PATIENT-REPORTED OUTCOME SURVEY POST PRIMARY CLEFT LIP AND PALATE SURGERY IN HOSPITAL KUALA LUMPUR USING CHILD ORAL HEALTH IMPACT PROFILE (COHIP) QUESTIONNAIRE

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Introduction: Patient-reported outcome survey is one of the modality to assess the outcome of surgical intervention from patient's perspective. Cleft lip and palate is one of the most common congenital abnormality encountered in plastic surgery field. The primary surgical repair for both cleft lip and palate were among the commonest procedure done in treating cleft lip and palate patients. The outcome of the surgery includes the aesthetic results, speech, functionality self image and quality of life. Child Oral Health Impact Profile questionnaire is one of the reliable and valid assessment tools in the form of questionnaire to evaluate patient's reported outcome post primary cleft lip and palate surgery.

Objectives: The aims of this study is to evaluate the patients reported outcome post primary cleft lip and palate surgery in Hospital Kuala Lumpur using Child Oral Health Impact Profile (COHIP) questionnaire. Specifically is to describe patient's perspective regarding their oral health, functional well-being, social/emotional well-being, school environment and self image post cleft lip and palate repair using the COHIP score. Second specific objective is to compare the mean difference of COHIP score between early and late timing of surgery. Third specific objective is to compare mean difference of COHIP score between 3 categories of surgeons.

Methodology: A cross-sectional study was carried out on patients with cleft lip and palate in Hospital Kuala Lumpur, Malaysia. All patients details with orofacial cleft operated between 1999 and 2005 that have been treated at the Department of Plastic and Reconstructive Surgery, Hospital Kuala Lumpur, Malaysia were retrieved from the operation theatre logbook to see the eligible patients in the age group of 8 to 14 years old to participate in the study. From the details collected, the patients that fulfilled the criteria were invited to participate in the study via a phone call. The appointment date was given to the patients. On the day of the appointment the patients and their parents/guardians were given a participant information sheet, contains the details of the study conducted. The researcher provided information and answered any question regarding the study. The consent form for parents/guardians and the child participant assent form were distributed after both parents/guardians and the patients agree to participate in the study. The COHIP Questionnaire was distributed to the participant after the researcher wrote down all the demographic details. The patient was given 30 minutes to complete the questionnaire.

Results: A total of 82 patients participated in the study. Of these, 41 (50.0%) were male and 41 (50.0%) were female with 1:1 ratio of male to female. Their age range is between 8 years old to 14 years old, with a mean age of 11.3 years old. The majority of the subjects had left unilateral cleft lip and palate *n*=44 (53.7%). About one-third of the subjects had bilateral cleft lip and palate *n*=25 (30.5%) and the remainder had right unilateral cleft lip and palate *n*=13 (15.9%). Majority of the subjects were operated by the specialist *n*=39 (47.6%), whereby consultants operated on 35 subjects (42.7%) and trainee operated on 8 subjects (9.8%). Early primary cleft palate repair less than 1 year old of age were done to 52 subjects (63.4%). Late primary cleft palate repair more than 1 year old of age were done to 30 subjects (36.6%). There was a good outcome of the oral health, functional well-being, social emotional well-being, school environment and self image in primary repair cleft lip and palate with the mean overall Child Oral Health Impact Profile (COHIP) score of 102.2. There were significant differences of the overall COHIP and the oral health well-being, school environment and treatment expectation subscales score between the consultants, specialists and trainee. There was no significant difference of overall COHIP and all the subscales score between early and late timing of surgery.

Conclusions: Results showed the overall satisfaction post primary cleft lip and palate repair was satisfactory and thus supported the continuation of current management protocol in HKL for the management of cleft lip and palate patients.

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IV LIST OF ABBREVIATIONS

COHIP	Child Oral Health Impact Profile
HKL	Hospital Kuala Lumpur
IQ	Intelligent Quotient
OHRQoL	Oral Health-related Quality of Life
SPSS	Statistical Package for Social Sciences
BCLP	Bilateral Cleft Lip and Palate
UCLP	Unilateral Cleft Lip and Palate
BCL	Bilateral Cleft Lip
UCL	Unilateral Cleft Lip
VPI	Velo Pharyngeal Insufficiency
ICC	Intraclass Correlation Coefficient

