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Retrospective research on the efficiency of ENT treatment and speech- language therapy for cleft palate patients.

Summary

Background: Children with clefts are at high risk for speech and language disorders, particularly when the palate is affected and insufficient velopharyngeal function is present. To prevent psychological and functional problems, the craniofacial team of the VUMC stimulates cleft palate children to reach their full articulation potential during their 6th year. Presumptions exist that discharge of ENT and speech- language therapy happens more and more often after the 6th year. The main purpose of this study is to investigate whether a trend is growing that cleft palate children are discharged from ENT and speech-language therapy at older age. An additional objective of this study is to delineate the impact of a unilateral versus bilateral, a complete or an incomplete cleft palate on the age of discharge from ENT and speech- language therapy, and to compare the ages of discharge of male versus female patients, monolingual versus bilingual patients and patients who registered in the craniofacial team immediately after birth and patients who registered at older age. Furthermore this study investigates whether the indication of a pharyngoplasty influences the age of discharge from ENT and speech-language therapy.

Methods: The sample consisted of 178 non-syndromic patients, all born between 1994 and 2004. Medical records of 109 male (61,2%) and 69 female (38,8) subjects were investigated. For every year between 1994 and 2004 the age (in months), at which the selected patients reached a sufficient articulation potential according to the speech-language therapist and the ENT specialist of the craniofacial team, was listed. Also the gender and mother tongue were listed and whether the patient had an indication for pharyngoplasty. Furthermore the type of cleft was registered, using the LAHSHAL system.

Results: Regression analysis showed a slightly significant relationship between the independent variable 'year of birth' and the dependent variable 'age of discharge', with a R2 of 0,032 and a significance level of 0,038 when counting the patients who were not yet discharged at 03-2011 as missing values. To overcome these missing values of subjects who were not yet discharged at the time of data collection, percentages of discharged patients between 1999 and 2004 are weighted with other years. Regression analysis showed that in this weighing R2 is 0,084 and the significance level is 0,000. Regression analysis showed a more significant relationship between the independent variable 'year of discharge' and the dependent variable 'age of discharge', with a R2 of 0,312 and a significance level of 0,000. None of the variables 'overt or submucous cleft', 'monolingual/bilingual', 'indication for pharyngoplasty' and 'registration at older age' had a significant relation to the dependent variable.

Conclusions: During the period between 1994 and 2004 cleft palate patients, treated by the VUMC craniofacial team tend to reach their optimal articulation level at older age. From 1994 on, the percentage of patients, who are older than 6 jears (84 months) at the moment of discharge from ENT and speech-language therapy, increases. This means that the achievement of the goal of the craniofacial team of the VUMC, to make sure the patient reaches an optimal articulation level at the age of 6, decreases over the years. Several factors that may be of influence on this development are highlighted in the discussion. Future studies are needed to more closely define the impact of the motivation of the patient and the parents on the success of the ENT and speech-language therapy.

1. Introduction

In young children, early detection and early treatment of speech- language disorders is important in order to avoid problems concerning social, behavioral development and school functioning (Silva et al. 1987, Hart et al. 2004, Finneral et al. 2009, Van Daal et al. 2007, McCabe 2005, Johnson et al. 2010). Speech- language disorders occur for a variety of reasons, including malformations of the speech organs. Among the most frequent malformations are craniofacial clefts of the lips, the palate and alveolus. Occurrence in Europe ranges from 6.3 newborns per 10.000 in Spain to 26.2 per 10.000 in Finland (Calzolari et al. 2000). The average value for Europe is 15.2 affected children per 10.000 births, the average in the Netherlands ranges from 16.0 per 10.000 in the South West and 22.7 per 10.000 in the North (Calzolari et al. 2000). Both studies of families with members with clefts and epidemiological studies have shown that clefts can be caused by environmental factors as well as genetic factors (Cristensen 1999). Over five different genes are held responsible for the development of cleft lip and palate (Mavroudi et al. 2007). In most cases the development of a cleft is originated by genetic diversity and is further influenced by genome-environment interaction (Mavroudi et al. 2007). Environmental factors during the first months of pregnancy, such as alcohol, smoking in combination with diabetes, viruses, medication, exposure to radiation and folic acid deficiency can increase the risk of giving birth to an affected child (Mavroudi et al. 2007). Clefts develop when, during the embryonic stage normal development is disturbed and merging of the developing facial components fails to occur (Profitt and Fields 2000). Normally, the maxillary components merge with the medial nasal and lateral nasal components during the 6th week of the pregnancy (Profitt and Fields 2000). When this fusion fails to occur, it could lead to a cleft lip and/or a cleft alveolus (Profitt and Fields 2000). Between the 6th and the 8th week of pregnancy, the closure of the secondary palate takes place. Incomplete fusion of the palatal components results in an overt or submucous cleft of the palate (Profitt and Fields 2000). The type of cleft varies from a complete bilateral cleft of the lip, alveolus, hard and soft palate to a microform cleft of the lip (Mavroudi et al. 2007). Therapeutic management of children with clefts is a long and complex procedure, demanding a team of experts from different scientific fields (Am. Cleft-Palate Craniofac. Assoc. 2004). In case of cooperation with external local health providers, the craniofacial team should coordinate any intervention (Am. Cleft-Palate Craniofac. Assoc. 2004). Evaluation by an experienced ENT specialist and a speech- language therapist is essential because children with clefts are at high risk for speech and language disorders (Schuster et al. 2005, Brunnegard and Lohmander 2007, Ruiter et al. 2009). Particularly when the palate is affected and insufficient velopharyngeal function is present, feeding and nutritional problems, developmental delays, hearing problems and speech and language problems often occur (Brunnegard and Lohmander 2007, Ruiter et al. 2009, Flynn et al. 2009, Beaumont 2008). Speech development depends on control of the lips, tongue and palatal muscles, while language development depends on understanding and using words and sentences (Goorhuis-Brouwer 1978). In cleft palate children, speech problems almost always occur as a result of anatomical anomalies, while language problems are more often related to syndromes associated with clefts (Harville et al. 2007). A significant portion of children have clefts as one feature of a broader pattern. Approximately 30% of the cases is associated with systemic defects and structural abnormalities, the remaining 70% of the cleft cases is nonsyndromic and occur as isolated conditions (Jones 1988, Rollnick and Pruzansky 1981). The incidence of a syndrome in combination with an isolated cleft palate is higher than in other types of clefting (Jones 1988). Almost 400 syndromes associated with clefts are listed (Thomas et al. 2008). Many cleft related syndromes also include mental retardation, developmental delays, learning difficulties and behavioral problems (Venkatesh 2009). Because speech-language development is based on cognitive development (Karmiloff-Smith 1992), these additional problems may have further negative effects on the speech-language development.

Articulation

To correctly produce speech sounds, both speech knowledge of the language system and sufficient motor control are required (Priester et al. 2009). The knowledge of the language system determines whether a particular speech sound is formed in the correct manner (Priester et al. 2009). In children with a nonsyndromic cleft, this ability is usually intact. The amount of speech motor control determines whether a particular speech sound can be made at the place of articulation (Priester et al. 2009). Articulation problems and hypernasality often occur as a result of the anatomical problems in children with a cleft palate (Brunnegard and Lohmander 2007, Ruiter et al. 2009, Flynn et al. 2009, Beaumont 2008). Children's articulation develops between the ages of 6 months and 6 years (see Table 1), and is based on a combination of linguistic knowledge that is acquired by the child, and the growth and maturing of speech motor control (Goorhuis-Brouwer and Schaerlaekens 2000).

Age	Minimal speech potential
1;0 – 1;6	Understands two-word commands Points out Much and varied babbling, accompanied by understandable utterances
1;6 – 2;0	Acquired five to ten words
2;0 – 2;6	Understands three- word sentences Two- word utterances with incomplete word construction
2;6 – 3;0	Three- word utterances with incomplete word construction
3;0 – 3;6	Three- to five- word utterances About 50% of the utterances is intelligible
3;6 – 4;0	Spontaneously tells tales 50 – 75% of the utterances is intelligible
4;0 – 5;6	Tells tales based on pictures Utters simple sentences Difficulties with conjugations and plurals 75 - 90% of the utterances is intelligible
> 5;6	Utters well constructed complex sentences All utterances are intelligible Concrete language use

Table 1; Language acquisition (Goorhuis-Brouwer 2007).

Dutch contains thirteen simple vowels and four diphthongs (Table 2 does not display the vowels /Y/ and /ə/). In normal speech development Dutch children between 1;3 and 1;8 have acquired five Dutch vowels (Beers 1995) (see Table 2). At age 3 normally developing Dutch children have mastered the entire set of mother tongue vowels (Stes and Elen 1991). Not much is known about the acquisition of the diphthongs /ɛɪ/, /œy/ and /ɔu/ (Gillis and Schaerlaekens 2000). Apart from nasality, cleft palate children usually do not have much trouble with pronouncing vowels.

Age	**Vowels**
1;3 – 1;8	i ɪ u ɛ a
1;9 – 1;11	ɔ ɑ
2;0 – 2;2	e o
2;3 – 2;5	
2;6 – 2;8	
2;9 – 2;11	
3;0 – 3;2	Œ
Older	ø y

Table 2; Acquisition order of the Dutch vowels (Beers 1995).

90% of the Dutch-speaking children at age 4 have mastered 64% (14/22) of all Dutch consonants and 75% have mastered 77% (17/22) of the consonants (Stes and Elen 1991). Children must be able to produce the majority of single consonants by the age of 4, with the exception of the /s/ and the /r/ (Priester et al. 2009). In Table 3 the acquisition of the Dutch consonants is displayed. The data are segmented in consonants occurring at the beginning of words and syllables and consonants occurring at the end of words and syllables (Gillis and Schaerlaekens 2000). At least 11 initial consonants (/p/, /t/, /k/, /m/, /n/, /s/, /χ/, /h/, /j/, /f/ and /ʋ/) and 6 at least final consonants (/p/, /k/, /s/, /χ/, /m/, /n/) are present in 80–100% of the normal developing preschool age children (see Table 3) (Beers 1995).

