STANDARDS OF CARE BY MULTIDISCIPLINARY TEAM FOR CLEFT LIP AND PALATE PATIENTS IN MALAYSIA (A PILOT STUDY)

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FACULTY OF DENTISTRY
UNIVERSITY OF MALAYA
KUALA LUMPUR

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THESIS SUBMITTED IN FULFILMENT OF THE REQUIREMENTS FOR THE DEGREE OF MASTER OF CLINICAL DENTISTRY (ORAL AND MAXILLOFACIAL SURGERY)

FACULTY OF DENTISTRY
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ORIGINAL LITERARY WORK DECLARATION

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Title of Research Report: Standards of Care by Multidisciplinary Team for Cleft Lip and Palate Patients in Malaysia (A Pilot Study)

Field of Study: Oral and Maxillofacial Surgery

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ABSTRACT

Introduction: The management of cleft lip and palate (CLP) patients are complex and require coordinated care of multidisciplinary team (MDT). In Malaysia, cleft combined clinic (CCC) only available in small numbers and situated in urban centres. To date, there were limited number of research conducted to investigate standard of care for CLP patients among CCC in Malaysia. **Objective:** The aims of this study is to investigate the standards of care provided for management of cleft lip and palate (CLP) patients in Malaysia in comparison to the recommendations made by Clinical Standards Advisory Group (CSAG) UK 1998. **Methods:** This is a cross sectional questionnaire-based survey study. The self-administered postal questionnaire comprises of few components that was related to the cleft team services which include overall cleft services, team formation, team members, clinical sessions & infrastructure and audit practice. **Results:** All questionnaires were send to three university hospitals which provide cleft services. It was found that the composition of each cleft team varies through the MDT model in every centre. Oral & maxillofacial surgeon (OMFS) and orthodontist were the only specialties that attended most of the time in CCC in all cleft teams. All centres provided complete treatment for cleft patients starting from baby until adulthood. There was variation in surgeons that involved in primary lip and palate repair while alveolar bone graft (ABG) surgery was done mainly by OMFS. None of the cleft team used database for their record keeping and actively involved in clinical audit program. **Conclusions**: Management for CLP patients among university hospital in Malaysia was provided through a MDT model care as recommended by CSAG UK 1998 with some of the recommendations fulfilled by the cleft team. However, improvement are needed especially in the area of forming a database for record keeping and having a routine clinical audit program.

KEYWORDS: Cleft lip and palate, Clinical Standards Advisory Group, Multidisciplinary team

ABSTRAK

Pengenalan: Pengurusan pesakit rekahan bibir dan lelangit (RBL) adalah kompleks dan memerlukan penjagaan khusus dan diselaraskan melalui sepasukan multidisiplin. Di Malaysia, Klinik Klef Bersepadu (KKB) hanya terdapat dalam bilangan kecil dan terletak di pusat bandar. Sehingga kini, terdapat kajian yang terhad dijalankan untuk menyelidik ukuran penjagaan bagi pesakit RBL di Malaysia. **Objektif:** Matlamat kajian ini adalah untuk mengkaji amalan semasa bagi pengurusan pesakit RBL di Malaysia dan pematuhan terhadap cadangan yang dibuat oleh Kumpulan Penasihat Piawaian Klinikal (KPPK) UK 1998. **Kaedah:** Ini adalah kajian borang kaji selidik lintang. Borang kaji selidik yang dihantar terdiri daripada beberapa komponen yang berkaitan dengan perkhidmatan pasukan klef. Data yang dikumpul adalah berkaitan tentang perkhidmatan klef, pembentukan pasukan, ahli pasukan, sesi & infrastruktur klinikal, audit, penyimpanan rekod, rawatan yang disediakan dan kes rujukan yang diterima oleh pasukan klef. **Keputusan:** Semua soal selidik telah dikembalikan dari tiga hospital universiti yang menyediakan perkhidmatan klef. Komposisi setiap pasukan klef melalui model pasukan multidisiplin adalah pelbagai. Pakar bedah mulut & maksilofasial dan ortodontik adalah satu-satunya kepakaran yang menghadiri kebanyakan masa di KKB dalam semua pasukan klef. Semua pasukan memberikan rawatan lengkap untuk pesakit klef bermula dari bayi sehingga dewasa. Terdapat variasi dalam pakar bedah yang terlibat dalam pembaikan bibir dan lelangit manakala pembedahan tulang pinggul dilakukan terutamanya oleh pakar bedah mulut & maksilofasial. Tiada pasukan klef yang menggunakan pangkalan data untuk menyimpan rekod mereka dan terlibat secara aktif dalam program audit klinikal. Kesimpulan: Pengurusan untuk pesakit KKB di kalangan hospital universiti di Malaysia telah disediakan melalui penjagaan model pasukan multidisiplin seperti yang disyorkan oleh KPKK UK 1998 dengan beberapa cadangan yang dipenuhi oleh pasukan klef. Walaubagaimanapun, penambahbaikan diperlukan

terutamanya dalam bidang membentuk pangkalan data bagi penyimpanan rekod dan mempunyai program audit klinikal yang rutin.

KATA KUNCI: Rekahan bibir dan lelangit, Kumpulan Penasihat Piawaian Klinikal, Pasukan Multidisiplin

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LIST OF SYMBOLS AND ABBREVIATIONS

ABG	:	Alveolar bone graft
BCLP	:	Bilateral cleft lip and palate
CLAPAM	:	Cleft lip and palate association of Malaysia
CCC	:	Cleft combined clinic
CL	:	Cleft lip
CLP	:	Cleft lip and palate
CL/P	:	Cleft lip with or without palate
CSAG	:	Clinical Standards Advisory Group
CP	:	Cleft palate
ENT	:	Ear, nose and throat
IIUM	:	Islamic International University Malaysia
MDT	:	Multidisciplinary team
MOH	:	Ministry of Health
MOHE	:	Ministry of Higher Education
NICE	:	National International Clinical Excellence
OMFS	:	Oral and Maxillofacial Surgeon
SLT	:	Speech and Language Therapist
UCLP	:	Unilateral cleft lip and palate
UK	:	United Kingdom
UKM	:	Universiti Kebangsaan Malaysia
UKMMC	:	Universiti Kebangsaan Malaysia Medical Centre
UM	:	University of Malaya
USM	:	University Science of Malaysia

LIST OF APPENDICES

CHAPTER 1: INTRODUCTION

1.1 Background

Cleft lip with or without palate (CL/P) deformity has become a major public health problem affecting one in 700 to 1000 births around the world (Murray, 2002). CL/P is considered one of the most common birth defects that possess significant medical, psychological, social, and financial implications on the affected individuals and families (Allam, Stone, & Windsor, 2014). In addition to the aesthetic disfigurement, a child with CL/P suffers substantial functional morbidity such as restricted maxillofacial growth, speech anomalies, difficulty in swallowing and feeding, hearing loss and/or recurrent ear infections.

