FACIAL AESTHETICS AND PSYCHOSOCIAL OUTCOME ASSESSMENT FOLLOWING TREATMENT OF NON-SYNDROMIC CLEFT PATIENTS

Doctor of Clinical Dentistry (Orthodontics)

Manuscript

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Acknowledgements

This thesis would not have been possible without the invaluable assistance of the following people.

Professor Wayne Sampson, P.R. Begg Chair in Orthodontics, the University of Adelaide, for his expert advice, guidance and editorial opinion throughout this project.

Dr Rachel Roberts, Senior Lecturer in Psychology, the University of Adelaide, for her expert advice, guidance and editorial opinion throughout this project.

Dr Lisa Jamieson, Research Fellow at the Australian Research Centre for Population Oral Health, the University of Adelaide, for her expert assistance in statistical analysis, interpretation of the results, advice and guidance throughout this project.

Associate Professor Craig Dreyer, Orthodontics, the University of Adelaide, for his guidance and useful feedback.

Signed Statement

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Summary

The main aims of this study of treated South Australian adult patients with the diagnosis of non-syndromic cleft, was to evaluate the effect of long-term cleft treatment on general health-related quality of life (HRQoL) and oral health impact, to determine associations by age and gender, and to compare against the South Australian population norms. Furthermore, the study aimed to evaluate the opinions of a group of professionals and a group of lay people regarding the facial appearance of South Australian adult patients treated for orofacial clefting, as well as their perceptions whether further surgery was required to correct the facial appearance. It also set out to determine whether there are differences in opinion within professional groups as well as between lay people with and without a cleft.

Participants (n=88) were recruited from cleft patients treated at the Children, Youth and Women's Health Service under the Australian Craniofacial Unit over the last 34 years (1975 to 2009). Participants all had surgery to correct their unilateral or bilateral cleft lip, cleft palate, cleft lip and palate, and to correct jaw size discrepancies. Inclusion criteria were non-syndromic cleft patients aged 18 years or over who had completed their cleft treatment at this centre. Participants completed a questionnaire that included information pertaining to age, sex, HRQoL and oral health impact. State-based and national norms were used for comparative purposes. HRQoL was measured by the SF-36 questionnaire with high values indicating a good level of HRQoL. Oral health impact was measured by OHIP-14 questionnaire where high values indicated a poor level of oral health.

Photographic records of 80 of the above participants were obtained with their consent. The photographs were taken following the completion of all treatment including orthognathic surgery as well as revision surgery. The photographs were standardised using computer software (Adobe Photoshop Windows PC version CS8.0) for size, background and brightness. Frontal, left profile and right profile views were available for each patient. These

images were cropped, re-scaled and projected onto a screen for assessment by a panel of professional and lay people raters. Professionals (2 plastic surgeons, 1 dentist, 1 orthodontist, 1 psychologist) and lay people (1 male, 1 female adult without a cleft; 1 male, 1 female adult with a cleft) were recruited. The raters were asked to rate the photographs according to attractiveness of each patient's nose, lips and overall facial appearance. The raters were also asked whether they thought further surgery was required. Facial aesthetics was measured by Visual Analogue Scale (0-100mm) with high values indicating good aesthetics. Necessity for further treatment was measured by Visual Analogue Scale (0-100mm) where high values indicated high perceived need for further treatment.

There were no significant age or sex differences in the cleft sample's SF-36 and OHIP-14 scores. When compared against South Australian 2002 state-level norms, cleft participants scored higher on physical functioning and physical role function but lower on vitality and mental health. The prevalence of having experienced one or more of OHIP–14 items 'fairly often' or 'very often' was 2.7 times higher than national-level estimates, while extent was 2.8 times and severity 1.7 times higher.

The professionals rated facial aesthetics significantly lower and had a lower perception of need for further treatment than the lay people with and without a cleft. The lay people with a cleft rated facial aesthetics significantly higher and had a lower perceived need for further treatment than the lay people without a cleft. The non-surgical professionals rated facial aesthetics significantly lower and had a lower perceived need for further treatment than the surgical professionals.

Oral health among cleft patients included in our study was poor compared with population-level estimates. The HRQoL showed mixed results, with the vitality and mental health components being poorer in the cleft group compared with population-level estimates.

These results indicate that treatment for orofacial clefting does not entirely remove the factors contributing to poor HRQoL and oral health.

Differences exist in the facial aesthetics ratings and perceived need for further surgery between professionals, lay people with and without a cleft. This has profound implications in the assessment of the cleft deformity and management of treatment expectations.

Chapter 1

Literature Review

Introduction

Orofacial clefts, including cleft lip with or without cleft palate, are among some of the more visible birth defects with a prevalence of 8.83 per 10000 births in Australia (World Health Organisation, 2003). The prevalence of cleft lip with or without cleft palate has been reported as 9 per 10000 births between 1987 and 1996 (Hurst et al., 1999). For the same period, the prevalence of cleft palate has been reported as 6.1 per 10000 births (Hurst et al., 1999). Due to the varying manifestations and severity of cleft lip and palate, treatment may vary pending the timing and technique of the surgery carried out. Aesthetic and functional results, including speech, should be accounted for in the long-term goals of treatment (Jeffery and Boorman, 2001; Marcusson et al., 2002). It has been suggested by Sinko et al. (2005) that the final outcome of cleft treatment is very difficult to predict at the commencement of treatment. This is in part due to the variations in growth and development as well as the compliance of the patients. Hence it is not until the patient has reached twenty years of age that the final outcome can fully be discerned (Sinko et al., 2005).

There is a direct correlation between aesthetic appeal and the psyche of a person in social interactions. A review of the literature demonstrates that several studies have shown an association between aesthetics and social categorisation (Clifford et al., 1972; Corter et al., 1978; Adams and Crane, 1980; Sigelman et al., 1986; Langlois et al., 1987; Langlois et al., 1995). This was also reported by Strauss and Broder in their literature review (Strauss and Broder, 1991). The concept of “stigma” was used to show how people respond to other people with different appearances and conditions (Goffman, 1963). As a result of these differences in people, either in aesthetics or congenital conditions, stereotypes and prejudice may arise (Ablon, 1981). It has been suggested that one aspect of cleft research may use the concept of stigma to assess the impact of facial appearance and speech on the patients’

psyche and social interaction. This may range from the patients' life achievements to sociodemographic status (Strauss and Broder, 1991). It is necessary to introduce the concept of stigma as it does affect how the treated cleft patients have interacted with society and, to a certain degree, how it has affected their life.

Patients who have undergone facial surgery and those who have facial disfigurements are affected psychosocially and experience changes in the way in which they conduct social interaction (Macgregor, 1970). Ramstad and co-workers (1995) have demonstrated that even minor scars from surgery can have significant social impacts on the patients. Previous research have been conducted on the impact of orofacial surgery on self-perceptions (Clifford et al., 1972; Richman, 1983; Richman et al., 1985; Ramstad et al., 1995; Marcusson et al., 2001a; Marcusson et al., 2001b; Marcusson et al., 2002; Sinko et al., 2005; Landsberger et al., 2006; Meyer-Marcotty and Stellzig-Eisenhauer, 2009).

There is also growing evidence that some persons hold negative expectations for individuals with clefts and that they express these expectations in negative treatment (Albino et al., 1990), suggesting that individuals with clefts may be a socially stigmatised group.

As studies have shown that Western society tends to favour facial aesthetics and conversational abilities (Dion et al., 1972; Albino et al., 1990), this would have significant impact on a cleft patient and their psyche. It has been demonstrated that low self-perceived facial aesthetics as well as low self-confidence can affect areas such as expectations of performance and success in life (Phillips, 1984), body image issues and confidence (Richman, 1983). Essentially, people who have a low perception of their self-image tend to be less confident and tend to achieve a lower level of success in life. It appears that a significant proportion of individuals with clefts may fall into this category.

Review of the literature on psychosocial effects of cleft lip and palate

It is important to understand that physical appearance is not the only factor to consider when assessing the success of cleft treatment. Psychosocial factors must also be accounted for when evaluating the outcome of cleft treatment. These factors range from satisfaction with results (including speech and aesthetics), self-confidence as well as social functioning. Some studies have shown gender differences in the above outcomes, where females were more affected than males (Kapp, 1979; Leonard et al., 1991; Sinko et al., 2005).

In relation to satisfaction with treatment results, many studies show variations in the levels of satisfaction. Based on some of the previous research, it appears the majority of adult patients are satisfied with their facial and dental results following cleft treatment (Clifford et al., 1972; Noar, 1991; Ramstad et al., 1995), with Marcusson and colleagues (2002) demonstrating that the cleft cohort was significantly less satisfied with their facial aesthetics than the control cohort. However, there appears to be some ambiguity when examining the patients' desire for further surgical treatment. Marcusson et al. (2002) found 47% of the cleft patients would like further surgery; Noar (1991) found 54% of their patients were not satisfied with certain individual aspects of their facial appearance; yet another study showed 35% of their patients desired further surgery (Ramstad et al., 1995).

Regarding satisfaction with speech, studies have shown an overwhelming number of patients with clefts being satisfied (Noar, 1991), particularly those patients who were happy with their facial appearance to begin with. Ramstad and co-workers also showed that 15% of their cleft cohort, who were satisfied with their facial aesthetics, showed dissatisfaction with their speech (Ramstad et al., 1995).

With regards to specific facial aesthetics, it was found that individuals with bilateral cleft lip and palate were not as happy with the aesthetics of their nose and upper lip (Oosterkamp et

al., 2007). Marcusson and co-workers (2002) found that in general, several of their cleft patients had a higher expectation of the results of their surgery compared to what was actually achieved and this was more pronounced with nasal surgery results. When looking at facial aesthetics rating differences between raters, Sinko and co-workers (2005) showed that women tended to have significantly lower ratings for their facial aesthetics compared to professional raters. This was more pronounced between the ages of twenty-four and thirty years (Sinko et al., 2005). In terms of a third party perspective, regarding bilateral cleft lip nasal deformity correction, it was found that there were no significant differences between the aesthetic ratings of the professional and lay raters (Lo et al., 2002).

It has been shown that attractive individuals were viewed in a more positive light than less attractive individuals, including getting better treatment and being considered smarter (Dion et al., 1972). Previous researchers have also shown that children with cleft lip and palate were teased about their facial aesthetics or speech (Bernstein and Kapp, 1981; Heller et al., 1981; Noar, 1991). In addition, one study found that the cleft cohort was observed to experience emotional and social issues as a result of their cleft condition (Bernstein and Kapp, 1981). Based on these findings, it would seem that the psychosocial wellbeing of cleft patients would be somewhat affected by their condition. Following a review of the literature, Hunt and co-workers (2005) made a similar assumption regarding distressing effects of the cleft condition on the patient's psychosocial wellbeing.

