THE IMPACT OF EAR AND HEARING PROBLEMS ON QUALITY OF LIFE IN PATIENTS WITH CLEFT LIP AND/OR PALATE

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1. ABSTRACT

**DUTCH**

**Inleiding:** Schisis is een aangeboren afwijking die zich kenmerkt door een spleet in de bovenlip, kaak en/of verhemelte. Deze aandoening gaat gepaard met een divers aantal gezondheidsproblemen op het gebied van spraak, voeding, gehoor, het gebit en eveneens sociale en psychologische problemen. De incidentie van middenoorproblemen bij kinderen met enkel een gespleten lip is niet hoger dan in de normale populatie zonder schisis. Echter, bij kinderen met een gespleten verhemelte komen middenoorproblemen, meer specifiek otitis media met effusie (OME), wel veel frequenter voor. Dit is te wijten aan een slecht functionerende buis van Eustachius. Een gekende complicatie van otitis media met effusie is het optreden van een conductief gehoorverlies welke de spraak- en taalontwikkeling van het kind negatief kan beïnvloeden. Het doel van deze studie is om te onderzochten in welke mate de oor- en gehoorproblemen een impact hebben op de levenskwaliteit bij kinderen met schisis.

**Method:** De studiepopulatie bestaat uit 77 niet-syndromale schisispatiënten van 6 t.e.m. 18 jaar die opgevolgd worden door het multidisciplinair schisissteam in het UZ Gent. Voor de opbouw van de studie werd zowel een kwalitatieve als kwantitatieve aanpak gehanteerd. Een nieuwe vragenlijst die specifiek peilt naar de impact van oor- en gehoorproblemen op de levenskwaliteit bij kinderen met schisis werd gegenereerd. De volgende domeinen van levenskwaliteit werden in rekening gebracht: communicatie, gehoorverlies, lichamelijke klachten, beperking van activiteiten en het sociaal-emotionele luik. Voor elk van deze topics werd de invloed van type schisis, leeftijdscategorie, geslacht, financiële status en opleidingsniveau van de ouders onderzocht.

**Resultaten:** De meerderheid van de kinderen met een verhemeltespleet (met/зоnder lipspleet) rapporteerde geen majeure psychosociale problemen ten gevolge van hun oor- en gehoorproblemen. Volgens de patiënten zelf is er weinig tot geen sprake van pesten, schaamte of verminderd zelfvertrouwen en de oorproblemen ondermijnen de dagelijkse activiteiten niet. Tevens geeft het merendeel aan een goede band te hebben met leeftijdgenoten. Het gehoorverlies heeft een grotere impact op de levenskwaliteit dan eventuele oorklachten zoals oorpijn, een vol gevoel in het oor of loopoor aangezien deze klachten eerder beperkt zijn. Veel schisispatiënten hebben moeite om duidelijk en verstaanbaar te spreken. Naar mening van de ouders, leiden deze communicatiemoeilijkheden bij 1 op 5 kinderen tot gedragsproblemen en sociaal isolement. Er is dus een discrepantie tussen de levenskwaliteit gerapporteerd door de kinderen enerzijds en hun ouders anderzijds. Wat betreft prestaties op school kunnen zowel
gehoorverlies als slaapproblemen ten gevolge van oorklachten de schoolresultaten negatief beïnvloeden.

**Conclusie:** De resultaten van deze studie benadrukken het belang van een goed functionerend schisisteam aangezien de problemen bij schisispatiënten variabel en complex zijn en tevens een grote zorg met zich mee brengen voor zowel het kind als zijn/haar familie. Om een beter beeld te krijgen over de impact van oor- en gehoorproblemen op de levenskwaliteit bij kinderen met schisis en bijgevolg de zorg te optimaliseren is er nood aan grote multicentrische studies waarbij deze thesis gebruikt kan worden als een pilootstudie.
Introduction: Cleft lip and/or palate (CLP) is associated with a large variety of health problems in terms of speech, feeding, hearing, dental anomalies, social and psychological issues. In children born with a cleft lip, we expect no more middle ear problems than in children without clefts. However, the incidence of middle ear problems - especially otitis media with effusion (OME) – is very high in children born with a cleft palate due to Eustachian tube dysfunction. The sequelae of OME, in particular conductive hearing loss, can negatively influence the speech and language development of the child. The goal of this thesis is to investigate to what extent these ear and hearing problems affect the quality of life in children with CLP.

Methods: The study population consists of 77 children from 6 to 18 years old with non-syndromic cleft lip and/or palate who are followed by the multidisciplinary orofacial cleft team at Ghent UH. The overall methodological approach consists of a combination of qualitative and quantitative research. In the absence of a standardized questionnaire on this subject, a new questionnaire was generated for this study. Following domains of quality of life are taken into account: communication, hearing loss, physical symptoms, limitation of activities and social-emotional impact. For each of these topics, the influence of type of cleft, age category, gender, financial status and level of education of the parents is examined.

Results: The majority of children with cleft palate ± lip does not suffer from major psychosocial problems as a result of their ear and hearing problems. No teasing, no limitation of activities, no shame, no impaired self-esteem and a good relationship with peers are reported by the children. The hearing loss has a greater impact on quality of life than ear complaints such as ear ache, ear fullness or ear discharge. Many children have difficulty speaking clearly and understandably. According to the parents, in 1 out of 5 children communication difficulties lead to behavioural problems and social isolation. So there is a discrepancy between the quality of life reported by the children and by their parents. Scholastic achievement is negatively influenced by two factors: hearing loss and sleeping problems due to ear complaints.

Conclusion: The results of this study emphasize the importance of a well-functioning multidisciplinary cleft team since the problems in patients with cleft lip/palate are variable and complex. To adequately comprehend the impact of ear and hearing problems on quality of life in children with CLP and improve medical care, large-scale multicentre studies are needed whereby this thesis can be used as a pilot study.
2. INTRODUCTION

Cleft lip and/or palate (CLP) affects 1 in 700 live births, making it the most common congenital defect in the head and neck region. The new-born baby with CLP is likely to have difficulty feeding but will also develop ear and hearing problems, speech problems, dental anomalies and may have associated social and psychological issues. Especially the presence of middle ear pathology is almost universal in children born with a cleft palate. Despite the fact that multiple studies have been conducted about the quality of life in children with cleft lip and palate, the specific impact of ear and hearing problems on the quality of life in these patients has not yet been investigated thoroughly.

2.1. Classification

Orofacial clefts represent a diverse group of disorders affecting the lips and oral cavity. When classifying this group of anomalies, a first important distinction is made on the presence or absence of additional malformations and/or syndromes. Orofacial clefts can occur as an isolated defect (non-syndromic) or can be associated with additional malformations. Hundreds of syndromes including clefts in a sequence (e.g. Pierre Robin sequence), clefts with multiple congenital anomalies (e.g. anomalies concerning the heart, brain, skin), clefts in a monogenic syndrome (e.g. Stickler’s syndrome) and clefts in a chromosomal aberration (e.g. 22q11 deletion syndrome or velocardiofacial syndrome) have been reported (1-5). Because of the complexity of this wide range of syndromes, this thesis will solely focus on the non-syndromic subgroup of patients.

Orofacial clefts are classified based on the portion of the face that is involved in the cleft (Figure 1). Orofacial clefts are often subdivided into two groups: cleft lip with or without cleft palate and isolated cleft palate in which the lip is not affected (6, 7). This subdivision is based on the fact that the aetiology of an isolated cleft palate is different from that of a combined cleft lip and palate, both genetically and embryologically (8). Cleft lip can occur on one side (unilateral) or on both sides (bilateral) (9). In the cleft palate group, distinction can be made between complete/incomplete and soft/hard palate clefts (10).

Besides the open forms of cleft palate, there exists a special kind of cleft known as submucous cleft palate. In this type of cleft, the bony and muscular abnormalities in the roof of the mouth are covered by a mucous membrane which makes it difficult to diagnose the cleft during physical examination (2).
Figure 1. Non-syndromic orofacial clefts
(A) Cleft lip and alveolus. (B) Cleft palate. (C) Incomplete unilateral cleft lip and palate. (D) Complete unilateral cleft lip and palate. (E) Complete bilateral cleft lip and palate.

2.2. Epidemiology
Clefts of the lip and/or palate belong to the most prevalent and well known birth defects (5, 7). Even more, cleft lip and/or palate is the most common congenital abnormality occurring in the craniofacial region. The overall prevalence of orofacial clefts is approximately 1 in 700 live births. The incidence of clefts varies between countries and racial or ethnic groups, as well with variability in environmental exposures and socioeconomic status. In general, the prevalence rate is highest in Asian and Amerindian populations (1/500) and lowest in African populations (1/2500). European populations have intermediate occurrence rates at about 1/1000 (5, 7, 9, 10).

The prevalence of orofacial clefts associated with syndromes and/or anomalies can differ among different populations. The reported incidence of associated malformations varies between different studies, resulting in inconsistent data. In general, the majority of orofacial clefts are considered to be non-syndromic (1, 7). Isolated cleft palate has been demonstrated to be more frequently associated with genetic syndromes and additional anomalies compared to combined cleft lip and palate (5, 8, 10). Within the CL±P group, the proportion of cases with additional malformations appears to be higher in the bilateral subgroup (10).

2.3. Pathogenesis and aetiology
Development of the lip and palate implies a complex series of events including cell migration, growth, differentiation, and apoptosis. These programmes require strict coordination (6). Clefts of the lip and palate result from a lack of fusion of the frontonasal and maxillary processes during embryogenesis (4, 6). The reason why these embryonic parts fail to fuse is complex and multifactorial. Most cases of CLP are caused by the interaction between environmental and genetic factors during the first weeks of pregnancy. The presence of at least one other affected family member strongly suggests that genetic factors are involved, while sporadic cases with no family history are likely to be more associated with environmental risks (4, 5, 7).
2.3.1. Genetic factors

Genetic factors include abnormal gene variants that are inherited from the father and/or mother. These aberrant genes can contribute to the aetiology of orofacial clefts in two ways. On the one hand, they may be directly responsible for the CLP, as is the case for most syndromic CLP. In some syndromes the underlying genetic aetiology has not yet been identified. On the other hand these genetic factors may create a susceptibility to an increased risk of developing a cleft (multifactorial aetiology). When environmental factors interact with a genetically susceptible genotype, a cleft may develop during embryogenesis (5). The importance of genetic susceptibility is also suggested by the predilection of left-sided clefts and the female excess of isolated cleft palate (6, 7). As the knowledge of genetic aetiology continues to grow, it will be important to distinguish between the sub-phenotypes and to attempt to correlate genotype and phenotype (10).

2.3.2. Lifestyle and environmental risk factors

Orofacial clefts are known to be influenced by environmental risk. Especially maternal risk factors are of great importance. Maternal smoking, maternal alcohol consumption and medication use in pregnancy such as corticosteroids and antiepileptic drugs have been linked consistently with increased risk of orofacial clefts. Maternal illness, obesity and diabetes may also elevate the chance of CLP. Other factors that might be associated with increased risk of orofacial clefts concern maternal nutrition, including poor vitamin B6 status, zinc and folate deficiency, but these associations are uncertain or inconsistent (5-7).

2.4. Examination and diagnosis

Orofacial clefts can be diagnosed prenatally by ultrasonography but sensitivity is low, particularly for cleft palate. Genetic testing could improve sensitivity and specificity of prenatal diagnosis for syndromic and non-syndromic orofacial clefts. Most cases of cleft lip and cleft palate are noticed immediately at birth by physical examination of the mouth, nose and palate and don't require special tests for diagnosis. However, sometimes certain types of cleft palate for example a submucous cleft of the soft palate might not be diagnosed until later in life (6).