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VII ABSTRAK

Kajian laporan pesakit adalah salah satu cara untuk mengesan keberkesanan sesuatu hasil pembedahan daripada pandangan pesakit itu sendiri. Rekahan bibir dan lelangit adalah satu bentuk kecacatan sejak lahir yang paling banyak dirawat dalam bidang plastik surgeri. Pembedahan pembetulan utama bagi rekahan bibir dan lelangit adalah prosedur rawatan yang paling kerap dijalankan dalam rawatan rekahan bibir dan lelangit. Hasil akhir daripada pembedahan termasuklah hasil estetik , pertuturan, fungsi, imej diri dan kualiti kehidupan. Soalan kajiselidik profil impak kesihatan oral kanak-kanak adalah suatu kaedah yang terbukti berkesan dan sahih untuk mengukur laporan pesakit selepas pembedahan utama rekahan bibir dan lelangit. Suatu kajian keratan rentas telah dijalankan ke atas pesakit rekahan bibir dan lelangit di Hospital Kuala Lumpur, Malaysia. Populasi kajian diperolehi dengan melihat semula rekod pembedahan rekahan bibir dan lelangit dari tahun 1999 sehingga 2005. Seramai 82 pesakit telah menyertai kajian ini. Daripada jumlah itu seramai 41 orang atau 50% adalah lelaki dan seramai 41 orang lagi atau 50% adalah perempuan. Nisbah lelaki kepada perempuan adalah 1:1. Umur mereka adalah dalam lingkungan 8 ke 14 tahun dengan min umur 11.3 tahun. Majoriti daripada subjek kajian mengalami rekahan bibir dan lelangit sebelah kiri. Satu pertiga mengalami rekahan bibir dan lelangit di kedua-dua belah dan 15.9% lagi mengalami rekahan bibir dan lelangit sebelah kanan. Secara keseluruhannya 39 subjek (47.6%) dibedah oleh pakar bedah plastik, manakala pakar perunding membedah seramai 35 subjek (42.7%) dan pakar dalam latihan membedah seramai 8 subjek (9.8%). Pembedahan pembetulan rekahan lelangit dilakukan sebelum umur satu tahun untuk seramai 52 subjek (63.4%) dan lewat iaitu melebihi satu tahun seramai 30 subjek (36.6%). Terdapat keputusan yang baik dalam kesihatan oral, fungsi, kesihatan sosial emosional, persekitaran sekolah dan imej diri dalam kalangan peserta kajian dengan min skor COHIP 102.2. Terdapat faktor statistik yang signifikan dalam skor kesuluruhan COHIP, kesihatan oral, persekitaran sekolah dan harapan ke atas rawatan apabila dibandingkan dalam kalangan kumpulan pakar bedah yang berbeza iaitu pakar perunding, pakar bedah plastik and pakar dalam latihan. Tiada faktor statistik yang signifikan dalam skor keseluruhan COHIP di antara pembedahan rekahan bibir yang dilakukan awal sebelum pesakit berumur satu tahun atau lewat selepas pesakit berumur satu tahun. Keputusan menunjukkan secara keseluruhan kepuasan pesakit adalah memuaskan dan keputusan yang diperolehi dapat digunakan untuk mengekalkan protokol dalam rawatan rekahan bibir dan lelangit supaya kualiti kehidupan pesakit dapat dikekalkan pada tahap yang baik dan memuaskan pada masa hadapan.

VIII ABSTRACT

Patient-reported outcome survey is one of the modality to assess the outcome of surgical intervention from patient's perspective. Cleft lip and palate is one of the most common congenital abnormality encountered in plastic surgery field. The primary surgical repair for both cleft lip and palate were among the commonest procedure done in treating cleft lip and palate patients. The outcome of the surgery includes the aesthetic results, speech, functionality self image and quality of life. Child Oral Health Impact Profile questionnaire is one of the reliable and valid assessment tools in the form of questionnaire to evaluate patient's reported outcome post primary cleft lip and palate surgery. A cross-sectional study was carried out on patients with cleft lip and palate in Hospital Kuala Lumpur, Malaysia. Study populations are obtained by reviewing operative records of cleft lip and palate done in Hospital Kuala Lumpur from 1999 to 2005. A total of 82 patients participated in the study. Of these, 41 (50.0%) were male and 41 (50.0%) were female with 1:1 ratio of male to female. Their age range is between 8 years old to 14 years old, with a mean age of 11.3 years old. The majority of the subjects had left unilateral cleft lip and palate n=44 (53.7%). About one-third of the subjects had bilateral cleft lip and palate n=25 (30.5%) and the remainder had right unilateral cleft lip and palate n=13 (15.9%). Majority of the subjects were operated by the specialist n=39 (47.6%), whereby consultants operated on 35 subjects (42.7%) and trainee operated on 8 subjects (9.8%). Early primary cleft palate repair less than 1 year old of age were done to 52 subjects (63.4%). Late primary cleft palate repair more than 1 year old of age were done to 30 subjects (36.6%). There was a good outcome of the oral health, functional well-being, social emotional well-being, school environment and self image in primary repair cleft lip and palate with the mean overall Child Oral Health

Impact Profile (COHIP) score of 102.2. There were significant differences of the overall COHIP and the oral health well-being, school environment and treatment expectation subscales score between the consultants, specialists and trainee. There was no significant difference of overall COHIP and all the subscales score between early and late timing of surgery. Results showed the overall satisfaction post primary cleft lip and palate repair was satisfactory and thus supported the continuation of current management protocol in HKL for the management of cleft lip and palate patients.

