well) is usually performed between the ages of six months and two years. Research indicates that an early operation, before seven months, is beneficial for the development of speech (§ 1.2). The psychosocial and cognitive development of a child can be relevant for speech and language but cleft palate children do not appear to be different in these aspects (§ 1.3). They do show a higher prevalence of middle ear infection, however. Although hearing loss as a result of OME has not been shown to lead automatically to speech and language problems, it is possible that in combination with another problem such as the deviant speech motor system due to the cleft palate that it does become important (§ 1.3). The anatomical problem does not greatly affect the production of vowels but the result is often hypernasality. Of the consonants the plosives /p/, /t/, /b/ and /d/ pose the greatest problems, the sonorants /m/, /n/, /v/, /w/, /j/ and /l/ the least. The labial plosives are the most difficult to articulate, but coronal and dorsal ones are also produced less (§ 1.4).

Chapters 2 and 3 look at both the early speech development before the first words appear (the prelexical stage) and the later language development. The first year of life is important for the development of speech perception. Hearing loss as a result of OME could be crucial for later speech and language production at this time (§ 2.1). In the first year the speech apparatus is in development (§ 2.2) and the speech production also develops quickly in this period. Speech sounds produced at this time can be considered as developmental patterns of speech movements and can be divided into movements of simple phonation, simple articulation, varied phonation, and babbling (analyzed according to Koopmans-van Beinum & Van der Stelt 1986). Varied phonation and babbling show the greatest differences in babies with cleft palate. Babbling occurs less and with less variation as compared with peers without a cleft (§ 2.3). The results of medical intervention in the first year are not immediately clearly observable; some results suggest a positive influence at a later age (§ 2.4).

As a group children with cleft palate appear to have problems with language even at school age (§ 3.1). Therefore, it is worthwhile to study speech as well as language. When the first words appear, the speech sounds are organized into a phonological system. Children with a cleft palate produce fewer sound segments correctly, particularly plosives. They are also different in phonological processes: they show more cluster reduction, more backing and more nasal assimilation (§ 3.2). Their vocabulary appears to be smaller (§ 3.3) and their
grammatical ability is less well developed, although relatively little is known about younger children (§ 3.4).

This study examines the production of speech and language in 30 cleft palate (CP) children at the age of two years. The influence of hearing loss and medical intervention is also studied (§ 4.1). The data are taken from a spontaneous language sample of mother-child interaction (§ 4.2). For some variables the children are compared with existing norm values; for other variables they are compared with a reference group (REF) of 9 children. Both groups of children had normal intelligence (§ 4.3.1.2). The children came from varying social-economic backgrounds (§ 4.3.2). Both groups were comparable in medical condition except for OME and hearing (§ 4.3.3). The evaluation of hearing loss at two years was done on the basis of field audiometry. The children were categorized in three groups of hearing loss: level 0: loss less than 20dB (CP: 2, REF: 7); level 1: loss betw. 20-40dB (CP: 19, REF: 2); level 2: loss betw. 40-55dB (CP: 9, REF: 0). The children were not selected on the basis of hearing loss and are therefore not equally distributed across hearing groups. The CP children were further categorized according to whether they had received a particular medical treatment (such as an oral plate, surgical closure of the palate or a drainage of the middle ear by tubes). When a treatment had been given, it could be considered to have been applied relatively early, in the first year of the children’s life, or late, thereafter. The study examines many different variables from speech production and language and also the relationship between scores on the variables (§ 4.4). Different types of analysis are used including an analysis procedure for morphology and syntax (GRAMAT, Bol & Kuiken 1988) and for phonology (FAN, Beers 1995). Because of the type of groups non-parametric statistics are used (§ 4.5).

Chapter 5 contains the results from general measures of speech and language, before phonology is studied in detail in Chapters 6 and 7. The CP children do not differ as a group from the REF children, neither in the amount of time they produced speech nor in the number of utterances produced, during twenty-minutes video-recording. The groups of mothers show also no differences. Communicatively the two groups are therefore comparable (§ 5.1). The CP children can be less well understood and, not surprisingly, have more characteristics symptomatic of CP speech such as nasality (§ 5.2). The number of spontaneous utterances and imitations the two groups produce do not differ significantly. Nor does the number of prelexical or lexical utterances differ (§ 5.3). It must be noted though that the CP group shows a great variation. The cleft palate children are lexically less far developed (§ 5.4) since as a group they produce significantly fewer content and function words (types and tokens). The analysis of grammatical development poses a methodological problem. The instrument GRAMAT requires at least 50
grammatically analyzable utterances, ideally 100. One third of the CP children cannot be analyzed since they produce less than 50 utterances. This indicates that a subgroup exists within the CP group with a problem in language development. Of the 20 children who can be analyzed, only 2 show up as delayed at this age (§ 5.5).

The analysis of the influence of hearing loss shows that it impairs intelligibility and increases CP speech as an additional factor alongside cleft palate. Hearing loss also negatively influences lexical ability and the length of the 5 longest utterances (MLUL) in the assessment of grammatical ability. Children with a hearing loss between 20-40dB and who had an early placement of drainage tubes score better than children with a loss between 40-55dB or with later or no drainage (§ 5.6). The results on all the variables show a great heterogeneity in the CP children as a group.