2.5. Clinical manifestations

Orofacial clefts are associated with a variety of health complications including speech problems, difficulty feeding, hearing loss, dental anomalies and may have associated social and psychological issues (5, 6, 11). Although recovery is possible with good quality care, orofacial clefts inevitably pose a burden to the individual, the family and society (6). Because this thesis investigates the impact of middle ear problems on quality of life, mainly the middle ear problems will be further discussed.
2.6. **Hearing and middle ear status**

Middle ear problems and hearing loss are well-known complications in cleft palate patients, however the importance of this problem is not always acknowledged. Especially otitis media with effusion (OME), the accumulation of non-purulent fluid in the middle ear space, is common amongst infants with an unrepaired cleft palate. Surgical cleft palate repair reduces the occurrence of OME, but many children continue to suffer from middle ear problems through childhood and even into early adulthood (12-14).

It is recommended to perform otologic and audiologic tests as soon as possible after birth. Regular appointments with an ENT (ear, nose and throat) specialist should follow (even after the palate has been repaired) so that any hearing loss can be treated properly. It is of great importance that these children are followed up from birth until adulthood by specialized cleft palate teams (9, 12, 14-17).

2.6.1. **Epidemiology**

Otitis media with effusion is a common disease of early childhood (16, 18). In the overall population, OME has been reported to occur in about 50% of children during the first year of life, increasing to about 60% by the age of two years. In children born with cleft lip only, we expect no more ear problems than in children without clefts. However, children born with a cleft palate do experience more ear problems. 90% of them will develop OME before their first birthday, increasing to 97% by the age of two years. Children born with a cleft palate are also more susceptible to recurrent OME in comparison to healthy children. More specifically, about 50% of cleft palate children will suffer from recurrent or persistent otitis media and have permanent conductive hearing loss. In comparison, the prevalence of conductive hearing loss is about 12.9% among young children without palatal clefts (14, 16, 17, 19).

2.6.2. **Middle ear anatomy**

The middle ear cavity is located in the mastoid process of the temporal bone. It is an air-filled space located between the tympanic membrane and the inner ear. Inside the middle ear, three small bones or ossicles (the malleus, incus and stapes) form a chain and transmit sound vibrations from the eardrum to the inner ear. The middle ear cavity is connected with the nasopharynx via a partly cartilaginous and partly bony tube, called the Eustachian tube(14). From the middle ear the Eustachian tube runs anteriorly, inferiorly and medially towards the nasopharynx. The tube opens in the nasopharynx just below the posterior end of the inferior turbinate. The Eustachian tube acts as an air pressure equalizer and permits aeration of the middle ear. In the normal resting phase the Eustachian tube is closed, but opens widely during yawning, chewing and swallowing. When the Eustachian tube opens, it ventilates the middle
ear, releases mucus and equalizes the air pressure between the outer and middle ear, which facilitates the transmission of sound through the eardrum (12, 14).

2.6.3. Pathogenesis of OME

Multiple factors cause OME in children with a cleft palate. Because of the important function of the Eustachian tube (i.e., ventilation, protection and clearance) the high incidence of middle ear problems in children with a cleft palate is often ascribed to Eustachian tube dysfunction. Pressure differentials are created by atmospheric pressure changes in the environment (e.g., during a plane landing) or by gaseous absorption by the mucous membrane of the middle ear (12, 15, 17). In cleft palate patients, the Eustachian tube is unable to equalize pressure and drain secretions. This lack of ventilation of the middle ear cavity causes a negative pressure inside the middle ear and retraction of the tympanic membrane. Mucous secretions are formed through osmosis and a collection of fluid becomes trapped inside the middle ear cavity. Eventually, this can result in OME, defined by the presence of middle ear effusion (3, 12, 13, 15, 18) (Figure 2).

![Figure 2. Eustachian tube function](https://healthwise.org/assets/images/ear Anatomy 02 01.png)

2.6.3.1. Anatomical and mechanical factor

In general, the Eustachian tube of children is not fully developed yet with the opening to the nasopharynx being narrower. Therefore, the narrow opening of the Eustachian tube is easily obstructed when upper respiratory tract infection causes swelling and inflammation of the respiratory mucosa. This results in negative pressure in the middle ear (17).

Specifically in cleft palate patients, the anatomic and structural defects can negatively affect the velopharyngeal function. Due to velopharyngeal insufficiency, the abnormal reflux of food
and fluid into the nasal cavity can set up chronic inflammatory changes around the Eustachian orifices with oedema and hypertrophy of the adenoid pads. This leads to obstruction of the Eustachian tube and secondary middle ear disease. In addition, the absence of a normal mechanical barrier between the oral and nasal cavity changes the bacterial flora of the region. Pathogenic bacteria may overgrow and pass easily to the middle ear cavity resulting in middle ear infection with effusion (12, 14, 17).

### 2.6.3.2. Dynamic factor

The dynamic factor in middle ear physiology depends upon the intact anatomy of the Eustachian tube and its extrinsic musculature. Tubal opening results from a synergistic action between the tensor veli palatini and levator veli palatini muscles. Several studies (12, 14) confirm the important role of the tensor veli palatini muscle as primary opener of the Eustachian tube. Thereby it has a key role in middle ear ventilation and drainage of the Eustachian tubes. The levator veli palatini muscle is also involved in tubal opening but rather at a different stage that precedes the actual opening.

The tensor veli palatini and levator veli palatini muscles are not intact in children with cleft palate. The muscle fibres do not have a normal course and midline palatal insertion, resulting in a lack of anchorage and the absence of a firm attachment of the muscles to the Eustachian tube in cleft palate patients. Thereby the regular opening function of the Eustachian tube is compromised (12, 14, 15, 17).

### 2.6.4. Clinical manifestations of OME

OME is not only more common in children with cleft palate, but middle ear effusions in these children are also likely to occur at an earlier age and be more persistent. Complications of OME include tympanic membrane retraction, tympanosclerosis, cholesteatoma, chronic otitis media and conductive hearing loss. Often these chronic middle ear problems persist into adulthood (8, 16, 18).

These sequelae of OME can in turn lead to delays in children’s speech and language development. Many studies indicate that even after surgery for CLP, language development depends on the extent by which hearing ability is maintained. So if OME is not treated right, long-term hearing loss can negatively influence the speech and language development of children (17). Note that, regardless of the middle ear problems, children with a cleft palate are at an immediate disadvantage for their speech development due to the anatomical aberrant connection between oral and nasal cavities, called velopharyngeal insufficiency. If the velopharyngeal sphincter does not close appropriately during speech, air escapes into the nose producing nasal air escape and hypernasal speech (2, 20).
2.6.4.1. Hearing loss

In general, hearing loss can be categorized by which part of the auditory system is damaged. The three main types of hearing loss are: conductive, sensorineural, and mixed hearing loss (Figure 3). Conductive hearing loss occurs when sound is not transmitted efficiently through the outer ear canal to the eardrum and the ossicles of the middle ear. Sensorineural hearing loss occurs when there is damage to cochlear hair cell function (‘sensory’) or damage to the cochlear nerve (‘neural’). Mixed hearing loss refers to a combination of both conductive and sensorineural hearing loss.

![Figure 3. Types of hearing loss: conductive, sensorineural and mixed hearing loss](image)

The most common type of hearing loss in children with cleft palate is conductive hearing loss caused by OME and its complications (18). It concerns a mild to moderate conductive hearing loss of about 25 to 30 dB, which is mostly temporary, variable in degree and frequently recurrent. It can affect one or both ears but it occurs usually bilateral. The hearing loss associated with OME is more severe in children with cleft palate than in children without cleft palate due to the higher viscosity of the middle ear secretions in cleft palate patients. Thicker middle ear secretions transduce sound less efficiently compared to the thinner secretions in children without clefts. Moreover, children with cleft palate have a 100- to 200-fold higher probability of developing cholesteatoma than children without CLP (13, 15, 17, 21).

Less frequently a sensorineural hearing loss occurs as a sequel of OME in non-syndromic cleft palate when the toxins produced by long-term inflammation pass through the round or oval window into the inner ear (17). Otherwise, syndromic cases of cleft palate, for example Stickler
syndrome, are more often associated with a sensorineural hearing loss. In contrast to most cases of conductive hearing loss, this type of hearing loss is not always reversible (5).

The hearing loss associated with OME negatively affects speech recognition which may lead to communication problems (13). Especially during the learning phase in these infants, hearing loss may lead to delayed speech and language development and can have a significant negative effect on their school performance and (social) behaviour (12, 16).

2.6.4.2. Long-term otologic and audiologic outcomes

The prevalence of OME decreases with increasing age due to morphological changes in the Eustachian tube which result in an improved tubal function. In children without clefts, middle ear disease often improves significantly after the age of 2 years, although a peak of OME at the age of 4 years is possible. However these improvements are seen later (about 3 years later) and less consistently in children with cleft palate (3, 8). For example, approximately 15-24% of children with cleft palate still experience OME in adulthood (13).

As children with CLP become older, their hearing improves in the lower to mid-frequencies (500 Hz to 4000 Hz) but not in the high frequencies (6000 and 8000 Hz). Eventually normal hearing is mostly acquired between the age of 10 to 15 years old in all cleft types, except in cases of complications of previous middle ear problems (1, 3). Whether the improvement of hearing levels is due to medical interventions, simply a result of the natural course of OME or both, remains unanswered (18).

2.6.5. Treatment of OME

Because of the earlier age of onset, prolonged course and higher rate of recurrence and complications of OME children with cleft palate require special consideration and a different approach (22). In 2008 NICE (National Institute for Health and Care Excellence; UK) published guidelines on the assessment and treatment of otitis media with effusion in children with cleft palate (Figure 4). The persistence of bilateral OME and hearing loss should be confirmed over a period of 3 months of active observation before intervention is considered. The child's hearing should be retested at the end of this period. If OME had not improved after three months of ‘watchful waiting’, ventilation tubes or hearing aids are recommended (16, 17).
Ventilation tubes (grommets) have become a standard procedure in the treatment of OME in children with clefts (9, 18). Grommets are generally inserted on 2 occasions: therapeutic insertion for children with symptoms of OME and prophylactic at the time of palatoplasty/lip closure (14, 19). Ventilation tube insertion (VTI) can be beneficial for the recovery of hearing in children with cleft palate and OME because it creates ventilation of the middle ear by removing the secretions (9). Early VT placement in children with clefts may reduce the incidence of OME, reduce hearing loss and improve the long term clinical and audiologic outcomes (9, 19, 23). For this reason, some authors advocate a more aggressive approach to VT placement at a younger age, even in asymptomatic cleft children. But grommets do not
always result in long-term benefits and may also cause pathologic changes in the eardrum such as persistent eardrum perforation, eardrum retraction and tympanosclerosis (8, 18). Therefore, other authors question the efficacy of aggressive and prophylactic management of OME using VT due to the complications of VTI and the questionable benefit they might provide during development (8, 19, 22). Szabo et al. (22) suggested that the risks of complications from VTI in cleft palate patients are few and manageable if standard sized ear tubes are used, while long-lasting tubes are associated with a higher risk of retained tube, sclerosis, retraction or ear drum perforation.

Thus today’s subject of debate is whether VT should be placed prophylactically in patients with cleft palate or should be reserved for symptomatic patients such as those with otalgia, recurrent ear infections or symptomatic conductive hearing loss as a result of OME (8, 16).

2.6.5.2. Hearing aids (HA)

The current NICE guidelines suggest that hearing aids are offered as an alternative for ventilation tube insertion when surgery is contraindicated or not acceptable. Hearing aids avoid the need for surgery and the risk of damage to the ear, however, HA may have a great impact on a child’s psychosocial wellbeing (17).

2.7. Treatment

The treatment of orofacial clefts is a challenging task due to the variety of problems that are associated with it (18). Because of the complexity of the disorder, an expert team dedicated to cleft treatment is needed (5). Care for cleft patients generally includes many disciplines: nursing, maxillofacial surgery, plastic surgery, ENT specialists, speech therapy, audiology, counselling, psychosocial interventions, genetics, orthodontics and dentistry (5, 6). Successful treatment of children born with orofacial clefts consists of multidisciplinary surgical and nonsurgical treatments that are performed from birth to adulthood (9). The aim of all treatments for a child born with a cleft lip and/or palate is to allow the individual to achieve the best possible outcome in order to live a balanced and fulfilling life (2).