Although not generally life-threatening, living with a cleft creates with a significant strain on health (Sinno et al., 2012). The treatment needs of children with CLP are numerous, complex, requiring specialized and coordinated care of a multidisciplinary team (MDT). Therefore, dealing with all its elements and making all the best choices for the patient is hard for a clinician. For this reason, a team approach is highly recommended.

A key element of this protocol is the availability of a Cleft Combined Clinic (CCC) to provide multi-disciplinary care to cleft patients. Treatment aims are to restore as normal anatomy (face and dentition) and normal function (speech, feeding and hearing) and to encourage normal physical and psychological development (Kasten et al., 2008). The interaction and consultation between various disciplines provides care for the cleft patients that can be systematically and comprehensively planned as well as enhances understanding of the possibilities and limitations of various treatment approaches (Ghani et al, 1996).

Controversy abounds with regard to the most appropriate methods of organization and management for cleft patients and large volume of literature continues to be produced by teams and individuals who advocate particular management regimes (Asher-McDade & Shaw, 1990). It is also evident that wide range of surgical techniques exist to correct this anomaly, but with no clear-cut guidelines for optimal timing or method (Atack, Hathorn, Mars, & Sandy, 1997).

Specific to Malaysia, CCCs are available only in a very small number of urban centres and treatment of cleft patients is done in main hospitals with only a few disciplines involved with no standardized protocol and recall appointments for further follow-up. Hence, a number of patients are left untreated or partially treated until adulthood (Ghani et al, 1996). To date, there are limited or almost no research has been conducted to investigate the standard of care for CLP patient among the CCC in Malaysia. Therefore, the purpose of this study is to evaluate the standards of care provided by the multidisciplinary cleft teams' in hospital of Malaysia. In the end, it is hoped that this project will provide proposals for the existing CCCs to be made more effective, provide the guidelines for the setting up of CCCs in more hospitals across Malaysia and the establishment of Cleft Registry in Malaysia.

1.2 Aim and Objectives

1.2.1 Aim

To compare the standards of care provided by multidisciplinary teams for cleft lip and palate patients in Malaysia to the recommendations made by Clinical Standards Advisory Groups (CSAG) UK 1998

1.2.2 Objectives

i. To investigate the structure of the CCC (team composition, clinical sessions & infrastructure
ii. To identify clinical audit practice
iii. To determine method of decision making and record keeping
iv. To identify range of treatment provided for cleft lip and palate in various centres
v. To compare all data collected with CSAG UK 1998

CHAPTER 2: LITERATURE REVIEW

2.1 Cleft lip and palate epidemiology

A cleft lip and palate is one of the most common facial birth defect, estimation worldwide is about one in every 700 babies. It depends on the geographic origin, racial, ethnic background and socioeconomic status (Mossey et al, 2009). Study done by Das et al 1995 reported that, among the ethnic groups, Native American and Asians have the highest prevalence (14/10 000 live births), this is followed by European descent or the Whites group (10/10 000 live births) and the least one is the African Americans populations (4/10 000 live births). For cleft palate only, there is no difference between ethnic groups. It is approximately 5/10 000 live births. There is a gender differences for cleft lip and palate. The ratio is 2:1 male to female involving the cleft lip with or without cleft palate and 1:2 ratio male to female when the cleft involving the palate only. The reason of this difference is, because the palatine shelves in girls are close 1 week later compare to boys.

2.2 Cleft lip and palate aetiology

Cleft lip and palate is a major congenital structural anomaly that is notable for significant lifelong morbidity and complex aetiology. The extensive psychological, surgical, speech and dental involvement emphasize the importance of understanding the underlying causes. In addition, cleft can be divided into non-syndromic and syndromic group. The majority of cleft is non-syndromic or isolated anomalies. It consists of 70% cases cleft lip with or without palate and 50% cases of cleft palate (CP) only (Murthy & Bhaskar, 2009). However, the others study reported that the prevalence are between 20% to 30% only (Calzolari et al., 2007; Monica Rittler et al., 2011; G. M. Shaw, Carmichael,

Yang, Harris, & Lammer, 2004). The most common associated anomalies with CL/P are congenital heart disease, limb and vertebral (Genisca et al., 2009; Ma'amon & Abu-Hawas, 2008).

Until now, the cause of non-syndromic cleft still unknown and there is no exact mechanism that had been discovered. It is believed, the cause is multifactorial. Any disturbance in the developmental sequence can result in cleft formation. The aetiology involves both genetic and environmental factors, which is highly complex with the molecular basics remains largely unknown (Brito et al., 2012).

The association between maternal smoking and CL/P has been assessed in many studies and a meta-analysis of these studies suggests a positive association (Little, Cardy, & Munger, 2004). Different studies have been conducted worldwide to evaluate the smoking association and often resulting in varying prevalence rate as high as 20% (Honein et al., 2007). (Chevrier et al., 2005) and (Bille et al., 2007) reported that, maternal alcohol consumption during early pregnancy (first trimester) is a well-known cause of foetal alcohol syndrome (FAS) with positive association with CL/P.

The relationship between maternal dietary intake and embryonic/foetal nutrition is not fully understood. Nutrient supply to the embryo can be influenced by a number of adaptive physiological changes that can occur during pregnancy, including alteration in maternal internal absorption (Hozyasz, 2010). Maternal intake of vitamin A supplements more than 10,000 IU has been shown to cause CL/P in additions to other malformations. It is suggested that, vitamin A intoxications results in a multitude of alterations in embryos and several genes involved in palate development interact or can be modified in expression by vitamin A and its analogues (Azaïs-Braesco & Pascal, 2000).

Folate, a general term for various forms of this naturally occurring B-vitamin, and folic acid, its oxidized and more bioavailable form found in multivitamins and food supplements, play important roles in the synthesis and methylation of DNA as well as in the metabolism of amino acids and their by-products, such as homocysteine (Forges et al., 2007).Many studies have been performed in an attempt to determine the role of folate in the aetiology of CL/P or CP only. A number of intervention studies suggesting a protective effect of folic acid on the recurrence of oral clefts have been performed (Tolarova & Harris, 1995). However, the data that suggest folic acid supplementation can reduce the incidence of cleft remain controversial (Hayes et al., 1996).