However, contrary to the above findings, some researchers have demonstrated that cleft patients seem to adjust reasonably well in terms of general psychological wellbeing (Noar, 1991; Marcusson et al., 2001a; Sinko et al., 2005). Similarly, following an assessment of several literature reviews of psychosocial effects on patients with clefts, Hunt et al. (2005) reported that patients with clefts do not seem to experience major adverse psychological effects. Although they did cite other articles which suggested that these results are not

conclusive and may be attributed to weaknesses in the research methodology of the previous research (Turner et al., 1998; Thompson and Kent, 2001).

Studies have shown conflicting findings regarding self-confidence. Berk et al. (2001) showed that adults with clefts had lower self-confidence, in a Chinese population. They also showed this cohort to experience significantly more anxiety. Yet other studies show that a majority of patients with clefts regardless of age showed adverse findings for self-esteem (Noar, 1991; Turner et al., 1997). While some research show that children with clefts have good self-esteem (Kapp, 1979; Leonard et al., 1991; Persson et al., 2002). Other studies show lower scores when examining specific self-concept components in isolation (Broder and Strauss, 1989) or specific cohorts such as girls compared to controls (Kapp, 1979).

Other aspects that have been studied in previous research include the relationship between the type of cleft as well as age and the prevalence and severity of psychosocial effects on the patient. In relation to effects of type of cleft on psychosocial impairment, it is likely that the more severe the cleft condition, the more severe the psychosocial effects are on the patient as facial appearance is more likely to be affected by the severity of the cleft condition. However, studies seem to show little correlation between the two variables. Several measures such as self-esteem (Starr, 1978; Kapp, 1979; Starr, 1980), behavioural issues (Millard and Richman, 2001), general development (Starr et al., 1977) and psychosocial functions (Heller et al., 1981) have been shown to be independent of the type of cleft condition. Conversely, different effects on several measures were found in patients with different clefting. Millard and Richman (2001) found that patients with cleft palate only, scored lower than the other types of cleft for parent and teacher ratings of depression and anxiety (ie. higher levels of depression and anxiety). Broder and co-workers (1994) found facial aesthetics satisfaction levels were lower in patients with more visible clefts such as cleft lip with or without cleft palate. Maris and co-workers (2000) showed that at 12 months, infants with cleft palate only

had decreased rates of attachment security to their mothers compared to the other cleft groups. Although by the 24 months stage, the difference between the cleft groups were no longer present.

It has been demonstrated that on its own, age does not appear to have any significant psychosocial effects on the cleft patient (Leonard et al., 1991). In their study, Leonard and co-workers found that self-concept scores were affected when age was combined with gender. Satisfaction with facial aesthetics has been shown to remain fairly constant with age among cleft lip and/or cleft palate patients (Broder et al., 1992). While other studies showed that as the patient gets older, their facial appearance satisfaction level improves (Thomas et al., 1997); they become more satisfied with their speech function (Broder et al., 1992); among females with cleft palate only, they become more dissatisfied with their facial appearance (Broder et al., 1994).

In relation to the research methodology, several studies (Kapp, 1979; Leonard et al., 1991; Persson et al., 2002) have employed questionnaires that are dated, some as old as thirty years. There have been several advances in the field of psychology and newer instruments have been developed since then. Examples of these newer instruments include the Medical Outcomes Study (MOS) 36-Item Short-Form Health Survey (Ware and Sherbourne, 1992) and Short-form Oral Health Impact Profile (Slade, 1997). Hunt and co-workers (2005) reported that the normative data yielded by older questionnaires may not be as valid when comparing against present day cohorts. The newer instruments may be more effective at measuring psychosocial effects than the older questionnaires as they may have more up to date normative data to compare against. Data, which have been gathered using standardised methodology, should be pooled from several cleft treatment centres for evaluation. This would allow for more meaningful findings (Hunt et al., 2005).

Surgical techniques for cleft treatment have also evolved and developed over the course of the last few decades (Reisberg, 2000; Shaw et al., 2001). As the bulk of the abovementioned studies carried out in this field have been published up to several decades ago (Clifford et al., 1972; Starr et al., 1977; Starr, 1978; Adams and Crane, 1980; Starr, 1980; Kapp, 1979; Bernstein and Kapp, 1981; Heller et al., 1981; Richman, 1983; Richman et al., 1985; Broder and Strauss, 1989; Leonard et al., 1991), they may not have had the benefit of improved surgical techniques or treatment protocol, which may deliver better treatment outcomes. This in turn may have a significant psychosocial impact on the cleft patients.

Furthermore, the majority of the research in this field has been conducted in Europe (Noar, 1991; Ramstad et al., 1995; Turner et al., 1997; Persson et al., 2002; Sinko et al., 2005; Meyer-Marcotty and Stellzig-Eisenhauer, 2009; Mani et al., 2010) or in the United States (Clifford et al., 1972; Kapp, 1979; Adams and Crane, 1980; Bernstein and Kapp, 1981. No Australian literature specifically related to the psychosocial aspect of treated cleft adult patients has been found in the current review.

Quality of Life Assessment

Quality of life assessment is increasingly being used in the modern era of medicine to evaluate treatment outcomes (Klee et al., 1997). Quality of life can be explained as an attribute that measures and sums all the different aspects of a person's life, including several aspects such as a person's well-being, health, self-perception, how they function, socioeconomic factors and how satisfied they are with life (Corson et al., 1999). Corson and colleagues (1999) examined an instrument termed health-related quality of life (HRQL). They suggested that health related quality of life is more specific in assessing how disease and its treatment can influence a person's psyche, social functioning as well as physical condition. There are several health-related quality of life instruments that have been

developed. However, it has been suggested that when examining quality of life outcomes in craniofacial treatment such as cleft treatment, an instrument that includes multiple measures and multiple items would be most useful (Bennett and Philips, 1999). One particular health-related quality of life instrument, the short form (SF) -36 (Ware and Sherbourne,1992), can be very useful in cleft research that examines psychosocial outcomes. The SF-36 is readily available, easily administered and has been established in Australia, as being valid and reliable (McCallum, 1995; Sanson-Fisher and Perkins, 1998), as well as in the United States (McHorney et al., 1993; McHorney et al., 1994) and the United Kingdom (Brazier et al., 1992).

Moreover, multidimensional questionnaires and instruments used for psychosocial evaluations and quality of life assessment for cleft patients have improved significantly. Several of the questionnaires used in contemporary medical psychology have been validated and their reliability has been established (Wu et al., 1991; Hörnquist et al., 1993; Marcusson et al., 2001b).

Several studies exploring the quality of life experienced by a cleft cohort with varying cleft conditions have found the quality of life to be within acceptable levels (Noar, 1991; Marcusson et al., 2001a; Sinko et al., 2005). Although, certain aspects that have shown negative impact include social interaction, family interaction, socioeconomics, life meaning, satisfaction with appearance and some patients feeling depressed and anxious (Heller et al., 1981; Ramstad et al., 1995; Marcusson et al., 2001a). Due to weaknesses in the research methodology (Turner et al., 1998; Thompson and Kent, 2001) as well as the lack of uniformity and consistency in the research methodology (Hunt et al., 2005), the previous studies do not appropriately demonstrate the effects of orofacial clefting on the psychosocial well-being of the patient. Moreover, while there have been several studies conducted on general health-related quality of life (Kapp, 1979; Heller et al., 1981; Noar, 1991; Ramstad et

al., 1995; Marcusson et al., 2001a; Persson et al., 2002; Sinko et al., 2005; Meyer-Marcotty and Stellzig-Eisenhauer, 2009; Mani et al., 2010), only a few of these studies have focused on the combination of both general health-related quality of life and oral health impact on the cleft affected individual (Heller et al., 1981; Noar, 1991; Sinko et al., 2005).

In a study with Swedish adults treated for unilateral cleft lip and palate (UCLP), it was found that based on the age and sex of the patient, the UCLP affected their health-related quality of life (QoL) differently (Mani et al., 2010). Mani and co-workers (2010) concluded that younger patients were more adversely affected in several aspects of their QoL than older patients. Some differences were also shown between male and female UCLP patients. However, apart from the mental health aspect of their QoL, compared to normative data, the UCLP cohort had similar health-related QoL. (Mani et al., 2010). These results cannot be extrapolated to patients affected by other types of clefts. In addition, the patients used in the study were not treated with the same cleft treatment protocol.

In summary, there is a plethora of studies conducted on all the above-mentioned aspects of orofacial clefting. However, an overwhelming theme is that contradictory results are shown depending on the studies. It has been suggested that psychosocial problems may present in patients with clefts, but limited evidence shows that they tend to adequately adjust and function (Hunt et al., 2005). This variation in research results is due to the difficulties involved in comparing research data on psychosocial effects arising from cleft treatment. None of the studies present long-term research data or they may have inconsistent research methodology and poor control cohort. In addition, there are very few studies that have examined cleft effects on both the general health related quality of life as well as oral health impact. Moreover, there does not appear to have been any such studies conducted in Australia.

Chapter 2

Research questions

In light of the conflicting evidence presented in previous literature reviews, this investigation sets out to support or refute the assumption that individuals with clefts experience greater psychosocial problems than those who do not have clefts.

The overall aim is to evaluate the scientific evidence linking clefts with an increased risk of psychosocial problems. The review will address a number of specific questions:

1. How do treated adult cleft patients compare with an age- and gender-matched group without a cleft in the general population, in terms of health-related quality of life?
2. How do treated adult cleft patients compare with an age- and gender-matched group without a cleft in the general population, in terms of oral health impact?
3. Is there a difference in how a group of professionals and a group of lay people rate the facial appearance of adult patients having been treated for orofacial clefting?
4. Is there a difference, within the different professional groups as well as lay people with and without a cleft, in how they rate facial appearance of adult patients having been treated for orofacial clefting?
5. Is there a difference between a group of professionals and a group of lay people in their perception of need for further surgical treatment for adult patients having been treated for orofacial clefting?
6. Is there a difference, within a group of professionals and a group of lay people with and without a cleft, in their perception of need for further surgical treatment for adult patients having been treated for orofacial clefting?

Aims/Objectives of the project

The main aims of this study of treated South Australian adult patients with the diagnosis of non-syndromic cleft, were to evaluate the effect of long-term cleft treatment on general

health-related quality of life and oral health impact, to determine associations by age and gender, and to compare against the South Australian population norms.

Furthermore, the study aims to evaluate the opinions of a group of professionals and a group of lay people regarding the facial appearance of treated South Australian adult patients having been treated for orofacial clefting, as well as their perceptions on whether further surgery is required to correct the facial appearance. It also sets out to investigate whether there are differences in opinions within the different professional groups as well as between lay people with and without a cleft.

Hypotheses

In relation to general health-related quality of life and oral health impact, the following hypotheses are made:

General health-related QoL:

- Lower HRQoL for the treated cleft cohort compared to the general population without clefts.
- Lower HRQoL for the younger group compared to the older group in the treated cleft cohort.
- Lower physical and emotional aspects of HRQoL for males compared to females in the treated cleft cohort.

Oral health impact:

- Poor oral health for the treated cleft cohort compared to the general population without clefts.