2.7.1. Surgical

There is no consensus about the optimal timing and sequence for the correction of orofacial clefts and there exists no universally accepted protocol. Cleft lip repair is commonly performed between 3-6 months of age, based on the ‘rule of 10s’: an age of at least 10 weeks, with a weight of 10 lb (4.5 kg) and haemoglobin level of 10 g/dl (24).

The timing of surgical repair of a cleft palate remains controversial but in order to obtain normal speech, the repair should be carried out in the first year of life (24). There are several surgical techniques for hard palate closure (e.g. von Langenbeck, Veau-Wardill-Kilner, two flap) and
soft palate closure (e.g. Furlow technique). The primary goal of each technique for hard palate closure is to ensure normal facial growth, while repair of the soft palate aims to normalize speech, swallowing and middle ear ventilation (9, 14, 20, 23).

Further operations to improve speech, correct jaw bone growth (bone grafting), orthognathic surgery and plastic surgery may all be required later in life (5).

2.7.2. Non-surgical

Besides surgical therapy, also non-surgical care is needed for each CLP patient. For the management of cleft speech disorders, it is essential that specialised speech and language therapists are involved to monitor and guide speech development. It should be remembered that an adequate velopharyngeal function is needed for therapy to be wholly successful (2, 20, 25). Because OME and hearing loss are common complications of orofacial clefts, careful audiologic and otologic assessment is necessary, as described above. Also, paediatric dental health is a mandatory part of early cleft care and later in life orthodontics may be needed to maximize dental health and appearance. At last, psychological therapy needs to be offered to children and their parents. Skilled psychologists may help dealing with any psychological issues or distress that are associated with cleft lip and palate (25).

2.8. Quality of life (QoL)

2.8.1. Orofacial clefts

Orofacial clefts and its treatment affect many domains related to a patient’s quality of life, including appearance, speech, social integration, self-image and physical and psychological functioning (26). Significant psychological and social burdens have been reported in relation to orofacial clefts. Children with orofacial clefts are confronted with aesthetic concerns, speech and hearing disabilities, and difficulty acquiring social skills. Problems of psychological well-being in cleft patients include lower self-concept, grade retention, low school achievement and low social competence which can lead to feelings of loneliness and social anxiety. Children and teenagers with orofacial clefts are more likely to feel like they look different, to be worried about what people think and experience bullying. In contrast, some studies have found that orofacial clefts have less of an impact on health related QoL in children and suggest that having a cleft palate/lip does not inevitably lead to psychosocial problems. These findings may be due to differences in sample size and/or study population, as well as the use of different questionnaires (11).

Despite the fact that multiple studies have been conducted about the quality of life in patients with orofacial clefts, there is still no comprehensive PRO (patient-reported outcomes) instrument that measures quality of life for CLP patients. This might reflect the complexity of
the disorder. As said, a lot of different domains of QoL are involved in cleft patients. The relative importance of these various domains might change over the period of a child’s development to adulthood (27). It is important that valid questionnaires are available to craniofacial surgeons because patient-reported outcome measures that include aesthetic results, speech, functionality, self-image, incorporation into society and quality of life would provide surgeons with a more comprehensive assessment of surgical outcomes (26).

2.8.2. OME

Otitis media is one of the most common disorders in childhood and may have a considerable impact on the quality of life of children and their parents. Children with OME may experience physical problems such as ear discomfort, difficulty in sleeping and eating and poor balance. Behavioural problems at school or nursery, as well as difficulties with hearing and speech are also common. Hearing loss may interfere with social communication resulting in emotional distress (28).

On the other hand, very little is known about the specific effects of middle ear problems on wellbeing and quality of life in patients with orofacial clefts. There is only one study that did research on this topic. Tierney et al. (29) focused on OME and the associated experiences of children with cleft palate and their parents. The study showed that the impact of OME spanned several areas of a child’s life and affected the wider family. Children with orofacial clefts and their parents reported a range of negative consequences associated with OME that could prevent the children from engaging fully with their social world, due to emotional distress, educational delay and communication problems.

2.9. Research objective

Despite the fact that the presence of middle ear problems is almost universal in children with cleft lip and palate (CLP), previous research has not thoroughly explored the views of children with CLP and their parents on living with hearing-related difficulties. It is important that their opinions are investigated because these problems inevitably pose a burden to the patient and its family, which is not life-threatening, but may cause considerable distress. The goal of this thesis is to investigate the impact of ear and hearing problems on the quality of life in children with cleft lip and palate by means of a quality of life questionnaire. Since there exists no cleft lip and palate-specific validated questionnaire to assess the impact of ear and hearing problems in these patients, a new questionnaire was generated for this study. In this way, we want to quantify the degree to which these ear and hearing problems impact the patient’s quality of life in a valid and reproducible way. The ultimate goal is to generate research-based knowledge to inform treatment decision making and to improve medical care for current and future patients.
3. METHODS

The Ethical Committee of Ghent University Hospital approved this study (Belgian registration number: B670201629264). The decision as to whether or not to take part in this study was completely voluntary. Participants were assured that all information is kept confidential. (Appendix I: opting-out information letter)

3.1. Literature research

An electronic bibliographic search was conducted to find relevant articles about orofacial clefts and more specific about ear and hearing problems and quality of life in these patients (Figure 5). The following search terms were used: orofacial clefts, cleft lip, cleft palate, quality of life, middle ear, hearing and otitis media. Searches were performed in the following databases: Pubmed, Embase and Google Scholar. Articles in other languages than English and articles with a publication date older than 15 years ago were excluded. Relevant articles were selected based on title and abstract. The impact factor of the journal and the number of citations of the article were also taken into account. Hence, an exception was made to also include an article from 1999 since it is highly-cited with great relevance to this study.

**Figure 5.** Flow diagram of literature search strategy
3.2. **Experimental research**

The overall methodological approach of this study consisted of a combination of qualitative and quantitative research. The qualitative part has provided insights into the problem and generated ideas for later quantitative research.

### 3.2.1. **Study population**

The severity and complexity of the defects in patients with cleft lip and/or palate requires treatment and care by a multidisciplinary cleft team. The multidisciplinary orofacial cleft team of Ghent UH (University Hospital) consists of dental care, orthodontics, otorhinolaryngology, head and neck surgery, oral and maxillofacial surgery, plastic surgery, speech therapy, genetics, nursing, psychology and social work. More than 500 patients (especially children, but also adolescents up to the age of 18 years and adults in case of specific problems) are followed by the orofacial cleft team in Ghent UH with approximately 35 new cleft patients every year. The target group for this study was retrieved out of this patient population. Each patient is invited on a regular basis for a joint consultation in Ghent UH. The patient will be seen by all the disciplines that are relevant for him/her at that moment. Such joint consultations take place every month.

Based on age and associated syndromes, the patient population was reduced to a number of 167 patients who were eligible for the study. The minimum age was set at 6 years, because younger children would not be able to understand and/or complete the questionnaire themselves. Due to the fact that the prevalence of ear and hearing problems in cleft patients differs remarkably between children and adults, it was decided to exclude patients older than 18 years (8). This group of children and adolescents was then subdivided into a group of patients of 6 to 11-year-olds (born in 2006-2011) and another group of 12 to 18-year-olds (born in 1999-2005) because in most cases the ear and hearing problems decrease with increasing age and may even disappear (if no complications occur). This trend can be explored in this study by comparing both patient groups with each other. Furthermore, syndromic clefts were excluded because the wide range of associated syndromes would complicate the interpretation of the results. Thus our study population consists of children from six to eighteen years old with isolated cleft lip and/or palate (CLP), followed by the multidisciplinary orofacial cleft team at Ghent UH.
3.2.2. **Questionnaire**

A systematic literature review was performed to ensure that there exists no cleft lip and palate-specific validated questionnaire to assess the impact of the ear and hearing problems in these patients. Only one other study of Tierney et al. has explored the experiences of children with cleft palate and their parents about daily life with otitis media with effusion (OME) and associated hearing loss. They investigated this on the basis of semi-structured interviews, which makes this study the first one to explore this subject by means of a questionnaire (29).

In order to create a comprehensive, valid and reliable health-related quality of life (HRQoL) questionnaire, the guidelines outlined by the Scientific Advisory Committee of the Medical Outcomes Trust were followed to the best extent possible (Figure 6) (26, 30, 31).

![Figure 6. HRQoL questionnaire development and validation](image)

3.2.2.1. **Conceptual model**

The first step was to develop a conceptual model consisting of a description of the concepts that the questionnaire is intended to assess, and, the relationship between these concepts. This model formed the conceptual basis of the content of the questionnaire. These concepts are all associated to middle ear problems in children with CLP and contribute to the quality of life of these patients. The following aspects were considered important: communication, hearing loss, physical symptoms, limitation of activities and social-emotional impact (Figure 7).
3.2.2.2. Item generation

Literature review along with expert opinion and patient interviews have provided an extensive pool of potential items for the first draft of the questionnaire and ensured that the most important areas are considered for inclusion. A thorough literature study about orofacial clefts was conducted to gain a better understanding of the birth defect and its associated symptoms, in particular the ear and hearing problems. Pre-existing standardised questionnaires were an important source of information. General health questionnaires as SF-36 (Short Form-36) and GHSI (Glasgow Health Status Inventory Questionnaire) may be reliable but they are not sensitive enough because they measure health-related quality of life in diverse patient populations. OM-6, OMO-22 and COMOT-15 are examples of questionnaires measuring quality of life in children with otitis media. These questionnaires provided relevant and valuable information, but were insufficient to use in this case because they are not sensitive to all of the cleft-specific domains. The otolaryngologists of Ghent University specialised in clefts were called upon to gather more knowledge about which aspects should be included in the questionnaire. These experts also reviewed the first draft of the questionnaire. Furthermore, the opinion of Professor Sara Willems, experienced researcher in health sciences at Ghent
University, was requested on the subject of questionnaire design. Based on her advice additional socio-economic determinants of health were added to the questionnaire.

Expert opinion and literature review are essential aspects of item generation, but also patient interviews are valuable sources of input for the creation of a HRQoL questionnaire. Patient interviews permit the identification of issues of most importance to patients that may not be considered significant by health care providers. During the monthly joint consultations with the multidisciplinary cleft team, 5 children and their parents were asked about the impact of the ear and hearing problems they might experience. Taking into account all information obtained from these different resources, a questionnaire measuring the impact of ear and hearing problems on quality of life in cleft children was drafted.

### 3.2.2.3. Item reduction

Item reduction is also a vital part of questionnaire development. It ensures that only statistically strong questions are included in the final measure. The first draft of the questionnaire was pretested (or piloted) on a small sample of patients by means of cognitive interviewing. Cognitive interviewing or cognitive testing is an evidence-based qualitative method specifically designed to detect difficulties that individuals may have with the questionnaire (32). This technique was applied to 6 children who visited Ghent UH for their annual consultation with the multidisciplinary cleft team. By observing the respondents while answering the survey questions, difficulties with the questionnaire were noticed and further discussed. In this way, items that were not (fully) understood by the respondents were detected. By conducting the cognitive interviewing in the development process, it was still possible to change and improve the survey questions. During this process, the questionnaire was revised several times until the final version was obtained (Appendix II: questionnaire).

### 3.2.3. Data collection

In order to determine which cleft patients were eligible for the study, patient information was consulted in the electronic health record. Data collection in this study was principally based on the use of questionnaires because it enables to collect data from a large number of patients quite quickly. The questionnaire used in this study was predominantly based on closed ended questions allowing the data to be analysed quantitatively for patterns and trends. After every particular topic the participants had the opportunity to provide comments and explain their answer in a free text box. The questionnaire was available as paper version and electronic version. The questionnaire was sent by post to each participant in paper version together with a stamped return envelope so that the patients could return the filled in questionnaire free of charge. The web link for the online survey was also sent along, so that the questionnaire could be completed online depending on the preference of the participant. In order to improve the
response rate a reminder email was sent with the kind request to fill in the questionnaire if this has not already been done. In this way a response rate of 43% (71 out of 167) was achieved. The patients who did not respond include illiterates, patients who don’t speak Dutch or patients who have recently moved. Other possible explanations for non-response are: lack of time, no incentive, excessive survey length and the absence of ear and hearing problems in children with isolated cleft lip. 5 patients refused to participate. Therefore, these data will not be used in this study.