In the other side, in a meta-analysis study reported the usage of maternal use of multivitamin supplements in early pregnancy can decrease the birth prevalence of orofacial cleft cases up to 25% (Johnson & Little, 2008). This is proven by the interaction between the multivitamin and maternal can reduce the hyperthermia during pregnancy, which lead to reduce risk the of cleft associated with hyperthermia (Botto, Erickson, Mulinare, Lynberg, & Liu, 2002).

In addition to environmental and genetic factors, there is an association between CL/P and consanguineous marriages (Leite & Koifman, 2009). Consanguinity is considered a significant factor in autosomal recessive diseases; it has also been associated with congenital anomalies such as hydrocephalus, polydactyly and CL/P (Mónica Rittler, Liascovich, López-Camelo, & Castilla, 2001). The risk of congenital is higher in subjects born of first degree consanguineous parents compared with those of non-consanguineous marriages (Kanaan, Mahfouz, & Tamim, 2008). For that reason, consanguineous marriages should be discouraged.

Maternal age is also considered as a risk factor for numerous chromosomal alterations. However, there is no consensus whether it represents a risk factor for CL/P. A study carried out in California population showed that women older than 39 years old had twice the risk of having a child with CL/P when compare to the age between 25 to 29 years old (G. M. Shaw, Croen, & Curry, 1991). Another study in Asian population, showed a relationship between advanced maternal age and bilateral CLP among males and CLP among females (Cooper et al., 2000). Current studies show that there is an association between maternal age and an increased risk for cleft lip and palate; however, the paternal age, pregnancy order and inter-pregnancy interval were not significant (Martelli et al., 2010).

Studies of gene-environment interaction effects have become increasingly important for complex traits such as oral cleft, whose aetiology probably involves both genes and environmental factors. With the advent of the genomics era and advances in both quantitative and molecular analysis techniques, there have been great improvements in the identification of causative genetic mutations and associations underlying syndromic forms of CL/P as shown in Table 1 (Dixon, Marazita, Beaty, & Murray, 2011). On the other hand, there is currently little progress in identifying and understanding of the genetic aetiology of isolated (non-syndromic) CL/P cases as shown in Table 2 (Marazita & Mooney, 2004)

Table 1: Cleft associated syndromes in which the mutated gene has been identified

Cleft lip ± cleft palate (CL/P)	Cleft palate (CP)
Deafness and dystonia — ACTB	Oculofaciocardiodental — BCOR
Familial gastric cancer and CLP — CDH1	CHARGE — CHD7
Craniofrontonasal — EFNB1	Stickler type 1 — COL2A1
Roberts — ESCO2	Desmosterolosis — DHCR24
Holoprosencephaly — GLI2	Smith–Lemli–Opitz — DHCR7
Hydrolethalus — HYLS1	Miller — DHODH
Van der Woude/popliteal pterygium — IRF6	Craniofrontonasal — EFNB1
Gorlin — PTCH1	Crouzon — FGFR2
CLP, ectodermal dysplasia — PVRL1	Apert — FGFR2
Holoprosencephaly — SHH	Otopalatodigital types 1 and 2 — FLNA
Branchiooculofacial — TFAP2A	Hereditary lymphedemadistichiasis — FOXC2
Holoprosencephaly — TGIF1	'Orofacialdigital' — GLI3
Ankyloblepharonectodermal dysplasiaclefting — TP63	Van der Woude/popliteal pterygium — IRF6
Tetraamelia with CLP — WNT3	Andersen — KCNJ2
	Cornelia de Lange — NIPBL
Midline cleft lip	Xlinked mental retardation — PQBP1
	Isolated cleft palate — SATB2
Opitz G/BBB — MID1	Diastrophic dysplasia — SLC26A2
Orofacialdigital type I — OFD1	Pierre Robin — SOX9
	DiGeorge — TBX1
	Treacher Collins — TCOF1

Table 2: Currently reported gene-environment interaction in cleft lip and palate

Gene-environment interaction in cleft lip and palate	
TGFA/Smoking	TGFB3/Alcohol
TGFA/Alcohol	RARA/Smoking
TGFA/Vitamins	MTHFR/Vitamins
MSX1/Smoking	P450/Smoking
MSX1/Alcohol	GST/Smoking
TGFB3/Smoking	EPHX1/Smoking

2.3 Multidisciplinary approach for treatment CLP

The currently accepted model for delivery of care for CL/P in the most appropriate way is through the multidisciplinary (MDT) cleft team. This is a group of an individual from different specialist background who work closely together, not only to bring each specialist's particular expertise to the patient in the optimum way, but also to develop an understanding of the requirements and specialist skill of the other team members to enhance the delivery of the total package (D Hodgkinson et al., 2005). It has been suggested that such an integrated system of delivery of care enables the individuals within the team to function in an interdisciplinary way so that all aspects of health care for the cleft condition can be delivered in a seamless way.

The available evidence suggests that there is a strong relationship between positive treatment outcome and the availability of centralized care by a high quality dedicated cleft team (Allam et al., 2014). The management of the CL/P patients in many parts of the world is undertaken by a number of the surgical specialties including OMFS, plastic surgery, ENT and even general surgery. OMFS are by virtue of the considerable training, uniquely qualified to manage both hard and soft tissue conditions affecting the orofacial region. In contrast, the majority of plastic surgeon is not trained in hard tissue surgery. Therefore, the collaboration of specialties in the management of CL/P patients is paramount with optimum patients care and improved outcome being the ultimate goal (Brennan et al., 2001).

Because of the complexity of sequelae of CL/P, comprehensive and coordinated care of MDT for the affected patients is needed. The specialties involved should include OMFS, orthodontist, plastic surgeon, ENT surgeon and speech & language therapist. The

others specialties such as audiologist, paediatric surgeon, clinical geneticist, paediatric dentist, psychiatrist, specialist cleft nurse and social worker have been mentioned in the literature but their services in the MDT are not universal (W. C. Shaw et al., 1992).

It is vital for this team to work together as a team to balance the many important factors in managing the care of a child who has a cleft. Team members must communicate effectively among themselves and also with the child and parents. Individuals on the team must respect one another's opinion and be flexible in planning and carrying out the best treatment for the child. Other than that, periodic evaluation and follow up is necessary to assess the effectiveness of the previous planning and surgery (Jones, Sadove, Dean, & Huebener, 2011). Care of the patient within a multidisciplinary team requires the development of a protocol of care or care pathway to enable the team members to function most effectively and to maximize the benefits of this system to the patient

2.4 Background Clinical Standards Advisory Group (CSAG) United Kingdom (UK) 1998

In the early 1990's, UK Health Ministers had asked the CSAG to investigate and make recommendations on the care for patients with CL/P, in response to concerns which had emerged about the variations between the standards of treatment for UK patients and those elsewhere in Europe. There are 2 earlier studies that had raised the concerns about the quality of cleft care in UK. The first study by Mars et al (1987) as cited by Sandy et al (2001) had develop a method for assessing dentolaveolar relations which demonstrated shortcomings in a comparison between UK and Norwegian cleft centre.