Age	**Initial consonants**	**Final consonants**
1;3 – 1;8	p t m n j	P
1;9 – 1;11	k	K
2;0 – 2;2	s χ h	t s χ
2;3 – 2;5	b f ʋ	m n
2;6 – 2;8	l r	
2;9 – 2;11	d	
3;0 – 3;2		
Older	ʃ	l r f ŋ

Table 3; Acquisition order of the Dutch consonants (Beers 1995).

At 2;11 the acquisition of initial consonants is almost completed (Gillis and Schaerlaekens 2000). The occlusives /p/, /t/, and the nasals /m/, /n/ and the approximant /j/ are acquired between 1;3 and 1;8, followed by the acquisition of /k/. After acquisition of these sounds the acquisition of the fricatives /s/ and /χ/, and the acquisition of /h/ starts (Gillis and Schaerlaekens 2000). At the age of 2;3 the first voiced consonants, /b/ and /ʋ/, together with the fricative /f/ are acquired. The liquids /l/ and /r/ will not be acquired before 2;6 and the acquisition of the voiced /d/ takes place even later. The /ʃ/ is acquired at older age because at 3;2 the production did not meet the criteria of the research (Gillis and Schaerlaekens 2000).

The voiced fricatives /z/ and /v/ are not listed in the table because the research took place in the Randstad (the area around Amsterdam, Rotterdam, Utrecht and The Hague), where voiced fricatives are devoiced in spontaneous speech (Gillis and Schaerlaekens 2000). The acquisition of consonants follows a strict pattern; occlusives are acquired before fricatives, followed by nasals and liquids (Gillis and Schaerlaekens 2000). There seems to be a difference in the acquisition of initial and final consonants; a consonant is first acquired in the initial position of a word or syllable and thereafter in final position (Gillis and Schaerlaekens 2000). The acquisition seems to be not entirely finished at the age of 4 (Gillis and Schaerlaekens 2000), but if the sounds, enlisted in table 2, are not acquired at this age, an articulation problem is considered to be present (Priester et al. 2009). Articulation disorders could also include backing, nasalization and the use of compensatory articulations (Priester et al. 2009). In normal development the majority of consonant clusters is present at age 4 or is thought to develop between 4 and 6 years of age. Exceptions are the production of /sch/ and consonant clusters with /s/ or /r/, which will develop later on (Priester et al. 2009). Within the area of speech management it is well known that there are certain speech characteristics that are typical for cleft palate patients who require treatment of their speech disorder (Henningson et al. 2008). One of the specialists participating in the rehabilitation of cleft palate patients is the speech-language therapist. At least half of all cleft palate patients requires speech-language therapy at some point in their lives (Peterson-Falzone et al. 2001). The specific treatment goals of speech-language therapy are to establish correct articulatory placement, to modify resonance disorders and to treat accompanying language and voice disorders (Henningson et al. 2008). For children with cleft palate, specific sounds are difficult to produce because of the functional and structural anomalies associated with clefting. Most obviously, the inability to build up intraoral air pressure due to the open oral-nasal passage interferes with the production of high-pressure consonants, such as /p/, /t/ and /b/ (Chapman et al. 2001). Studies of phonetic development of young infants with cleft palate during the first year of life have shown that a greater number of nasals, glides, and glottal fricatives and fewer oral stops are present in their early inventories. These children also showed a preference for the glottal and labial place of articulation (O'Gara et al. 1994). This avoidance of alveolar and palatal sounds may be related to the reduced palatal surface available for lingual contact in children with wide palatal clefts (Lohman-Agerskov et al. 1998). The repertoire of consonants available to children with cleft palate is reduced by the limitations in manner and place characteristics (Chapman et al. 2001). The speech problems of toddlers with a cleft palate mainly are articulation problems of /b/, /t/, /d/, /v/, /f/, /z/ and /s/ (Beers 1995). The consonants /t/ and /d/ are often palatalised and substituted by /k/ or a pharyngeal friction. The fricatives /f/ and /s/ are often nasalized or substituted by nasal friction. The /b/, /v/ and /z/ are often devoiced (Beers 1995).

ENT

Children with a cleft palate are in high risk of developing otitis media with effusion (OME) (Jansonius-Schultheis 1999, Doyle et al. 1980, Paradise et al. 1969). A high incidence of OME and hearing loss is found among children with cleft palate, even after surgical repair of the cleft (Paradise et al. 1969). OME is reported to be almost universal among infants with cleft palate (Paradise et al. 1969) and can be caused by many factors. The immune system can be influenced by factors such as age, genetic predisposition, and atopy, whereas environmental factors, such as siblings, day-care attendance, and season, relate to microbial load (Bluestone 1996). As seen in Figure 1, the Eustachian tube plays an important, central role (Rovers 2008). During a normal swallow movement, the Eustachian tube opens to refresh the air in the middle ear (Jansonius-Schultheis 1999). Due to a cleft of the palate, the tensor and levator veli palatini muscles can have functional defects. Due to the abnormal insertion of the tensor and levator palati muscles the Eustachian tube fails to open, resulting in functional tube obstruction, negative middle ear pressure and insufficient ventilation of the middle ear (Doyle et al. 1980). The main consequence of OME for most affected children is a collection of fluid that occurs within the middle ear space as a result of the negative pressure, causing a mild-to-moderate conductive hearing loss (Jansonius-Schultheis 1999). In hearing loss caused by OME, the infection-induced fluids reduce the intensity of sound vibrations, which reach the inner ear via the tympanic membrane. The vibrations are damped and this causes deteriorated perception of sound (Jansonius-Schultheis 1999). An episode of acute otitis media can cause two problems, recurrent acute otitis media or persistent middle ear effusion (Damoiseaux et al. 2005). In about 50% of the children under 2 years with acute otitis media, a persistent middle ear effusion is found within three months after occurrence (Damoiseaux et al. 2005). To prevent development of persistent otitis media, surgical treatment may be recommended to reestablish ventilation of the middle ear, keep the hearing at a normal level, and prevent recurrent infection. One of the strategies used to treat this condition is to place tympanostomy tubes after myringotomy to remove the middle ear fluid, which is drained with a ventilation tube (Testa et al. 2010).

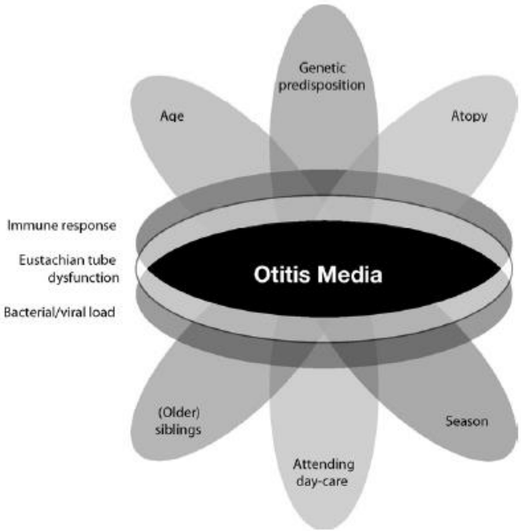

Figure 1; Risk factors involved in the pathogenesis of otitis media (Copied from Bluestone 1996).

Because the hearing loss associated with OME commonly affects children at an age of rapid speech- language development and acquisition of social skills, concerns have been raised regarding the possible long-term consequences of OME on speech, language, cognitive development, intelligence, and behavior (Sheahan 2002). But there continues to be considerable debate over whether a history of OME during the first few years of life causes later speech and language difficulties (Roberts, Rosenfeld and Zeisel 2004). Hearing loss has been hypothesized to cause a child to encode information inefficiently, incompletely, or inaccurately into the database from which language develops (Roberts et al. 2004). In a meta-analysis, Roberts, Rosenfeld and Zeisel (2004) found a slightly negative association between OME and preschoolers' receptive and expressive language in the group studies. Additionally, hearing was also related to receptive and expressive language in infancy (3%–9% of variance). Conversely, there were no significant findings for the analyses of OME during early childhood versus receptive or expressive language during the preschool years in the correlation studies. Similarly, there are no significant findings for OME versus vocabulary, syntax, or speech during the pre-school years. These results indicate no to very small negative associations of OME and associated hearing loss to children's later speech and language development (Roberts, Rosenfeld and Zeisel 2004). A multicenter, randomized, controlled trial (embedded in a cohort) with 2 treatment arms studied the effectiveness of ventilation tubes on the language development in Dutch speaking infants with persistent OME. Hearing loss and expressive and comprehensive language were assessed every 6 months, while tympanometry and otoscopy were performed every 3 months. No relevant differences were found in expressive or comprehensive language between the 2 groups after adjustment for educational level of the mother, IQ of the child, and differences at baseline (Rovers et al. 2000).

Apart from tympanostomy tubes, the most applied medical treatment in cleft palate children is a pharyngoplasty. In cleft palate children the levator palati muscles often have been shifted from their normal transverse arrangement to a longitudinal position (Stal et al. 1998). The muscles insert aberrantly on the bony free edge of the hard palate instead of forming a complete muscular sling, leading to inability of the posterior margin of the soft palate to fully contact the pharyngeal wall (Stal et al. 1998). The degree of velopharyngeal insufficiency depends on the anterior displacement of the muscles. When velopharyngeal insufficiency severely reduces the intelligibility, the ENT specialist has to decide whether a pharyngeal flap or a pharyngoplasty is needed to reduce the degree of nasality (Abdel-Aziz 2007). Pharyngeal flaps are usually superiorly based pedicle flaps of mucosa and underlying constrictor muscle. The goal of a phyngoplasty is to create lateral ports that can easily close to diminish nasality and improve intelligibility (Abdel-Aziz 2007).