3.2.4. Data processing

The statistical analysis was performed using IBM SPSS version 24 (IBM SPSS, Armonk, NY, USA). A database was set up in the SPSS system. For all the closed questions, data was coded and entered into the database. For the open-ended questions, all the specifications and remarks were extrapolated from the written questionnaire and transferred into the database. Both a descriptive and analytical approach was used in this study, with emphasis on the descriptive aspect.

3.2.4.1. Descriptive statistics

Firstly, the demographic characteristics of the participants in this study were reported. As mentioned before, the study aims to assess the impact of ear and hearing problems on quality of life by describing the 5 themes as presented in the conceptual model: physical symptoms, hearing loss, communication, limitation of activities and social-emotional. Frequency tables were requested for all variables to display counts and percentages for each distinct value. A scale of 0 to 10 was used to rate the socio-emotional aspects of quality of life whereby 0 equals ‘strongly disagree’, 5 corresponds to ‘neutral’ and 10 equals to ‘strongly agree’.

3.2.4.2. Statistical tests

A p-value lower than 0.05 was considered statistically significant meaning that the null hypothesis was rejected when p < 0.05 and not rejected when p > 0.05. All tests were two-tailed (two-sided). The appropriate test to explore the correlation between categorical variables is the chi-squared test when the following conditions are met: no more than 20% of the expected counts are less than 5 and all individual expected counts are 1 or greater. When these conditions were not met, the fisher's exact test was used instead. Since the questionnaire is not standardized, new variables and outcome measures needed to be defined. These outcome measures (social participation, school, hobbies (swimming), mental wellbeing and communication) were composed by combining multiple questions and calculating the mean score. Then, the mean scores were dichotomized into two categories: patient with problems versus patients without problems. In this way, the following hypotheses were
examined: correlation between type of cleft and the occurrence of ear problems, correlation between the age category and the number of ear infections in the past year, correlation between the side of the cleft and the side of the ear problems and finally it was examined whether the quality of life (the outcome measures) is influenced by gender, type of cleft, age category, financial status and level of education of the parents. Also, in order to substantiate the correlation between age and the occurrence of ear problems, a binary logistic regression analysis was conducted.
4. RESULTS

4.1. Study population

4.1.1. Demographic characteristics of the study population

Our initial study population consisted of 71 patients born with cleft lip and/or palate including 45 male (63.4%) and 26 female patients. As discussed in the methods section, it concerns children and adolescents aged 6 to 18 years with a mean age of 11.9 years (SD=3.9). Divided into the two age categories, this comes down to 31 patients (43.7%) in the group of 6 to 11-year-olds and 40 patients in the other group of 12 to 18-year-olds (Figure 8).

![Figure 8. Distribution of patients (n) by age (years)](image)

Five of the 71 (7.0%) participants were born in a country other than Belgium (Poland, China, Iraq, Kazakhstan and Thailand). Two people (2.8%) speak a language other than Dutch (Kurdish, Polish) and 3 people speak another foreign language besides Dutch (Arabic, French, Turkish) at home. In 12 cases, the parents have or had financial problems and in 4 cases medical treatment has been delayed sometimes for financial reasons. 50.0% of the fathers and 52.1% of the mothers had a higher education level (college of post-secondary education or university).

The study population can be classified into 7 groups according to the type and location of the cleft: left-sided unilateral cleft lip, right-sided unilateral cleft lip, bilateral cleft lip, cleft palate, left-sided cleft lip and palate, right-sided cleft lip and palate and bilateral cleft lip and palate (Table 1). Literature has shown that cleft lip with or without cleft palate is most typical in males, and isolated cleft palate is most frequent in females (6). In our study, this corresponds to 69.1%
of the cleft lip with or without cleft palate consisting of boys and 56.3% of isolated cleft palate consisting of girls. Among unilateral clefts, the left-sided clefts are reported to be more common than right-sided clefts (33). This is also the case in our study population. As defined in the methods section, all the cases are non-syndromic. 12 patients (16.9%) have indicated that the cleft lip and/or palate occurs within their family, meaning that there are other people in the family with clefts. Besides, 15 patients (21.1%) have other family members who also suffer from ear and hearing problems, predominantly age-related hearing loss (presbycusis).

Table 1. The number of patients (count and %) per type of cleft

<table>
<thead>
<tr>
<th>Type of Cleft</th>
<th>Frequency (n)</th>
<th>Percentage (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Left-sided unilateral cleft lip</td>
<td>13</td>
<td>18.3</td>
</tr>
<tr>
<td>Right-sided unilateral cleft lip</td>
<td>3</td>
<td>4.2</td>
</tr>
<tr>
<td>Bilateral cleft lip</td>
<td>0</td>
<td>0.0</td>
</tr>
<tr>
<td>Cleft palate</td>
<td>16</td>
<td>22.5</td>
</tr>
<tr>
<td>Left-sided cleft lip and palate</td>
<td>16</td>
<td>22.5</td>
</tr>
<tr>
<td>Right-sided cleft lip and palate</td>
<td>9</td>
<td>12.7</td>
</tr>
<tr>
<td>Bilateral cleft lip and palate</td>
<td>14</td>
<td>19.7</td>
</tr>
<tr>
<td>Total</td>
<td>71</td>
<td>100.0</td>
</tr>
</tbody>
</table>

4.1.2. Correlation between the type of cleft and the occurrence of ear problems

Literature has shown that the incidence of ear and hearing problems in patients with isolated cleft lip is the same as in the controlled population (without clefts). Otitis media with effusion (OME) may occur in children with isolated cleft lip, as in any other child, but not as a result of the cleft (34). Before proceeding to further analysis, we tested whether this finding is also applicable to our study population using a chi-squared test. Currently only one person, equal to 7.1% (95% CI 1.3 ; 31.5%), of all patients with an isolated cleft lip indicated that she suffers from ear and hearing problems. On the other hand, 67.9% (95% CI 54.5 ; 78.9%) of the other group patients with cleft palate ± cleft lip is experiencing problems with their ears and hearing.
For a significance level of $\alpha=0.05$ and 1 degree of freedom, the Pearson chi-squared value is 16,546 with a significance (p-value) < 0.001. This results in a highly significant difference, meaning that in this study ear problems are significantly less common in patients with isolated cleft lip in comparison with patients with cleft palate (with or without cleft lip) (Figure 9). This confirms what already has been shown in previous research. Thus, it stands to reason to exclude the cases of isolated cleft lip for further analysis.

![Figure 9](image). This bar chart shows the % of patients with ear problems and without ear problems in isolated cleft lip compared to cleft palate ± cleft lip

Due to this fact, the study population is reduced to 55 patients including 34 boys (61.8%) and 21 girls. The age varies between 6 and 18 years with a mean age of 12.2 years (SD=3.9). Subdivided into the two age categories, it comes down to 23 children from 6 to 11 years old (mean age=8.3 ; SD=1.69) and 32 adolescents from 12 to 18 years old (mean age=15.0 ; SD=2.1). 16 children (29.1%) were born with only a cleft palate, while the other 39 children had a cleft lip and palate. In these two groups, the prevalence of ear problems is almost evenly distributed, namely 68.8% in isolated cleft palate patients and 67.6% in combined cleft lip and palate patients. This finding substantiates the fact that the ear and hearing problems result from the cleft palate (and not the cleft lip) due to anatomical reasons (see introduction).

### 4.1.3. Comparison of the two age groups
We divided our study population into two groups (a group of patients of 6 to 11-year-olds and another group of 12 to 18-year-olds) to explore whether the ear problems decrease with increasing age. The parents of the participating patients were asked how many times their child had a middle ear infection in the past year, ranging from none to four or more. 56.5% (95% CI 36.8 ; 74.4%) of the 6 to 11-year-olds had none or 1 ear infections, compared to the other 43.5% (95% CI 25.6 ; 63.2%) who had 2 or more ear infections in the past year. These figures are remarkably different in the other age group of which merely 3.2% (95% CI 0.6 ; 16.2%) of the 12 to 18-year-olds had 2 or more ear infections in the past year compared to the other 96.8% (95% CI 83.8 ; 99.4) who had none or 1 ear infection. A Fisher’s Exact-test was used to explore the correlation between the age of the patient (6 to 11 years versus 12 to 18 years old) and the number of middle ear infections in the past year (none or 1 versus 2 or more). For a significance level of α=0.05, the Fisher’s exact value is 13.189 with a significance (p-value) < 0.001. This means that in this study population the number of middle ear infections is significantly lower in the older age group (Figure 10). As cited in the introduction section, this finding does not specifically apply to children born with clefts, but it is a general fact that the prevalence of OME decreases with increasing age due to morphological changes in the Eustachian tube which result in improved tubal function (3).

Figure 10. This bar chart shows the % of patients with none or 1 and 2 or more ear infections in the past year in 6 to 11-year-olds compared to 12 to 18-year-olds
Additionally, a binary logistic regression analysis was performed to examine to what extent the probability of ear problems in general is influenced by the age of the children in our study population. It shows that with increasing age the probability of ear problems decreases. More specifically, the odds of ear problems decreases with 9.0% (95% CI -6.5 ; 22.4) for each year the child gets older. This result applies to this study population, but it cannot be extended to the general population since it is not statistically significant (p=0.240).

4.1.4. Correlation between the side of the cleft and the side of the ear problems

Before proceeding to further analysis of quality of life in our study population - the main research goal - it was examined whether there is a correlation between the side of the cleft (left, right or both sides) and the side of the ear problems (left, right or both sides) in patients with combined cleft lip and palate. In these patients there is an overall impaired function of the palate but we expect greater ear and hearing problems in the ear alongside the cleft due to anatomical features concerning the Eustachian tube (see introduction).

In our study population of 55 participants, 17 patients (30.9%) didn’t have any ear problems at that time and among the other 38 patients the distribution of the ear problems according to localization is the following: 2 missing values, 6 left, 8 right and 22 on both sides. In all the isolated cleft palate patients the ear problems are located both left and right. In case of the combined lip and palate clefts, the correlation between the side of the cleft and the side of the ear problems was examined by means of a Fisher’s exact test. For a significance level of α=0.05 the Fisher’s exact value is 9.338 with a significance (p-value) =0.036. As shown in Figure 11, this means that in patients born with a right-sided cleft lip and palate the ear problems are also mainly on the right. The same applies to the left side and both sides.

![Figure 11](image_url)

**Figure 11.** This chart shows the % of patients with ear problems located on left, right and both sides according to the side of the cleft
4.2. Physical symptoms

86.8% of patients have ever had tympanostomy tubes (grommets) inserted into the eardrum but only a small minority had a bad experience with it. More specifically, they did not like the operation or the tubes had fallen out too early. The others did not mind having tympanostomy tubes.

According to their parents, 10 children (18.2%) suffered from one ear infection and 11 children (20.0%) suffered from two or more ear infections in the past year, meaning that more than half of the children in our study population had no ear infections in the past year. Following complaints were associated with the ear infection in descending incidence: ear ache, ear discharge, impaired hearing, fever and general discomfort. Two children had no complaints. Of the 21 patients who have experienced an ear infection, 19 patients (90.5%) visited a doctor. With the exception of one case, all doctors had prescribed medications: ear drops with almost every patient and to a lesser extent also painkillers and oral antibiotics.

In general, the majority of our patients had no ear-related physical complaints like ear ache, ear fullness or ear discharge during the past month. In the patients who did indicate that they had physical complaints, these symptoms usually occurred a few times a month, rarely more frequent.