1.0 Introduction

Cleft lip and/or palate is one of the most common congenital craniofacial anomalies encountered in plastic surgery practice worldwide. The prevalence of cleft lip and/or palate varies between 1.5 to 25/10000 births (Mossey & Little, 2002). The average occurrence rate is approximately 1 in 700 live birth (Bell et al., 2013; Bellis & Wolgemuth, 1999). In Malaysia, the incidence of congenital birth defects is 14.3/1000 births and 11.9% is cleft lip and palate (Thong, Ho, & Khatijah, 2005). The occurance of cleft lip, cleft palate and a combination of both conditions in Malaysia was reported in 1 out of 941 births (NOHSS, 1998). The rate of occurance of cleft in a Maternity Hospital, Kuala Lumpur over 2-year period was 1.24 per 1000 livebirth (Boo & Arshad, 1990). Cleft lip and palate are associated with a multiple variety of health complications such as feeding, speech, growth and physical health problems such as recurrent middle ear infections. Patients with cleft lip and palate undergo various modalities, including surgical, medical, speech, dental and other health interventions (Nackashi, Rosellen, & Dixon-Wood, 2002)

In cleft lip and palate, there are significant psychological and social burdens have been reported. Children with cleft lip and palate often having a challenging psychological adjustment due to aesthetic concern, speech and hearing disabilities and difficulty acquiring the social skills necessary for adjustment (Havstam, Lohmander, Dahlgren Sandberg, & Elander, 2008; Kapp-Simon, 1986). Children with cleft lip and palate also have a higher risk of developmental problems including cognitive performance. Treatment of cleft lip and palate started from newborn and extends throughout adolescent. Psychological adjustment has been reported to be a problem during an adolescent period of treatment (Havstam, Laakso, & Ringsberg, 2011; Kapp-Simon, 1986).

The plastic surgery services in Malaysia started in Hospital Kuala Lumpur in 1970 and the cleft surgery is among the early procedure performed, however till date there is no proper patient-reported outcome studies for post primary lip and palate repair conducted.

Patient’s reported outcome are critical for assessment of patient’s care. It is important for surgeon to evaluate the outcome of the treatment from patient’s perspective. The goal of cleft lip and palate treatment are excellent aesthetic and functional outcomes. This is important to improve the patient’s quality of life. The ultimate aim of cleft lip and palate care is for the child to be able to achieve his or her full potential and able to function well in the society. Patient-reported outcomes measure include aesthetic results, speech, functionality, self image, incorporation into society, and quality of life will provide surgeons with more comprehensive assessment of surgical outcomes (Eckstein, Wu, Akinbiyi, Silver, & Taub, 2011).

These outcomes measurement will provide a valuable information for the surgeon in terms of their surgical outcomes, patients satisfaction and will lead surgeon to understand the limitation or flaws that need to be addressed in order to improve the services. It is important for a leading institution in the country such as Hospital Kuala Lumpur to conduct a research on the patient’s satisfaction and evaluation of the outcome post primary cleft lip and palate repair and evaluate the result in order to audit the services and improvement is to be carried out based on the result obtained.

Earlier studies showed the cleft lip and/or palate patients were perceived to have lower self esteem, difficulty in the learning process, and a tendency to be more depressed and anxious. They were less social and having difficulty in meeting new friend because of their deformity (Feragen & Borge, 2010). Some of them also suffered psychosocial problems. Functional and aesthetic problems frequently arise and later in life some of them will experience difficulties with oral health and speech. Cleft lip and palate patients were teased because of their cleft and it affected their self-confidence (Noor & Musa, 2007).

Many methods were used to evaluate patient's reported outcome or satisfaction. Health-related Quality of Life is one of the tool used to assess the impact of disease severity on a child's physical, psychological, social and emotional well-being. Studies have found that children with visible facial differences such as cleft lip and palate have lower quality of life and that their quality of life is similar to that of children with other chronic disorders (Topolski, Edwards, & Patrick, 2005). Pre-adolescent children with cleft lip and palate perceived their quality of life diferently at different age group and these have been reported. For children with isolated cleft palate their quality of life is gradually improved towards adolescence but for children with cleft lip, the quality of life decrease as they approach adolescence. These is mainly when acceptance by their peers becomes more critical (Damiano et al., 2007).

The use of reliable, valid and responsive patient questionnaire is vital to evaluate the impact of cleft lip and palate surgery on the lives of these patients. The existing cleft lip and palate specific instrument have been studied and showed none of the existing instrument evaluate thoroughly all the issue related to cleft lip and palate patients. Five valid and reliable questionnaires have been analyzed and were found sufficiently valid. Among those five identified, the Child Oral Health Impact Profile (COHIP) is well-developed, valid, reliable and assesses many social-emotional and oral-functional aspects of clefts (Eckstein et al., 2011).

Children and adolescent have been found to provide reliable information regarding their own oral health-related quality of life when appropriate questionnaire techniques are used although few scales specifically designed for them have been developed (Filstrup et al., 2003; Jokovic et al., 2002; Jokovic, Locker, Tompson, & Guyatt, 2004).

COHIP contains domain for oral health, functional well-being, social-emotional well-being, school environment, self image, treatment expectancy and global health. All of these domains also provide information regarding facial appearences, speech and satisfaction of treatment (Broder & Wilson-Ganderson, 2007).

The quality of life measure such a facial appearances and speech are mainly the goals of the surgery for cleft lip and palate. Surgeon with certain experience in managing cleft lip and palate have shown to have better outcome (Shaw, Semb, & Nelson, 2001). Hospital Kuala Lumpur is the training center for plastic surgeon in the country. The surgeon involved in managing cleft lip and palate are consultant with more than 5 years experience, the specialist less than 5 years, and the trainee. Each of the group significantly involved in the cleft and palate surgery. This study also aim to see whether surgeons' experience is affecting the patient overall COHIP score.