Social- behavioral aspects

The goal is for cleft palate children to have a sufficient articulation potential by the age of 6 in order to prevent psychological and functional problems. In a study of 44 children with clefts from age 4 to 12, was found that rates of internalising problems increased at age 7 (Richman and Millard 1997). Children with clefts have, from the age of 7, raised rates of teacher-reported social problems and anxious and depressed behavior. Direct observations and child representations also revealed that children with cleft palate have more difficulties in social relationships (Murray et al. 2010). The percentages of 7 year old children in cleft and control groups scoring in the borderline/clinical range on teacher reports (TRF) and child problems reported by mothers (CBCL) of overall internalising and externalising problems,

	% borderline clinical range			
	Control	Cleft	χ^2	p
a) TRF	n = 72	n = 89		
Internalising	9.7	20.2	3.34	.05
Externalising	11.1	14.6	0.42	.34
Social problems	5.6	18.0	5.64	.01
Anxious depressed	1.4	11.2	6.06	.01
Withdrawn depressed	1.4	10.1	5.19	.02
b) CBCL	n = 75	n = 91		
Internalising	10.7	16.5	1.16	.20
Externalising	13.3	16.5	0.31	.37
Social problems	4.0	8.8	1.52	.18
Anxious depressed	8.0	7.7	0.01	.58
Withdrawn depressed	2.7	8.8	2.72	.09

Table 3; Group differences on child emotional adjustment (TRF & CBCL) (Copied from Murray et al. 2010).

and of anxious-depressed, withdrawn-depressed and social problems are shown in Table 3 (Murray et al. 2010). The most consistent predictor of the difficulties experienced by children with cleft palate is their capacity to communicate effectively, and such problems include both speech-language and pragmatic difficulties (Richman and Millard 1997). Children with language difficulties show significantly more withdrawn behavior in the playground and spend few time interacting than their peers (Fujiki et al. 2001). Child communication problems largely accounted for these effects, especially in children with cleft palate as well as cleft lip. So children with clefts are at raised risk for socio-emotional difficulties in the school years; clinical interventions should focus on communication problems; specific interventions around the transition to school may be required (Murray et al. 2010). School functioning is also in danger when speech- language skills are insufficient. Law et al. (1998) state that: ‘‘Speech and language development is intimately related to all aspects of educational and social development’’[1]. In cleft palate children anatomical problems often cause speech problems, while hearing problems often cause problems with perception and delay language acquisition. This could, in combination with psychological problems, result in communication difficulties. Communication impairment that first presents in childhood may be associated with activity limitations and participation restrictions that extend across the lifespan (Law et al. 1998). Intervention of ENT specialist, speech-language therapist should start as early in life as possible if there is a possibility for developing these problems. Systematic reviews of the literature have shown that children, who are not treated before their school years, can continue to have educational, occupational and social difficulties until at least 28 years of age (McCormack et al. 2009). Having a communication problem can impact on educational outcomes in both the short and long term. Children with communication problems typically require more school-based remedial assistance than normally developing peers, and may achieve lower grades. They can have difficulty with mathematics and literacy, including difficulty with phonological awareness, spelling, reading comprehension, and reading accuracy (Mackie et al. 2004, McLeod 2009). To prevent these potential problems, the craniofacial team of the VUMC stimulates cleft palate children to reach their full articulation potential during their 6th year. But presumptions exist that discharge of ENT and speech-language therapy happens more and more after the 6th year. The main purpose of this study is to investigate whether a trend is growing that cleft palate children are discharged from ENT and speech-language therapy at older age. An additional objective of this study is to delineate the impact of a unilateral versus bilateral, complete or incomplete cleft, on the age of discharge from ENT and speech- language therapy, and to compare the ages of discharge of male versus female patients and monolingual versus bilingual patients and if registration at older age is of influence. Furthermore this study investigates whether the indication of a pharyngoplasty influences the age of discharge from ENT and speech- language therapy.

[1] Law et al. (1998) p. 2

2. Methods

Subjects

Every year approximately 40 new patients with an orofacial cleft register in the craniofacial team of the VUMC. The sample consisted of 178 non-syndromic patients, all born between 1994 and 2004. Medical records of 109 male (61,2%) and 69 female (38,8) subjects were investigated. 10 patients, 7 boys and 3 girls, (5.6%) attended the cleft palate team not immediately after birth and were considered as an apart group of subjects. All 10 subjects were adopted from China and registered in the craniofacial team at older age.

Treatment

All patients have been treated from birth until the age of 21 by the VUMC craniofacial team. The craniofacial team of the VUMC uses the following treatment protocol:

0 mnd	orthodontie	plastische chirurgie	
3 mnd	orthodontie	plastische chirurgie	
9 mnd	orthodontie	plastische chirurgie	
1 jaar		KNO / logopedie	team
1,5 jaar		kindertandheelkunde	
2 jaar		KNO / logopedie	
2,5 jaar		kindertandheelkunde	
3 jaar		KNO / logopedie	
4 jaar		KNO /logopedie / kindertandheelkunde	team
5 jaar	orthodontie	KNO / logopedie	
6 jaar	orthodontie	KNO / logopedie	team
9 jaar	orthodontie	kaakchirurgie	team
12 jaar	orthodontie		team
15 jaar	orthodontie		team

Table 4; Treament protocol of the VUMC craniofacial team. (Copied from: vumc.nl/afdelingen/schisisteam).

In all cases speech- language development was judged as insufficient and from the 3th year treatment of ENT (if not before) and speech- language therapy started. After referral by the craniofacial team, speech therapy was given by a speech-language therapist in the residence of the patient. Controls on speech-language development took place at least twice a year by the ENT specialist and the speech-language therapist of the craniofacial team, and assessment was done by means of the Nederlandstalig Schisis Articulatie Onderzoek. When the patient reached a sufficient articulation level in which no errors associated with cleft palate occur, the speech-language treatment was considered completed and the patient was discharged from speech-language therapy.

The goal of the craniofacial team of the VUMC is to make sure the patient reaches an optimal articulation level at the age of 6, just before the patient starts reading and writing. Between 1994 and 2004 the same treatment protocol has been maintained, and all patients have been treated and assessed by the same ENT specialist and speech-language therapist.

Assessment

From the age of 3 until discharge of ENT and speech-language therapy, the patients articulation was evaluated at least twice a year. After the patient has turned 6, assessment took place to judge whether the patients has reached his optimal articulation level. The patients articulation potential was assessed by the speech-language therapist and the ENT specialist of the craniofacial team. Assessment consist of the following:

1. Assessment of consonant formation in words and sentences.
2. Assessment of nasal air escape during syllable, word and sentence production.
3. Subjective assessment of the intelligibility.
4. Assessment of the nasal resonance by means of nasometry.
5. Assessment of facial grimaces during speech production.
6. Comparison of speech in test setting and spontaneous speech.
7. Investigation whether there are any circumstances in which the child is able to produce the difficult sounds.

When the patients articulation potential was assessed to be sufficient, he/she was discharged from speech-language therapy. When the articulation potential was judged insufficient, the patient continued to receive speech-language therapy until he/she reached a sufficient articulation potential and passed the articulation test. When the patient, at age 6 or older, showed, at least tree times in a row, no improvement at the articulation test, a ceiling effect was assumed and the patient was discharged.

Material

All patients born between 1994 and 2004, who met the criteria and reached their full articulation potential were investigated. All medical records of the selected cleft palate patients were examined. Then for every year between 1994 and 2004, the age (in months), at which the selected patients reached a sufficient articulation potential according to the speech-language therapist and the ENT specialist of the craniofacial team, was listed. Also the gender and mother tongue were listed and whether the patient had an indication for pharyngoplasty. Furthermore the type of cleft was registered, using the LAHSHAL system. The LAHSHAL system uses the initials of the English terms Lip, Alveolus, Hard and Soft palate in one line to describe the cleft (see Figure 2). Thus, a complete bilateral cleft of the lip, alveolus, soft and hard palate is described as "LAHSHAL" (O'Kriens 1989), whereas a cleft of the lip and alveolus of the left side is described as "LA…..". Incomplete clefts are recorded using small letters whereas complete clefts are described with capital letters (O'Kriens 1989). Because the medical records showed no specificity other than type of cleft, clefts were classified by the following categories: "LAHSHAL", "LAHS…", "…SHAL", " ..HSH.." and "submucosal"

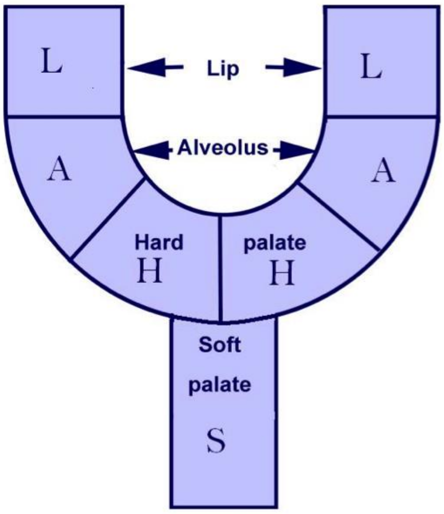

Figure 2; LAHSHAL system of cleft lip alveolus and palate documentation (Copied from Kriens 1989).

Analysis

To discover whether nowadays cleft palate children reach their full articulation potential at later age than before, the following H0 hypothesis should be rejected in favor of the H1 hypothesis:

H0: Between 1994 and 2004 all cleft palate children reached their optimal articulation level at the same age.

H1: Between 1994 and 2004 a trend is grown that cleft palate children reach their optimal articulation level at older age.

To do so, the correlation between the year, during which a cleft palate child is born, and the age, at which the child is discharged, was calculated. The relation between the independent variable 'year of birth' and the dependent variable 'age of discharge' was investigated though linear regression analysis. Also, the correlation between the year, during which a cleft palate child is discharged from ENT and speech-language therapy, and the age, at which the child is discharged, was calculated. Furthermore the relation between the dependent variable 'age of discharge' and the independent variables 'gender', 'type cleft', 'overt or submucous cleft', monolingual/bilingual', 'indication for pharyngoplasty' and 'registration at older age' was investigated trough multiple regression analysis.

3. Results

'Age of discharge' per 'year of birth'

Regression analysis showed a slightly significant relationship between the independent variable 'year of birth' and the dependent variable 'age of discharge', with a R2 of 0,032 and a significance level of 0,038 when counting the patients who were not yet discharged at 03-2011 as missing values. Figure 3 displays a scatterplot of all discharged patients, born between 1994 and 2004. As seen in Figure 3, the regression line steadily runs upward. Patients who were not yet discharged at 03-2011 were labeled as missing values, because their age of discharge was still unknown at time of data collection and it is uncertain at what age the subjects would have been discharged from ENT and speech-language therapy. If discharge of all patients with an

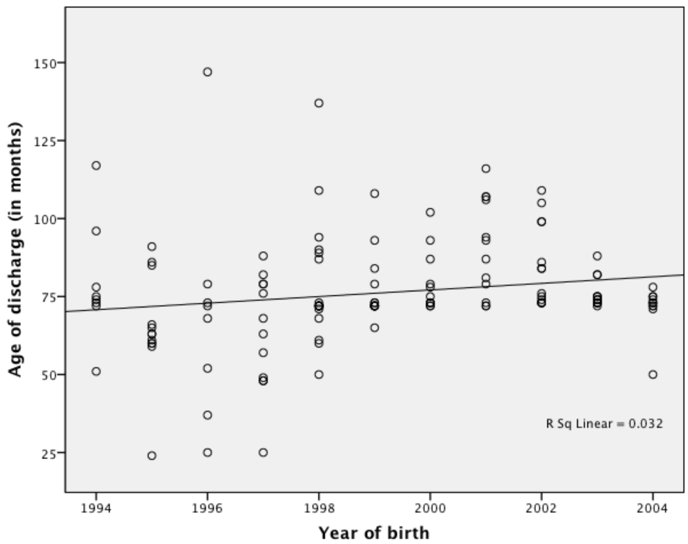

Figure 3; Scatterplot of 'the age of discharge' per 'year of birth', leaving out all subjects not discharged at 03-2011.