The second initiative reported by Shaw et al (1992) as cited in Sandy et al (2001) was an inter centre comparison of outcomes by six European centres. The study showed, two UK centres who participated in the study were the weakest on almost every aspect of care, including organization and outcomes such as dental arch relations. It was suggested at that time, these poor outcomes were related to the fact that UK centres had little standardization of technique, the surgeons were operating intermittently on as small numbers of patient and the treatment was provided in an un coordinated manner by isolated general surgeons, speech therapist and occasionally dentist. Other than that, the large number of cleft team existed in UK were poorly organized (Williams, Shaw, & Devlin, 1994).

Mindful of this, the UK Health Minister set up the CSAG commissioned as an independent source of expert advice to the Health Ministers to improve the clinical standards and services outcomes services for CL/P patients. In 1998, the CSAG UK study results were confirming poor outcomes of surgery for children born with CLP and the CSAG committee made few recommendations to the UK government (Sandy et al., 1998). Currently, CSAG 1998 recommendations have been broadly implemented in all

centralized cleft service in UK (Scott et al., 2014). The CSAG UK 1998 comes out with recommendations in relation to the planning of CLP services for the UK. The recommendations are:

i. Cleft care should be provided by 8 – 15 services

ii. Cleft care should be provided through MDT

iii. Each MDT "must have" specialty members

- Primary cleft surgeon
- Secondary cleft surgeon
- Orthodontist
- Speech and language therapist
- Paediatrician
- ENT surgeon / audiology physician
- Coordinator / manager

4) The MDT should liaise closely with (all of the following)

- Special nurse
- Psychologist
- Genetics
- Paediatric dentist
- Restorative dentist

5) Each service should provide care for 100 -120 new cleft cases (of all type) per year

6) Each primary surgeon should operate on at least 40-50 cases annually

7) Each cleft service should provide paediatric inpatient care

8) Each cleft service should provide outpatient care in a paediatric setting

9) Each cleft service should be involved in a program of clinical audit, to monitor outcomes for their service and in comparison with other service

10) Each cleft service should have an active program of clinical research

The CSAG report emphasized the successful management of children born with CL/P requires a MDT approach and highly specialized treatment from birth to teens and early twenties. It is also suggested that treatment of clefts in the UK would be better organized if services, particularly primary surgery could be concentrated so that, a smaller number of surgeons would be involved in primary repairs surgery. Although high volumes are not in themselves a guarantee of high-quality outcomes, the quality of outcome can be satisfactory assessed only in a unit with large caseload. The CSAG result showed some beneficial effects of volume on outcome. Therefore, all surgeons involved in the care of cleft patients recommended performing surgery at least 40 – 50 annually.

Centralization needs to be balanced with population needs and accessibility. The report found that, there are about 57 cleft services in UK. With the coordination of services, expertise and resources were concentrated and this will enable a team to treat a high volume of patients to a standardized protocol, long term follow up and to be able to compare results with other centres. Centralized care in regional centres has also been demonstrated to improve results and lead to fewer revisions. Recommendations also made on young specialists (surgeons, orthodontists, speech and language therapist and etc) of the cleft team. They must be able to undergo a properly structured cleft care training program. Training program for all specialist cleft clinicians should be approved only in cleft centres where high volume and high-quality clinical expertise is available. This will help the team to manage cleft patients through the holistic approach. Other than that, the

CSAG report recommends the cleft surgical site needs to be adjacent to other specialized paediatric services, so that adequate paediatric in-patient facilities, paediatric anaesthetists and paediatric nurses are available.

CHAPTER 3: MATERIAL AND METHODS

3.1 STUDY DESIGN

This study is a cross sectional survey which included 3 University Hospitals in Malaysia namely:

i. Cleft Combined Clinic, Faculty of Dentistry, University of Malaya (UM)
ii. Cleft Combined Clinic, Department of Oral & Maxillofacial Surgery, Universiti Kebangsaan Malaysia Medical Centre (UKMMC)
iii. Cleft Combined Clinic, School of Dental Sciences, University Science of Malaysia (USM)

These centres were chosen as they fulfil all the requirements needed in this study, were willing to participate and ethical approval were obtained from all centres. The inclusion and exclusion criteria for this study are:

i. Inclusion criteria: CCC established at least 1 year (before 31st December 2016)

ii. Exclusions criteria: CCC composed of less than 2 disciplines

3.2 MATERIAL

3.2.1. QUESTIONNAIRE

The questionnaire used in this research was adopted from a series of specialty-specific questionnaires based on data collected from the 1998 CSAG Study (Scott et al., 2014). This questionnaire was then modified to suit the practice in Malaysia, validated and pre tested prior application to these centres. (Appendix A and B)

This included the general data and all the parameters needed for this study. The parameters were divided into 7 components which namely:

a) Team formation and Service structure

b) Team members

c) Clinical sessions and infrastructure

d) Audit

e) Combine clinic decision making and record keeping

f) Treatment provided

g) Referrals

These parameters were to investigate mainly on the surgical and medical specialties involved, infrastructure as well as patients load on each centre. Each centre were also inquired on records and availability of audit practiced by the team.

3.3 Data Collection Method

The questionnaires were distributed to all CCC involved in this study via email and conventional mail, directed to the coordinator or representative of the CCC. Feedback of the questionnaires were obtained within 1 month after distribution.

3.4 Data Analysis

Data obtained from these centres were analysed using SPSS Version 20.0. Descriptive analysis was performed on most data.

3.5 Ethical approval and funding

This research had received ethical approval and funding from:

i. Medical Ethics Committee, Faculty of Dentistry University of Malaya
[Reference Number: DF OS1702/009(P)] (Appendix C)

ii. Human Research Ethic Committee (HREC), University Science of Malaysia
[Reference Number: USM/JEPeM/17080361] (Appendix D)

iii. Research Ethic Committee, The National University of Malaysia
[Reference Number: UKM PPI/111/8/JEP-2018-220] (Appendix E)

iv. Dental Research Management Centre (DRMC), Faculty of Dentistry University of Malaya
[Reference Number: DPRG/02/17]