The timing of the surgery is one of the factor that contribute significantly in the outcome of cleft lip and palate surgery. If cleft lip repair is done at the age of 3 to 4 months old followed by palate surgery less than 1 year old, the speeh outcome is better in early cleft palate closure (Dorf & Curtin, 1982). In this study we will categorize cleft palate surgery based on the timing which is early and late and to correlate with the score given by the patients.

This study will used the COHIP questionnaires and analyzed the score of each domain to determine the children and adolescent oral health-related quality of life post primary cleft lip and palate repair in Hospital Kuala Lumpur. The correlation of the epidemiology data of the subject such as the surgeon factors and the timing of the surgery with their overall COHIP score will also be study.

1.1 Justification of the Study

It is becoming increasingly difficult to ignore the patient's reported outcome for the assessment of patient's care in the field of the plastic surgery. Especially when it is involved the treatment of congenital deformities such as cleft lip and/or palate where the goals of the treatment are excellent aesthetic and functional outcomes. Therefore a comprehensive assessment of the surgical outcomes must includes the patient's reported outcome (Eckstein et al., 2011).

The patient's reported outcome is an important component of the surgical assessment outcome and plays a key role in patients's quality of life. It will provides patient's perspective of the surgical outcomes. The information gathered from the study will provide surgeon to understand the limitation or flaw in their treatment protocol and will lead the surgeon to make an amendment if necessary.

So far, however, as the leading institution in the country for cleft lip and/or palate treatment, Hospital Kuala Lumpur have not evaluated the patient's reported outcome post primary cleft lip and palate surgery. This study will focus on this matter generally by using the Child Oral Health Impact Profile (COHIP) as an asessment tool.

The Child Oral Health Impact Profile (COHIP) is identifed as a tool to determine patient's reported outcome in this study as it contains domain for oral health, functional well-being, social-emotional well-being, school environment, self image, treatment expectancy and global health. These domains provide evaluation of facial appearances, speech treatment satisfaction (Broder & Wilson-Ganderson, 2007). COHIP is a well developed, valid and reliable. It assesses many oral-funtional and social emotional aspects of cleft patients (Eckstein et al., 2011)

Specifically the timing of the surgery and the surgeon's catergory also played a signifficant role in the outcomes of the surgery. The speech outcome is better in early cleft palate closure compare to late palatal closure (Dorf & Curtin, 1982). Thus, this study aim to compare the mean difference of the COHIP score between early and late timing of surgery for palatal closure.

Surgeon's experiences have shown to effect the outcome of the cleft lip and palate surgery (Shaw et al., 2001). This study will compare the mean difference of the COHIP score between the surgeon's category based on their experience. Three category were identified, the consultant with more than five years experiences, specialist with less than five years experiences and the trainee.

The results will help to identify whether the current practise in Hospital Kuala Lumpur should be maintained or amendment should be made to provide high quality and more efficient services to improve the outcome of the surgical interventions based on the patient's perspective.