4.3. Hearing loss

The patients were asked about everyday situations to investigate the impact of potential hearing loss on their quality of life. More than 90% of children experience no difficulty in hearing the doorbell, telephone, alarm clock or traffic. More difficulties arise when it comes to hearing what is said on television or radio and understanding the teacher but this remains limited to 26.0% and 18.6% of the patients, respectively. At the same time, some children are having considerable trouble understanding people in the following situations: when too many people talk at once (43.6%), when there is a lot of background noise e.g. on the playground or at restaurant (40.0%), when someone whispers (32.7%), when there is a larger distance between oneself and the person who is speaking (25.5%) and in the classroom (20.0%). 21 patients (38.2%) get to hear sometimes that the television volume is too high. One fourth of the children often needs to ask someone to repeat what he/she said and 18 children (32.7%) do not always react when someone talks to them. 3 out of 55 patients wear a hearing aid. None of them are ashamed of it or are laughed at it, but one child finds it bothersome that background noises are too loud.
4.4. **Social-emotional**

A scale of 0 to 10 was used to rate the socio-emotional aspects of quality of life whereby 0 equals ‘strongly disagree’, 5 corresponds to ‘neutral’ and 10 equals to ‘strongly agree’. The frequencies (%) described below are based on the cases who are considered problematic meaning they scored average more than 5/10. In general, the ear and hearing problems do not seem to have a major impact on the state of mind and social functioning of the children in this study. 2 children did indicate that they are less self-confident because of their ear and hearing problems. This feeling of embarrassment is a bit more prominent at school than when they are with friends or during their hobbies. Altogether, the impact on the self-confidence is limited and not in a way that it would prevent them from getting to know new people and making new friends. Also, it does not appear that the children in this study are being excluded by others. 7.2% of the patients sometimes feel sad because of the ear and hearing problems they have. If they have something on their mind, they can mostly talk about it with their friends but even more with their parents. 5.4% of the children are jealous of others who have no ear and hearing problems and feel different from other children making them feel angry or annoyed. The most significant finding is that 13.1% of the children in this study are more or less afraid that their ear and hearing problems will get worse in the future. To conclude, the lives of the children in this study are not remarkably different on account of their ear and hearing problems.

Notice that participants were asked to keep in mind that all their answers should be based on the underlying ear and hearing problems. So everything described above, should be related to the ear and hearing problems.

4.5. **Limitation of activities**

4.5.1. **School**

Children are not less motivated to go to school because of their ear and hearing problems but no less than 3 out of 4 children have already missed classes or activities at school because of it. The most common reasons for absence include appointments with an ENT-specialist, the (annual) consultation in Ghent UH with the multidisciplinary cleft team and also ear infections and the complaints that are associated with it. One mother suggested that it would be good for children to miss little to no classes at school because of doctor’s appointments because these children already constitute a vulnerable group and the more absent at school, the more vulnerable they become. No additional difficulties are seen with making homework or studying for a test. Meanwhile, due to the ear and hearing problems some children are having difficulties understanding the teacher (21.8%) and concentrating in class (12.7%). 1 out of 5 patients in
this study had to repeat a grade, especially in kindergarten and also the first year of primary school. None of them had to repeat a grade in secondary education. The same number of children needs or needed tutoring, for example for mathematics, Dutch or reading.

4.5.2. Leisure

The ear and hearing problems do not seem to influence social participation like meeting friends, attending birthday parties or going to hobbies, except for swimming. Nearly half of the participants in this study experienced problems with their ears while swimming, for example earache. Some children are afraid to get water in their ears and avoid swimming for that reason. Someone also mentioned the earache she experiences while partying. Other hobbies like youth movement, music school, playing an instrument, sports, playing videogames, listening to music or going to the cinema are not hampered by possible ear and hearing problems in this study. As mentioned above, a notable number of children get to hear sometimes that the television volume is too high and also in this case 8 children (14.5%) indicated that they had difficulty understanding what was said on television.

Participants were asked whether they cannot sleep at times because of earache. 9 out of the 55 children (16.4%) responded positively of which only one person described it as a significant burden. The child’s mother added that there has been trouble falling asleep for a long time due to earache and noises in the ears that are scary. However, with respect to the total study population, these sleeping problems remain limited and they do not significantly affect the functioning of the child at school (Fisher’s Exact p-value=0.628). The proportion of children with problems at school is 7.2% (95% CI -3.9 ; 18.3) higher in the group of children whose ear problems (ear ache) undermine their sleep, compared to the group of children who did not report to have sleeping problems. Since the confidence interval for the difference contains zero, it is confirmed once again that this finding may not be generalized.

4.6. Communication

As it is difficult for children to evaluate their own speech, we asked their parents about any issues with pronunciation and communication. Speech seems to be a problem for many children. According to their parents, 2 out of 3 children have a poor pronunciation and also 58.2% speaks more or less unclear. It is therefore not surprising that more than half of the patients are often misunderstood by others. Slow speech is also a mild problem in our study population, but to a much lesser extent. 48 out of 55 patients (87.3%) needed speech therapy. In addition, we also asked the parents if their children often misunderstand other people but in 80% this is not the case. It is also interesting to note that as a result of these communication problems about 20% of all children have, according to their parents, mild to moderate problems
concerning troubled, hyperactive or even aggressive behaviour and are isolating themselves from their environment.

4.7. Role of gender, type of cleft, age category, financial status and parental level of education

In the final part, it was examined to what extent the quality of life is influenced by type of cleft, age category, financial status and level of education of the parents. Following quality of life outcome variables were taken into account: mental wellbeing, school, hobbies (swimming) and communication. As already described above, the ear and hearing problems do not seem to influence social participation in this study population. Type of cleft makes a distinction between isolated cleft palate and combined cleft lip and palate. A group of patients of 6 to 11-year-olds and another group of 12 to 18-year-olds form the two known age categories. Financial status is defined by the fact if the parents have trouble making ends meet at the end of the month or not. The level of education is divided into three categories: low (primary education or lower secondary education), medium (higher secondary education) and high (higher education or university). To investigate the interrelationship between these variables, the Fisher’s Exact test was used (Appendix III: table of Fisher’s Exact test significance values).

4.7.1. Mental wellbeing

The performed statistical tests show that the mental wellbeing of the children in this study is not influenced by the type of cleft and neither by the financial status or parental level of education. There are only two children (equal to 3.8% with a 95% CI 1.1 ; 13.0) whose mental wellbeing is affected by their ear and hearing problems to a considerable degree (mean score >5 on a scale of 10) and these two children are both girls and both belong in the 6 to 11 years old age category. However, this finding is not statistically significant.

4.7.2. School

Problems at school like grade retention, need for tutoring and general problems (including difficulty concentrating in class, doing homework, studying for a test and understanding the teacher) do not seem to be influenced by type of cleft nor age category, nor financial status. We see that 17.0% (95% CI -2.1 ; 36.4) more boys need to repeat a grade compared to girls. Furthermore, there is a statistically significant correlation between grade retention and the level of education of the parents. More specifically, the lower the parental level of education in this study, the more likely their children have to repeat a grade. This was examined for both the mother and the father, by means of a Fisher’s Exact test with the corresponding significance values being p=0.012 (level of education of the mother) and p=0.002 (level of education of the father).
4.7.3. **Communication**

A higher proportion of children with combined cleft lip and palate needed speech therapy in comparison to the children with an isolated cleft palate. These proportions amount to 92.3% (95% CI 79.7 ; 97.3) in combined cleft lip and palate and 75.0% (95% CI 50.5 ; 89.8) in isolated cleft palate patients. Similar proportions apply to boys (94.1%) compared to girls (76.2%). So in our study population, the need for speech therapy is slightly higher in children with combined cleft lip and palate and male patients. But this is not confirmed by a statistically significant Fisher’s Exact test. Meanwhile, a statistically significant correlation ($p=0.047$) was found between the level of education of the mother and problems with speech. A post-hoc contingency table analysis using the Bonferroni correction showed that this significant result relates to the group of children whose mothers are low-educated. In this study population it means that there are less problems with speech in the group of children whose mothers are low-educated compared to middle- and high-educated mothers. In concrete figures, the percentage of children with speech problems are 14.3% (95% CI 2.6 ; 51.3), 70.0% (95% CI 48.1 ; 85.5) and 59.3% (95% CI 40.7 ; 75.5) respectively for a low, middle and high level education of the mother.

4.7.4. **Hobbies: swimming**

Neither gender, type of cleft, age category, financial status nor parental level of education have an influence on the occurrence of trouble with swimming.
5. DISCUSSION

5.1. Discussion of the results

Most children are able to have a happy youth and social life after reconstruction of their cleft. The results of this study suggest that the psychosocial well-being of children and adolescents with palatal clefts is not greatly affected by the ear and hearing problems they might experience. Adolescence is a time of change for everyone and sensitive issues will come up but in this study there are no major psychosocial issues related to the ear and hearing problems in children born with a palatal cleft. No teasing, no limitation of activities, no shame, no impaired self-esteem and a good relationship with peers were reported. However, it is important that if psychosocial problems arise in these children and adolescents, they can easily find professional help. Yet there are some difficulties as a result of possible ear and hearing problems in particular areas of functioning in children born with a palatal cleft. In this study, difficulties are seen in relation to physical health, hearing loss, school and communication.

5.1.1. Physical health and hearing loss

The hearing loss itself seems to have a greater impact on quality of life than the physical complaints (ear ache, ear fullness, ear discharge) since the latter were rather limited. More than half of the patients had no ear infections in the past year and the majority had no ear-related physical complaints at all during the past month. In certain situations like swimming, the physical complaints may increase. Ear problems (in particular ear ache) hindered the involvement in swimming in nearly half of the children. It is therefore advisable that children wear ear plugs at time when water could get into their ears.

Children experienced the hearing loss mainly when they were watching television and to a noticeably lesser extent upon hearing the doorbell, telephone, alarm clock or traffic. Situationally, the hearing loss was mainly manifest when too many people talk at once, when there is a lot of environmental noise and when someone whispers or there is a large distance between oneself and the person who is speaking. Children at school are exposed to a lot of environmental noises within the classroom. Research has shown that these classroom noises can negatively influence children’s performance at school, including reduced memory, motivation and reading ability (35). Linked to the high prevalence of conductive hearing loss in children with cleft palate, it is not surprising that school performance of some of these children is affected.

5.1.2. School

In this study, scholastic achievement seemed to be negatively influenced by two factors: hearing loss and sleeping problems due to ear complaints. In 1 out of 5 cases hearing
impairment led to difficulties in understanding the teacher resulting in difficulties concentrating in class in approximately 1 out of 8 children. Secondly, children with sleeping problems due to ear complaints had 7.2% more problems at school than children without sleeping problems. However, this finding is not statistically significant and may therefore not be generalized.

Concerning grade retention, 1 out of 5 children had to repeat a grade once. Since this cumulative rate relates to all children aged 6 to 18 years old, it is difficult to compare this figure with average figures in Flanders (Belgium) but on the basis of a careful assessment it does not seem that the number of repeaters is significantly higher in our study population than average (statistical yearbook 2016-2017 - Flemish Ministry of Education and Training). Also 1 out of 5 children needed tutoring, for example for mathematics, Dutch or reading. The ear and hearing problems and consequential difficulties in understanding the teacher and concentrating in class can indirectly contribute to this finding, however this is debatable.

3 out of 4 study participants have already missed classes or activities at school because of appointments with an ENT-specialist or ear complaints. The demands of ongoing medical care in children with orofacial clefts may remove them from normal peer-group activities and further accentuate their differences. In this study, one mother explicitly mentioned this issue but practically it is almost unfeasible to schedule all doctor appointments outside school hours.

5.1.3. Communication

The results of this study show that speech is a problem for many children. Good hearing is vital to the overall wellbeing of a child but hearing also plays an important role in speech and language development. The accumulation of fluid in the middle ear (OME) in children with palatal clefts can cause a conductive hearing loss which makes it more difficult to mimic the sounds of speech. If children are having an extremely difficult time hearing and understanding language, they may even start to ignore conversations. About 20% of individuals in this study exhibited self-isolating behaviour (internalizing behaviour) or otherwise troubled, hyperactive and even aggressive behaviour (externalizing behaviour) as a result of communication problems, according to their parents. The acting out behaviour can be explained by the theory that it is a reflection of frustrations while the self-isolating behaviour can represent a self-defence mechanism to minimize peer rejection as stated by Endriga et al (36). However, the children themselves have not reported any kind of major impaired social functioning or social withdrawal as a result of their ear and hearing problems.