In relation to facial aesthetics ratings and perceived need for further surgery, the following hypotheses were made:

Facial aesthetic ratings:

- No difference in ratings between the professional and lay raters.
- No difference in ratings within the professional group of raters.
- Lower ratings from the female compared with the male lay raters.
- Lower ratings from the lay raters with a cleft with those without a cleft.

Perceived need for further treatment:

- No difference in perception between the professional and lay raters.
- No difference in perception within the professional group of raters.
- Higher perceived need from the female compared with the male lay raters.
- Higher perceived need from the lay raters with a cleft compared with those without a cleft.

Significance/Contribution to the discipline

There is conflicting evidence in the available literature when it comes to establishing whether adults with repaired cleft experience increased psychosocial problems as a result of their cleft. On the basis of currently available evidence, it is impossible to state the extent of the problem with any certainty. For every study reporting psychosocial problems among those with cleft, there are others which refute this finding. Where difficulties have been established, they are mostly related to behavioural problems, dissatisfaction with facial appearance, and difficulty with specific aspects of social functioning.

Moreover, a significant amount of the studies in this field were published before 1990. Surgical techniques in the area of cleft treatment have become more refined in the past 30 years and, therefore, the visible deformity associated with this anomaly may not be as

pronounced today. In addition, there is a greater awareness of the difficulties associated with being visibly different and it is likely that the clinician's approach to such children and adults has changed for the better. In addition, multidimensional questionnaires and instruments used for psychosocial evaluations and quality of life assessment for cleft patients have improved significantly. Several of the questionnaires used in contemporary medical psychology have been validated and their reliability has been proven (Marcusson et al., 2001a; Marcusson et al., 2001).

The proposed research will help improve knowledge in this field and either confirm or refute the findings from existing literature. Furthermore, the majority of the research in this field has been conducted in Europe or in the United States. Hence, the data gleaned from these studies may not apply to cleft patients in Australia. The proposed research will elucidate information which is more relevant to the Australian cleft cohort. It will also help determine whether the current cleft treatment protocols in Australia, more specifically in South Australia, are adequate in addressing the needs of the individuals affected by a cleft.

Although previous studies have assessed cleft treatment outcome on QoL by looking at health-related QoL (Mani et al., 2010; Sinko et al., 2005), to the best of our knowledge, there has been no study to date which has examined the impact of cleft lip and/or palate treatment outcome on the quality of life (QoL) as assessed by a combined generic health and generic oral health approach. Moreover, there has been no study to date which has compared perceptions of facial aesthetics in treated adult cleft patients between a panel of professional and lay people raters. Nor have there been any studies on the difference in perceptions between lay people with and without a cleft, as well as studies on any gender differences between these groups.

Not only is it important for the cleft patient to be satisfied with the treatment results, the perception of the layperson in the society with which they interact can determine how

successful these cleft individuals integrate into society, either in work, education, or social situations. In addition, insight should be gained into the perceptions of the cleft individuals themselves. A better understanding of the differences in facial aesthetics perceptions between the professional member of a cleft team, the lay person and the individual with a cleft, would be an invaluable aid in cleft treatment planning, discussion of treatment outcomes with the patient, as well as management of patient expectations in order to achieve optimal treatment results and patient satisfaction.

Outline of research methods

Participants were recruited from cleft patients treated at the Children, Youth and Women's Health Service under the Australian Craniofacial Unit over the last 34 years (1975 to 2009). Participants all had surgery to correct their unilateral or bilateral cleft lip, cleft palate, cleft lip and palate, and to correct jaw size discrepancies. Inclusion criteria were non-syndromic cleft patients aged 18 years or over who had completed their cleft treatment at this centre.

Potential participants were contacted initially by mail, in order to introduce the research project. They were given contact details to inform the hospital should they not wish to participate (in which case their names were removed from the list and no further contact was made). A consent form, the questionnaire and a self-addressed reply-paid envelope were included for participants to complete and return.

General health-related quality of life and oral health impact

These two aspects were measured and assessed by the use of a self-report questionnaire included in the mail-out. The self-report questionnaire comprised of two sections, the SF-36 and OHIP-14. Descriptions of the SF-36 and OHIP-14 have been included in Chapter 3.

Responses from the participants were then analysed and compared against the population norms. Further details are described in the "Materials and Methods" section of Article 1 in Chapter 4.

Facial aesthetics and perception of need for further surgery

With the consent of the patients, photographic records were obtained. The photographs were taken following the completion of all treatment including orthognathic surgery as well as revision surgery. The photographs were de-identified, standardized and projected onto a screen to be assessed by a panel of expert and lay person raters. The raters were asked to rate the photographs according to attractiveness of the patient's nose, lips and overall facial appearance. The raters were also asked whether they thought further surgery was required. Both measures were rated on a Visual Analogue Scale. Further details are described in Chapter 3 and in the "Materials and Methods" section of Article 2 in chapter 4.

Chapter 3

Health-related quality of life

A widely used general health-related quality of life instrument is the Short-Form 36 (SF-36) health survey (Ware et al., 1993). This was designed in the United States for use in clinical practice and research, health policy evaluations and general population surveys. The instrument is widely used to assess health-related quality of life at a population-level, thus enabling comparison across population groups. The Oral Health Impact Profile (OHIP) is the most widely used oral health impact instrument, being used among different population groups as well as in population-level surveys. OHIP is a standardised questionnaire that measures people's perceptions of adverse impacts of oral conditions on wellbeing and quality of life (Slade and Spencer, 1994). It was developed in South Australia and has been tested and validated in Australia (Slade, 1997) and several other countries (John et al., 2002). The Short Form Oral Health Impact Profile questionnaire is a condensed version of the original 49-item survey. The short form version is a 14-item health survey (OHIP-14) and has been shown to have good reliability, validity and precision (Slade, 1997).

The self-report questionnaire used in the present study included items pertaining to age and sex, the SF-36 and OHIP-14 (Appendix 4).

SF-36

The SF-36 instrument was designed for self-administration, comprised 36 items and took five to ten minutes to complete. The multi-item scale assessed eight subscales:

1. Physical Function: Limitations in physical activities because of health problems
2. Social Function: Limitations in social activities because of physical or emotional problems
3. Physical Role Function: Limitations in usual role activities because of physical health problems

4. Bodily Pain: Concerns amount of pain and limitations due to body pain
5. Mental Health: General mental health, deals with questions about depression and nervousness
6. Emotional Role: Evaluates limitations in usual role activities because of emotional problems
7. Vitality: Deals with feelings of energy or tiredness
8. General Health: Measures the subjective evaluation of general health status

Responses to the SF-36 items were re-coded, summed and transformed to provide the eight dimensions with scores between 0 and 100, with higher scores indicating better health (Ware et al., 1993). Five of the subscales (Physical Functioning, Role Physical, Bodily Pain, Social Functioning and Role Emotional) define health status as the absence of limitation or disability. For these subscales, the highest possible score of 100 is achieved when no limitations or disabilities are observed. Three of the subscales (General Health, Vitality and Mental Health) measure a wider range of negative and positive health states. For these subscales, a score in the mid-range is earned when respondents report no limitations or disability. A score of 100 on these bipolar subscales is only earned when respondents report positive states and evaluate their health favourably (Ware et al., 1993). The SF-36 has been tested for validity and reliability in Australia (McCallum, 1994; McCallum, 1995; Sanson-Fisher and Perkins, 1998), the United States of America (McHorney et al., 1994) and the United Kingdom (Brazier et al., 1992). The “limitations in physical functioning” subscale is the most valid subscale among the physical subscales and “general mental health” subscale is the most valid among psychological subscales (Sullivan, 2002).

OHIP-14

The OHIP-14 assessed the oral health impact on seven subscales:

1. Functional limitation (e.g., difficulty in chewing)
2. Physical pain (e.g., toothache)
3. Psychological discomfort (e.g., self-consciousness of oral condition)
4. Physical disability (e.g., avoiding certain foods due to oral condition)
5. Psychological disability (e.g., concentration affected by oral condition)
6. Social disability (e.g., being less tolerant of others due to oral condition)
7. Handicap (e.g., being totally unable to function)

Responses for each item were made on a Likert-type scale and ordinal values were coded for each question ranging from zero for a response of "never" through to four for a response of "very often". Three summary variables were then computed:

1. Prevalence: the percentage of people reporting one or more items "fairly often" or "very often".
2. Extent: the number of items reported "fairly often" or "very often".
3. Severity: the sum of ordinal responses, thus taking into account impacts experienced occasionally or hardly ever, and could range from 0 to 56.

Statistical analysis

Comparison with population-level norms

For comparative purposes, the SF-36 data for the cleft cohort were compared with normative data from the South Australian general population without clefts (Dal Grande, 2002). The South Australian general population survey involved a multistage, systematic, clustered area sample of South Australian households, with approximately 75% of the sample selected from

the Adelaide metropolitan area and the remainder from country centres with a population of 1000 or more. Hotels, motels, hospitals, nursing homes and other institutions were excluded from the sample. Survey participants were aged 15 years or over. The survey yielded 3012 completed interviews with a response rate above 70%. The data were weighted to the South Australian population by age, sex, area (metropolitan Adelaide and country South Australia) and the probability of selection within the household. The population data used were either the most recent Australian Bureau of Statistics Census or Estimated Residential Population (Dal Grande, 2002).

The OHIP-14 cleft data were compared with those of the National Survey of Adult Oral Health (NSAOH); a cross-sectional study of oral health among Australians aged 15 years or more living in all states and territories (Slade et al., 2007). NSAOH utilised a three-stage, stratified clustered sampling design. The first stage selected postcodes, the second stage selected households within sampled postcodes, and the third stage selected one adult from each sampled household. Because of the differences in the probability of participation, data were weighted to ensure estimates were representative of the Australian population from which survey participants were selected. Weights were calculated to reflect probabilities of selection and to adjust for different participation rates across postcodes, and among age and sex categories. Our study comprises the 4170 participants age-matched against the cleft study who completed the computer-assisted telephone interview, a dental examination and a subsequent self-report questionnaire (Slade et al., 2007).

Data analytic approach

Mean scores of the eight components of SF-36 will be determined, together with their 95% confidence intervals (CI). Univariate distributions of OHIP-14 prevalence, extent and severity will be determined. Demographic variables are to be classified into age- and gender-

matched groups. Due to the possibly limited number of participants in each age group, the patients will be dichotomised by the median age split into two groups. Findings are considered to be statistically significant when 95% CIs are not over-lapping. Statistical analyses for the NSAOH sub-section will take into account the clustered sampling design to yield unbiased standard error estimates and design effects using the 'complex sampling' tool in SPSS PC version 17.0; Chicago, IL, USA; thus producing weighted population estimates. For purposes of this analysis, data for NSAOH participants aged 18–65 who completed a questionnaire containing the OHIP-14 items will be included.