1.2 Conceptual Framework

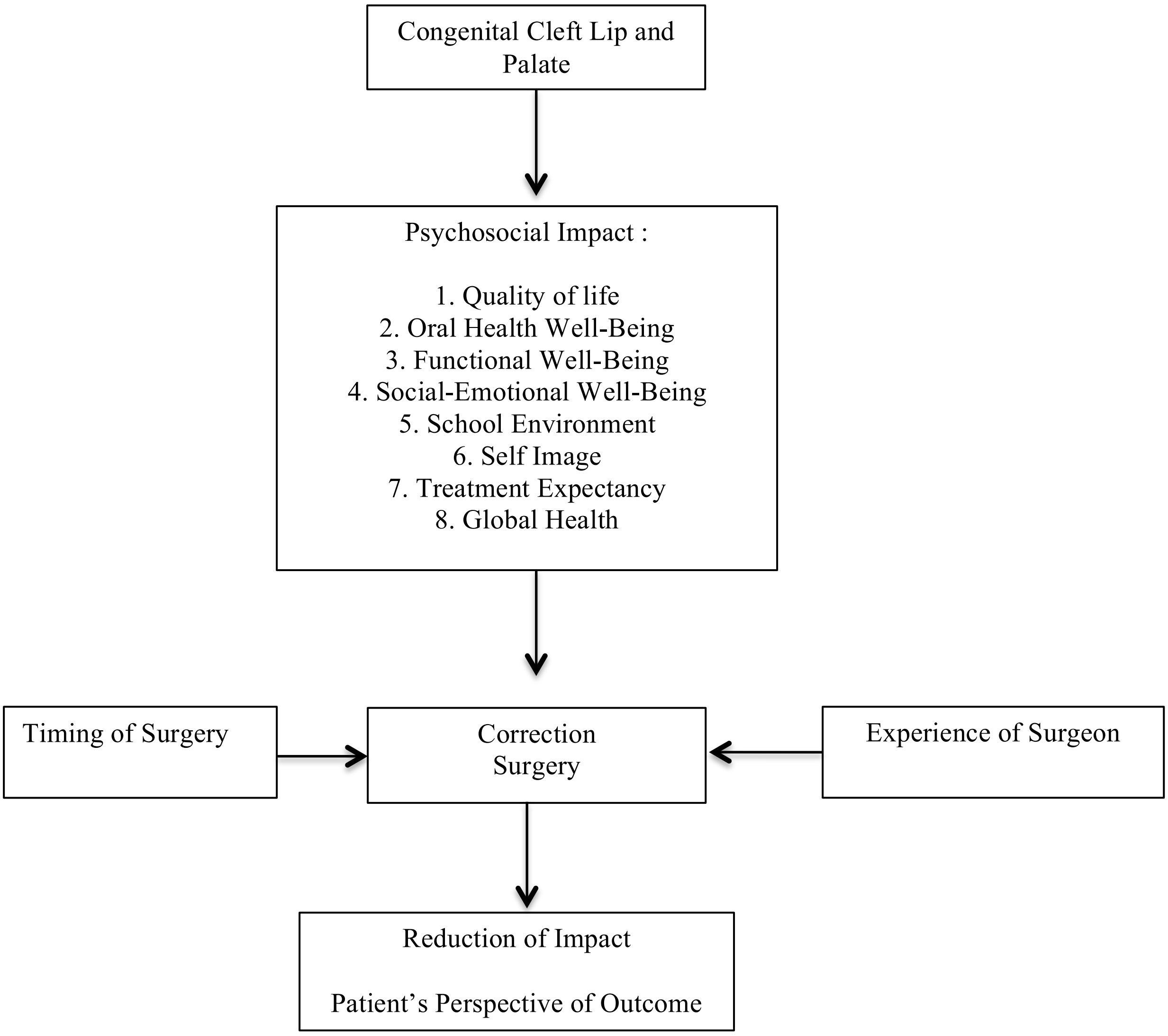

Figure 1 Conceptual framework

2.0 Literature Review

2.1 Epidemiology of Oral Clefts

Cleft lip and/or palate are collectively term as oral clefts. The overall incidence of oral clefts is approximately about 1 in 750 live births and making the oral clefts the second most common congenital defect after clubfoot (Sadove, Aalst, & Culp, 2004). The most common type of oral cleft is a bifid uvula, occurring in aprroximately 1 to 2% of the population (Watkins, Meyer, Strauss, & Aylsworth, 2014).

About 21% of cases had isolated cleft lip, 46% had cleft lip and palate, and 33% had isolated cleft palate. Most cases of cleft lip and/or palate are unilateral in 80 to 85% of patients and bilateral in 15 to 20% (Setó-Salvia & Stanier, 2014). As for site of occurrence cleft lip and palate effected left side more often, left-sided clefts were twice as frequent as right-sided clefts and 6 times more frequent than bilateral clefts, for a 6:3:1 ratio (Watkins et al., 2014). Isolated cleft lip without cleft palate is typically unilateral approximately about 80% and it is on the left side in 70% of the cases.

Approximately 10% of the cleft lips are incomplete (Coleman & Sykes, 2001). Bilateral cleft lip is associated with cleft palate in 86% of cases, whereas 70% of unilateral cleft lips are accompanied by cleft palate (Watkins et al., 2014). Cleft lip with or without cleft palate shows evidence of racial heterogeneity with the incidence of cleft lip with or without cleft palate ranged from 0.71 to 1.29 per 1000 births in Caucasians and from 0.43 to 0.80 in those of African descent (Tanaka, Mahabir, Jupiter, & Menezes, 2012). The race was the only demographic variable associated with cleft lip and the incidence is highest among Asians, followed by Caucasians, and lowest in people of African descent (Tanaka et al., 2012). It is likely that environmental factors play a role in the incidence and distribution of cleft types among different populations with varying susceptibilities.

There appears to be less racial heterogeneity in isolated cleft palate, which has an average incidence of 0.5 in 1000 births among the races. The incidence of submucous clefts of the palate is 1 in 1000 to 1 in 2000 births (Watkins et al., 2014). Unlike other isolated clefts of the secondary palate, the incidence of uvular clefts varies with race, being highest in Native Americans 1 in 9 births and relatively low in whites about 1 in 80 births (Coleman & Sykes, 2001).

Cleft lip and/or palate occur more frequently in males, whereas cleft palate occurs more frequently in females. Males predominate in isolated cleft lip without cleft palate in about 60% of cases and cleft lip with cleft palate in 67% of the cases (Coleman & Sykes, 2001). In contrast, complete clefts of the secondary palate are twice as common in females as in males, and the incidence of isolated soft palate clefts is approximately the same for the both sexes. Unequal gender distribution of the cleft lip and/or palate is attributed to the different timing of embryological process between males and females (Davidson, 2012).

2.2 Embryology of the Oral Clefts

The understanding of craniofacial embyrogenesis and the pathogenesis of orofacial clefting are important. Traditionally facial development has been described with the classical theory of embryogenesis by the formation, migration, and fusion of five facial prominences or processes; the frontonasal, the bilateral maxillary, and the bilateral mandibular (Durscy, 1869; His, 1874). It describes formation of the primary palate as a fusion of the medial and nasal prominences of the frontonasal process with the maxillary prominences during weeks 4 to 7 of gestation. The formation of the secondary palate is described as the fusion of the two lateral palatal processes of the maxillary prominences during gestational weeks 5 to 12. This model of facial development involves the assembly of formed structures also known as processes based on a simplified description of external morphology.

Recently the facial prominences or processes were described as complex arrangements of developmental fields under genetic control, not single autonomous or anatomic units (Carstens, 2002). Early embryonic development is genetically controlled through the production of growth factors that target specific embryonic cell populations and guide their differentiation, migration and morphogenesis (Marazita & Mooney, 2004). In addition to the presence, concentration gradients and diffusion patterns of growth factors, normal development is regulated by intercellular communication and selective cell membrane permeability.