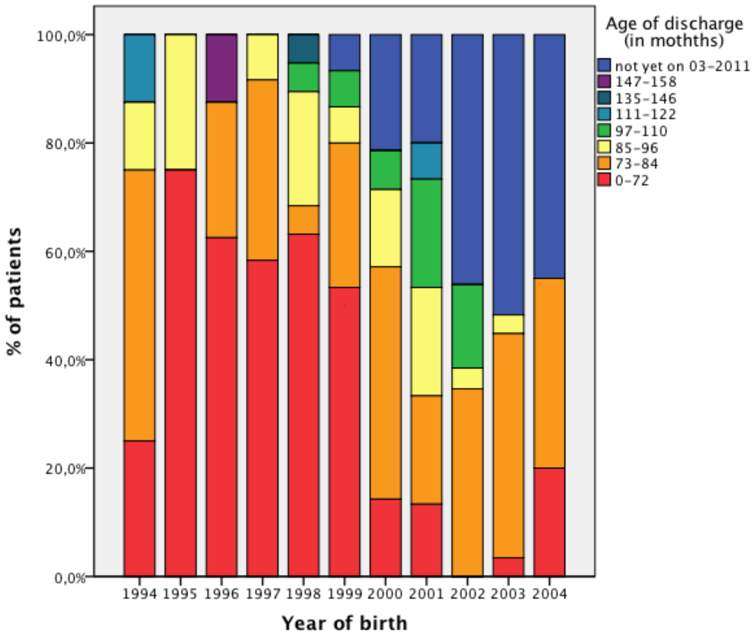

Figure 4; Bar graph of the percentage of patients per year of birth, per category of age of discharge.

unknown value would have taken place at 03-2011, age of discharge would have ranged from 75 to 135 months. Unfortunately, when labeling patients who are still receiving ENT and speech-language therapy as missing values, the results become biased because the graph shows a far slighter slope then it would if the age of discharge of all patients would have been known. The distribution of the number of patients who were not yet discharged at 03-2011 is displayed in the following graph. As seen in Figure 4 the percentage of patients who are not yet discharged at the moment of counting starts growing in 1999 at 7% and peaks in 2003 with 52%. At the same time, the percentage of patients who were discharged at or before 72 months drops. This percentage is highest in 1995 with 75% and lowest in 2002, when not a single patient was discharged before or at the age of 72 months.

To overcome these missing values of subjects who were not yet discharged at the time of data collection, percentages of discharged patients between 1999 and 2004 are weighted with other years. For every year with missing values (1999–2004) the percentage of discharged patients was calculated and compared to a equated percentage subjects of all other years. This comparable percentage of subjects, consisted of the earliest discharged patients per year, with as result that only the 'best' patients of every year between 1994 and 2004 are compared. Figure 5 shows the

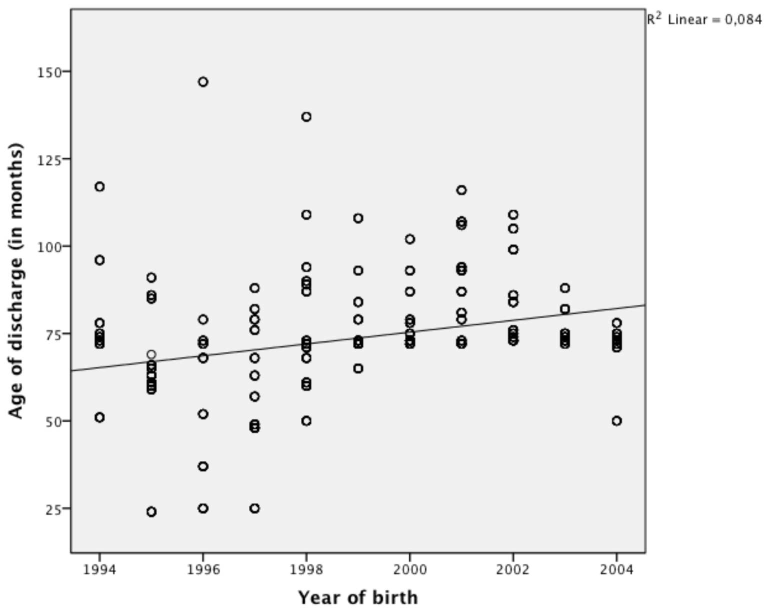

Figure 5; Scatterplot the weighted cases of 'the age of discharge' per 'year of birth'.

distribution of the weighted cases. Regression analysis showed a R2 of 0,084, while the significance level is 0,000. Figure 5 obviously shows a regression line with a steeper slope than Figure 1, revealing an even faster increase of age of discharge per year.

'Age of discharge' per 'year of discharge'

Regression analysis showed a more significant relationship between the independent variable 'year of discharge' and the dependent variable 'age of discharge', with a R2 of 0,312 and a significance level of 0,000. This relation is displayed in Figure 6 with an even steeper slope than Figure 5, implicating an increase of 'age of discharge' with every year. Extreme outliers are seen in Figure 6 between 1995 and 2000 far beneath the regression line, and between 2005 and 2010 far above the regression line. The exact distribution of subjects is displayed in Figure 7. All subjects who were still receiving ENT and speech-language therapy at the time of data collection are represented by the grey bar, which shows that all subjects will be discharged in 2012 or later, at the age of 75 months or older. Red bars represent the patients who passed the articulation test at, or before, 72 months. As clearly seen in Figure 7, the percentage of patients who are discharged at older age than 72 months increases swiftly. In 1997 - 1999 all discharged patients were 72 months or younger, while in 2011 all patients were 97 to 110 months old when ENT and speech- language therapy ended.

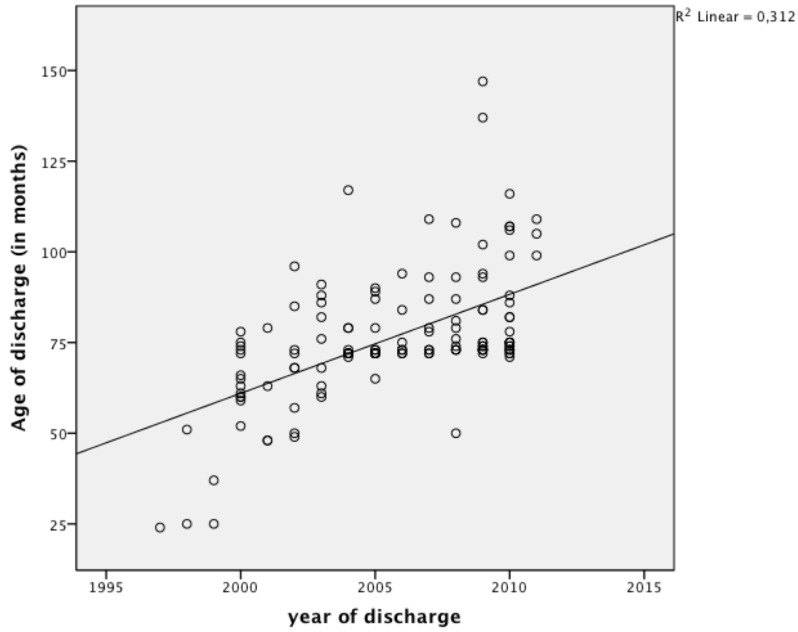

Figure 6; Scatterplot of 'the age of discharge' per 'year of discharge', leaving out all subjects not discharged at 03-2011.

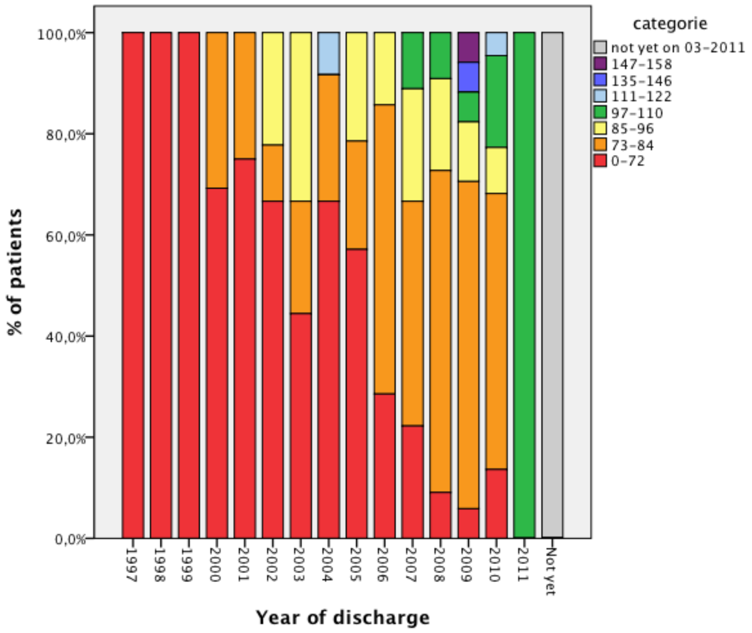

Figure 7; Bar graph of the percentage of patients per 'year of discharge', per category of 'age of discharge'.