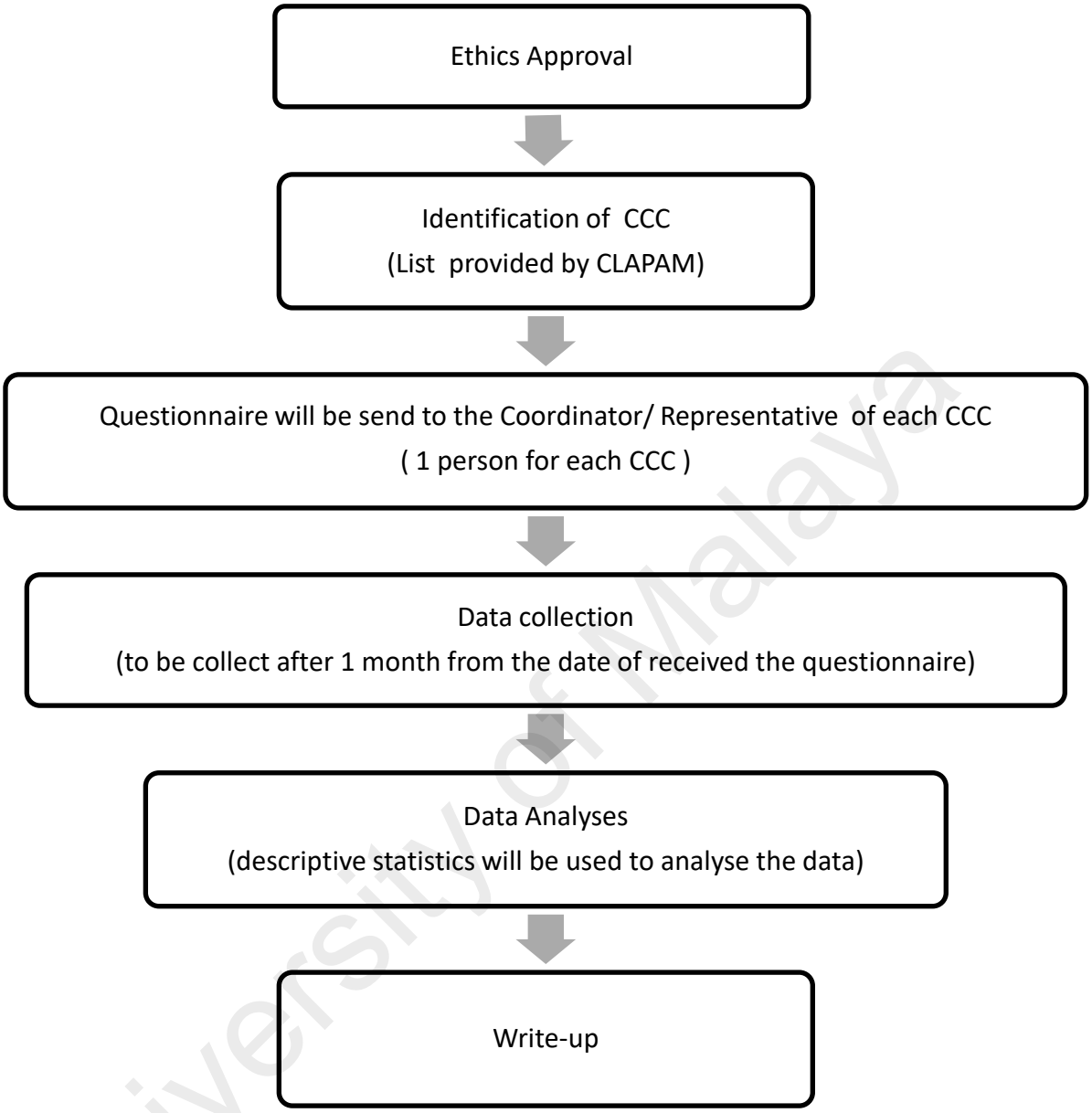

Figure 1: Flow chart of the study methodology

CHAPTER 4: RESULTS

4.1 Team formation

Table 4.1: Team formation

	UM	UKM	USM
Year of establishment	2000	2013	2012
Year of centralized services	2000	2013	2012

Table 4.1 shows the history of the CCC for each university involved in this study. There are only three university hospitals in Malaysia that provide cleft team. The questionnaires were distributed to all of the cleft services with 100% respond rate. UM has been the pioneer of the formation of the CCC, followed by USM and UKM.

4.2 Team members

Table 4.2.1: MDT members and CSAG recommendations in CCC

Specialty	UM	UKM	USM	CSAG
Audiologist	✓	✓	X	X
Clinical Director	✓	✓	✓	✓
Clinical Geneticist	✓	X	X	✓
ENT Surgeon	✓	✓	X	✓
Medical Photographer	✓	X	X	X
OMFS	✓	✓	✓	✓
Orthodontist	✓	✓	✓	✓
Paediatric Dentist	✓	✓	✓	✓
Paediatrician	✓	X	X	✓
Plastic Surgeon	✓	✓	X	✓
Psychiatrist	X	X	X	✓
Restorative Dentist	X	X	X	✓
SLT	✓	✓	✓	✓
Special cleft Nurse	X	X	X	✓
Total	**11**	**8**	**5**	**12**

Table 4.2.1 shows the specialty in the cleft team of each university. Almost all of the universities followed the CSAG 1998 recommendations. UM cleft team has the biggest number of specialties, followed by UKM and USM. The only cleft team that having clinical geneticist and paediatrician was UM cleft team but lacking in involvement of the psychiatrist, restorative dentist and special cleft nurse.

Table 4.2.2: Frequency of attendance at the combine clinic and years of experience for each specialty

	Attendance at Combine Clinic			**Years of Experience**
UM	Most of the time (>50%)	Some of the time (50-30%)	Rarely/ Never (<30%)	
Audiologist		✓		>10 years
Clinical Geneticist		✓		>10 years
ENT Surgeon	✓			>10 years
Medical Photographer	✓			>10 years
OMFS	✓			>10 years
Orthodontist	✓			>10 years
Paediatric Dentist	✓			
Paediatrician		✓		>10 years
Plastic	✓		✓	5 years
SLT		✓		>10 years
UKM				
Audiologist	✓			8 years
ENT Surgeon	✓			>10 years
OMFS	✓			8 years
Orthodontist		✓		>10 years
Paediatric Dentist		✓		5 years
Plastic Surgeon	✓			5 years
SLT	✓			5 years
USM				
OMFS	✓			>10 years
Orthodontist	✓			>10 years
Paediatric Dentist			✓	5 years
SLT	✓			5 years

The frequency of attending MDT clinic and years of experience for each specialty is shown in Table 4.2.2. It shows that, the "must have" specialties in all of the three team attend the MDT clinic most of the time as recommended by the CSAG UK 1998. UM having the most number of specialties, more than half of the specialty presents at all the time during CCC with more than 10 years of experience handling the CL/P patients and SLT, Paediatrician and Audiologist attended half of the time.