Disruption of gene-controlled, growth-factor mediated cell differentiation, migration, and fusion may result in congenital malformations (Carstens, 2002). In Carstens's neuromeric model of developmental fields, the face is conceptualized as a series of genetically defined developmental fields, each with a specific cellular content and a recognizable functional matrix (Carstens, 2004).

Each field develops from a specific anatomic zone of the embryo called a neuromere, which is based on a segmented model of the embryonic nervous system. Unique patterns of gene expression determine the anatomic boundaries of each zone within the neural tube of the embryo.

Many of the genes within a specific zone share an identical base pair sequence called a homeobox (hox). The neuromeric zones can be mapped during the course of development by their hox and other zone specific genes.

Facial development is described as the formation, migration, coalescence, and interaction of separate genetically based developmental fields (Carstens, 2002).

Disruption of a neuromeric zone will result in abnormalities in the developmental field originating from that zone and will mechanically disrupt normal interactions with adjacent fields, resulting in field mismatch.

2.3 Aetiology of Cleft Lip and Palate

Oral clefts can occur as a result of teratogenic exposures, single-gene disorders, or chromosomal abnormalities and are classified as syndromic when they occur with other congenital defects as one of over 400 described syndromes (Gorlin, Cohen, & Hennekam, 2001). In approximately 50% to 70% of cases there is no recognized pattern of malformation and the cause of the disorder is unknown (Gorlin et al., 2001).

Such cases are classified as non-syndromic, and may either be isolated or non-isolated, depending on whether they occur with other congenital defects (Mitchell et al., 2002). Many syndromic clefts have simple Mendelian patterns of inheritance. Isolated clefts, however, are usually genetically complex traits (Lidral & Murray, 2004).

Molecular studies have shown that mutations in genes such as IRF6 (Kondo, Schutte, Richardson, Bjork, & Knight, 2002) and MSX1 (Mossey & Little, 2002; Salahshourifar, Halim, Wan Sulaiman, & Zilfalil, 2011) can cause orofacial clefts. The study done in Malay population in Kelantan also shown a contribution of MSX1 genes in aetiology of cleft lip and palate (Salahshourifar et al., 2011). In addition to genetic factors, a number of other risk factors have been identified during early pregnancy when the lip and palate are developing. Teratogens such as anticonvulsant drugs and corticosteroids (Park-Wyllie, Mazzotta, Pastuszak, Moretti, & Beique, 2000) as well as maternal smoking (Honein, Rasmussen, & Reefhuis, 2007), alcohol use (Romitti et al., 2007), and exposure to organic solvents and agricultural chemicals have all been associated with orofacial clefts (Shaw, Nelson, Iovannisci, Finnell, & Lammer, 2003). Vitamin deficiencies (Munger et al., 2004) and viral infections (Acs, Banhidy, Puho, & Czeizel, 2005) can also increase the risk of orofacial clefts.

2.4 Psycosocial Factors

Numerous studies have demonstrated the negative impact that orofacial clefts can cause on the child's psychosocial development. Children with orofacial clefts have lower self-concept scores than children without clefts (Broder & Strauss, 1989). They are also more likely to have learning disabilities, grade retention, and low school achievement (Broder, Richman, & Matheson, 1998).

Children with orofacial clefts are at risk for dyslexia related to fluency and naming speed (Richman & Ryan, 2003) and 30-40% of them have early reading problems (Richman, Wilgenbusch, & Hall, 2005). Children with orofacial clefts report more problems with social well-being when compared to children with dental caries (Locker, Jokovic, & Tompson, 2005). Adolescents with orofacial clefts often demonstrate social inhibition (Kapp-Simon & McGuire, 1997). Low social competence in teens with clefts can lead to feelings of loneliness and social anxiety (Pope & Ward, 1997).

Males with cleft lip and palate are more likely to have a midline brain anomaly and lower IQ than those without clefts (Nopoulos et al., 2001). Males with orofacial clefts are likely to have a smaller orbitofrontal cortex, which is associated with lower social functioning scores (Nopoulos et al., 2005).

2.5 Health-related Quality of Life

Quality of life is define as an “individual’s perception of their position in life in the context of the culture and value system where they live, and in relation to their goals, expectations, standards, and concerns.” Health-related quality of life is a patient-reported outcome that helps clinicians to assess how patients are affected by their illness or condition by quantifying a child or parent’s perspective of the child’s functional and psychosocial well-being. Measuring health-related quality of life is becoming an increasingly important mechanism for evaluating different aspects of disease management. Children with a chronic disease and poor health-related quality of life had average monthly healthcare costs that were twelve times higher than children who had a chronic disease but better health-related quality of life (Seid, Varni, Segall, & Kurtin, 2004).

Hence, health-related quality of life might be used to identify patients that could benefit from different treatment modalities or more aggressive preventative care. Increasing their health-related quality of life could potentially decrease their overall healthcare costs. Health-related quality of life can also help clinicians make more informed treatment recommendations as they are able to better understand the patient’s perspective and how the disease impacts their life (Ralstrom, 2009).