Other variables

The relation between independent variable ‘age of discharge’ and the dependent variables ‘gender’, ‘type cleft’, ‘overt or submucous cleft’, ‘monolingual/bilingual’, ‘indication for pharyngoplasty’ and ‘registration at older age’ were investigated trough multiple regression analysis. Correlation between the independent variable and these dependent variables may have provided a possible explanation of the relation between ‘year of birth’ or ‘year of discharge’ and ‘age of discharge’. Medical records of 109 male (61,2%) and 69 female (38,8) subjects were investigated. 48 subjects (27%), 32 male, 16 female suffered from a cleft palate on the left side (see Table 5). 25 (14%) of the subjects suffered from a cleft of the right side, 19 of then were male and 6 female. 28 (15.7%) patients suffered from a double cleft (17 male and 11 female) and 77 (43.3%) patients had a cleft of the hard and soft palate (41 male and 36 female). Of all patients, 7 male and 7 female subjects (7.9%) had a submucous cleft of the palate. 10 patients, 7 boys and 3 girls, (5.6%) were adopted from China and registered in the craniofacial team at older age.

variable	Male (109)	Female (69)
Type cleft	17 double 32 left side 19 right side 41 hard and soft palate	11 double 16 left side 6 right side 36 hard and soft palate
Submucous cleft	7	7
Indication pharyngoplasty	32	19
Registration at older age	7	3

Table 5; Summary of the dependent variables per gender.

None of the previously mentioned variables had a significant relation to the dependent variable. Table 6 shows the R2 and significance level per variable, leaving out the variable ‘monolingual/bilingual’. This variable is not displayed because the medical records of the investigated patients lacked information on this subject. The mother tongue of patients was not systematically mentioned (bilingualism was mentioned in 26 cases but suspected in more) and could thereby not provide reliable results. In all cases of registration at older age the mother tongue was reported. All patients who did not join the craniofacial team immediately after birth were adopted from China and spoke any form of Chinese as first language. The remaining patients, whose mother tongue is known, spoke several different languages as first language.

Variable	R square	Significance level
Gender	0,004	0,480
Type cleft	0,001	0,699
Submucous	0,006	0,374
Indication pharyngoplasty	0,010	0,255
Registration at older age	0,006	0,188

Table 6; Regression analysis of the dependent variables.

As seen in Table 6, boys and girls are discharged at the same age, within the range of 24 to 147 months. The type of cleft, whether this is a double, right sided, left sided cleft or a cleft of the hard and/of soft palate has no significant influence on the age of discharge. Neither changed a submucous cleft or an indication to apply a pharyngoplasty the outcome of the ENT and speech- therapy. Even a registration in the craniofacial team at older age, as result of adoption, made no difference to the age at which the child finished receiving ENT and speech-language therapy.

4. Discussion

Nederlandstalig Schisis Articulatie Onderzoek

Speech characteristics related to cleft palate typically can be and are identified perceptually by ENT specialist and speech-language pathologists who are experienced in working with patients with cleft palate. Several systems have been proposed as outcome measures for speech of individuals with cleft palate (Henningson et al. 2008, Fuchs et al. 2008, Schuster et al. 2007). Thus, there is a great deal of variability among the various systems that are being used to collect and analyze data related to speech. A State-of-the-Art article presented a thorough review of methods used for description of speech characteristics related to cleft palate during the last 50 years. The authors stated that ‘‘we must strive for a more standardized protocol for describing articulation . . . ’’[2] regarding the different aspects of speech in clefts (Keuhn and Moller 2000). The ENT specialist and speech-language therapist of the VUMC craniofacial team used the same criterions to assess the patients articulation (as seen in Chapter 2, Paragraph 3 ‘Assessment’) during the period between 1994 and 2004. Based on these criterions the speech-language therapist of the VUMC craniofacial team developed a standardized articulation test, with respect to articulation errors in cleft palate speech. The Nederlandstalig Schisis Articulatie Onderzoek was developed in cooperation with the Nederlandse Vereniging voor Schisis en Craniofaciale Afwijkingen (NVSCA) in order to constitute an adequate assessment of cleft palate errors. Since introduction in 2002, the standardized articulation test has been used by the craniofacial team of the VUMC, and thereafter by all Dutch craniofacial teams, to assess the patients articulation potential of cleft palate patients. This test is, since 2002, commonly used to assess the level and progress of the patients speech development, whether ENT treatment and/or speech-language therapy is

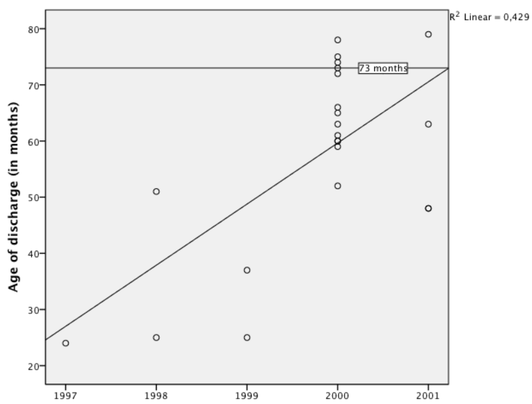

Figure 8; Scatterplot of ‘the age of discharge’ per ‘year of discharge’ from 1997 untill 2001.

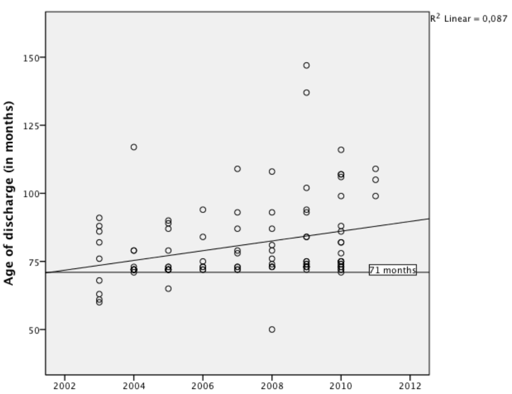

Figure 9; Scatterplot of ‘the age of discharge’ per ‘year of discharge’ from 2003 onwards, leaving out all subjects not discharged at 03-2011.

[2] Keuhn and Moller (2000); p. 353

required and to decide whether the patient will be discharged from ENT and speech- language therapy. Introduction of the Nederlandstalig Schisis Articulatie Onderzoek during the analysed period between 1994 and 2004 may have influenced the results by alternating the assessment tools. Figure 8 shows a scatterplot of the 'age of discharge' per 'year of discharge', of all patients, discharged before the introduction of the Nederlandstalig Schisis Articulatie Onderzoek in 2002. Figure 9 shows a scatterplot of all patients, discharged in 2003 or later. As seen in Figure 8 and 9, the increase of age at time of started during the period before the introduction of the articulation test, and continues during the period after 2002. The introduction of the test does not seem to influence the data, because the regression line in Figure 8 halts at 73 months in 2001, while in continues in Figure 9 in 2003 at 71 months. This means that, during 2002, no notable shifts have taken place. It might even be the case that, during the first period after the introduction of the Nederlandstalig Schisis Articulatie Onderzoek patients were discharged at yonger age than before the introduction. This means that criterions have probably became a little less severe after introduction of the Nederlandstalig Schisis Articulatie Onderzoek as articulation test. If this is true, the regressionline of Figure 6 (Chapter 4, Paragraph 'Age of discharge' per 'year of discharge') would be even steeper.

Language

Between 1996 and 2005 (data from 1994 and 2004 were not available) the percentage of immigrants in the Netherlands increased with 19.98% from 2498715 to 3122717 (see Figure 10). Most immigrants originate from Morocco, the (former) Dutch Antilles, Suriname and Turkey. As seen in Table 7, the number of immigrants of the previously mentioned countries increased swiftly over the period between 1996 and 2005. It is reasonable to assume that the same increase applies for the period between 1994 and 2004.

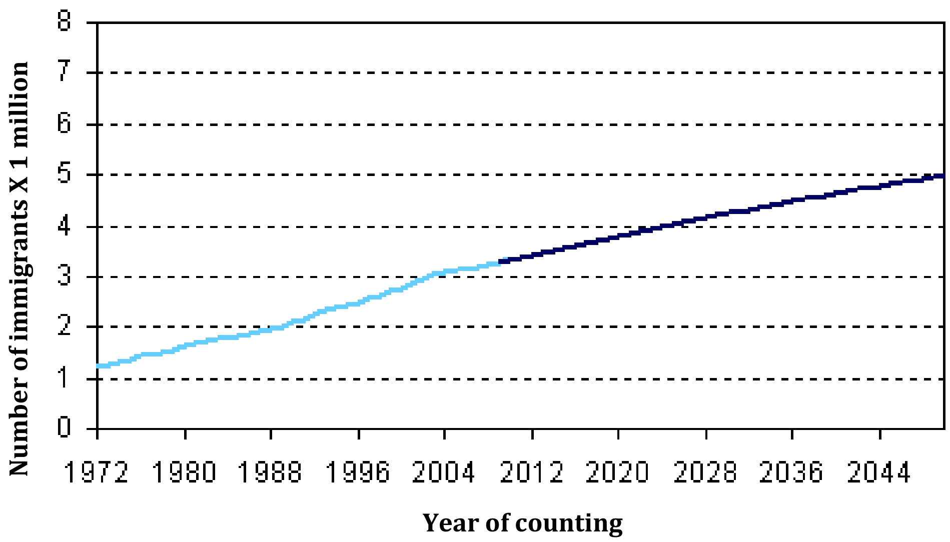

Figure 10; Graph of the number of immigrants in the Netherlands between 1972 and 2010 (counted at the 1th of januari) and prognosis of 2011-2050 (Copied from CBS Bevolkingsstatistiek, CBS Allochtonenprognose in: Sanderse et al. 2010)..

Country of origin	1996	2005	Increase (%)
Morocco	225088	315821	28.7%
(former) Dutch Antilles and Aruba	86824	130538	33.5%
Suriname	280615	329430	14.8%
Turkey	271514	358846	24.3%

Table 7; Increase of immigrants between 1996 and 2004, classified in the four most common countries of origin. (Copied from CBS Bevolkingsstatistiek).

