

A Malaysian National Cleft Registry: An Orthodontic Point of View

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ABSTRACT

Cleft lip and palate (CLP) is one of the most prevalent birth defects. CLP can have a profound physical impact on the child and a psychological impact on both the parents and child. The systemic collection and audit of data on CLP has traditionally been an integral part of comprehensive cleft care. This requires the development of a national cleft registry. This article describes the current orthodontic cleft care, the benefits of a registry and assesses the challenges faced in developing a national cleft registry in Malaysia from an orthodontic point of view.

Keywords: *Cleft lip and palate; registry; orthodontic; Malaysia*

INTRODUCTION

Cleft lip and palate (CLP) is one of the most common forms of craniofacial birth defects (World Health Organization, 2003). The aetiology of CLP is not fully understood but it has been proven that there is a strong genetic connection. It is further influenced by environmental and lifestyle factors (Murray, 2002). CLP has a detrimental effect on the facial features and well-being of children in terms of their health, speech, hearing, appearance, dentition and quality of life. Having a team of specialists works on the issue is considered the best option for treatment (Hodgkinson *et al.*, 2005).

The systemic collection and audit of data on CLP has traditionally been an integral part of comprehensive cleft care (Bearn

et al., 2001; Shaw *et al.*, 2001). This requires the establishment of a national cleft registry. Reliable and up-to-date data on phenotypes, birth prevalence and outcomes of treatment are essential for the accurate auditing of children born with CLP. Health professionals who manage children with CLP act as sources of information to contribute to the cleft registry. The collection of information may be done on a voluntary basis or under the authority of legislation. The type of reporting used is critical as compulsory reporting will yield a higher amount of data than voluntary outcome reporting (Thong, 2014).

The specialities needed for comprehensive cleft care are plastic surgery, audiology, oral and maxillofacial surgery, orthodontics, otolaryngology/head and neck surgery,

paediatrics, paediatric dentistry, psychology, speech-language pathology, nursing and genetics. Orthodontists play a crucial role as record keepers on the cleft and craniofacial team. One of the reasons orthodontists have been given the task of record-keeping is because they treat children with CLP most frequently and for the longest duration. There are diverse ranges of orthodontic-related outcome measures for CLP (Haque *et al.*, 2015a). A simple and accurate outcome measure is essential to the success of a cleft registry.

This article describes the current status of orthodontic cleft care in Malaysia including the benefits and challenges in creating a cleft registry from an orthodontic point of view.

CLEFT CARE IN MALAYSIA

The prevalence of CLP in Malaysia is approximately one in 941 live birth each year (Oral Health Division, Ministry of Health of Malaysia, 1998). CLP occurs more commonly in females than males, and clefts involving the lip and palate are the most prevalent (Ali Shah *et al.*, 2015). The aetiology of CLP in Malaysia is multifactorial (Ayu *et al.*, 2003; Rahman *et al.*, 2008; Haque *et al.*, 2015b). The Ministry of Health of Malaysia has conducted various health promotion programmes and initiatives to increase women's awareness of the risk of exposure to environmental factors such as tobacco, alcohol, poor nutrition, medicinal drugs and teratogens, and lack of folic acid during pregnancy. Furthermore, there is a strong cultural belief about the cause of CLP among the Malaysian population. However, this cultural belief does not hinder individuals with CLP from seeking treatment from the Malaysian healthcare system (Hussin, 2017).

The Malaysian healthcare system comprises government-managed primary care healthcare centres and hospitals and the still-developing private care, which is primarily situated in physician clinics and hospitals in

metropolitan areas. Multidisciplinary CLP treatment is mainly delivered in government hospitals and at some university, military, and private hospitals.

Children with CLP in Malaysia receive the necessary treatment, but there is limited study on the overall outcome of the care being provided (Lee *et al.*, 2004; Zreaqat *et al.*, 2009; 2012; Mohd Ibrahim *et al.*, 2015; Cheong *et al.*, 2016; Zulkipli *et al.*, 2018). The overall outcome of care might vary between regions because some centres have a well-coordinated, multidisciplinary team that meeting on a monthly basis, whereas in some regions, cleft care is poorly centralised and provided by individual clinicians (Mohd Ibrahim *et al.*, 2015).