Facial aesthetics and perception of need for further surgery

The second part of the data collection involves the assessment of the facial aesthetics of the patients following cleft repair. Photographs of the patients following the completion of all treatment, including orthognathic surgery, as well as revision surgery will be obtained. The photographs will be standardised using computer software (Adobe Photoshop Windows PC version CS8.0) for size, background and brightness. For each patient, three views will be presented; including a frontal, left profile and right profile view. These images will be cropped, re-scaled and projected onto a screen to be assessed by a panel of expert and lay people raters. The names of the photographs will be removed and a random number assigned to each patient's photograph for identification purposes by the researcher (PF). The raters will be asked to rate the photographs according to attractiveness of the patient's nose, lips and overall facial appearance. The raters will also be asked whether they think further surgery is required. Both measures were rated on a Visual Analogue Scale.

The rating panel will be asked the following questions in relation to the cleft cohort's photographs (Appendix 5):

1. With regards to the face, how attractive would you rate the following?

2. With regards to the face, do you think further surgery is required to change the appearance of the following?

Facial aesthetics rating

The Visual Analogue Scale was preferred over the Likert Scale because the former employs a 100 mm scale with a broad range of distinctive possibilities, whereas the latter uses ratings from 1 to 5 or from 1 to 10 (Sinko, 2005). According to Jaeschke et al. (1990), both methods of presenting responses options show the same level of construct validity and responsiveness. In this study, high scores indicate good aesthetics for the first measure and high perceived need for the second measure.

The members of the rating panel comprised health professionals (a non-surgical group and a surgical group), adult lay people above the age of 18 years (a lay group without cleft and a lay group with cleft). The groups are as follows:

1. Non surgical group: a male orthodontist and a female dentist from Adelaide Dental Hospital; a female psychologist who is in the private sector.
2. Surgical group: a male plastic surgeon with extensive (5 years) cleft experience and regularly performs cleft surgery at the Australian Craniofacial Unit; a female plastic surgeon with little involvement with cleft surgery from the private sector.
3. Lay people with cleft: a female and a male volunteer above the age of 18 years.
4. Lay people without cleft: a female and a male volunteer above the age of 18 years.

The professional raters will not have been involved in the treatment of the cleft patients who were assessed. Panel assessment of facial aesthetics using this method has been conducted in several studies, and has been shown to be both valid and reliable (Howells and Shaw, 1985; Tobiasen et al., 1991; Lo et al., 2002; Marcusson et al., 2002; Sinko et al., 2005;

Tatarunaite et al., 2005; Kenealy et al., 2007). Other investigations also confirm that a high agreement on facial aesthetics exists among panels with different backgrounds (Lundström et al., 1987; Peerlings et al., 1995).

Statistical analysis

Mean scores will be generated from the Visual Analogue Scales for each of the components measured (nose, lips and face), with high scores indicating good aesthetics for the first measure and high perceived need for the second measure. The following groupings will be made: professional (dentist, orthodontist, psychologist, 2 plastic surgeons), lay (2 lay people without a cleft [male and female], 2 lay people with a cleft [male and female]).

The professional group is further sub-divided into a non surgical group: dentist, orthodontist and psychologist, versus a surgical group: 2 plastic surgeons (one with extensive cleft experience and the other with less cleft experience. This would enable comparisons of ratings between surgeons with different backgrounds). Student's t-tests will be used to compare group scores, with findings considered statistically significant when P values are 0.05 or less.

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Chapter 4

Statement of Purpose

In light of the conflicting evidence presented in previous literature reviews, this investigation sets out to support or refute the assumption that individuals with clefts experience greater psychosocial problems than those who do not have clefts.

The aim of this study of treated South Australian adult patients with the diagnosis of non-syndromic cleft, is to compare differences in the health-related quality of life as well as the degree of adverse impacts of oral disease on wellbeing and quality of life between these patients and an age- and gender-matched group without cleft in South Australia.

Furthermore, the study aims to compare the treated cleft patients' self-reported satisfaction with facial appearance as well as self-perception of facial attractiveness with those of a panel of expert and layman judges. The patients' desire for further treatment is also compared with the perception of the judges.

The results of the study are presented in the form of two papers:

1. General Health-Related Quality of Life and Oral Health Impact Among Australians with Cleft Compared with Population Norms; Age and Gender Differences. (Provisionally accepted for publication by the Cleft Palate Craniofacial Journal, pending minor revisions. The revised article has been re-submitted and is included in this thesis)

2. Evaluation of Facial Aesthetics and Perceived Need for Further Treatment Among Adults with Cleft as Assessed by Cleft Team Professionals and Lay Persons. (Submitted for publication by the Cleft Palate Craniofacial Journal)

Article 1

GENERAL HEALTH-RELATED QUALITY OF LIFE AND ORAL HEALTH IMPACT AMONG AUSTRALIANS WITH CLEFT COMPARED WITH POPULATION NORMS; AGE AND GENDER DIFFERENCES

Doctor of Clinical Dentistry (Orthodontics)

Article 1

Written in the format for submission to:

The Cleft Palate-Craniofacial Journal

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Foo, P., Sampson, W.J., Roberts, R.M., Jamieson, L.M. and David, D.J. (2011) General Health-Related Quality of Life and Oral Health Impact Among Australians with Cleft Compared with Population Norms; Age and Gender Differences. *The Cleft Palate-Craniofacial Journal, In-Press*

Article 2

EVALUATION OF FACIAL AESTHETICS AND PERCEIVED NEED FOR FURTHER TREATMENT AMONG ADULTS WITH CLEFT AS ASSESSED BY CLEFT TEAM PROFESSIONALS AND LAY PERSONS

Doctor of Clinical Dentistry (Orthodontics)

Article 2

Written in the format for submission to:

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ABSTRACT

Objectives: To compare the ratings of professionals and lay people with and without a cleft regarding the facial aesthetics of adult patients previously treated for orofacial clefting. The necessity for further treatment, as perceived by the respective groups, is also compared.

Design: Cross-sectional study.

Participants/setting: Professionals (2 plastic surgeons, 1 dentist, 1 orthodontist, 1 psychologist) and lay people (1 male, 1 female adult without a cleft; 1 male, 1 female adult with a cleft) were recruited to rate photographs of 80 non-syndromic cleft patients treated by the Australian Craniofacial Unit from 1975 to 2009.

Main Outcome Measures: Facial aesthetics was measured by a Visual Analogue Scale (0-100mm). High values indicate good aesthetics. Necessity for further treatment was also measured by a Visual Analogue Scale (0-100mm). High values indicate high perceived need for further treatment.

Results: The professionals rated facial aesthetics significantly lower and had a lesser perception of need for further treatment than the raters with and without a cleft. The lay people with a cleft rated facial aesthetics significantly higher and had a lesser perceived need for further treatment than lay people without a cleft. The non-surgical professionals rated facial aesthetics significantly lower and had a lower perceived need for further treatment than the surgical professionals.

Conclusions: Differences exist in the facial aesthetics ratings and perceived need for further surgery between professionals, lay people with and without a cleft. This should be considered when managing cleft treatment outcome expectations.

Key words: facial appearance, cleft, perceived need for treatment, aesthetics

INTRODUCTION

Orofacial clefts, including cleft lip with or without cleft palate, are among the most common visible birth defects, occurring in one out of every 500-1000 live births worldwide (Murray, 1995). With a prevalence of 14.7 per 10000 births with orofacial clefts in Australia (Lancaster and Pedisich, 1995), orofacial clefts are the most frequent congenital craniofacial deformities (Sinko et al., 2005). Cleft lip and palate vary greatly in terms of the width of the cleft and other characteristics. The timing of surgery and technique of reconstruction may differ. Moreover, the face being the site of the deformity ensures attention is drawn to the problem. In the long term, the treatment of cleft lip and palate should provide good aesthetic and functional results, including speech and occlusion (Jeffery and Boorman, 2001). An issue associated with the management of cleft lip and palate patients is that it may be up to two decades before the definitive results can be discerned. Due to the patient's physical development and variability in the level of cooperation, it is difficult to predict the final outcome when the cleft treatment is started. It has been suggested that the final result can only be assessed when the patient is about 20 years old (Sinko et al., 2005). In order to assess the final outcome of cleft treatment, studies would have to be conducted on adult cleft patients.

The relationship between appearance, social stereotyping, and expectations has been established as one of the most consistent research findings in social science (Byrne, 1971; Clifford et al., 1972; Clifford and Walster, 1973; Boukydis, 1978; Corter et al., 1978; Adams, 1980; Adams and Crane, 1980; Berscheid, 1980; Berscheid and Gangestad, 1982; Sigelman et al., 1986; Langlois et al., 1987). Differences in facial appearance are readily noticeable and central in impression formation. Attractiveness has an important effect on psychological development and social relationships. The bodily signs of being different, known as stigma, often carry a moral evaluation, usually a negative one. Goffman's classic work "Stigma:

Notes on the management of spoiled identity" (1963) has provided a theory of stigmatisation and handicap useful in understanding the social responses to human difference and health conditions. Persons may be seen as different when judged by cosmetic norms or with prejudices about the cause and nature of congenital conditions (Ablon, 1981). Myths, fiction, and legends reveal a cross-culturally shared distress with deformity (Shaw, 1981). Stigma theory has not, to date, been a principal focus of research into facial disorders. A broadly defined social science approach to cleft research might employ stigma theory and examine how difference in speech and appearance may alter social interaction and identity. Studies of school performance, career attainment, dating, peer social relations, teasing and discrimination may be done to determine how stigma experience results in altered interactions and self-concept (Broder and Strauss, 1991). It is necessary to introduce the concept of stigma as it does affect how the treated cleft patients have interacted with society and, to a certain degree, it has affected their life thus far.

Anxiety and depression have been reported to be twice as prevalent in adults with clefts compared with normal controls (Ramstad et al., 1995). Dissatisfaction with facial appearance has been found to be a predictor of depression among adults with clefts and controls (Marcusson et al., 2002). Berk et al. (2001) examined social anxiety among adults with clefts and found significantly more social anxiety and avoidance among those with a cleft than among siblings and controls.

In summary, it appears that aesthetics in general, particularly facial aesthetics, plays a significant role in the individual's general perception of life, especially between the ages of 18 and 30 years (Jacobson, 1984; Harris and Carr, 2001). The individual's facial appearance is one of the most relevant measures of the success of treatment for cleft lip and palate (Asher-McDade et al., 1991). The aesthetic outcome can be judged by the patient's satisfaction, as well as by the verdict of independent experts and laymen. Together with the

complexity of the cleft deformity, various parts of the face may be involved. This can range from a scarred lip, a nasal deformity, missing tooth/teeth, malocclusion, hypoplastic maxilla to a nasal voice or a combination of these features.

It was found that bilateral cleft lip and palate patients were significantly less satisfied with the appearance of the upper lip and nose (Oosterkamp et al., 2007). Marcusson and co-workers (2002) found that many adults with repaired clefts generally expect better results from surgery, particularly the nose. It was also found that women rated their facial appearance significantly poorer than the opinion of experts, with the difference especially marked in women aged 24 to 30 years (Marcusson et al., 2002). A large proportion of adults with clefts expressed the need for further surgical treatment (Marcusson et al., 2002). From a a third party perspective of bilateral cleft lip nasal deformity correction, it was found that there were no significant differences between the aesthetic ratings of the professional and lay raters (Lo et al., 2001).