Most immigrants have a mother tongue other than Dutch and are therefore bilingual. Generally speaking, a bilingual (or, in more general terms, a multilingual) is someone in the possession of two (or more) languages (Brasileiro 2009). There is a certain amount of agreement in the literature that it is not necessary for the bilingual to be equally proficient in all of his or her languages to be considered a bilingual. It is actually very rare for a child to acquire native-like proficiency in more than one language (Pearson et al. 1993). In most cases, bilinguals have greater proficiency in one language, which is referred to as their dominant (or stronger) language, the other language being referred to as their non- dominant (or weaker) language (Brasileiro 2009). This has consequences for Dutch spoken language in children of non-Dutch spreaking immigrants and adopted children. Lenneberg (1967) hypothesized that becoming fluent in a language could be acquired only within a critical period, extending from early infancy until puberty. Research supports the conclusion that a critical period for language acquisition has not only consequences for first language, but extends its effects to second language acquisition (Johnson and Newport 2004). The children, mentioned above, start acquiring Dutch immediately after birth or at school age, but before the critical period of language acquisition is over. In order to acquire the speech sounds of their languages, bilingual infants have to deal with the same challenges as monolingual children and have to deal with various additional factors, such as structural differences between their languages, frequency of each sound in each language and the level of overlap between different categories. (Bosch & Sebastián-Gallés 2003). Specifically bilinguals' input is more complex than that of monolinguals. In the case of vowel acquisition, for instance, bilingual children have to acquire a greater number of vowels than monolingual children, since their input presents them with two vowel systems (Brasileiro 2009). The relative frequency of exposure to each of these categories is different than when learning one language at the same time. Generally speaking, dual exposure results in reduction of bilingual children's input. Even assuming the ideal 50%-50% exposure to each of the languages, bilingual input is reduced to half of the input a monolingual child receives (Brasileiro 2009). One could in this case hypothesize a delay in the acquisition of a contrast by children acquiring two languages. A brief review of the literature in bilingual sound acquisition seems to confirm this hypothesis (Brasileiro 2009). These characteristics of bilingual acquisition applies to immigrants in the Netherlands as well, resulting in a difference between native Dutch children and immigrant children who learn Dutch as a second language.

As mentioned in the previous chapter (Chapter 3, Paragraph 3 'Other variables') the mother tongue of patients could not be systematically documented. Bilingualism was mentioned in 26 cases but suspected in more. Figure 11 displays the distribution of the documented bilingual patients, born during the period between 1994 and 2004. This distribution reflects more or less the increase of immigrants in the Netherlands, as described in Figure 9.

Figure 11; Graph of the distribution of the 26 documented bilingual patients, born during the period between 1994 and 2004.

For cleft palate children speech acquisition is impeded by malformations of the speech organs and bilingualism may interfere with a positive outcome of language acquisition. This means that the increase of age of discharge of ENT and speech-language therapy may be influenced and perhaps partly explained by the increase of immigrant children in the Netherlands. This increase of immigrants is predicted to grow, as seen in Figure 10, and may, in the future become of more importance to the outcome of the ENT and speech-language therapy of the craniofacial team. As seen in in the previous chapter (Chapter 3, Paragraph 3 'Other variables) the increase of adopted Chinese children in the dataset had no influences on the outcome of the research.

Velo-Cadio-Facial Syndrome

The Velo-Cardio-Facial Syndrome (VCFS), also known as 22q11.2 deletion syndrome is caused by the deletion of a small segment of the long arm of chromosome 22 (specified as 22q11.2 deletion), and is one of the most common genetic disorders in humans (Ardinger and Ardinger 2002). VCFS is typically characterized by cleft palate, heart abnormalities, learning disabilities, and over 180 other clinical findings (Vrtička 2007). The communicative impairment in VCFS involves voice, language, articulation and hearing. Speech therapy is influenced by the mental retardation, behavioral problems and attention deficits. Whether there exists a syndrome-specific articulation pattern in VCFS is not yet fully decided, but it shows that a limitation in the range of motion of articulatory movements and the variety of articulatory postures is a salient characteristic of speech in VCFS (Van Lierde et al. 2001). It must be considered that VCFS has a variable expression. The degree of mental retardation, the presence of hearing loss, type and severity of palatal anomaly can determine particular articulation patterns (Van Lierde et al. 2001). The treatment of VCFS patients is identical to that of any patient with velopharyngeal dysfunction. However, delay in cognitive development, behavior problems and muscular weakness in VCFS patients may negatively influence the outcome of the treatment. The speech-language therapist must be aware of the learning disabilities and concentration problems present in the majority of the VCFS children.

Their cognitive level tends to stay at a concrete level causing problems with abstract thinking and organization or planning (Vantrappen et al. 1998). In order to keep this study as unbiased as possible, all patients of the VUMC craniofacial team with a cleft, combined with a syndrome were excluded from the dataset. This is also the case for VCFS patients. VCFS can be detected by Fluorescence In Situ Hybridization (FISH) and Array Comparative Genomic Hybridization (Array CHG) (see Figure 12). FISH uses fluorescent staining in order to identify chromosomes involved in multifaceted rearrangements (Bishop 2010). In array CGH, genomic DNA from the patient and DNA from a reference are labeled with different fluorescent colours and co-hybridized to the array matrix glass containing clone DNA. By calculating the ratio of the fluorescence intensity of the test to that of the reference DNA, the copy number changes for a particular location in the genome can be measured. (Lu et al. 2007). Even though Array CHG was introduced in the early 1990 FISH was first used in 1994, the detection of VCFS is not guaranteed in all patients included in this study. FISH is not completely suitable for whole genome scans in routine clinical testing because it lacks the necessary resolution (Bishop 2010). Unlike FISH, array CHG does not include uniform coverage across the arms of each chromosome (Cheung et al. 2005). Apart from inadequacy of the methods, detection is dependent on clinical referral, and therefore patients with atypical or minimal phenotype might be overlooked (Kobrinski and Sullivan 2002). This means for the results of this study that some VCFS patients with minimal phenotype might be included in the sample. It is more likely that diagnosis patients with VCFS was omitted during the period when the FISH and Array CHG were still developing and medical practice was still habituating to the methods. This means that during the first years, analysed in this study, some VCFS patients may have influenced the results. If this is the case, the steepness of the regressionline of Figure 3, 5 and 6 (Chapter 3) would have increased even faster.

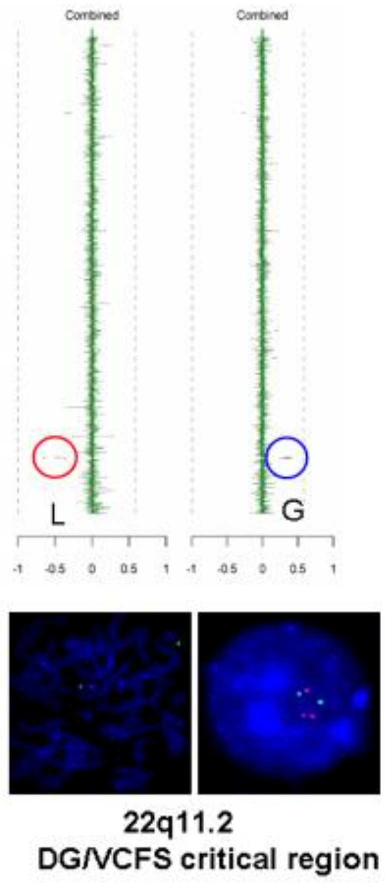

Figure 12. Example of array CHG ratio plots with FISH validation. The plot shows reciprocal microdeletion and microduplication at DGS/VCFS critical regions on chromosome 22q11.2. The vertical lines represent the clones interrogating targeted regions of the genome, aligned according to their chromosomal position with the signal intensity ratios. Gains (G) are to the right and losses (L) to the left. The red circle highlight the signal ratio revealing genomic losses whereas the blue circle indicates copy number gains. The FISH validation results are displayed underneath. Control probes were labeled with a green chromophore and clones detecting deletions or duplications were labeled in red for DG/VCFS deletion/duplication (Copied from Lu et al. 2007).

Role of parents

For children to achieve appropriate communication, it is necessary that they live with someone with the mode of interaction capable of encouraging them to naturally learn how to communicate and adopt the adult social model (Stern 1985). Children and their parents are involved in predictable daily routines and enhance their communications skills in these contexts. Speech development is influenced by the quantity and quality of the social interactions in which the child participate (Pamplona et al. 2001). Especially parents of children with communication disorders, such as a cleft palate, should adopt strategies to encourage their child's speech and language development. The acceptance of speech and language therapy for children with speech and language delays by the parents and the child is an important factor for the effectiveness of therapy (Keilmann et all. 2004). The involvement of parents in the delivery of treatment is increasingly sought by professionals. According to John (1998), in recent years there has been a shift in the provision of health care to take greater account of the views of health care users. She concluded that there is a need to take note of the view of the clients/ patients and their caretakers and where practicable to tailor therapy to meet their needs (John 1998). According to Pamplona et al (1996, 2001, 2001) parents should be included in the speech therapy in order to adopt the proper communication strategies to effect the speech development of their child in a positive way. It was suggested that the positive outcome of the treatment of articulation disorders during a summer camp (Pamplona et al. 2004) was caused by the active role of the parents of the cleft palate children. Jobe et al. (2008) found that training of parents to support speech therapy at home has a very positive influence on the speech development of cleft palate children. Finally Al-Sadhan et al. (2008) concluded that cleft palate children and their parents have very similar attitudes and concerns about speech problems and speech therapy. These outcomes suggest that the parents of the cleft palate children could be of great help in the speech- language therapy when they are included in the therapy sessions and instructed for training at home.

Good cooperation between parents and therapists is of enormous importance as parents not only have to bring their children to therapy regularly, they often participate in the therapy doing exercises with their child at home and modify their everyday way of using language to optimize the circumstances for speech and language learning (Keilmann et all. 2004). This is time consuming and parents need to facture their every-day routine around it. As seen in Table 8, the number employed women in the Netherlands increased swiftly over the period between 1996 and 2005. At the same time the number of employed men remained stable. It is reasonable to assume that the same increase applies for the period between 1994 and 2004.

Gender	1996	2005	Increase (%)
Men	76.7%	77.7%	1.0%
Women	49.9%	58.4%	8.6%

Table 8; Percentage of emloyed men and women in the Netherlands in 1996 and 2005 (Based on CBS Arbeidsdeelname en werkeloosheid.).

At the same time increased the number of divorces between 1980 and 2008 with 20.17% (CBS huwelijksontbindingen) and the number of single parents increased between 1995 and 2005 with 22.77% (CBS huishoudens). Households with both parents working and single parent households have become more common in the period between 1994 and 2004. It is plausible to suggest that these social changes are of negative influence on the role parents play in the speech language- therapy, during therapy as well as at home. This may effect the outcome of the ENT and speech-language therapy of cleft palate children, as these patients are not fully supported in their learning process.