For many aspects of the analysis of phonology the FAN instrument was used (§6.1). Again one third of the CP group could not be analyzed since they produced too few words; 19 of the 30 CP children could. The comparison was made with the REF children with good hearing (N=7) (§6.2). As a group the 19 CP children are delayed; they achieve contrast level 2 as opposed to level 3, the level expected at the age of two years. The phonological contrast [fricative] is often absent; the contrasts [sonorant], [labial], [coronal] and [dorsal] are present. At level 2 fewer phonemes are produced than in normal children at this phonological level. Seven CP children have not acquired a single phoneme in final position which shows a large delay. Of the total of 19 CP children 10 have reached level 3 or higher but often with fewer phonemes; six children are in general delayed and three have an abnormal development. If the sound systems of the delayed and abnormal children are studied from the perspective of which sounds are almost acquired, then development can be seen to be moving for some children, but not all, in a normal direction (§ 6.4).

Hearing has an influence on the phonological system (§6.5). The children with a severe or moderate hearing loss and no early drainage had a greater chance of being excluded from the FAN analysis because they produced too few words. The CP children with a moderate loss and early drainage had the most normal phonological development. Hearing also influences the number of phonemes acquired. Hearing loss is clearly not the only factor which explains phonological delay, however. The two CP children with normal hearing both show a delayed acquisition.

Within the normal phonological acquisition the contrast grade which is reached correlates with the number of phonemes acquired. The CP score, representing the amount of deviances in articulation due to the cleft palate, although a speech measure, does not correlate with phonological variables. Phonological acquisition is related to some other language variables. A CP
child who has not acquired the contrasts [explosive] and/or [dorsal] and possesses only a limited number of phonemes, also has few types of function words, a shorter MLUL and speaks for a shorter amount of time. During acquisition a child applies different types of phonological processes in word production. Segments can, for example, be deleted or substituted. Some processes are frequent in the early acquisition and are seen as normal, others are unusual and are symptomatic of a disordered acquisition. The CP group (N=19) was compared with the REF group with normal hearing (N=7) for these analyses. The CP children show a very normal pattern in their processes, even those children with a delayed or abnormal phonology (§7.5). The same processes are applied on the whole. The difference lies in more frequent use of cluster reduction and final consonant deletion (§7.2), and unusual substitution processes such as backing, frication, and nasalization (§7.4). The CP children often choose to produce target words which do contain the sounds which they have acquired, such as nasals. Children in the REF group however use more plosives. The cleft palate definitely has an influence on phonological processes and selection, but hearing loss is also a factor (§7.6).

Chapter 8 considers the effects of medical intervention on selected speech and language variables (§8.1), in particular those which have been shown to be characteristic of CP children. The oral plate has the most influence on the phonological acquisition (§8.2). Children who were not treated with an oral plate were more likely to be in the group which had such poor language that they could not be analyzed. The children who were treated with the oral plate for a long time also had a better chance of having a normal contrast level and a higher number of phonemes acquired. The longer the plate had been worn, the more normal certain phonological processes were. More correct clusters were produced. The closure of the palate was also shown to have a positive effect on intelligibility and the CP speech score. Other effects were also seen in the phonological processes (§8.3). Middle ear drainage had a positive effect on the acquisition of contrasts as on certain processes. Early drainage, in the first year of baby’s life, has more positive effects than later drainage (§8.4). Although a factor analysis could not be executed because of the unequal distribution of the children across cells, inspection of the data strongly suggests that combined as well as early intervention has the most positive results on speech and language.

In conclusion the results from CP children at two years of age show as a group that they are statistically different in their intelligibility, the presence of CP speech deviancies and lexical ability (§9.1). The heterogeneity of the group is, however, very clear. One third of the children hardly speak and are therefore hugely delayed. One third are normal in their lexical and grammatical development but have phonological problems. One third is normal in lexicon, grammar and phonology. Hearing loss due to middle ear infection
has a negative effect on phonological and lexical ability. Medical intervention can alleviate the negative effects of both hearing loss and the cleft palate, especially if carried out early, in the first year of life.

The results have also produced some findings which go further than the research questions ($\S$ 9.2). The target sounds which the children aim at producing are different for the CP group as a whole. This finding is not interpreted as avoidance but as a result of lack of oral tactile-kinaesthetic and auditive information. When an oral plate has been placed from early on and early middle ear drainage has taken place, then the selection of these target sounds becomes more normal. It is suggested, that due to these early interventions, the development of speech perception is enhanced, leading to more normal mental representations of speech and language. The CP children are able to produce speech and language in a more normal way, and in a shorter time. Another finding is that, although the mothers of the CP children as a group were not different in the amount that they spoke, the mothers of the CP children who barely speak are different. The interpretation is that this is a result of the child's slow speech and language development. This point requires further research.

The CP speech score of the children does not relate to other variables in language; intelligibility however does relate. This suggests that the deviant phonetic production has little effect on the higher levels of organization of language ($\S$ 9.3).

Middle ear infection or OME is a long disputed factor in the speech and language development. This study of cleft palate children shows that it does have an effect in these children on both their phonological and lexical ability. An effect on grammatical ability is not clearly found. If children with a cleft differ in MLUL, there is an effect of the severity of hearing loss and/or the abnormal phonological development without [plosive] and/or [dorsal]. This is due to insufficient medical intervention, leading to insufficient oral speech perception and it is not necessarily due to OME. However, early drainage has a positive effect on this variable ($\S$ 9.4).

The study concludes with a brief look at other possible factors which might explain the behavior in the group of CP children who barely speak at two years of age. It does not seem possible to find an explanation in the mother-child interaction, nor in the children's cognitive development. In this group of children more severe hearing loss is found (mostly a level 2 loss of 40–55 dB) with insufficient intervention. Maybe this group of children shows the effect of less language input due to more auditory deprivation than other groups of cleft palate children. The type of cleft (the cleft palate only) may be relevant. Sex of the child may also be important since most were boys. These are points which need further study.