4.3 Treatment Provided

Table 4.3.1: Type and timing of treatment provided by cleft team

Treatment	Average age		
	UM	UKM	USM
Primary lip repair	3 months old	3 months old	3 months old
Primary palate repair	9 months old	6 months old	9 months old
ABG	9 years old	12 years old	9 years old
Speech therapy	<1 year old	<1 year old	<1 year old
Hearing assessment	<1 year old	<1 year old	<1 year old
Lip revision	6 years old	17 years old	17 years old
Rhinoplasty	17 years old	17 years old	17 years old
Orthodontic treatment	9 years old	9 years old	9 years old
NAM	< 1 month	< 1 month	< 1 month
Orthognathic surgery	18 years old	18 years old	21 years old

Table 4.3.1 shows the type of treatments that provided by the cleft team in the hospital university. The surgery for primary lip repair was done at the average of 3 months by all of the team. Meanwhile, the average time for primary palate repair was between the age of 6 – 9 months and the alveolar bone graft was done at the age of 9 to 12 years old. Speech therapy and hearing assessment was done at the age of less than 1 year by all

team. UM cleft team did the lip revision at earlier age, at around 6 year old compared to UKM and USM team where the lip revision was done at the age of 17 and above. Treatment for rhinoplasty and orthodontic also provided by all team and there was no difference between the average of age among the team. Orthognathic surgery performed when orthodontic team had completed the alignment of the teeth and usually done at the age of 18 years old and above.

Table 4.3.2: Specialty involved in surgery for CL/P patients

Treatment	Specialty		
	UM	**UKM**	**USM**
Primary lip repair	OMFS/Plastic	Plastic	OMFS/Plastic
Primary palate repair	OMFS/Plastic	Plastic	OMFS/Plastic
ABG	OMFS	OMFS	OMFS
Lip revision	OMFS/Plastic	Plastic	OMFS/Plastic
Rhinoplasty	ENT/OMFS	ENT	ENT
Orthognathic	OMFS	OMFS	OMFS

Table 4.3.2 shows the specialties involved with the surgery for cleft patients. There were three main surgeons involved in the surgery: OMFS, plastic surgeon and ENT surgeon. ABG and orthognathic cases were done by OMFS. While rhinoplasty cases were done by ENT surgeon. There were different surgeons involved in treatment of primary lip and palate in each team

4.4 Clinic sessions and infrastructures

Table 4.4: Clinical sessions and infrastructure of CCC

	UM	UKM	USM
Session per year (2016)	24	12	24
Venue of CCC	OMFS clinic	ENT clinic	Dental specialist clinic
Average number of patients	15	5	5
Average number of first visits	34	20	8
Total number on follow up outpatient appointment	320	40	110
Number of inpatients	*	24	*
Number of readmissions	*	*	*
Total number of bed-days			
- primary lip repair	3 days	3 days	3 days
- primary palate repair	3 days	3 days	3 days
- Alveolar bone graft (ABG)	3 days	4 days	3 days
Written protocol/guidelines	No	Yes	No

* - No data available

Table 4.4 shows the clinic sessions and infrastructure of each CCC involved in this study. UM and USM held CCC sessions twice a month while UKM only held the clinical sessions once a month. CCC of UM had the highest number of average patients seen per session. This included new patients and follow up patients. All the CCC did not have proper documentation on inpatients that required early readmission prior to the surgery. All teams had 3 bed-days for each type of surgery. However, UKM had extra one day of bed-days for ABG procedure compare to the other two centres. The UKM was the only team that has written guidelines or protocol for the management of patients with CL/P from birth until adulthood.

4.5 Combine clinic decision making and records keeping

Table 4.5: The CCC decision making and records keeping

	UM	UKM	USM
Clinic decisions recorded	Folder	Folder	Folder
Team decisions communicated to outreach care	Referral letter	Referral letter and phone call	Referral letter and phone call
Family provided with written reports of the team assessment	Yes	No	No
Specialty clinical records integrated	Yes	No	No
Clinical records kept	Oral surgery Clinic Unit record	Hospital Unit record	Hospital Unit record
Data base	No	No	No

Table 4.5 shows the decision making and record keeping by the cleft team. All of the decisions recorded in a folder. Among the team, UM cleft team was the only one that provided written reports of the team assessment after every clinic session to patients and their family members. The clinical records were integrated across all specialty. The clinical records were kept in record unit record, except for UM. In UM, the clinical records were kept in the record unit in Oral Surgery clinic. Up to date, there was no database used by all cleft teams to keep all the records regarding the patients.

4.6 Referrals

4.6: Referral case for the past 1 year (2016)

	UM	UKM	USM
Total referral	34	20	8
Babies (birth <1 year)	23	17	5
Babies (birth <1 year) with UCLP	17	16	5
Babies (birth <1 year) with BCLP	4	0	0
Babies (birth <1 year) with submucous cleft	2	1	0
New children (1 <18 years) referred	9	0	1
New adults (≥18 years) referred	2	3	2

Table 4.6 shows the pattern of cases that was referred to the cleft team in year 2016. UM cleft team received the highest case followed by UKM and USM. Most of the cases were referred at the age of less than 1 year old and UCLP is the commonest type of cleft. There was no patient transferred to the team from the other cleft team.

4.7 Audit and research programs

None of the cleft teams that involved in this study had conducted regular audit and research programs.

CHAPTER 5: DISCUSSION

5.1 Cleft combined clinic and ethical approval

Based from the list given by CLAPAM, there were 9 centres throughout the whole country that provide treatment and services for CL/P patients through MDT model. CLAPAM is a non-profit, non-government organisation and a support group consisting of volunteers from parents with children born with CL/P, adults with CL/P and healthcare professional. First established in 1992, CLAPAM provide various support services and also functions as a link between members and medical professional. The CCC available in Malaysia at the year of 2016 were:

i. Hospital Kuala Lumpur, WP Kuala Lumpur
ii. Hospital Sg. Buloh, Selangor
iii. Hospital Sultan Haji Ahmad Shah, Pahang
iv. Hospital Sultanah Nur Zahirah, Terengganu
v. Hospital Wanita & Kanak – Kanak, Sabah
vi. Klinik Pergigian Besar Kota Bharu, Kelantan
vii. Pusat Perubatan Universiti Malaya, WP Kuala Lumpur
viii. Pusat Perubatan Universiti Kebangsaan Malaysia, WP Kuala Lumpur
ix. Hospital Universiti Sains Malaysia, Kelantan

Out of 9 centres, 3 centres were university hospitals and the remaining are general hospital under Ministry of Health (MOH). Different approval was needed from different ministry in order to conduct this survey. Even under Ministry of Higher Education (MHE), separate ethical approval needed for each university hospital. This was the most time-consuming stage and within this short period of time, we managed to obtain ethical approval from 3 university hospital and no centre from MOH was included in this study.