It is not surprising that most of the patients followed speech therapy, since in Ghent UH speech therapy is usually recommended to all children born with palatal clefts. These figures, however, confirm that the need for speech therapy in these individuals is high. This issue emphasizes the importance of a well-functioning multidisciplinary cleft team in which the speech-language
pathologist can work closely with the audiologist and ENT-specialist in order to anticipate these communication problems as optimal as possible. Furthermore, it is important that parents stimulate their child's speech and language at home and speak clearly and at an appropriate volume.

5.1.4. Role of gender, type of cleft, age, financial status and level of education of the parents

A statistically significant correlation was found between the parental level of education and grade retention. More specifically the lower the level of education of the parents, the more likely their child had to repeat a grade with the father’s education level being more significant (p=0.002) than the mothers’ one (p=0.012). Literature (37, 38) usually describes it the other way around, namely that the mothers’ level of education has a greater influence than the fathers’ one. Secondly, in this study population children of middle- and high-educated mothers have statistically significant more problems with speech compared to children of low-educated mothers. This is contrary to what is found in literature (39), namely that a higher level of parental education is associated with better oral health related quality of life (OHRQoL) in children, including speech.

Furthermore, a number of trends were observed, but not to the extent that it led to a statistically significant result. 17% more boys (26.5%) needed to repeat a grade compared to girls (9.5%). It is generally the case in Flanders that more boys need to repeat a grade than girls (statistical yearbook 2016-2017 - Flemish Ministry of Education and Training). We also found that the need for speech therapy is slightly higher in children with combined cleft lip and palate compared to isolated cleft palate. The lips are, just like the palate, an important part of the speech apparatus (40).

As regards the psychosocial problems, the mental well-being of only two children was affected to a considerable degree (means score >5 on a scale of 10) and these two children were both girls who belong to the younger age category (6 to 11 years old). According to Pope et al. (41), young children and girls may be more emotionally vulnerable but it is difficult to give a ruling on the role of age and gender on mental well-being since it involves only two patients in this study.

5.2. Comparison with results available in the literature

Since the results of this study are based on a self-generated questionnaire, it is more difficult to compare our own findings with those from other studies who applied different research methods and with different patient populations.
5.2.1. **QoL in patients with cleft lip and/or palate in general**

As mentioned in the introduction section, orofacial clefts have an impact on several areas of a child’s life: physical, psychological and social health. Findings on whether CLP increases the risk of having a significant psychopathology have varied between studies but it is beyond dispute that there are psychological and social challenges among children and adolescents with orofacial clefts. It seems that it is not necessarily the presence of the cleft itself that causes psychosocial problems but the experiences that often accompany it, such as negative social responses from peers about their appearance and speech for instance (42). Teasing, difficulties with peer relationships, rejection, low self-esteem, anxiety and depression are often reported in children with CLP but are absent in this study meaning that these problems don’t seem to result from the ear and hearing problems. Medical and functional limitations as a result of the cleft lip/palate may hinder the involvement in sports or other types of activities but in our study this was only limited to problems with swimming. Poorer scholastic achievement, impaired social functioning and behavioural problems are reported both in patients with CLP in general and in this study. Social withdrawal was present in 20% according to the parents and resulted specifically from communication problems while the extent of the problem is presumably greater in patients with CLP in general since negative social responses to facial appearance may contribute as well to an impaired social functioning. The same applies to behavioural problems. Learning difficulties can occur because of hearing impairments but the association of CLP with other congenital problems or syndromes increases the risk for cognitive impairment (36). However, all cases in this study were non-syndromic.

Facial appearance is an important part of quality of life in patients with CLP in general which was not taken into account when examining the specific impact of the ear and hearing problems on quality of life in this study. Because of the facial differences, individuals with cleft palate/lip are susceptible to additional psychosocial problems especially those relating to self-esteem, peer relationships and appearance. They may develop negative self-perceptions and isolate themselves from peers. Especially in adolescence, a time wherein social interactions increase and physical attractiveness becomes an important characteristic. Research has shown that particularly girls are more likely to develop problems relating to self-concept and appearance, while adolescent boys typically deal with anxiousness-depression and aggressive behaviour. Age also appears to be an influencing factor. As children with clefts grow older (and their social interactions increase), they tend to report more dissatisfaction with peer relationships and higher levels of social anxiety (27, 43-45).
5.2.2. **QoL in patients with OME without clefts**

Otitis media with effusion is reported to have a considerable impact on various domains of quality of life in children. Research (46) found that physical symptoms like ear ache or discomfort, ear drainage and difficulty in sleeping are prominent in children with OME, especially in those who had ventilation tubes inserted. OME not only affects physical functioning but also the general well-being of a child and its family. Behavioural problems at nursery or school like attention deficits, as well as difficulties with hearing and speech were also common. Hearing loss is an important factor in regard to quality of life in patients with OME, since it can interfere with social communication, as is clear from our study. Learning difficulties can occur because of hearing impairment. Therefore it is important that the school is informed and the student is assisted within the learning environment. The impact on quality of life appears to depend upon the severity of the condition, but not upon the gender while this is the case in CLP in general (28).

The impact of OME on daily functions and social lives of children without CLP as described in the literature is similar to what was found in this study, except for behavioural problems that were less present in this study. Cleft palate is reported to place children at increased risk for speech, language or learning problems due to otitis media. Therefore, children with palatal clefts are considered high risk and require closer monitoring for OME and associated hearing loss (47).

5.3. **Reflection**

Very little research has been done on this topic making this study the first to investigate the specific impact of ear and hearing problems on quality of life in children with CLP by means of a questionnaire. The strength of this study lies in the combined quantitative and qualitative research approach. In the initial phase of this experimental research, a qualitative approach by means of patient interviews was used (in combination with literature research and expert opinion) to explore the territory and identify the key concepts of quality of life in the study population. This information was then used to design a questionnaire that permits to do quantitative research. There exists no standardized questionnaire that covers all the aspects of quality of life concerning ear and hearing problems in patients with CLP. Despite the well-known benefits of using a standardized questionnaire, preference was given to a self-made questionnaire because we wanted to reflect the full range of perceptions and feelings that patients might hold concerning their ear and hearing problems. The small study population didn’t allow to do a pilot study in advance but within the available time and resources we tried to validate the self-designed questionnaire as much as possible (see methods section).
However, the detailed techniques for achieving validity, reliability, and standardisation are beyond the scope of this thesis.

The main limitation of this study was the rather small sample size (55 patients) and the consequential low statistical power. There was probably insufficient data to detect any but the largest differences. However, results of high clinical relevance are not automatically unimportant if there is no statistical significance. The results of this study are considered relevant since the availability of large-scale datasets that include measures of these outcomes is generally limited.

In addition, some other critical remarks were made. Although it was clearly stated to complete the questionnaire in function of the ear and hearing problems the patients might experience, it can be difficult for children to differentiate between the consequences of the ear and hearing problems on the one hand and the consequences of the cleft in general on the other hand. It is also possible that parents completed the questionnaire (whether or not together with their child) because the child was incompetent to fill in the questionnaire independently. It must be considered that children and parents may have different perspectives on quality of life and that these differences in parental reporting may produce different results. Response bias may result from the parent’s desire for their children to seem competent and well-socialized resulting in an under-reporting of the problem or on the opposite response bias may result from parental anxiety that could lead to a lower (worse) parent-report of HRQoL compared to the child-report (over-reporting) (48).

5.4. Conclusion
The objective of this study was to examine the impact of ear and hearing problems on the quality of life in children with cleft lip and/or palate. Since an extensive literature research has shown that children born with an isolated cleft lip experience no more ear and hearing problems than normal children and since this was statistically confirmed (p<0.001), the original study population was reduced to 55 patients born with a cleft palate ± lip, aged 6 to 18 years old.

Overall, the majority of children and adolescents with cleft palate ± lip do not appear to experience major psychosocial problems as a result of their ear and hearing problems, although difficulties in particular areas of functioning are reported: physical health, hearing loss, school and communication including speech. Gender, age, type of cleft, financial status and parental level of education does not appear to influence the occurrence or severity of these problems, except for the incidence of ear infections (otitis media) which decreases with increasing age. Based on literature research, the quality of life in children with palatal clefts seems more negatively influenced by ear and hearing problems than in children with OME without clefts. Cleft palate is reported to place children who have otitis media with effusion at
increased risk for speech, language or learning problems. On the other hand, the quality of life seems better compared to the quality of life in children with CLP in general since facial differences makes individuals with CLP susceptible to additional psychosocial problems especially those relating to self-esteem, peer relationships and appearance.

To conclude, the concept of quality of life in patients with orofacial clefts is complex and consists of multiple components that cannot be summarized into one outcome variable. To investigate this topic in the best possible way, a large study population is required based on a well-founded sample size calculation. The challenge will be to identify large samples of affected and unaffected individuals. A larger number of participants would increase the reliability of the study. Therefore, this thesis can be used as a pilot study for a large-scale multicentre study.
6. REFERENCE LIST

APPENDICIES

I. Opting-out information letter

Informatiebrief voor deelnemers aan de studie over gehoorproblematiek bij lip- en verhemeltespleet

1. Titel van de studies
1. Inventarisatie van audiometrische gegevens bij palatoschisis
2. Otologische problemen bij palatoschisis
3. Quality of life studie bij palatoschisis

2. Doel van de studies
1 op 700 kinderen wordt geboren met een lip- of verhemeltespleet. Het is de meest voorkomende congenitale aandoening van het hoofd- en halsgebied. Deze kinderen kunnen te maken krijgen met tal van bijkomende problemen zoals voedingsproblematiek, afwijkingen in de groei van het gelaat, spraak- en taalproblemen, tandafwijkingen en emotionele problemen. Ook oor- en gehoorproblemen behoren hierbij.

Wij starten op heden drie studies met elk een zwaartepunt omtrent deze aandoening. Een eerste studie gaat de gehoorproblematiek verder onderzoeken. Een tweede studie onderzoekt de frequentie van het optreden van middenoorproblemen (oorontstekingen, geleidingsslechthorendheid) en de nood aan chirurgische interventies. Ten slotte starten wij een studie om de invloed op de kwaliteit van leven verder na te gaan.

Voor de eerste studie omtrent de impact op het gehoor, is de uiting hiervan en wat de gevolgen hiervan zijn, nog niet met zekerheid geweten. Met deze studie willen we dan ook de gehoorproblemen bij kinderen met een gespleten lip of verhemelte in kaart brengen.

Voor de tweede studie omtrent de opvolging van de middenoorproblemen en chirurgische interventies, is een gedetailleerd overzicht nodig van de oor- en gehoorproblemen bij deze patiëntengroep. Zo hopen we een beter inzicht te krijgen in het verloop van de gehoorproblemen alsook in de prevalentie van de verschillende problemen.

Ten slotte is er een derde studie die de impact van schisis op de levenskwaliteit nagaat. Deze impact van schisis en meer bepaald van de schisisgerelateerde oor- en gehoorproblemen op de levenskwaliteit van deze patiënten is nog niet grondig onderzocht. Zo is het onder meer onduidelijk of kinderen bv. vaker extra begeleiding op school nodig hebben of andere hobby’s uitoefenen. Ook voor volwassenen is het moeilijk om in te schatten in welke mate een bepaald gehoorverlies invloed heeft op de levenskwaliteit. Met deze studie willen we de impact van oor- en gehoorproblemen bij patiënten met schisis nagaan op hun eigen dagelijkse leven en dat van hun omgeving.
Met deze informatie, verkregen uit deze drie studies, kunnen we in de toekomst nog beter inspelen op de verschillende oor- en gehoorproblemen zodat we de nieuw gediagnosticeerde patiënten een optimale zorg kunnen verlenen, op alle aspecten, zowel het gehoor, de medische opvolging en chirurgische interventies alsook het psychologisch luik.