Not only is it important for the cleft patient to be satisfied with the treatment results, the perception of the layperson in the society with whom they interact can influence how successful these individuals with clefts integrate into society, either in work, education, or social situations. A better understanding of the differences in facial aesthetics perceptions between the professional member of a cleft team, the lay person and the cleft individual, would be an invaluable aid in cleft treatment planning. Discussion of treatment outcomes with the patient, as well as management of patient expectations would be improved.

To the best of our knowledge, there has been no study to date which has compared perceptions of facial aesthetics in treated adult cleft patients between a panel of professional and lay people raters. Nor have there been any studies on the difference in perceptions between a cleft affected lay person and a lay person without a cleft, including studies on any gender differences between these groups. The aim of this study was to evaluate the opinions

of professionals and of lay people regarding the facial appearance of adults treated for orofacial clefting, as well as their perceptions about whether further surgery was required to correct the facial appearance. The study also investigates whether there are differences in opinions within the different professional groups, between lay people with and without a cleft as well as gender differences within the lay people with and without a cleft.

MATERIALS AND METHODS

Participants were recruited from cleft patients treated at the Children, Youth and Women's Health Service under the Australian Craniofacial Unit over the last 34 years (1975 to 2009). Participants all had surgery to correct their unilateral or bilateral cleft lip, cleft palate, cleft lip and palate, and to correct jaw size discrepancies. Inclusion criteria were non-syndromic cleft patients aged 18 years or over who had completed their cleft treatment at this centre. With the consent of the patients, photographic records were obtained. The photographs were taken following the completion of all treatment including orthognathic surgery as well as revision surgery. The photographs were standardised using computer software (Adobe Photoshop Windows PC version CS8.0) for size, background and brightness. For each patient, three views were presented; including a frontal, left profile and right profile view. These images were cropped, re-scaled and projected onto a screen to be assessed by a panel of expert and lay person raters. The names on the photographs were removed and a random number was assigned to each patient's photograph for identification purposes by one researcher (PF). The raters were asked to rate the photographs according to attractiveness of the patient's nose, lips and overall facial appearance. The raters were also asked whether they thought further surgery was required. Both measures were rated on a Visual Analogue Scale.

Facial aesthetics rating

The Visual Analogue Scale was preferred over the Likert Scale because the former employs a 100 mm scale with a broad range of distinctive possibilities, whereas the latter uses ratings from 1 to 5 or from 1 to 10 (Sinko, 2005). According to Jaeschke et al. (1990), both methods of presenting responses options show the same level of construct validity and responsiveness. In this study, high scores indicate good aesthetics for the first measure and high perceived need for further surgery for the second measure.

The nine members of the rating panel comprised health professionals (a non-surgical group and a surgical group), adult lay people above the age of 18 years (a lay group without cleft and a lay group with cleft). The rating members were recruited with a purposive sampling method. The groups are as follows:

1. Non surgical group: a male orthodontist and a female dentist from Adelaide Dental Hospital; a female psychologist who is in the private sector.
2. Surgical group: a male plastic surgeon with extensive (5 years) cleft experience and regularly performs cleft surgery at the Australian Craniofacial Unit; a female plastic surgeon with little involvement with cleft surgery from the private sector.
3. Lay people with cleft: a female and a male volunteer above the age of 18 years. Both volunteers were recruited from the Australian Craniofacial Unit.
4. Lay people without cleft: a female and a male volunteer above the age of 18 years. Both volunteers were recruited through the University of Adelaide staff.

The professional raters were not involved in the treatment of the cleft patients who were assessed, but had clinical experience with cleft patients. Assessment of facial aesthetics using this method has been conducted in several studies, and has been shown to be both valid and reliable (Howells and Shaw, 1985; Tobiasen et al., 1991; Lo et al., 2002; Marcusson et

al., 2002; Sinko et al., 2005; Tatarunaite et al., 2005; Kenealy et al., 2007). Other investigations also confirm that a high agreement on facial aesthetics can be found among panels with different backgrounds (Lundström et al., 1987; Peerlings et al., 1995).

Statistical Analysis

Mean scores were generated from the Visual Analogue Scales for each of the components measured (nose, lips and face), with high scores indicating good aesthetics for the first measure and high perceived need for the second measure. The following groupings were made: professional (dentist, orthodontist, psychologist, 2 plastic surgeons), lay (2 lay people without a cleft [male and female], 2 lay people with a cleft [male and female]). The professional group was further sub-divided into a non-surgical group: dentist, orthodontist and psychologist, versus a surgical group: 2 plastic surgeons (one with extensive cleft experience and the other with less cleft experience. This would enable comparisons of ratings between surgeons with different backgrounds). Students' t-tests were used to compare group scores, with findings considered statistically significant when P values were 0.05 or less.

ETHICAL APPROVAL

Ethics approval was granted by the Children, Youth and Women's Health Service Human Research Ethics Committee.

RESULTS

Of the 112 patients who satisfied the recruitment criteria, 88 patients agreed to participate in the study; a response rate of 79 percent. 3 patients declined to participate in the study, 2 patients were deceased, and 19 patients were not contactable (Possibly due to a change of address and/or deceased). Of the 88 participants, photographs of 80 participants were used

for the facial aesthetics assessment because there were incomplete photographic records for 5 patients and 3 patients did not have standardized profile photographs. Of the 80 participants, 41 were female and 39 were male. There were 44 participants with bilateral cleft lip and palate, 33 participants with unilateral cleft lip and palate, one participant with isolated cleft palate, one participant with submucous cleft palate, and one participant with unilateral cleft lip. The participants ranged from 18 years to 64 years of age. Mean age was 31 years (SD = 11.34) and the median age was 29 years.

The mean scores for the aesthetic evaluations by the raters of the 80 adults with treated clefts are shown in Table 1. In the attractiveness ratings, the professionals rated the treated adults with cleft as significantly less attractive in all components of the face [nose (44.5); lips (45.4); face (50.0)] compared with the lay raters [nose (57.7); lips (56.1); face (63.1)]. The lay people with a cleft rated the participants as significantly more attractive in all components of the face [nose (69.7); lips (66.2); face (72.2)] compared with the lay people without a cleft [nose (45.7); lips (46.1); face (54.1)]. Amongst the raters with a cleft, the female rated the nose as significantly less attractive, although she rated the face as significantly more attractive [nose (66.6); face (76.1)] compared with the male rater with a cleft [nose (72.8); face (68.3)]. Amongst the professional raters, the non-surgical professionals (dentist, orthodontist and psychologist) rated all components of the face as significantly less attractive [nose (42.9); lips (40.1); face (45.3)] compared with the plastic surgeons [nose (49.0); lips (51.0); face (57.1)].

The mean scores for the perceived need for further surgery of the 80 adults with treated clefts are shown in Table 2. There was significant disagreement between the professional raters and the lay raters with regards to perceived need for further surgery. The professional raters believed surgery was not required. While the lay raters perceived the opposite. Amongst the lay raters, those with a cleft considered further surgery was required

less often than the raters without a cleft. Interestingly, there was significant disagreement between the female and male raters with a cleft, with the female rater perceiving that further surgery was required less often than the male rater. There was also significant disagreement amongst the professional raters. The non-surgical professionals (dentist, orthodontist and psychologist) considered further surgery to be needed less compared with the surgical professionals (plastic surgeons).

DISCUSSION

The findings of the present study showed that there were significant differences between the perceptions of facial aesthetics between professionals and lay people. The treated cleft cohort was rated as significantly less attractive by the professionals compared with lay people, indicating that the professionals have a different concept of facial aesthetics. This is in contrast to previous studies where no difference in ratings was shown between the professionals and the lay people (Tobiasen et al., 1991; Lo et al., 2002 Sinko et al., 2005). In a cohort of 70 unilateral and bilateral cleft lip and palate participants with an age range of 18 to 30 years, Sinko et al. (2005) found significant agreement in facial aesthetic ratings between the medical staff and non-medical staff on the rating panel. Similarly, the current findings differed from those of Tobiasen and co-workers (1991), in which children, adolescents and medical students rated the facial aesthetics of cleft patients the same way as plastic surgeons. A study by Lo et al. (2002), with 64 bilateral cleft lip patients, revealed that both the professional and lay people groups rated their nasal aesthetics similarly. The difference between these findings and those of the present study could be due to the difference in rating groups. The rating panel in the former study comprised five surgeons and five lay people, whereas the raters in the present study included other professionals such as an orthodontist, dentist, psychologist, and the lay raters included cleft affected individuals.

This difference in rating panel composition may have influenced the aesthetic ratings. However, the rating panel composition of the present study may produce a more representative aesthetic rating, as it takes into account the different professionals of a cleft team whom, with their different expertise, may influence the type and or course of treatment, as well as the general public's perception. The inclusion of the raters with a cleft in the present study ensures that further insight is gained into the perceptions of the person whom the cleft treatment ultimately affects.

The ratings by the lay people with a cleft were significantly higher than those lay people without a cleft. Individuals with a cleft are familiar with orofacial clefting and its effects on facial appearance and may not perceive it in as negative a light as the individual without a cleft. Since no previous studies have investigated the facial aesthetics perceptions of an adult with a cleft, further investigations with a greater number of raters with a cleft are warranted, in order to strengthen the validity of the findings. Such an insight would be of great assistance in the planning of the cleft treatment and discussion of treatment outcomes with the patient, as well as managing the expectations of the cleft patient from the treatment.

The attractiveness ratings of the female rater with a cleft were significantly higher than those of the male rater with a cleft. Although no previous studies have investigated the perceptions of an individual with a cleft on facial aesthetics, the findings from some studies on cleft self ratings may be extrapolated for comparison. The facial aesthetics self ratings for a cohort of female participants with clefts in the 24 to 30 year age group were significantly lower compared with other male participants with clefts (Sinko et al., 2005). Interestingly, in the same study, female participants with clefts in the 18-23 year age group had the highest self ratings among all the participants with clefts. If the results from Sinko and co-workers are to be extrapolated, it would indicate that female individuals with a cleft between 24 to 30 years old would tend to have a more critical perception of facial aesthetics compared to male

individuals with a cleft, which should then translate to a lower aesthetic rating than the male raters with a cleft when assessing other individuals with a cleft. However, this was not consistent with the findings of the current study. Further investigations are required to gain further insight into the gender difference in perceptions.

The ratings from the surgical members of the professional group were significantly higher than that of the non-surgical members. In an assessment of facial aesthetics of 70 cleft patients, Sinko et al. (2005) showed that there was no significant difference between medical and non-medical raters. The panel of raters comprised a maxillofacial surgeon, orthodontist, psychologist, dental assistant and speech therapist, although it was unclear which members were considered “medical” and which were “non-medical”. A possible explanation for the present study’s findings may be that the surgical professionals are experienced in dealing with the surgical corrections of clefts, and hence may rate the cleft facial aesthetics higher in light of the potential complications and technical difficulties in achieving optimal aesthetic results.