The questionnaire was sent to the centres involved and the response rate was 100%. For this study, data collection was done from on 2016 to 2017 through communication with the cleft coordinator from these centres. The study was conducted at 3 different cleft centres. The centres involved are:

i. Department of Oral & Maxillofacial Clinical Sciences Faculty of Dentistry, University of Malaya (UM)
ii. Department of Oral & Maxillofacial Surgery, Universiti Kebangsaan Malaysia Medical Centre (UKMMC)
iii. Department of Oral & Maxillofacial Surgery, School of Dental Sciences, University Science of Malaysia (USM)

5.2 Structure of CCC

5.2.1 CSAG recommendations

CSAG is an independent source of expert advice for Health Minister in the UK. It was initially formed to improve the clinical standards and service outcome for the CLP patients. In 1998, CSAG made some recommendations in relation to the planning of CLP services in UK. The recommendations are:

i. Cleft care should be provided by 8 – 15 services
ii. Cleft care should be provided through MDT
iii. Each MDT "must have" specialty members
iv. The MDT should liaise closely with others specialty members
v. Each service should provide care for 100-120 new cleft cases per year
vi. Each primary surgeon should operate on at least 40-50 cases annually
vii. Each cleft service should provide paediatric inpatient care
viii. Each cleft service should provide outpatient care in a paediatric setting

ix. Each cleft service should be involved in a program of clinical audit

x. Each cleft service should have an active program of clinical research

5.2.2 Team composition

Our data showed, for the team composition of each CCC universities that involved, all cleft team was comply to the recommendations. Most of them comprise of the 7 "must have" specialties in the MDT clinic. The "must have" specialties are plastic surgeon, OMF surgeon, orthodontist, speech & language therapist, paediatrician, ENT surgeon and coordinator/manager. This is because all the teams involved in this study were set in university hospital setting. Hence, almost all specialty needed to form a MDT were available. The specialties that rarely present in CCC are the psychiatrist, restorative dentist and special cleft nurse. However, according to the recommendations, these are the team that MDT should liaise in the management of cleft patients.

UM was the first one to develop management of cleft patients through MDT model in 2004 and already established more than 10 years. This was followed by the establishment of cleft team at USM in 2012 and UKM in 2013.

5.2.3 Clinical sessions and infrastructures

For the clinical sessions and infrastructure, our data showed all of the centres were actively managing patient with CL/P over the year with varieties of 1 or 2 sessions per month. The average number of patients per sessions for each cleft centre are different. The cleft team at UKM and USM attended an average of 5 patients, whereby UM cleft team attended average of 15 patients per session. This shows, UM cleft team had highest working load in managing CL/P patients compare to UKM and USM cleft team. This is because, UM established first and located in the Klang valley area which is capital of our country and located near few tertiary hospitals in the region. Before 2012, USM were

joining the CCC located at Klinik Pergigian Besar, Kota Bharu Kelantan. In 2012, USM started a separate CCC in their university setting. Due to logistic reason, the CCC mainly are receiving referrals from certain part of Malaysia (especially from the state of Kelantan and Terengganu). This may be the reason contributing to their low number of patients seen in their centre.

In general, this study showed the use of paediatric facilities in cleft outpatient care was not implemented by the three cleft services as recommended. The CCC mainly held in general outpatient department environments (Dental clinic: UM and USM, ENT clinic: UKM). CSAG recommended every cleft centre provide outpatients care in a paediatric setting or in paediatric clinic. In this study, all CCC were held in general outpatient department. UM and USM CCC were held in dental clinic, while UKM cleft patients were seen in ENT clinic. The reason for all the centres decided to hold the combined cleft session in non-paediatric setting was because the 'leading' specialty was by OMFS or ENT. Setting up the combined clinic at non paediatric clinic didn't mean there were no involvements from the paediatricians. In UM, paediatricians and paediatric geneticist were always present in all sessions of combined clinic ever since the establishment of CCC. However, due to some issue regarding cost and manpower, they decided to review cleft patients in their own clinic. They do see cleft patients during same session as the combined clinic as to avoid multiple visits by the patients.

5.3 Clinical audit practice and clinical research

The CSAG reported that each cleft team should involve in a program of clinical audit. This is important for the cleft team to monitor the outcome of their services. The CSAG also recommend for each cleft centre to have separate audit and clinical research. None of the team in this study followed this recommendation.

Audit is a key aspect of everyday clinical care and essential for the safe as well as efficient functioning of any clinical environment. According to the National Institute for Clinical Excellence (NICE, 2002), a clinical audit is a quality improvement process that seeks to improve patient care and outcomes through a systematic review of care against explicit standards and the implementation of change with a further monitoring is used to confirm improvement in healthcare delivery. A subsequent re-audit can be done to confirm improvement of healthcare delivery.

From the clinical audit, the team can identify and promotes good practice, leads to improvement in patient care, provides information about the effectiveness of a service, highlights problem and helps with solutions and also will improves team working and communication among team member.

The CSAG also recommended for each cleft centre to have an active program of clinical research. None of the centre had done any clinical research in relation to cleft lip/palate. Clinical audit and research are closely related, but distinct disciplines. Research is creating new knowledge about whether new treatments work and whether certain treatments work better than others. Research forms the basis of nationally agreed clinical guidelines and standards – it determines what the best practice is (Modayil, Panchikkeel, & Alex, 2009).

The most frequently cited barrier to a successful audit and research are the failure of an organization to provide sufficient funds and protected time for healthcare teams (Modayil et al., 2009). In this study, there was no dedicated staff to manage the combined

clinic. All are clinical service staff that attends to patient, treat patient, and at the same time collecting data, keeping records and manage the patients' appointment

5.4 Decision making and record keeping

Clinical recordkeeping is an integral component in good professional practice and the delivery of quality healthcare. Good clinical record keeping should enable continuity of care and should enhance communication between different health professionals.

Each cleft team should have good clinical record keeping for all patients that attend the cleft clinic. All data that are kept in the record will be very useful not only for audit and research program to assess the outcomes of treatment techniques but also for research resources. CLP patients received treatment till adulthood. Hence, all team need good record keeping to follow through the treatment progress of cleft patient. Our data showed, all of cleft team involved used hard copy folder to record the progression of the patient. Referral letter and phone call were used for the team to communicate for outreach care.

Medical record keeping for CL/P patients is very important not only to the patients and family members, but also to the cleft team itself. The records tell us what, where and when something was done and why a decision was made. When record keeping was adequately performed, it improves the coordination and continuity of care, reinforces decision-making capacities, augments staff accountability and achieves more accurate vital statistics (Wong & Bradley, 2009).