3. Wat wordt verwacht van de deelnemer?
U wordt gevraagd deel te nemen aan een klinische studie. Dit houdt in dat u ons toestemming geeft om de gegevens van uzelf of uw kind, die reeds voorhanden zijn, te verwerken. Bij het ontbreken van gegevens zal er met u contact opgenomen worden om deze aan te vullen, en zal toestemming hiervoor schriftelijk gevraagd worden. Indien het relevant blijkt voor de studie kan er gevraagd worden langs te komen op het UZ Gent voor bijkomende of het opnieuw uitvoeren van gehoortesten, ook dan wordt u aangeschreven en wordt uw toestemming hiervoor gevraagd.

Voor de studie omtrent de invloed van schisis op de levenskwaliteit is het invullen van een vragenlijst essentieel. Ook aan uw kind wordt gevraagd om zelf een gelijkaardige vragenlijst in te vullen. Het invullen van de vragenlijst duurt ongeveer een half uurtje. U kan ons de vragenlijsten daarna terugbezorgen in de daartoe voorziene voorgefrankeerde envelop.

4. Deelname en beëindigen
Deelname aan deze studie vindt plaats op vrijwillige basis. Deelname aan deze studie brengt voor u en voor uw kind geen onmiddellijk voordeel. Uw deelname kan wel helpen om in de toekomst patiënten beter te kunnen helpen. U kan weigeren om deel te nemen aan de studie, en u en uw kind kunnen zich op elk ogenblik terugtrekken uit de studie zonder dat u hiervoor een reden moet opgeven en zonder dat dit op enigerlei wijze een invloed zal hebben op uw verdere relatie en/of behandeling met de onderzoeker. Als u en uw kind deelnemen, hoeft u geen actie te ondernemen.

5. Risico’s en voordelen
Deelname aan dit onderzoek houdt geen verwachte risico’s in voor uw kind. Dit onderzoek brengt geen onmiddellijk voordeel voor u of uw kind mee. Uw deelname aan deze studie kan bijdragen om in de toekomst objectieve resultaten en adviezen te formuleren ten aanzien van nieuw gediagnosticeerde patiënten en hun omgeving.

Deze studie werd goedgekeurd door een onafhankelijke Commissie voor Medische Ethiek verbonden aan UZ Gent en wordt uitgevoerd volgens de richtlijnen voor de goede klinische praktijk (ICH/GCP) en de verklaring van Helsinki opgesteld ter bescherming van mensen deelnemend aan klinische studies. In geen geval dient u de goedkeuring door de Commissie voor Medische Ethiek te beschouwen als een aanzet tot deelname aan deze studie.
De experimentenwet van 7/05/2004 verplicht ons om deelnemers aan wetenschappelijke projecten te verzekeren voor de deelname en het risico (hoe klein ook) dat men loopt. De waarschijnlijkheid dat u door deelname aan deze studie enige schade ondervindt, is extreem laag. Indien dit toch zou voorkomen, wat echter zeer zeldzaam is, werd er een verzekering afgesloten conform de Belgische wet van 7 mei 2004, die deze mogelijkheid dekt.

6. Kosten
Uw deelname aan deze studie brengt geen extra kosten mee voor u.

7. Vergoeding
Er is geen vergoeding voorzien voor deelname aan deze studie

8. Vertrouwelijkheid
In overeenstemming met de Belgische wet van 8 december 1992 en de Belgische wet van 22 augustus 2002, zal de persoonlijke levenssfeer van u en uw kind worden gerespecteerd en zal u toegang krijgen tot de verzamelde gegevens. Elk onjuist gegeven kan op uw verzoek verbeterd worden.

Vertegenwoordigers van de opdrachtgever, auditoren, de Commissie voor Medische Ethiek en de bevoegde overheden hebben rechtstreeks toegang tot de gegevens van uw kind om de procedures van de studie en/of de gegevens te controleren, zonder de vertrouwelijkheid te schenden. Dit kan enkel binnen de grenzen die door de betreffende wetten zijn toegestaan.

Als u en uw kind akkoord gaan om aan deze studie deel te nemen, zullen persoonlijke en klinische gegevens van uw kind tijdens deze studie worden verzameld. Verslagen waarin uw kind wordt geïdentificeerd, zullen niet openlijk beschikbaar zijn. Als de resultaten van de studie worden gepubliceerd, zal de identiteit van uw kind vertrouwelijke informatie blijven.

9. Contactpersoon
Als u aanvullende informatie wenst over de studie of over uw rechten en plichten, kunt u in de loop van de studie op elk ogenblik contact opnemen met:

Prof. dr. Els De Leenheer, Staflid Dienst Neus-, Keel-, Oorheelkunde en Otoloog schisisteam UZ Gent. Els.Deleenheer@Ugent.be

Dr. Helen Van Hoecke, Staflid Dienst Neus-, Keel-, Oorheelkunde UZ Gent. Helen.vanhoecke@ugent.be
Geachte heer, mevrouw,

Graag hadden wij uw medewerking gevraagd om mee te doen aan deze drie studies met als titel:
1. Inventarisatie van audiometrische gegevens bij palatoschisis
2. Otologische problemen bij palatoschisis
3. Quality of life studie bij palatoschisis

Vanuit het elektronisch patiëntendossier willen we de volgende informatie, hierboven beschreven, retrospectief opzoeken.

De verwachte totale duur van de studie is de tijd die we nodig hebben om de informatie op te zoeken in de dossiers van de opgenomen patiënten voor de jaartallen 2000 tot 2016. Er zullen maximaal 500 personen aan deze studie deelnemen.

Deze studie werd goedgekeurd door een onafhankelijke Commissie voor Medische Ethiek verbonden aan dit ziekenhuis, en zal worden uitgevoerd volgens de richtlijnen van ICH/GCP opgesteld in de verklaring van Helsinki opgesteld ter bescherming van individuen deelnemend aan klinische studies.

Als u akkoord gaat om aan deze studie deel te nemen, zullen uw persoonlijke en klinische gegevens tijdens deze studie worden geanonimiseerd (hierbij is er totaal geen terugkoppeling meer mogelijk naar uw persoonlijke dossier). In overeenstemming met de Belgische wet van 8 december 1992 en de Belgische wet van 22 augustus 2002, zal uw persoonlijke levenssfeer worden gerespecteerd. Als de resultaten van de studie worden gepubliceerd, zal uw anonimiteit aldus verzekerd zijn.

U hoeft voor deze studie geen actieve medewerking te verlenen. Wanneer U er geen bezwaar tegen heeft dat we uw medisch dossier consulteren met als enige doelstelling op een anonieme wijze nota te nemen van de nodige beschikbare gegevens, hoeft U niets te doen.

Wanneer U bezwaar heeft dat we uw dossier consulteren voor dit onderzoek, dan vragen we U bijgevoegd document terug te sturen. Op dit document geeft U te kennen geen toestemming te geven om uw dossier te consulteren.

Met de meeste hoogachtig,
Prof. dr. Els De Leenheer, Staflid Dienst Neus-, Keel-, Oorheelkunde en Otoloog schisisteam UZ Gent.
Dr. Helen Van Hoecke, Staflid Dienst Neus-, Keel-, Oorheelkunde UZ Gent.

Opting Out:
1. Inventarisatie van audiometrische gegevens bij palatoschisis
2. Otologische problemen bij palatoschisis
3. Quality of life studie bij palatoschisis

Ik,......................................................, ben niet akkoord om mijn toestemming te verlenen om mijn patiëntengegevens te gebruiken voor deze studie.

Datum:
Handtekening:
II. Questionnaire

Hallo,


Als je een vraag niet begrijpt, kan je aan je mama of papa vragen om mee te helpen. Er is ook plaats voorzien om iets meer uitleg te schrijven in de kantlijn of onderaan de vragen bij ‘opmerkingen’. Het is erg belangrijk dat je deze vragen eerlijk en serieus invult. Probeer alle vragen volledig te beantwoorden. Er zijn geen juist of foute antwoorden. Hoe meer je opschrijft, hoe meer wij eruit kunnen leren.

**Hoe vul je de vragenlijst in:**

Bij de meeste vragen kan je het antwoord aankruisen of omcirkelen. Meestal mag je maar 1 antwoord geven. Als je meerdere antwoorden mag geven staat het erbij geschreven. We weten dat dit soms moeilijk is, maar probeer het antwoord te nemen dat het beste bij je past. Bij sommige vragen moet je zelf een antwoord opschrijven.

**Vergeet niet dat de vragen specifiek gaan over de problemen met jouw oren en gehoor.**

Neem je tijd en alvast bedankt om mee te doen!

Professor Dr. Els De Leenheer, Neus-Keel-Oorarts UZ Gent
Dr. Frederick Dochy, arts-specialist in opleiding Neus-Keel- en Oorheelkunde
Mej. Justine De Paepe, student arts

Bij vragen kan steeds contact opgenomen worden met Frederick Dochy via Frederick.dochy@ugent.be
DEEL 1

1. Hoe vaak had je de afgelopen maand last van... (Omcirkel)

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<th>Geen</th>
<th>Een paar keer per maand</th>
<th>Een paar keer per week</th>
<th>Elke dag</th>
<th>Continu</th>
</tr>
</thead>
<tbody>
<tr>
<td>Oorpijn</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>Het gevoel dat je oren dicht zitten</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>Minder goed horen</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>Vocht dat uit je oor loopt</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>Hoofdpijn</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
</tr>
</tbody>
</table>

2. Heb je ooit buisjes in je oren gehad?
   □ Nee
   □ Ja

   Zo ja: (Er zijn meerdere antwoorden mogelijk)
   □ Ik vind het niet leuk
   □ Ik voel de buisjes zitten / De buisjes doen pijn / De buisjes vallen er uit
   □ Ik schaam mij ervoor
   □ Ik vind het niet erg
   □ Andere: .................................................................................................................................

3. Heb je ooit een hoorapparaat gehad?
   □ Nee
   □ Ja

   Zo ja: (Er zijn meerdere antwoorden mogelijk)
   □ Ik vind het niet leuk omdat ik sommige geluiden veel te luid hoor
   □ Ik schaam mij er voor
   □ Het hoorapparaat zit niet goed aan mijn oren
   □ Anderen lachen mij er mee uit
   □ Ik vind het niet erg
   □ Andere: .................................................................................................................................

Opmerkingen:
.................................................................................................................................................
.................................................................................................................................................
.................................................................................................................................................
DEEL 2

1. **Heb je het soms moeilijk met het horen van ...** (Omcirkel)

<table>
<thead>
<tr>
<th></th>
<th>Nee, niet moeilijk</th>
<th>Ja, een beetje moeilijk</th>
<th>Ja, heel moeilijk</th>
<th>Niet van toepassing</th>
</tr>
</thead>
<tbody>
<tr>
<td>De deurbel</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>De telefoon</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>De televisie of radio</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>De wekker</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>De juf of meester</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Het verkeer (auto’s, fietsbel...)</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
</tbody>
</table>

2. **Heb je het soms moeilijk om andere mensen te verstaan...** (Omcirkel)

<table>
<thead>
<tr>
<th></th>
<th>Nee, niet moeilijk</th>
<th>Ja, een beetje moeilijk</th>
<th>Ja, heel moeilijk</th>
<th>Niet van toepassing</th>
</tr>
</thead>
<tbody>
<tr>
<td>In de klas</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Als er veel lawaai is</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>bv. op de speelplaats, op restaurant</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Als mensen door elkaar praten</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Als iemand tegen je fluistert</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Als iemand tegen je babbelt die verder weg van je staat</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
</tbody>
</table>

3. **Zeggen ze soms tegen jou dat:**

je veel ‘wablieft’ zegt.

- Ja
- Nee

de televisie zeer luid staat.