2.6 Health-related Quality of Life in Children with Orofacial Clefts

Recent health-related quality of life studies have improved our understanding of how orofacial clefts affect children. Children with visible facial differences have lower quality of life scores compared to children with no facial differences, but when compared to children with chronic disorders, their scores are similar, and slightly better in the area of family relationships (Topolski et al., 2005).

Children with orofacial clefts showed few differences in health-related quality of life scores, oral symptoms, and emotional well-being when compared to children with dental caries (Locker et al., 2005). Health professionals who work with children with orofacial clefts report these children have a slightly lower health-related quality of life (Wehby, Ohsfeldt, & Murray, 2006).

Adults with cleft lip and palate had significantly lower health-related quality of life scores in the subscales social functioning and emotional role, but not in the subscales physical functioning, physical role, bodily pain, general health, vitality, and mental health (Sinko et al., 2005). Adults with cleft lip and palate who desired further treatment had significantly lower health-related quality of life scores compared to those who did not desire further treatment in the subscales physical role and vitality, and trends toward significant differences.

One study (Damiano et al., 2007) used the pediatric quality of life survey and telephone interviews with parents to determine health-related quality of life in pre-adolescent (2-12 years old) children with non-syndromic oral clefts. This study had found that children with cleft lip and/or palate scored significantly lower than healthy children on the psychosocial health component scale. They found no difference on any of the pediatric quality of life scales when comparing children according to type of cleft. Higher household income was associated with higher total quality of life scores, as well as with high physical and psychosocial scores. A higher number of household members, a higher health status rating, and the mother's is not a single mother were also associated with higher scores. The parent's perception of how happy the child was with his or her facial appearance was associated with higher total and psychosocial health scores. Quality of the child's speech was highly correlated with the total, physical, and psychosocial health rating of the child. After controlling for demographics, children with less severe speech problems had higher overall health-related quality of life as well as higher physical and psychosocial health domain scores. Children with cleft palate had significantly higher health-related quality of life than children with cleft lip and palate. Both categories of younger children ages 2-4 and 5-7 had higher health-related quality of life than children who were 8-12 years old. The health-related quality of life was better among children with cleft lip and cleft lip and palate than among those with cleft palate until age 7, then declined for 8-12 year olds. Children with cleft palate had better health-related quality of life observed in the oldest age group. This suggests that the difference in outward appearance related to lip involvement has greater relative importance, as the child gets closer to adolescence.

Another study (Warschausky, Kay, Buchman, Halberg, & Berger, 2002) examined parent's perceptions of health-related quality of life in children ages 5 to 18 years with cleft and other craniofacial anomalies using the child health questionnaire. In this study, parents reported that children with cleft lip and/or palate had physical and psychosocial scale scores that fell within normal limits.

2.7 Oral Health-related Quality of Life

The impact of oral health on quality of life is usually referred to as oral health-related quality of life. Oral health-related quality of life is recognized as a multi-dimensional concept, comprising both the presence and absence of oral disease as well as psychological aspects of oral health (Gift, Atchison, & Dayton, 1997). Two major survey instruments have been developed to measure oral health-related quality of life in children: the child perceptions questionnaire and the child oral health impact profile.

The child perceptions questionnaire was developed to assess children's perception of the impact of oral disorders on physical and psychosocial functioning. Many studies have shown mixed results regarding the child perceptions questionnaire ability to detect oral health-related quality of life differences between children with orofacial clefts and controls. In one study (Jokovic et al., 2002), the child perceptions questionnaire was used for 11 to 14 year-old children, was found to be valid and reliable and able to discriminate oral health-related quality of life differences between children with orofacial clefts versus those in the orthodontic and pediatric dentistry groups. Internal reliability and retest reliability were excellent with Chronbach's alpha of 0.88 and Intraclass Correlation Coefficient (ICC) of 0.88. In another study (Jokovic et al., 2004), the child perceptions questionnaire was used for 8 to 10 year-olds, detected a slightly higher level of impact in the orofacial than in the pediatric dentistry group, but overall did not demonstrate discriminative validity with Intraclass Correlation Coefficient (ICC) of 0.67.

2.8 Child Oral Health Impact Profile

The child oral health impact profile is a 38 questions survey designed to measure self-reported oral health-related quality of life in school age children ages 8-14 years. Its readability score has been assessed at a 3.5 grade reading level, making it suitable for children in third grade or higher. The child oral health impact profile contains 34 items to measure oral health-related quality of life in five domains: Oral Health, Functional Well-Being, Social-Emotional Well-Being, School Environment and Self Image. It also contains 4 items to assess Treatment Expectations and Global Health of the child.

The questionnaire contains items to assess both positive and negative aspects of oral health-related quality of life following the World Health Organization concept that health is more than the absence of disease (Broder, 2007; Broder, McGrath, & Cisneros, 2007; Broder & Wilson-Ganderson, 2007).