When asked whether further surgery was required to improve the facial appearance of the cleft cohort, the professionals considered surgery to be required significantly less so compared to the lay people. However, it should be noted that the mean score of the professionals was 39.1, indicating that they tended to disagree that further surgery was needed, whilst the mean score of the lay people was 52.6, which indicated that they neither agreed nor disagreed that further surgery was needed. Sinko et al. (2005) found no difference between the medical and non-medical members of the rating panel in their perception of the need for further surgery. Their findings also indicated that the rating panel tended to deem further treatment unnecessary. The difference in the present findings may be due to the difference between the size and composition of the rating panels. In the current study, there was a greater number of lay people and non-medical raters, as well as the inclusion of lay

people with clefts. In the former study, there were no lay people, only "non-medical" members of the assessment panel. A possible reason for the professionals' lower perceived need for further surgery may be due to their better understanding of the limitations of surgery in correcting facial deformities. In many instances, despite having rated facial aesthetics lower, the professional members of the assessment panel may have deemed that reasonable aesthetic results have been achieved for the cleft patients, hence the higher disagreement on the need for further surgery. Amongst the professional raters, despite a lower attractiveness score, the non-surgical members deemed surgery was needed significantly less than the surgical members. Again, the difference in perceived need for further surgery may be due to the difference in surgical understanding of one group over the other. It is also interesting that there was no significant difference in the perceived need for further surgery between the female and male lay people.

On the other hand, the lay people with clefts deemed further surgery to be needed significantly less than lay people without clefts. This was consistent with their significantly higher attractiveness ratings. Moreover, the non-cleft lay people had the highest agreement that further surgery was required compared to the other groups of raters, which was also reflected in their low attractiveness ratings. It has been suggested that many patients with clefts tend to be tired of seeking further treatment (Sinko et al., 2005), especially having experienced a protracted course of cleft treatment from childhood to adulthood. It may be possible that the cleft lay people on the rating panel projected their own experiences to the assessment of the cleft cohort, hence the low agreement with regards to the need for further surgery.

The female rater with a cleft deemed further surgery to be needed significantly less than the male rater with a cleft. This was also consistent with and reflected in the higher attractiveness ratings by the female rater with a cleft. This finding contrasts with previous

findings where the female participants with a cleft expressed a desire for further treatment twice as often as the male participants with a cleft (Sinko et al., 2005). However, it must be kept in mind that the previous findings were based on the self reports of individuals with clefts, which may not apply to the individual's perception of other patients with clefts. Further studies regarding the perception of an individual with a cleft of other patients with clefts are warranted.

In the current study, 80 of the initial 112 patients were included in the aesthetic assessment. It may be argued that non-participants may have somehow skewed the ratings from the panel of raters. Three of the patients declined to participate. It is possible that their refusal was due to the adverse outcome of their cleft treatment and their inclusion in the study may have produced significantly lower aesthetic ratings. Alternatively, they may not have any problems from their cleft treatment and their inclusion may have produced significantly higher aesthetic ratings. Future investigations with a higher response rate would overcome this confounder. However, whether the aesthetic ratings were skewed higher or lower, it would still not influence the differences in ratings between the different raters.

The number of members within each subgroup of the rating panel may be considered small. However, the size and composition are comparable to or greater than previous studies (Lo et al., 2002; Marcusson et al., 2002; Sinko et al., 2005). It is possible that the aesthetic ratings and perceived need for further treatment may not be fully representative of the respective subgroups. It would have been ideal to have a greater number of members within each subgroup in order to overcome this potential confounder. However, it should be noted that there was a large number of ratings made by each rater. This contributed to the strength of the current study.

CONCLUSION

Differences exist between professionals who are part of the cleft treatment team, lay people and individuals with a cleft in the perception of facial aesthetics of treated adult cleft patients. The professionals rated the facial aesthetics as significantly less attractive compared with the lay people with and without clefts, although further surgery was also deemed less necessary. The non-surgical professionals reported lower facial aesthetics ratings and also deemed further surgery to be less necessary compared with their surgical counterparts. Among the lay people, the members with clefts reported higher facial aesthetics ratings and had a lower perception of necessity for further surgery compared with the non-cleft lay people. In addition, the female layperson with a cleft reported higher facial aesthetics ratings and also deemed further surgery to be needed less than the male layperson with a cleft.

Since there are very few studies that have investigated the subject at hand, comparisons of the findings from the present study have been made with some extrapolations from previous studies. This further emphasizes the need for further studies to be conducted in this area. Future research should focus on gaining further insight into the differences between the perceptions of the cleft and non-cleft affected individual and the professionals involved with the cleft treatment.

ACKNOWLEDGEMENTS

The authors would like to thank Ms Joanna Chen and Ms Beverly Ellis for their assistance in data entry and data collection.

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Table1. Mean aesthetic evaluations of adults with treated clefts
(Scores are in "mm", with a possible range of 0 to 100. Higher scores reflecting "very attractive")

How attractive would you rate the following:	**Nose** mean (standard error)	**Lips** mean (standard error)	**Face** mean (standard error)
Professional (n=5)	44.5 (1.7)*	45.4 (1.6)*	50.0 (1.4)*
Lay; cleft, non-cleft (n=4)	57.7 (1.7)	56.1 (1.8)	63.1 (2.4)
Lay; cleft (n=2)	69.7 (1.6)*	66.2 (1.9)*	72.2 (1.5)*
Lay; non-cleft (n=2)	45.7 (2.4)	46.1 (2.2)	54.1 (4.2)
Cleft; female (n=1)	66.6 (1.9)*	66.2 (2.1)	76.1 (1.1)*
Cleft; male (n=1)	72.8 (2.2)	66.2 (2.4)	68.3 (2.3)
Professional; dentist, orthodontist, psychologist (n=3)	42.9 (1.7)*	40.1 (1.8)*	45.3 (1.6)*
Professional; plastic surgeons (n=2)	49.0 (1.6)	51.0 (1.7)	57.1 (1.4)

*P<0.05

Table2. Mean perceived need for surgery for adults with treated clefts
(Scores are in "mm", with a possible range of 0 to 100. Higher scores reflecting "strongly agree")

Do you think further surgery is required to change appearance of the following:	**Nose** mean (standard error)	**Lips** mean (standard error)	**Face** mean (standard error)
Professional (n=5)	46.9 (2.1)*	45.2 (2.0)*	39.1 (1.8)*
Lay; cleft, non-cleft (n=4)	57.8 (1.5)	58.7 (1.3)	52.6 (1.3)
Lay; cleft (n=2)	46.3 (1.4)*	46.2 (1.2)*	39.7 (1.3)*
Lay; non-cleft (n=2)	69.3 (2.5)	71.2 (2.3)	65.5 (2.5)
Cleft; female (n=1)	28.3 (2.1)*	30.4 (2.2)*	18.1 (1.4)*
Cleft; male (n=1)	64.4 (2.5)	62.0 (2.3)	61.4 (2.5)
Professional; dentist, orthodontist, psychologist (n=3)	37.3 (2.3)*	41.8 (2.4)*	34.1 (2.1)*
Professional; plastic surgeons (n=2)	57.1 (1.9)	54.6 (2.0)	46.6 (1.7)

*P<0.05

Chapter 5

Concluding Remarks

The aim of this study of treated South Australian adult patients with the diagnosis of non-syndromic cleft, was to compare differences in the health-related quality of life as well as the degree of adverse impacts of oral disease on wellbeing and quality of life between these patients and an age- and gender-matched group without cleft in South Australia. Furthermore, the study aimed to compare the treated cleft patients' self-reported satisfaction with facial appearance as well as self-perception of facial attractiveness with those of a panel of expert and layman judges.

There were no detectable age or sex differences in either general health-related quality of life or oral health impact among participants with treated orofacial clefting included in our sample. This refuted the study's hypotheses that there would be differences in the HRQoL between different age groups, gender, as well as between the cleft cohort and the general population. When compared with population-level estimates, the oral health impact among these Australians with treated orofacial clefting was poor, which confirmed the initial hypothesis. However, the general health-related quality of life showed mixed results, with the vitality and mental health components being poorer in the cleft group compared with population-level estimates. Our findings indicate that treatment for orofacial clefting does not entirely remove the factors contributing to poor general health-related quality of life and oral health.

Differences exist between professionals who are part of the cleft treatment team, lay people and individuals with a cleft in the perception of facial aesthetics of treated adult cleft patients. This refuted the initial hypotheses of no differences in ratings and perceived need for further surgery between the professionals and lay raters. The professionals rated the facial aesthetics as significantly less attractive compared with the lay people with and without clefts, although further surgery was also deemed less necessary. The non-surgical

professionals reported lower facial aesthetics ratings and also deemed further surgery to be less necessary compared with their surgical counterparts, which was contrary to the initial hypothesis. Among the lay people, the members with clefts reported higher facial aesthetics ratings and had a lower perception of necessity for further surgery compared with the lay people without a cleft. In addition, the female layperson with a cleft reported higher facial aesthetics ratings and also deemed further surgery to be needed less than the male layperson with a cleft. These results refute the initial hypotheses that lay raters with a cleft and female lay raters would have lower aesthetic ratings and higher perceived need for further surgery compared to the lay raters without a cleft and male lay raters.

There were some limitations to the present study. These are as follows:

- Inability of OHIP-14 to elicit the extent of the effect of clefting on the patient's general oral health.
- Limitations relating to the use of historical normative data for comparisons.
- The study's assumption that the self-report questionnaires were completed by the patients without influence or input from anyone else.
- The limited sample size within each cleft type.
- The limited sample size within each age group.
- The limited sample size within each rating sub-group.

These limitations have been discussed in detail in Article 1 and 2 in Chapter 4.

Some of the strengths of the current study are as follows:

- All the patients were treated at a well-established cleft centre, under the same standardized protocol.
- Surgery for all the patients were carried out primarily by one surgeon.

- The patients have been reviewed over a long period of time, with some up to 34 years.
- There was a high number of participants and a high response rate.

These factors have contributed to the strength of the study and have been discussed in detail in Article 1 and Article 2 in Chapter 4.

Despite the limitations of the study, the aims of the study have all been achieved. The results refuted all the initial hypotheses with the exception of one. The hypothesis that the treated cleft cohort would have a worse oral health impact compared to the general population without a cleft was confirmed.

Since there are very few studies that have investigated the subject at hand, comparisons of the findings from the present study have been made on some extrapolations from previous studies. This further emphasizes the need for further studies to be conducted in this area. Further investigations with larger sample sizes for each cleft type as well as each age group are warranted. This would allow meaningful comparisons between the different types of clefts as well as comparisons within and between different age groups. With regards to the significant differences in QoL found in the current study, future research should focus on the reason for these differences. Perhaps a smaller-scale qualitative approach could be undertaken in future research in order to explore the differences in QoL.