- Ja
- Nee

je niet reageert als iemand iets tegen je zegt.

- Ja
- Nee
**Opmerkingen:**

…………………………………………………………………………………………………………………………………………………………….
…………………………………………………………………………………………………………………………………………………………….
…………………………………………………………………………………………………………………………………………………………….

**DEEL 3**

Omcirkel een cijfer tussen nul en tien dat het best bij jou past. 0 wil zeggen het helemaal niet waar is wat er geschreven staat. 10 wil zeggen dat je helemaal eens bent.

<table>
<thead>
<tr>
<th>Helemaal oneens</th>
<th>Helemaal eens</th>
</tr>
</thead>
<tbody>
<tr>
<td>0    1    2    3    4    5    6    7    8    9    10</td>
<td></td>
</tr>
</tbody>
</table>

Ik ben bang dat mijn oorproblemen erger zullen worden in de toekomst. 0 1 2 3 4 5 6 7 8 9 10

Ik voel mij verdrietig door mijn oorproblemen. 0 1 2 3 4 5 6 7 8 9 10

Ik heb minder zelfvertrouwen door mijn oorproblemen. 0 1 2 3 4 5 6 7 8 9 10

Ik ben beschaamd door mijn oorproblemen:
1. Op school 0 1 2 3 4 5 6 7 8 9 10
2. Bij mijn vrienden en vriendinnen 0 1 2 3 4 5 6 7 8 9 10
3. In de jeugdbeweging/sportclub/muziekschool 0 1 2 3 4 5 6 7 8 9 10
4. Als ik iemand nieuw leer kennen 0 1 2 3 4 5 6 7 8 9 10

Door mijn oorproblemen voel ik me anders dan de anderen. Dit ergert me of maakt me kwaad. 0 1 2 3 4 5 6 7 8 9 10

Ik ben jaloers op anderen die geen oorproblemen hebben. 0 1 2 3 4 5 6 7 8 9 10

Ik word uitgesloten door mijn oorproblemen. 0 1 2 3 4 5 6 7 8 9 10

Door mijn oorproblemen vind ik het moeilijk om nieuwe vrienden te maken. 0 1 2 3 4 5 6 7 8 9 10

Ik vind dat ik veel vrienden heb. 0 1 2 3 4 5 6 7 8 9 10

Als ik met iets zit, kan ik hier altijd over praten met mijn vriendjes of vriendinnetjes. 0 1 2 3 4 5 6 7 8 9 10

Als ik met iets zit, kan ik hier altijd over praten met mijn mama of papa. 0 1 2 3 4 5 6 7 8 9 10

Mijn leven is anders door de oorproblemen die ik heb. 0 1 2 3 4 5 6 7 8 9 10

Door mijn oorproblemen ga ik soms niet naar:
1. uitstappen van school bv. sportdag 0 1 2 3 4 5 6 7 8 9 10
2. activiteiten van mijn jeugdbeweging/sportclub/muziekschool 0 1 2 3 4 5 6 7 8 9 10
3. verjaardagsfeestjes 0 1 2 3 4 5 6 7 8 9 10
4. afspraakjes met vrienden/vriendinnen 0 1 2 3 4 5 6 7 8 9 10
Door mijn oorproblemen heb ik soms geen zin om naar school te gaan.

Opmerkingen:

DEEL 4

Heb je door je oorproblemen soms problemen met... (Omcirkel)

<table>
<thead>
<tr>
<th>Hobby's</th>
<th>Nee, helemaal geen problemen</th>
<th>Ja, een beetje problemen</th>
<th>Ja, veel problemen</th>
<th>Niet van toepassing</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sport</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Zwemmen</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Muziekschool</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Jeugdbeweging</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Videogames spelen</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Naar muziek luisteren</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Naar televisie kijken</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Naar de cinema gaan</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Andere:</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Op school</th>
<th>Nee, helemaal geen problemen</th>
<th>Ja, een beetje problemen</th>
<th>Ja, veel problemen</th>
<th>Niet van toepassing</th>
</tr>
</thead>
<tbody>
<tr>
<td>Je concentreren in de les</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Huiswerk maken</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Leren voor een toets</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
</tr>
</tbody>
</table>
De juf of meester verstaan 1 2 3 4
Andere: ........................................ 1 2 3 4
....................................................
Niet kunnen slapen door de oorpijn 1 2 3 4

Heb je al eens moeten zittenblijven op school (je jaar moeten overdoen)?

☐ Nee
☐ Ja

Zo ja: in welk leerjaar? .......................................................... ..........................................................

Volg je bijles of heb je vroeger bijles gevolgd?

☐ Nee
☐ Ja

Zo ja: voor welk vak/welke vakken: .......................................................... ..........................................................

Volg je logopedie of heb je vroeger logopedie gevolgd?

☐ Nee
☐ Ja

Heb je al lessen of activiteiten op school gemist:
(Er zijn meerdere antwoorden mogelijk)

☐ Omdat je een afspraak bij de neus-keel-oorarts had
☐ Omdat je een oorontsteking (oorpijn) had
☐ Andere: .......................................................... ..........................................................
☐ Ik heb nog nooit lessen of activiteiten op school gemist door problemen met mijn oor/gehoor

Opmerkingen:
...........................................................................................................................................................
...........................................................................................................................................................
...........................................................................................................................................................

DEEL 5

Zou je dit deel van de vragenlijst door iemand anders (bijvoorbeeld mama of papa) willen laten invullen?

In welke mate vormen onderstaande zaken een probleem voor uw kind?

<table>
<thead>
<tr>
<th></th>
<th>Geen probleem</th>
<th>Een klein probleem</th>
<th>Een behoorlijk probleem</th>
<th>Een ernstig probleem</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
Hij/zij heeft een slechte uitspraak 1 2 3 4
Hij/zij spreekt traag 1 2 3 4
Hij/zij spreekt onduidelijk 1 2 3 4
Hij/zij wordt verkeerd verstaan door anderen 1 2 3 4
Hij/zij verstaat anderen verkeerd 1 2 3 4

**Ten gevolge van eventuele communicatieproblemen:**

Hij/zij gedraagt zich onrustig, hyperactief of zelfs agressief 1 2 3 4
Hij/zij isoleert zich van zijn omgeving 1 2 3 4
Hij/zij heeft leerproblemen 1 2 3 4

Door wie werd dit deel van de vragenlijst ingevuld? ……………………………………………………………………………………………………………………………………………………………

**Opmerkingen:**

…………………………………………………………………………………………………………………………………………………………
…………………………………………………………………………………………………………………………………………………………
…………………………………………………………………………………………………………………………………………………………

**DEEL 6**

Voor mama en papa: gelieve nog onderstaande gegevens in te vullen.

**Gegevens van uw kind**

- Voornaam: ....................................................................................................................
- Achternaam: ....................................................................................................................
- Geslacht: ..........................................................................................................................
- Geboortedatum: ..............................................................................................................
- Geboorteland: ............................................................................................................... 
- Hoeveelste kind in rij: ..................................................................................................
- Uw kind is geboren met:
  - ☐ Enkel een gespleten lip links / rechts / beide kanten (omcirkel)
  - ☐ Enkel een gespleten verhemelte
  - ☐ Een gespleten lip en een gespleten verhemelte links / rechts / beide kanten (omcirkel)
- Aan welke kant situeren de oorproblemen zich? links / rechts / beide kanten (omcirkel)

- Maakt de gespleten lip/verhemelte van uw kind deel uit van een syndroom (omcirkel): Ja / Nee

  Zo ja, welk syndroom? ……………………………………………………………………………………………………………

- Hoeveel keer heeft uw kind het afgelopen jaar een oorontsteking gehad?

  □ 0  □ 1  □ 2 of 3  □ 4 of meer

  Welke klachten had hij/zij toen door deze oorontsteking?
  *(Er zijn meerdere antwoorden mogelijk)*

  □ Geen klachten  □ Oorpijn  □ Vocht dat uit je oor loopt  □ Minder goed horen  □ Koorts  □ Je voelde je ziek  □ Andere: ……………………………………………………………………………………………………………

  Bent u toen naar de dokter gegaan?

  □ Nee  □ Ja

  Zo ja: heeft de dokter toen medicatie voorgeschreven?

  □ Nee  □ Ja  □ Ik weet het niet

  Zo ja: welke medicatie? *(Er zijn meerdere antwoorden mogelijk)*

  □ Oordruppels  □ Pijnstillers  □ Antibiotica  □ Ik weet het niet  □ Andere: ……………………………………………………………………………………………………………

- Gebruikt hij/zij een hoortoestel? (omcirkel wat past): Ja / Nee

  Zo ja, welk? (kruis aan)

  □ Hoortoestel  □ Cochleair implant (CI)  □ FM-systeem  □ Botverankerd hoortoestel (BAHA: bone-anchored hearing aid)  □ Andere: ……………………………………………………………………………………………………………
Gegevens van het gezin

- Aantal kinderen in het gezin:  
  
- Andere familieleden met gespleten lip en/of verhemelte (omcirkel wat past): Ja / Nee

- Andere familieleden met oor- of gehoorproblemen (omcirkel wat past): Ja / Nee
  Zo ja, welke problemen?

- Geboorteland van de moeder:

- Geboorteland van de grootmoeder:

- Geboorteland van de vader:

- Geboorteland van de grootvader:

- Welke taal wordt er voornamelijk gesproken thuis:

- Hoogst behaald diploma van de moeder:
  - Lager onderwijs
  - Lager secundair onderwijs
  - Hoger secundair onderwijs
  - Hoger onderwijs: hogeschool
  - Hoger onderwijs: universiteit
  Hoogst behaald diploma van de vader:
  - Lager onderwijs
  - Lager secundair onderwijs
  - Hoger secundair onderwijs
  - Hoger onderwijs: hogeschool
  - Hoger onderwijs: universiteit

- Kunnen jullie op het einde van de maand de eindjes financieel aan elkaar knopen? Ja/ Nee/ Soms

- Werd medische zorg al eens uitgesteld omwille van financiële redenen? Ja / Nee

Op welk telefoonnummer of e-mailadres kunnen wij jou of jouw ouders contacteren indien we nog bijkomende vragen hebben?
III. Table of Fisher’s Exact test significance values

<table>
<thead>
<tr>
<th></th>
<th>Gender</th>
<th>Type of cleft</th>
<th>Age category</th>
<th>Financial status</th>
<th>Level of education</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Mother</td>
</tr>
<tr>
<td>Mental wellbeing</td>
<td>0.158</td>
<td>0.525</td>
<td>0.174</td>
<td>0.331</td>
<td>0.633</td>
</tr>
<tr>
<td>School</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>- grade retention</td>
<td>0.174</td>
<td>0.712</td>
<td>0.326</td>
<td>0.346</td>
<td>0.012</td>
</tr>
<tr>
<td>- tutoring</td>
<td>1.000</td>
<td>1.000</td>
<td>1.000</td>
<td>1.000</td>
<td>0.785</td>
</tr>
<tr>
<td>- general problems</td>
<td>1.000</td>
<td>1.000</td>
<td>1.000</td>
<td>1.000</td>
<td>0.587</td>
</tr>
<tr>
<td>Communication</td>
<td>1.000</td>
<td>0.342</td>
<td>0.768</td>
<td>0.409</td>
<td>0.125</td>
</tr>
<tr>
<td>- understanding people</td>
<td>0.559</td>
<td>0.205</td>
<td>0.256</td>
<td>0.131</td>
<td>0.917</td>
</tr>
<tr>
<td>- speech</td>
<td>1.000</td>
<td>0.550</td>
<td>1.000</td>
<td>0.720</td>
<td>0.047</td>
</tr>
<tr>
<td>- speech therapy</td>
<td>0.092</td>
<td>0.175</td>
<td>0.116</td>
<td>0.334</td>
<td>0.212</td>
</tr>
<tr>
<td>Swimming</td>
<td>0.397</td>
<td>0.359</td>
<td>0.528</td>
<td>0.474</td>
<td>0.856</td>
</tr>
</tbody>
</table>