Broder et al. established reliability and validity of the child oral health impact profile in a study of 523 children of diverse ethnicities recruited from pediatric dentistry clinics, orthodontic clinics, craniofacial clinics and elementary schools (Broder & Wilson-Ganderson, 2007). It was found that the child oral health impact profile had excellent scale reliability and test-retest reliability with Cronbach's alpha of 0.91 and Intraclass Correlation Coefficient (ICC) of 0.84 and there was no significant change in child oral health impact profile scores over time. The survey's discriminate validity (ICC = 0.84) was supported by the finding of significant differences in child oral health impact profile scores between the different clinical groups, with the craniofacial patients reporting lower oral health-related quality of life than pediatric patients, orthodontic patients or non-clinical subjects. Within the orthodontic group, children with more severe orthodontic problems were also found to have lower child oral health impact profile scores. Hence, the child oral health impact profile was sensitive (ICC = 0.84) to differentiate between groups as well as to differentiate in disease severity impact for a single clinical condition (Broder & Wilson-Ganderson, 2007). Convergent validity testing found that a significant positive relationship between Global Health and child oral health impact profile scores existed (ICC = 0.82). The acceptable reliability and validity with Chronbach's alpha of 0.91 demonstrated preliminary support for the child oral health impact profile as a useful psychometric tool (Slade & Reisine, 2007).

COHIP questionnaire has also been used to assess the Asian population in Korea and was found to be reliable and valid (Ahn et al., 2012). Cleft evaluation profile have been translated and used in the study of cleft lip and palate outcome measures in Hospital Universiti Sains Malaysia (Noor & Musa, 2007).

2.9 Craniofacial Patients and Child Oral Health Impact Profile

Broder and Wilson-Genderson (2007) compared child oral health impact profile scores among children recruited from pediatric, orthodontic, and craniofacial clinics. Their sample included 157 pediatric dentistry patients, 152 orthodontic patients, 110 patients with craniofacial anomalies, and 104 community-based participants. They found that the craniofacial group reported greater negative impact on their oral health-related quality of life than either the general pediatric or the orthodontic patients. The craniofacial patients had significantly lower scores for overall child oral health impact profile, as well as the subscales functional well-being and school environment. The craniofacial group also had lower scores, which approached significance for the subscale Social/emotional well-being.

(Noor & Musa, 2007) used Cleft Evaluation Profile (CEP) in sixty cleft lip and palate patients (12 to 17 years of age) from Hospital Universiti Sains Malaysia and find patients were teased because of their clefts and felt their self-confidence was affected by the cleft condition. The patients were frequently teased about cleft-related features such as speech, teeth, and lip appearance and they showed a significant level of satisfaction with the treatment provided by the cleft team. The features that were found to be most important for the patients were teeth, nose, lips, and speech. (Noor & Musa, 2007) concluded that cleft lip and/or palate patients were teased because of their clefts, and it affected their self-confidence and the Cleft Evaluation Profile (CEP) is a reliable and useful tool to assess patients' level of satisfaction with treatment received for cleft lip and/or paiate and can identify the types of cleft-related features that are most important for the patients.

2.10 Timing of Surgery for Cleft Lip and Palate

The timing of cleft surgery is controversial due to the opposing factors of speech and growth. It is generally acknowledged that early cleft repair results in a more normal speech pattern. It is also accepted that delayed cleft surgery resulted in more normal facial growth (Rohrich & Byrd, 1990). This fact is also supported by the recent studies by (Kobus & Kobus-Zaleśna, 2013) that the speech pattern is normal in 90% of the patients with early cleft palate surgery and similar observation of more normal facial growth in patients with delayed cleft palate surgery.

Although the timing of palate surgery is most frequently debated, cleft lip repair is also implicated as a cause of facial growth impairment. Further debate concerns the influence of an inherent maxillofacial growth deficiency, which may vary according to cleft type.

The modern trend is for early complete closure of the lip and palate to achieve the best speech outcome with minimal growth disturbance. Abnormal speech patterns are more difficult to correct later than facial growth deficiencies, which can be treated surgically.

Unfortunately, surgical treatment protocols vary widely among and within cleft centers, and outcome interpretation is extremely difficult due to the presence of uncontrolled confounding variables, numerous anatomic subtypes, multiple interventions, different surgical techniques, multiple surgeons, inadequate follow-up, small sample size, and low disease prevalence.

Most surgeons agree to perform cleft lip repair when the child is about 3 months of age. Early cleft palate repair increases the likelihood of normal speech development. Normal speech development is contingent on velopharyngeal competency and soft palate closure. Speech development begins with preparatory oropharyngeal movements in utero and progresses to infant babbling in the first 6 months of life. Consonant-vowel sequences normally emerge between 6 and 9 months (Smith & Oller, 1981). Infants with cleft palate have inadequate velopharyngeal valving and abnormal lingual-palatal contact which results in maladaptive compensatory articulation patterns during early speech development. To prevent the development of abnormal speech patterns, most speech pathologists advocate early palate repair.

The frequency of compensatory articulations in children who underwent early and late palate repair was compared, early is before 12 months, and late is later than 12 months. Compensatory articulations were significantly fewer in the early repair group (Dorf & Curtin, 1982). The benefit of early palatal closure in term of speech outcome was later proves by (Lohmander, Friede, & Lilja, 2012) and supported by (Willadsen, 2012).

The speech of children who were operated on at 3–7 months of age versus 12–18 months of age were compared and children whose palates were repaired earlier had better speech and needed fewer secondary pharyngoplasties than those whose surgeries were delayed beyond the first 12 months (Randall, 1983). In a long term survey by (Lohmander et al., 2012) also observed that the need of secondary pharyngoplasties procedure were markedly reduced when the palatal closure was performed early whih is less than 12 months.