ORTHODONTIC CLEFT CARE IN MALAYSIA

Orthodontic care for children with CLP is usually provided in a regional specialist dental clinic. This system requires the orthodontist and patient to travel to the hospital for a joint multidisciplinary clinic. According to the European consensus recommendation (Shaw *et al.*, 2001), orthodontists should treat a minimum of 40 new cases yearly to maintain their experience and specialist skills and to allow audits of their performance. Due to the decentralisation of cleft care in Malaysia, certain orthodontists would not come close to treating 40 new patients annually.

The European consensus recommendation stated that orthodontic care on CLP patients should only be provided by an experienced orthodontist who has undergone special training in cleft care. Currently, there is no restriction about the minimum experience or training a dentist must have to orthodontically treat children with CLP in Malaysia. This lack of regulation will result in considerable variation in the quality of orthodontic treatment offered to children with CLP.

Due to the lack of official government policy or guidelines, orthodontists use a range of different protocols in their treatments. The general lack of information and policy about treatment protocol and the difference in orthodontists' experience result in a great variation in treatment outcomes. A streamlined protocol should be provided based on the current best evidence. Research is required to assess the treatment outcome and determine the effectiveness of a given protocol. Zreaqat *et al.* (2009) conducted a retrospective study to establish the treatment outcome based on the dentoalveolar relationship among Malaysian children born with non-syndromic complete unilateral CLP. They concluded that the outcome of treatment is intermediate by utilising the GOSLON Yardstick.

The cost of treatment can vary considerably between centres. The private insurance system in Malaysia does not cover orthodontic treatment costs for cleft care, hence most cleft patients seek care in government-funded centres. The government provides financial support for cleft care in public centres, but only part of the treatment cost is free. This cost will undoubtedly further increase the strain on parents of children with CLP.

THE BENEFITS OF REGISTRIES

The most obvious function of a registry is to act as a tool for the management of patients who have an ongoing need for medical care (Solomon *et al.*, 1991). A registry particularly useful in cleft care because patients require long-term therapy and a multitude of specialities are involved. Registries can improve orthodontic care for children with CLP and encourage multicentre collaboration in Malaysia by being the apparatus for a community-wide cooperative approach to the handling of cleft patients (Hammond and Stassen, 1999). The establishment of a registry will enable an exchange of knowledge of techniques and the standardisation of protocol based

on best evidence, which is currently lacking in Malaysia. The registry also encourages audits and facilitates research by tracking the results of interventions throughout the CLP population. Having a registry will indirectly standardise the record-keeping in Malaysia. A standardised record will facilitate easy comparison between centres nationally or abroad (Shaw *et al.*, 2001).

THE ESTABLISHED REGISTRIES

The following are good examples of internationally well-established and validated cleft registries that keep records of orthodontic-related outcomes.

The Swedish Quality Registry for CLP started as a collaboration between six Swedish CLP centres in 1999 (Pegelow *et al.*, 2020). The orthodontists in the teams agreed to record the dental occlusions and treatment outcomes in the registry at 5, 10, 16, and 19 years of age. In the case of orthognathic surgery, study models for records are taken one-year post-surgery. The outcomes of treatments are assessed by using the Modified Huddart and Bodenham scoring system, 5-year-olds' index and GOSLON Yardstick. Other data recorded related to orthodontics are the cephalometrics value, any orthodontics prior to bone grafting and assessments of bone level in the cleft area.

CleftSiS was built on the foundation laid by the Scottish Association for Cleft Lip and Palate (SCALP). CleftSiS has a record-and-audit protocol based on the recommendations of the Craniofacial Society of Great Britain and Ireland (Clark, 2007). Assessment of the surgical outcomes of primary surgery and secondary alveolar bone grafting, facial profile, skeletal pattern and speech for 5-, 7-, 10-, 15- and 20-year-old patients forms the core of the audit. The clinical data entered into the system are displacement, centrelines discrepancy, overjet, GOSLON Yardstick, 5-year-olds' index, Modified Huddart and Bodenham

scoring system and any pre-surgical orthopaedics done.