Motivation

Another aspect of ENT and speech-language therapy might be influencing the results. Even when there is sufficient time, the parents need to be motivated to participate in the therapy and committed to carry out intructions and practice exercises with their child. Without speech-language therapy at least twice a week and regular practice twice a day it is difficult for a patient to reach a sufficient articulation level in which no errors associated with cleft palate occur. But it is of the utmost importance that the patient is impelled to improve his speech and language. Without a motivated patient who consideres a positive outcome of the treatment important and is willing to work for it, all favorable cincumstances are useless. There may have been some changes in the attitude of patients and parents towards ENT and speech-language therapy over the past few years. This is, however, difficult to prove, therefore future studies are needed to more closely define the impact of the motivation of the patient and the parents on the succes of the ENT and speech-language therapy.

5. Conclusion

To discover whether nowadays cleft palate children reach their full articulation potential at later age than before, the following H0 hypothesis was rejected in favor of the H1 hypothesis:

H0: Between 1994 and 2004 all cleft palate children reached their optimal articulation level at the same age.

H1: Between 1994 and 2004 a trend is grown that cleft palate children reach their optimal articulation level at older age.

During the period between 1994 and 2004 cleft palate patients, treated by the VUMC craniofacial team tend to reach their optimal articulation level at older age. From 1994 on, the percentage of patients, who are older than 6 years (84 months) at the moment of discharge from ENT and speech-language therapy, increases. This means that the archievement of the goal of the craniofacial team of the VUMC, to make sure the patient reaches an optimal articulation level at the age of 6, decreases over the years. None of the variables ‘overt or submucous cleft’, ‘monolingual/bilingual’, ‘indication for pharyngoplasty’ and ‘registration at older age’ could explain this increase of age at moment of discharge. Several factors, which may be of influence on this development, were perviously highlighted in the discussion. Apart from the increase of immigrants, the lack of time and possibilities, lack of motivation from both the patients as the parents may be of great impact on the outcome of the treatment. Future studies are needed to more closely define the impact of the motivation of the patient and the parents on the succes of the ENT and speech-language therapy.

References

Abdel-Aziz, M. Treatment of submucous cleft palate by pharyngeal flap as a primary procedure. International Journal of Pediatric Otorhinolaryngology, 71(3) (2007), 1093-1097.

Al-Sadhan, R., Al-Motlaq, I., Kapala, J., Attitudes and concerns of parents of cleft palate children about speech problems and therapy in their children. Pakistan Oral & Dental Journal, 28(1) (2008), 9-16

American Cleft Palate-Craniofacial Association. Parameters for the evaluation and treatment of patients with cleft lip/palate or other craniofacial anomalies. Revised Edition. Chapel Hill: American Cleft Palate-Craniofacial Association, 2004.

Ardinger, H.H., Ardinger, R.H., Clinical presentation of velo-cardio-facial syndrome. *Progress in pediatric cardiology*, 15(2) (2002), 93-97.

Beaumont, D. A study into weight gain in infants with cleft lip/palate. Paediatric nursing, 20(6) (2008), 20-24.

Beers, M. The phonology of normally developing and language-impaired children. Dissertation, University of Amsterdam, Amsterdam, 1995.

Bishop, R., Applications of fluorescence in situ hybridization (FISH) in detecting genetic aberrations of medical significance. *Bioscience Horizons*, 1(3) (2010), 85- 95.

Bley-Vroman, R., The logical problem of foreign language learning. Linguistic Analysis, 20 (1990), 3-49.

Bluestone, C.D. Pathogenesis of otitis media: role of Eustachian tube. Pediatric Infectious Disease Journal, 15(4) (1996), 281-291.

Bosh, L., Sebastián-Gallés, N., Simultaneous bilingualism and the perception of a language-specific vowel contrast in the first year of life. Language and Speech 46 (2-3) (2003), 217-243.

Brasileiro, I., The effects of bilingualism on children's perception of speech sounds. Dissertation, Utrecht, LOT 2009.

Brunnegard, K., Lohmander, A. A cross-sectional study of speech in 10-year-old children with cleft palate: results and issues of rater reliability. Cleft Palate Craniofacial Journal, 44 (1)(2007) 33–44.

Calzolari, E., Rubini, A., Neville, A.J., Bianchi, F. Special Report: EUROCAT and Orofacial Clefts: The Epidemiology of Orofacial Clefts in 30 European Regions. WHO International Collaborative Research on Craniofacial anomalies Meeting in Geneva, 2000.

Chapman, K.L., Hardin-Jones, M., Schulte, J. Vocal Development of 9-Month-Old Babies With Cleft Palate. Journal of speech, language and hearing research, 44(6) (2001), 1268-1284.

Cheung, S.W., Shaw, C.A., Yu, W., Li, J., Ou, Z., Patel, A., Yatsenko, S.A., Cooper, M.L., Furman, P., Stankiewicz, P., Lupski, J.R., Chinault, A.C., Beaudet, A.L., Development and validation of a CGH microarray for clinical cytogenetic diagnosis. Genetics in Medicine, 7(6) (2005), p 422-32.

Christensen, K. The 20th century Danish facial cleft population-Epidemiological and genetic-Epidemiological Studies. Cleft Palate Craniofacial Journal, 36(2) (1999), 96-104.

Daal, J., van, Verhoeven, L., Van Balkom, H. Behaviour problems in children with language impairment. Journal of child psychology and psychiatry and allied disciplines, 48(11) (2007), 1139-1148.

Damoiseaux, R.A.M.J., Rovers, M.M., Balen, F.A.M., van, Hoes, A.W., Melker, R.A., de, Long-term prognosis of acute otitis media in infancy: determinants of recurrent acute otitis media and persistent middle ear effusion. Family Practice, 23(8) (2005), 40-45.

Doyle, W.J., Cantekin, E.I., Bluestone, C.D. Eustachian tube function in cleft palate children. Annals of Otology, Rhinology and Laryngology, 89(68) (1980), 34–30.

Finneran, D.A., Francis, A.L., Leonard, L.B. Sustained Attention in Children With Specific Language Impairment (SLI). Journal of Speech, Language and Hearing Research, 52(4) (2009), 915-930.

Flynn, T., Moller, C., Jonsson, R., Lohmander, A. The high prevalence of otitis media with effusion in children with cleft lip and palate as compared to children without clefts. International journal of pediatric otorhinolaryngology, 73(10) (2009). 1441-1446.

Fuchs, M., Gugsch, C., Dannhauer, K. Evaluation of the Progress of Therapy in Patients with Cleft Lip, Jaw and Palate, Using Voice Analysis – a Pilot Study. Journal of Orofacial Orthopedics, 69(4) (2008), 257-267.

Fujiki, M, Brinton, B., Isaacson, T., Summers, C. Social behaviors of children with language impairment on the playground: A pilot study. Language, Speech and Hearing Services in Schools, 32(2) (2001), 101-113.

Gillis, S., Schaerlaekens, A. Kindertaalverwerving. Een handboek voor het Nederlands. Groningen, Nijhoff, 2000.

Goorhuis-Brouwer, S. De ontwikkeling van het praten. Rotterdam, Kooyker Wetenschappelijke Uitgeverij, 1978.

Goorhuis-Brouwer, S. Taalontwikkeling en taalstimulering van peuters en kleuters. Amsterdam, SWP Amsterdam, 2007.

Goorhuis-Brouwer, S.M., Schaerlaekens, A.M. Handboek Taalontwikkeling, Taal- pathologie en Taal therapie bij Nederlandssprekende Kinderen, Utrecht, De Tijdstroom, 2000.

Hart, K.I., Fujiki, M., Brinton, B. The Relationship Between Social Behavior and Severity of Language Impairment. Journal of Speech, Language and Hearing Research, 47(3) (2004) 647-663.

Harville, E.W., Wilcox, A.J., Terje Lie, R., Abyholm, F., Vindenes, H. Epidemiology of cleft palate alone and cleft palate with accompanying defects. European Journal of Epidemiology, 22(5) (2007), 389–395.

Henningson, G., Kuehn, D.P., Sell, D., Sweeney, T., Trost-Cardamone, J.E., Whitehill, T.L. Universal Parameters for Reporting Speech Outcomes in Individuals with Cleft Palate. Cleft Palate- Craniofacial Journal, 45(1) (2008), 1-17.

Jansonius-Schultheiss, K. Twee jaar taal en spraak bij schisis. Dissertation, Universiteit van Amsterdam, Amsterdam, 1999.

Johar, A.A., Ravichandran, K. Subhani, S.N., Jarba, E.A. Cleft Lip/ Palate and Craniofacial Anomalies, 1999-2005. King Faisal Specialist Hospital and Research Centre, Riyadh, Saudi Arabia. 2006.

Jobe, A.L., D'Mello, J., Kumar, S., Speech understandability of repaired cleft palate patients pre and post caregiver training. Indian Journal of Plastic Surgery, 40(2) (2008), 122-128

John, A., Measuring client and carer perspectives. International Journal of Language and Communication Disorders,33(suppl) (1998), 132–137.

Johnson, C.J., Beitchman, J.H., Brownlie, E.B. Twenty-Year Follow-Up of Children With and Without Speech-Language Impairments: Family, Educational, Occupational, and Quality of Life Outcomes. American journal of speech-language pathology, 19(1) (2010), 51-66.

Johnson, J.S., Newport, E.L., Critical period effects in second language learning: The influence of maturational state on the acquisition of English as a second language. Cognitive Psychology, 21(1) (2004), 60-99.

Jones, M.C. Etiology of facial clefts: Prospective evaluation of 428 patients. Cleft Palate Journal, 25(1) (1988), 16-20.

Karmiloff-Smith, A. Beyond modularity: A developmental perspective on cognitive science. Cambridge, the MIT Press, 1992.

Keilmann, A., Braun, L., Napiontek, U., Emotional Satisfaction of Parents and Speech-Language Therapists with Outcome of Training Intervention in Children with Speech and Language Disorders. Folia Phoniatrica Logopaedica, 56 (2004), 51–61.

Keuhn, D.P., Moller, K.T. Speech and Language Issues in the Cleft Palate Population: The State of the Art. Cleft Palate–Craniofacial Journal, 37(4) (2000), 348- 383.