Future research should focus on gaining further insight into the differences between the perceptions of the cleft and non-cleft affected individual and the professionals involved with the cleft treatment. A potential study could compare the treated cleft patients' self-reported satisfaction with facial appearance as well as self-perception of facial attractiveness with those of the expert and lay people raters. The patients' desire for further treatment can also be compared with the perception of the raters. A further aim of future research could set out to determine whether psychological aspects influence the health-related quality of life of adult patients with a repaired cleft lip and palate.

Chapter 6

Appendices

Appendix 1

Information letter to potential participants

Thursday 14th of May, 2009

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Facial Appearance and Well-Being Following Treatment of People with Clefts

Dear Sir/ Madam,

The doctors and team at the Australian Craniofacial Unit, Women's and Children's Hospital, in a joint effort with the Orthodontic Department, Adelaide Dental Hospital would like to invite you to consider participating in an important project that is looking at how facial appearance and psychosocial outcome is affected following treatment of people with clefts.

Success of cleft treatment is often assessed in terms of oral and facial function and facial appearance. However, the way in which people with clefts adjust and adapt psychologically following cleft treatment may be just as useful in measuring the success of treatment. The overall goal of treatment by cleft teams is to improve the medical, dental, speech, educational, and cosmetic needs while encouraging positive self-esteem, and interaction with other people in the community.

We are extremely keen to know how people with clefts, are getting on in life with respect to the aspects mentioned above. Aside from assessing the result of the facial appearance and patient satisfaction with the cleft repair, the research will also help us understand the psychosocial aspect and quality of life for people with clefts. By gaining a better understanding of how people with clefts are getting on in life following the completion of treatment, the cleft team can learn to be more effective and thorough in the early stages of treatment as well as understand the psychological needs of the patient.

We would like to invite you to be part of this study by answering some questions about your thoughts on your facial appearance, your satisfaction levels with your treatment, how you feel about certain areas of your health and how you function in your daily activities, your oral health and function. We also need some general information about you. The questionnaire should take about an hour for you to complete.
In addition, we would like you to give consent to the use of the existing photographic records that were taken during your treatment here at the Women's and Children's Hospital. A set of the photos of your face, taken at the completion of your treatment, will be shown to a panel of nine judges for assessment. We will ask the judges some questions in regards to their thoughts on the relative attractiveness of the faces shown on the photos.
All photographic records will be de-identified, and in no way will your name or your identity be revealed to the judges or any other members of the public.
All information gathered will be kept confidential and in no way will you be identified in the analysis or reporting of this research.
Below I have answered some frequently asked questions in regards to the research.

Do I have to participate?
NO. Although we would be grateful for any information you could provide us, and by participating you will be contributing to research on improving the lives of people with clefts, you are under no obligation to complete the questionnaire.
Will I be able to remain anonymous?

YES. You will not be identified in any way in the analysis. Data gathered from your responses in the questionnaire will be grouped with responses from other cleft patients and reported together. Although there will be some identifying information on the questionnaires that will allow us to contact you, we can guarantee that any information you supply will remain completely confidential.

What will happen with the information I supply?
All information will be coded and entered into a database and analysed by the researchers so that we can determine how the treated cleft patients are getting on in life in the community. This information may also have an impact on how cleft patients are treated and managed by health professionals.

Who will be able to see my photographs?
Following de-identification of the photographs, only the panel of judges and the researchers involved with the study will be able to see your photographs. The photographs will only be displayed for the panel of judges during the assessment period.

Who is on the panel of judges?
The panel of judges will comprise of both experts and lay-people. The experts on the panel will comprise of 5 health professionals involved with treating clefts. However, in order to prevent bias in the study, none of the health professionals would have been directly involved with your treatment at the Women's and Children's Hospital. There will be 4 laypeople making up the rest of the judging panel.

Will the judges be able to identify me from my photographs?
No. All names will be removed from the photographs and they will be de-identified. Your identity will not be revealed to the judges or any member of the public.

Will I have to do anything else beside complete the questionnaire?
NO. Once you have completed the questionnaire and returned it to us by mail, there will be nothing further for you to do.

Are there any risks?
We believe that the measure to be used in this study will not cause you any distress or anxiety. You are reminded that participation is not compulsory and you are able to withdraw from this study at any time without prejudice to your future treatment or relationship with the health service.

Will I be informed of the results of this study?
A summary of the results of the study will be mailed out to you upon request. Please indicate that you would like to receive a copy by ticking the box at the end of the questionnaire.

How do I know this research is legitimate?
This research has been approved by the **Children, Youth & Women's Health Service** and the Human Research Ethics Committee, the **University of Adelaide**. Should you wish to discuss the approval process with a representative of the Ethics committee, or have any concerns about the ethics of this study, please contact Ms Brenda Penny, Secretary of the CYWHS Research Ethics Committee, on 8161 6456.

Will I be paid to participate?
No, your participation in the first phase of the study is entirely voluntary.

Yes, I want to participate. What do I do now?
If you agree to take part, please fill out the enclosed Consent Form and Questionnaire. Once signed and completed, please send the consent form and questionnaire back to us in the self-addressed reply paid envelope provided.

Thank you for taking the time to consider this request. Your participation in this research is highly appreciated and will provide valuable information for the health and well-being of cleft patients. Should you require any additional information regarding this research, please contact Dr Peter Foo at the Orthodontic Department, Adelaide Dental Hospital on (08) 83035153 or **peter.foo@adelaide.edu.au**.

Yours sincerely

Professor David David

Head of Department
Australian Craniofacial Unit,
Women's and Children's Hospital

Appendix 2

Participant consent form

CONSENT FORM

I (name)_____

hereby consent to my involvement in the research project entitled:

Facial Appearance and Well-Being Following Treatment of People with Clefts

	Please Tick ✓
• I have read and understood the Information Sheet on the above project and understand that I am being asked to provide details of myself.	☐
• I understand that I may not directly benefit by taking part in this research.	☐
• I understand that while information gained in the study may be published, I will not be identified and all individual information will remain confidential.	☐
• I understand that I can withdraw from the study at any stage up until the end of the collection of data.	☐
• I understand that there will be no payment for taking part in this part of the study.	☐
• I am aware that I should retain a copy of the Information Sheet and Consent Form for future reference.	☐
• I **agree** to have photographic records of my face displayed to an assessment panel of judges for their feedback.	☐

Signed: _____

Date: _____

CONSENT FORM TO BE RETURNED TO RESEARCHER – PLEASE PLACE IN THE REPLY-PAID ENVELOPE PROVIDED AND MAIL BACK TO THE FOLLOWING ADDRESS:

UNIVERSITY OF ADELAIDE
ORTHODONTIC UNIT
SCHOOL OF DENTISTRY
SA, 5005

Appendix 3

Second follow-up letter to potential participants

Date

Prof David David
Australian Craniofacial Unit
Women's & Children's Hospital
Lv2 Good Friday Building
72 King William Street
North Adelaide SA 5006
Tel 08 81617235

www.cywhs.sa.gov.au

Facial Appearance and Well-Being Following Treatment of People with Clefts

Dear Sir / Madam,

Previously, we sent you some information with regards to the research project we are conducting. We requested for your participation by signing and returning the consent form and questionnaire provided. The doctors and team at the Australian Craniofacial Unit, Women's and Children's Hospital and the Orthodontic Department, Adelaide Dental Hospital, would like to thank you for agreeing to participate in the study.

Our records show that you have not returned the completed consent form and questionnaire sent to you, dated "Thursday, 14 May 2009". This is just a friendly reminder for you to return the signed and completed consent form and questionnaire in the reply paid envelope provided as soon as possible. It would be very helpful for us to obtain all the data from the completed questionnaires as soon as possible for the success of the study. If you have not received the questionnaire by mail, please find enclosed a copy of the questionnaire and a self-addressed reply paid envelope. Please complete and sign the consent form and questionnaire, and return by mail to us. If, for some reason, you have decided to withdraw from the study, or should you require any additional information regarding this research, please contact Dr Peter Foo at the Orthodontic Department, Adelaide Dental Hospital on **(08) 83033102** or via email **peter.foo@adelaide.edu.au** and we will be able to answer any of your questions, or alternatively, remove your details from the study.

On behalf of the team at the Australian Craniofacial Unit, Women's and Children's Hospital, and the Orthodontic Department, Adelaide Dental Hospital, I would like to thank you for taking the time to consider this request. Your participation in this research is highly appreciated and will provide valuable information for the health and well-being of cleft patients.

Yours sincerely
Professor David David

Head of Department
Australian Craniofacial Unit,
Women's and Children's Hospital

Appendix 4

Patient Questionnaire

Facial Appearance and Well-Being Following Treatment of People with Clefts

Questionnaire

Date: _____

Name: _____

Date of Birth: _____

Sex: _____

(Your name is required for the purpose of matching the questionnaire findings to your treatment details from the casenotes. It shall remain confidential and you will not be identified in any way in the analysis)

INSTRUCTIONS

THE QUESTIONNAIRE.
This questionnaire asks how troubles with your teeth, mouth or dentures may have caused problems in your daily life. We would like you to complete the questionnaire even if you have good dental health. We would like to know how often you have had each of the 14 listed problems during the LAST YEAR.

HOW TO ANSWER THE QUESTIONS.
Each question on the left hand side of the page asks you about a particular dental problem. You should think about each question in turn, and circle the answer to the right of the question, to indicate how often you have had the problem during the last year.

EXAMPLES

If you occasionally had painful aching in your mouth, you would circle the answer as shown in this example.

3. Have you ever had painful aching in your mouth? VERY OFTEN FAIRLY OFTEN OCCASIONALLY HARDLY EVER NEVER DON'T KNOW

If you have never had the problem during the last year, circle "NEVER" as follows.

3. Have you ever had painful aching in your mouth? VERY OFTEN FAIRLY OFTEN OCCASIONALLY HARDLY EVER NEVER DON'T KNOW

HOW OFTEN have you had the problem during the last year? (circle your answer)

1. Have you had trouble pronouncing any words because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
2. Have you felt that your sense of taste has worsened because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
3. Have you had painful aching in your mouth?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
4. Have you found it uncomfortable to eat any foods because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
5. Have you been self conscious because of your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
6. Have you felt tense because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
7. Has your diet been unsatisfactory because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
8. Have you had to interrupt meals because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
9. Have you found it difficult to relax because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
10. Have you been a bit embarrassed because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
11. Have you been a bit irritable with other people because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
12. Have you had difficulty doing your usual jobs because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
13. Have you felt that life in general was less satisfying because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW
14. Have you been totally unable to function because of problems with your teeth, mouth or dentures?	VERY OFTEN	FAIRLY OFTEN	OCCAS-IONALLY	HARDLY EVER	NEVER	DON'T KNOW

Continue next page…

This survey asks for your views about your health. This information will help keep track of how you feel and how well you are able to do your usual activities.

Please answer these questions by "check-marking" your choice. Please select only one choice for each item.