Due to varied outcome measures and the recognised necessity for consensus in this area, the International Consortium for Health Outcome Measurement (ICHOM) created an international, multidisciplinary working group to establish a standard set of outcome measures for the comprehensive assessment of cleft care (Allori *et al.*, 2017). Occlusion is evaluated at 5 and 12 years of age, and again at the end of treatment or at 22 years of age, whichever comes first. ICHOM adopted a clinical examination of overjet based on the GOSLON scale. Patient-reported outcomes are incorporated into the outcome standard. The function of mastication is evaluated by the patient-reported CLEFT-Q Eating-and-Drinking scale at 8 and 12 years, and again at the end of treatment or at 22 years of age, whichever comes first.

DISCUSSION

The establishment of a cleft registry requires meticulous and thorough planning involving comprehensive discussions between experts and open collaboration with a myriad of professional groups and healthcare providers. The development and operation of a registry, therefore, requires a long-term commitment. In many cases, it can take a couple of years to see the full benefits. One challenge in setting up a cleft registry is having a complete data capture. Without legislative authority for data entry, the registry will undoubtedly face difficulty in data collection. The other major problem in the establishment and maintenance of a registry is a cost. The potential benefits of the registry should be evaluated in light of what it will cost to develop and maintain.

In light of the orthodontists' involvement in the registry, standards of assessment and recording should be clear and unambiguous so that orthodontists involved in cleft care will record patient data in a consistent

way. There should be a consensus on what outcomes are important. The data and outcome measures should be well-defined and easy to measure.

The most popular and widely accepted outcome index is the GOSLON Yardstick (Mars *et al.*, 1987; Atack *et al.*, 1997). The GOSLON Yardstick provides an outcome index to assess the result of the primary surgery in the early permanent dentition. In Malaysia, providing the professional calibration required for orthodontists to use both indices is a tall order. The requirement to be professionally calibrated in the use of GOSLON Yardstick has been criticised. Consequently, the Modified Huddart and Bodenham scoring system has been suggested as an alternative. It is reliable, quick and easy to perform and its main advantage for orthodontists in Malaysia is that no assessor training is necessary (Gray and Mossey, 2005; Haque *et al.*, 2015a).

ICHOM has elected to use a clinical assessment of the overjet. The ICHOM development group recognised that there is limited evidence for the validity and reliability of this method, but in this case, the group prioritised simplicity and sustainability (Allori *et al.*, 2017). If orthodontists in Malaysia recorded the overjet for every patient at each visit, it would be simpler to compare this data.

Recently, patient-reported outcomes have become a priority in outcome assessments. In an attempt to evaluate patient-reported outcomes, CLEFT-Q has been developed specifically for CLP patients to assess their quality of life (Tsangaris *et al.*, 2017). Malaysia is not a native English-speaking country, therefore, the CLEFT-Q would need to be translated and validated before use.

At the beginning of registry development, it may be a good idea to have fewer variables. Too much data entry should be avoided because in a busy orthodontic practise, data collection is often perceived as extra, unnecessary work. It is important, to keep

the number of variables to a minimum. Therefore, the initial step in overcoming the challenges is to determine what information should be added to the registry. The cleft teams from various centres should reach a consensus on what outcome measure to be included. Once the majority has agreed, then only is developing a registry worthwhile because the success of a cleft registry is highly dependent on the support from the cleft team members.

Developing a centralised national registry to collect all the data is a huge task due to the number of individuals and centres involved. The first thing that must be done is to centralise the treatment for CLP children to centres with a multidisciplinary team of specialities. Next, an individual centre database must be developed to store all the data from each centre. The database of each centre would then be linked to a national registry. Having individual centres first develop their own database will lessen some of the work of developing a national registry into a smaller, more achievable task.

CONCLUSION

The systemic collection and analysis of data on children with CLP using a national registry is an essential part of comprehensive cleft care. Orthodontists have an obligation to review the success and shortcomings of treatment. A registry offers significant advantages by providing tools for comparing treatment outcomes between centres. Orthodontists in Malaysia must be aware of the advantages and limitations of each outcome before deciding what should be included in the registry. Careful consideration should be made of what will be included in the registry so as avoid endless data entry, which could deter orthodontists from contributing to it.

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