Kobrynski, L.J., Sullivan, K.E., Velocardiofacial syndrome, DiGeorge syndrome: the chromosome 22q11.2 deletion syndromes. Lancet (British ed.), 370(9596) (2007), p 1443-1452.

Kriens, O. LAHSHAL: An easy clinical system of cleft lip alveolus and palate documentation in Kriens O. (ed) Proceedings of the advanced workshop "What is a cleft?", Stuttgart G, Thieme 1989.

Law, J., Boyle, J., Harris, F., Harkness, A., Nye, C. Screening for speech and language delay: A systematic review of the literature. Health Technology and Assessment, 2(9) (1998), 1–183.

Lenneberg, E., In: Biological foundations of language, Wiley, New York, 1967

Lierde, K.M., van, Borsel, J., van, Cauwenberge, P., van,. Speech Patterns in Children with Velo-Cardio-Facial Syndrome. Two Case Studies. *Folia Phoniatr Logop* 53(4) (2001), 213-221.

Lohman-Agerskov, A., Soderpalm, E., Friede, H., Lilja, J. A Comparison of Babbling and Speech at Pre-Speech Level, 3, and 5 Years of Age in Children with Cleft Lip and Palate Treated with Delayed Hard Palate Closure. Folia Phoniatica et Logopaedica, 50(6) (1998), 320-334.

Lu, X., Shaw, C.A., Patel, A., Li, J., Cooper, M.L., Wells, W.R., Sullivan, C.M., Sahoo, T., Yatsenko, S.A., Bacino, C.A., Stankiewicz, P., Craig Chinault, C.O.A., Beaudet, A.L., Lupski, J.R., Cheung, S.W., Ward, P.A. Clinical Implementation of Chromosomal Microarray Analysis: Summary of 2513 Postnatal Cases. *PLoS ONE*, 2(3) (2007), 327-338.

Mackie, C., Dockrell, J.E. The Nature of Written Language Deficits in Children With SLI. Journal of speech, language and hearing research, 47(6) (2004), 1469, 1484.

Mavroudi, S., Rekopoulou, L., Papadopulos, N.A., Papadopoulos, M.A. Etiology, classification, diagnosis and treatment of cleft lip and palate. Hellenic Orthodontic Review, 10(2) (2007), 103-128.

McCabe, P.C. Social and behavioral correlates of preschoolers with specific language impairment. Psychology in the schools, 42(4) (2005), 373-388.

McCormack, J., McLeod, S., McAllister, L., Harrison, L.J. A systematic review of the association between childhood speech impairment and participation across the lifespan. International Journal of Speech-Language Pathology, 11(2) (2009), 155-170.

McLeod, S. Access to appropriate health care and education is a basic human right. In R. Body, L. McAllister (Eds.) Ethics in speech and language therapy. Chichester, UK, Wiley & Sons., 2009.

Murray, L., Arteche, A., Bingley, C., Hentges, F., Bishop, D.V.M., Dalton, L., Goodacre, T., Hill, J. The effect of cleft lip on socio-emotional functioning in school-aged children. Journal of Child Psychology and Psychiatry 51(1) (2010), 94–103.

O'Gara, M.M., Logemann, J.A., Rademaker, A.W. Phonetic features by babies with cleft lip and palate. Cleft Palate-Craniofacial Journal, 31(11) (1994),446–451.

Pamplona, M.C., Ysunza, A., Active participation of mothers during speech therapy improved language development of children with cleft palate. Scandinavian Journal of Plastic Reconstruction Hand Surgery, 34 (2001), 231–236.

Pamplona, M.C., Ysunza, A., Espinoza, J., A comparative trial of two modalities of speech intervention for compensatory articulation in cleft palate children, phonological approach versus articulatory approach. International Journal of Pediatric Otorhinolaryngology, 49 (1999), 21—26.

Pamplona, M.C., Ysunza, A., Jimenez-Murat, Y., Mothers of children with cleft palate undergoing speech intervention change communicative interaction. International Journal of Pediatric Otorhinolaryngology, 59 (2001), 173–179.

Pamplona, M.C., Ysunza, A., Uriostegui, U. (1996) Linguistic interaction: the active role of parents in speechtherapy for cleft palate patients. International Journal of Pediatric Otorhinolaryngology, 37 (1996), 17-27

Pamplona, M.C., Ysunza, A., Patiño, C., Ramırez, E., Drucker, M., Maźon, J.J., Speech summer camp for treating articulation disorders in cleft palate patients. International Journal of Pediatric Otorhinolaryngology, 69 (2004), 351—359.

Paradise, J.L., Bluestone, C.D, Felder, H. The Universality of otitis media in 50 infants with cleft palate. Pediatrics, 44(1) (1969), 35–42.

Pearson, B.Z., Fernández, S.C., Oller, D.K., Lexical Development in Bilingual Infants and Toddlers: Comparison to Monolingual Norms. Language Learning 43 (1993), 93-120.

Peterson-Falzone, S.K., Hardin-Jones, M.A., Karnell, M.P. Cleft Palate Speech. Missouri, Mosby, 2001.

Priester, G.H., Post, W.J., Goorhuis-Brouwer, S.M. Problems in speech sounds production in young children. An inventory study of the options of speech therapists. International Journal of Pediatric Otorhinolaryngology, 73(5) (2009), 1100-1104.

Proffit, W.R., Fields, H.W. Early Stages of Development. In: W.R. Proffit, H.W. Fields. Contemporary Orthodontics. Missouri, Mosby, 2000.

Richman, L., Millard, T. Cleft lip and palate: Longitudinal behaviour and relationships of cleft conditions to behaviour and achievement. Journal of Paediatric Psychology, 22(4) (1997), 487–494.

Roberts, J., Hunter, L., Gravel, J., Rosenfeld, R., Berman, S., Haggard, M., Hall, J., Lannon, C., Moore, D., Vernon-Feagans, L., Wallace, I., Otitis media and language learning: controversies and current research. Journal of Developmental and Behavioral Pediatrics, 25(2) (2004), 110-122.

Roberts, J., Rosenfeld, R., Zeisel, S. Otitis Media and Speech and Language: A Meta-analysis of Prospective Studies. Pediatrics, 113(3) (2004), 238-148.

Rollnick, B.R., Pruzansky, S. Genetic services at a center for craniofacial anomalies. Cleft Palate journal, 18(4) (1981), 304-313.

Rovers, M.M. The burden of otitis media. Vaccine, 26(7) (2008), G2-G4.

Rovers, M.M., Straatman, H., Ingels, K., Wilt, van der, G.J., Broek, van den, P., Zielhuis, G.A., The effect of ventilation tubes on language development in infants with otitis media with effusion: a randomized trial. Pediatrics, 106(3) (2000), E42.

Ruiter, J.S., Korsten-Meijer, A.G.W., Goorhuis-Brouwer, S.M. Communicative abilities in toddlers and in early school age children with cleft palate. International Journal of Pediatric Otorhinolaryngology, 73(5) (2009). 693-698.

Sanderse, C., Verweij, A., Beer, J., de, Wat zijn de verwachtingen voor de toekomst? In: Volksgezondheid Toekomst Verkenning, Nationaal Kompas Volksgezondheid. RIVM, Bilthoven, 2010.

Schuster, M., Paal, S., Reulbach, U., Strobel-Schwarthoff, K., Nkenke, E. Evaluation of Speech Disorders in Children with Cleft Lip and Palate. Journal of orofacial orthopedics, 66(4) (2005), 270-278.

Schuster, M., Vogt, B., Maier, A., Batliner, A., Noth, E., Nkenke, E., Eysholdt, U. Numeric quantification of intelligibility in schoolchildren with isolated and combined cleft palate. HNO, 55(11) (2007), 891-898.

Sheahan, P., Blayney, A.W., Sheanhan, J.N., Early, M.J. Sequelae of otitis media with effusion among children with cleft lip and/or cleft palate. Clinical Otolaryngology, 27(6) (2002), 494–500.

Silva, P.A., Williams, S., McGee, R. A longitudinal study of children with developmental language delay at age three: later intelligence, reading and behavioral problems. Developmental Medicine and Child Neurology, 29(5) (1987) 630—640.

Slabakova, R., The Fundamental Difference Hypothesis Twenty Years Later. Thematic issue of Studies in Second Language Acquisition, 31(2) 2009, 155-173.

Stal, S., Klebuc, M., Taylor, T.D. Algorithms for the treatment of cleft lip and palate, Clinics in Plastic Surgery, 25(4) (1998) 493-507.

Stes, R., Elen, R. Aspecten betreffende normatief onderzoek van de articulatie- ontwikkeling bij kinderen tussen 3 en 10 jaar. Omtrent logopedie 8. Herentals, Vlaamse Vereniging voor Logopedisten, 1991.

Stern, D., The interpersonal world of the infant: a view from psychoanalysis and developmental psychology. Basic Books, New York, 1985, 114- 125.

Testa, J.R., Dimatos, S.C., Greggio, B., Duarte, J.A. Evaluation of the Results and Complications of the Ventilation Tubes' Setting Surgery in Patients with Serous Otitis Media. Archives of International Otorrinolaringology, 14(1) (2010), 90-94.

Thomas, A.M. Chopra, S., Singh, N., Simratvir, M., Moghe, G. Syndromes associated with labiopalatine clefting: A report of three cases. Journal of Indian Society of Pedodontics and Preventive Dentistry, 26(2) (2008), 88-91.

Vantrappen, G., Rommel, N., Cremers, C.W.R.J., Devriendt, K., The velo-cardio-facial syndrome: - the otorhinolaryngeal manifestations and implications. *International journal of pediatric otorhinolaryngology,* 45(2) (1998), 133-141.

Venkatesh, R. Syndromes and anomalies associated with cleft. Indian Journal of plastic surgery, 42(3) (2009), 51-55.

Vrtička, K., Present-Day Importance of the Velocardiofacial Syndrome. Folia Phoniatrica et Logopaedica, 59(3) (2007), 141-146.

Sites:

CBS Bevolkingsstatistiek: http://www.cbs.nl/nl-NL/menu/home/default.htm
Last visited at: 04-06-2011

VUMC: www.vumc.nl/afdelingen/schisisteam
Last visited at: 12-02-2011.