Only cleft team in UM had the clinical records integrated, the guardian and parents of patients was provided with written report of the team assessment. Recording system are very important, because the family will feel more appreciated and they will have the feeling as part of the team. The other benefits of integrated report - all specialists are

informed and updated regarding the treatment progress of the patient. This too can avoid miscommunications between all specialty in MDT.

For each centre that having MDT model, it was recommended the team to develop a common database for all cleft patients with the specifications on the information to be collected as well as the timing of record collection. With the database, comparative inter centre studies in between different cleft service will be available. However, the finding of our study revealed that, there was no database used to keep the records of all patients that attend the clinic. Wong & Bradley (2009) reported that the implementation for data base record was simple and inexpensive, and the results indicate significant improvement in accessibility, completeness and physician satisfaction in the medical records system. The full support from hospital leadership and staff was the key to success.

5.5 Treatment provided

Due to the scarcity of multicentre studies on cleft lip and palate rehabilitation, protocol variation between studies has yet prevented the establishment of a unified, evidence-based clinical practice for cleft lip and palate treatment that accounts for the suitable rehabilitation protocol for each patients growth pattern and cleft type (de Ladeira & Alonso, 2012). Our data showed, all of the cleft services involved provided more than one type of surgery. For the treatment of primary cleft lip repair, we found that all of the cleft team done the surgery at the average of 3 months old. None of the centres provide primary lip repair during neonatal period. Research showed that neonatal period is no longer done due to high risk of mortality. This is in line with worldwide practice, where cleft lip repair was done at the age of 3 – 6 months of age (Slade, Emerson, & Freedlander, 1999). In most centres, the cleft lip repair was done at the age of 3 to 6 months of age.

For the primary palate repair, we reported that there was 2 different time of treatment. Cleft team at UKM done the surgery at the average age of 6 months, whilst the cleft team

at UM and USM, at the age of 9 months. There are varying schools of thought about the timing, staging, and implications on speech, facial growth and development. It was believed that the muscle is developing well at the age of 6 months. Hence, some cleft centres started primary palate repair at this age. It is found that cleft palate repair at the age of 9 – 12 months, has a more positive influence on speech problem and development of the maxilla (Pradel et al., 2009).

There are many surgical techniques and modifications and all protocols focus on the speech development, facial growth and velopharyngeal function (Farronato, Kairyte, Giannini, Galbiati, & Maspero, 2014). In this study, all centres practice the palatoplasty in one-stage repair protocol, whereby the soft & hard palate was closed in the same surgery. Even though there was a good result on maxillary growth of delayed hard palate repair in the two-stage palatal repair, this technique is highly being abandoned. The main reason is speech defects with velopharyngeal insufficiency (Farronato et al., 2014). The data showed either plastic surgeon or OMFS were involved in the primary lip and palate surgery. UM is the only one who had OMFS as the primary surgeon (primary lip &palate repair) as the others are done by plastic surgeon. This may be OMFS in UM being the oldest CCC and with the greatest number of patients had their surgeons trained in primary surgery.

In comparison to ABG procedure, all cleft team involved were done by the OMFS with the average age of 9 – 12 years old. There are three type of surgical procedures currently practiced by the cleft team worldwide to repair the cleft alveolus: gingivoperiosteoplasty, primary bone grafting and secondary bone grafting (Farronato et al., 2014). Our data showed the secondary bone grafting was broadly applied by all of the cleft team. This is because, the result or outcome is better than the two others technique. The advantages are, it minimizes the growth disturbances of the upper arch and gives

maxillary arch integrity with periodontal support for the teeth proximal to the cleft (Lip, 2001) . This protocol is now widely used and considered as a standard procedure for cleft alveolar repair.

Children born with a cleft involving the palate are at high risk of developing abnormal speech, feeding, hearing and language. Hearing and speech quality is important for futures of the child as it will affect their school performance ability in securing a place at work. With poor speech, the cleft individuals will have problem in normal conversation, and this will affect their self-esteem. Cleft patient also at high risk of developing middle ear effusions or otitis media with effusion (glue ear). Middle ear effusions are associated with conductive hearing loss because of the eustachian tube malfunction (Flynn, Möller, Jönsson, & Lohmander, 2009).

Assessment and treatment by Speech & Language Therapist for cleft children especially presented with CP was very crucial and early assessment was recommended. Our findings showed, the children were seen for hearing assessment and speech therapy at the average age of less than 1 year old in all cleft team. It is recommended the patient to received ENT care within the first 6 months of life through adolescence and audiologic care within the first 3 months of life with continued evaluation as determined by the child history of ear disease and hearing loss. Audiologic and hearing should be evaluated annually due to important interplay with speech& language development during childhood (Thomas, 2000). It is estimated that 50 to 100% of patient with CP have significant recurrent middle ear pathology even with early palatal closure (Paradise, Elster, & Tan, 1994).

5.6 Referrals

In this study the number of new case referral for the past one year was obtained in year 2016. Each of cleft centres in this study had their own book that registered all of patient that attended the clinic by referral letter or appointments. However, in UM they accepted walk-in to their clinic without any appointments or referral later. This will lead the actual number was not captured in their data. All cleft team in this study received new cleft case at the range from 8 – 34 patients per year. Compared to the recommendation made by CSAG, these centres are operating at low volume scale. CSAG recommended each service should provide care for 100 – 120 new cleft cases (of all type) per year. There are few factors that might contribute to the small number of referrals, such as (1) improper documentation/record in the registration book, (2) years of experience operating CCC, for example USM and UKM cleft team just started their CCC which is operated less than 5 years during the data collection done and (3) possibility of the child that born with CL/P was referred to the nearby hospital which is also provide a cleft care services either through the MDT model or private hospital especially. As well as number of populations in UK as compare to Malaysia.

CHAPTER 6: CONCLUSION

6.1 Conclusion

This study only comprised three centres that were all set up in university hospitals setting. In general, the cleft services in these three centres comply with few of the recommendations made by CSAG such as the cleft care was provided through MDT model and composition of team members were as recommended.

However, all centres showed inadequacy in recording clinical record in database and clinical audit. The recommendations for cleft care provided paediatric in-patient care and the number of new cases per year may not applicable to the cleft services in Malaysia. However, the other recommendations are still worth to be considered.

6.2 Limitations of study

This study did not include all CCC in Malaysia as we did not obtain ethical approval from CCC under Ministry of Health (MOH). Time is the major limitation in this study as separate applications for ethics were needed for each centre, especially it involved two different government organization (MOH and MOHE).

6.3 Recommendations

For future study, we would like to suggest:

i. Appropriate time given for ethics application to all centres
ii. Include all CCC in Malaysia

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