15. In general, would you say your health is:

□1. Excellent □3. Good □5. Poor
□2. Very good □4. Fair

16. Compared to ONE YEAR AGO, how would you rate your health in general NOW?
□1. MUCH BETTER than one year ago.
□2. Somewhat BETTER now than one year ago.
□3. About the SAME as one year ago.
□4. Somewhat WORSE now than one year ago.
□5. MUCH WORSE now than one year ago.

17. The following items are about activities you might do during a typical day. **Does your health now limit you** in these activities? If so, how much?

Activities	**1. Yes, Limited A Lot**	**2. Yes, Limited A Little**	**3. No, Not Limited At All**
a) **Vigorous activities,** such as running, lifting heavy objects, participating in strenuous sports?	□1. Yes, limited a lot	□2. Yes, limited a little	□3. No, not limited at all
b) **Moderate activities,** such as moving a table, pushing a vacuum cleaner, bowling, or playing golf?	□1. Yes, limited a lot	□2. Yes, limited a little	□3. No, not limited at all
c) Lifting or carrying groceries?	□1. Yes, limited a lot	□2. Yes, limited a little	□3. No, not limited at all
d) Climbing **several flights** of stairs?	□1. Yes, limited a lot	□2. Yes, limited a little	□3. No, not limited at all
e) Climbing **one** flight of stairs?	□1. Yes, limited a lot	□2. Yes, limited a little	□3. No, not limited at all
f) Bending, kneeing or stooping?	□1. Yes, limited a lot	□2. Yes, limited a little	□3. No, not limited at all
g) Walking **more than a mile**?	□1. Yes, limited a lot	□2. Yes, limited a little	□3. No, not limited at all
h) Walking **several** blocks?	□1. Yes, limited a lot	□2. Yes, limited a little	□3. No, not limited at all
i) Walking **one** block?	□1. Yes, limited a lot	□2. Yes, limited a little	□3. No, not limited at all
j) Bathing or dressing yourself?	□1. Yes, limited a lot	□2. Yes, limited a little	□3. No, not limited at all

18. During the **past 4 weeks**, have you had any of the following problems with your work or other regular activities *as a result of your physical health*?

	Yes	**No**
a) Cut down on the **amount of time** you spent on work or other activities?	□1. Yes	□2. No
b) **Accomplished less** than you would like?	□1. Yes	□2. No
c) Were limited in the **kind** of work or other activities?	□1. Yes	□2. No

d) Had **difficulty** performing the work or other activities (for example it took extra effort)?	□ 1. Yes	□ 2. No

Continue next page…

19. During the **past 4 weeks**, have you had any of the following problems with your work or other regular daily activities **as a result of any emotional problems** (such as feeling depressed or anxious)?

	Yes	**No**
a) Cut down on the **amount of time** you spent on work or other activities?	□1. Yes	□2. No
b) **Accomplished less** than you would like?	□1. Yes	□2. No
c) Didn't do work or other activities as **carefully** as usual?	□1. Yes	□2. No

20. During the **past 4 weeks**, to what extent has your physical health or emotional problems interfered with your normal social activities with family, friends, neighbours, or groups?

□1. Not at all
□2. Slightly
□3. Moderately
□4. Quite a bit
□5. Extremely

21. How much **bodily pain** have you had during the **past 4 weeks**?

□1. None
□2. Very mild
□3. Mild
□4. Moderate
□5. Severe
□6. Very severe

22. During the **past 4 weeks**, how much did **pain** interfere with your normal work (including both work outside the home and housework)?

□1. Not at all
□2. A little bit
□3. Moderately
□4. Quite a bit
□5. Extremely

23. These questions are about how you feel and how things have been with you **during the past 4 weeks**. For each question, please give the one answer that comes closest to the way you have been feeling. How much of the time during the **past 4 weeks**…

	1. All of the time	2. Most of the time	3. A good bit of the time	4. Some of the time	5. A little of the time	6. None of the time
a) Did you feel full of life?	□1. All of the time	□2. Most of the time	□3. A good bit of the time	□4. Some of the time	□5. A little of the time	□6. None of the time
b) Have you been a very nervous person?	□1. All of the time	□2. Most of the time	□3. A good bit of the time	□4. Some of the time	□5. A little of the time	□6. None of the time
c) Have you felt so down in the dumps that nothing could cheer you up?	□1. All of the time	□2. Most of the time	□3. A good bit of the time	□4. Some of the time	□5. A little of the time	□6. None of the time
d) Have you felt calm and peaceful?	□1. All of the time	□2. Most of the time	□3. A good bit of the time	□4. Some of the time	□5. A little of the time	□6. None of the time
e) Did you have a lot of energy?	□1. All of the time	□2. Most of the time	□3. A good bit of the time	□4. Some of the time	□5. A little of the time	□6. None of the time
f) Have you felt downhearted and blue?	□1. All of the time	□2. Most of the time	□3. A good bit of the time	□4. Some of the time	□5. A little of the time	□6. None of the time
g) Do you feel worn out?	□1. All of the time	□2. Most of the time	□3. A good bit of the time	□4. Some of the time	□5. A little of the time	□6. None of the time
h) Have you been a happy person?	□1. All of the time	□2. Most of the time	□3. A good bit of the time	□4. Some of the time	□5. A little of the time	□6. None of the time
i) Did you feel tired?	□1. All of the time	□2. Most of the time	□3. A good bit of the time	□4. Some of the time	□5. A little of the time	□6. None of the time

Continue next page…

24. During the **past 4 weeks**, how much of the time has your **physical health** or **emotional problems** interfered with your social activities (like visiting with friends, relatives, etc.)?

□1. All of the time
□2. Most of the time.
□3. Some of the time
□4. A little of the time.
□5. None of the time

25. How TRUE or FALSE is **each** of the following statements for you?

	1. Definitely true	2. Mostly true	3. Don't know	4. Mostly false	5. Definitely false
a) I seem to get sick a little easier than other people?	□1. Definitely true	□2. Mostly true	□3. Don't know	□4. Mostly false	□5. Definitely false
b) I am as healthy as anybody I know?	□1. Definitely true	□2. Mostly true	□3. Don't know	□4. Mostly false	□5. Definitely false
c) I expect my health to get worse?	□1. Definitely true	□2. Mostly true	□3. Don't know	□4. Mostly false	□5. Definitely false
d) My health is excellent?	□1. Definitely true	□2. Mostly true	□3. Don't know	□4. Mostly false	□5. Definitely false

Socio-demographic Status

In this section you will be asked questions with regards to your socio-demographic status.

26. Marital status. Are you:

□1. Married
□2. Separated
□3. Divorced
□4. Widowed
□5. Never married

27. How many children do you have:

□1. Younger than 15 years old? ___
□2. Older than 15 years old? ___

28. In what state is your usual residence?

□1. New South Wales
□2. Victoria
□3. South Australia
□4. Queensland
□5. Northern Territory
□6. Western Australia
□7. Tasmania
□8. Australian Capital Territory
□9. Other territories

29. What is the highest year of school you completed?

□1. Year 12 or equivalent
□2. Year 11 or equivalent
□3. Year 10 or equivalent
□4. Year 9 or equivalent
□5. Year 8 or below
□6. Did not go to school

Continue next page…

30. What non-school qualification level of education have you completed?

□ 1. Postgraduate Degree
□ 2. Graduate Diploma and Graduate Certificate
□ 3. Bachelor Degree
□ 4. Advanced Diploma and Diploma
□ 5. Certificate
□ 6. None

31. What is your employment status?

□ 1. Full-time employment
□ 2. Part-time employment
□ 3. Unemployed
□ 4. Full-time student
□ 5. Part-time student
□ 6. Home duties
□ 7. Other

32. What type of occupation are you in?

□ 1. Manager
□ 2. Professional
□ 3. Technician and trade worker
□ 4. Community and personal service worker
□ 5. Clerical and administrative worker
□ 6. Sales worker
□ 7. Machinery operator and driver
□ 8. Labourer
□ 9. Other (Please specify)

33. What is your gross individual weekly income?

□ 1. Negative/Nil income
□ 2. $1 - $149
□ 3. $150 - $249
□ 4. $250 - $399
□ 5. $400 - $599
□ 6. $600 - $799
□ 7. $800 - $999
□ 8. $1000 - $1,299
□ 9. $1,300 - $1,599
□ 10. $1600 - $1,999
□ 11. $2000 or more

34. Do you currently have a Health Care Card?

□ 1. Yes
□ 2. No

35. Is your current dwelling:

□ 1. Rented accommodation
□ 2. Being paid off
□ 3. Owned outright
□ 4. Rent-free accommodation
□ 5. Other

36. How many people were staying in your household on the previous night? _____

37. Do you own your own car?

□ 1. Yes
□ 2. No

38. How would you rate you own health? Would you say that it is …

□ 1. Excellent
□ 2. Very good
□ 3. Good
□ 4. Fair
□ 5. Poor

Continue next page…

39. In which country were you born?

□ 1. Australia
□ 2. England (UK)
□ 3. New Zealand
□ 4. Italy
□ 5. Vietnam
□ 6. China
□ 7. Greece
□ 8. Germany
□ 9. Philippines
□ 10. India
□ 11. Other (please specify)
_____

Satisfaction with facial appearance and self-perception of facial attractiveness

40. With regards to your cleft treatment, how satisfied are you with the result of the appearance of the following (Please indicate how satisfied you are by marking on the line with an "X"):

i. Nose
Very dissatisfied Very satisfied

ii. Lips
Very dissatisfied Very satisfied

iii. Teeth
Very dissatisfied Very satisfied

iv. Face
Very dissatisfied Very satisfied

v.

41. With regards to your face, how attractive would you rate the following (Please indicate how attractive by marking on the line with an "X"):

i. Nose
Very unattractive Very attractive

ii. Lips
Very unattractive Very attractive

iii. Teeth
Very unattractive Very attractive

iv. Face
Very unattractive Very attractive

42. With regards to your face, do you think further surgery is required to change the appearance of the following (Please indicate how strongly you agree by marking the line with an "X"):

i. Nose
Strongly disagree Strongly agree

ii. Lips
Strongly disagree Strongly agree

iii. Teeth
Strongly disagree Strongly agree

iv. Face
Strongly disagree Strongly agree

THANK YOU FOR COMPLETING THE QUESTIONNAIRE. PLEASE INDICATE IF YOU WOULD LIKE TO RECEIVE A SUMMARY OF THE RESULTS OF THE STUDY IN THE MAIL BY TICKING THE BOX BELOW AND PROVIDE A MAILING ADDRESS FOR US TO MAIL THE SUMMARY.

□ **Mailing Address:** _____

_____

_____

QUESTIONNAIRE TO BE RETURNED TO RESEARCHER – PLEASE PLACE IN THE REPLY-PAID ENVELOPE PROVIDED AND MAIL BACK TO THE FOLLOWING ADDRESS:

DR PETER FOO
ORTHODONTIC UNIT
THE UNIVERSITY OF ADELAIDE
REPLY PAID 498
ADELAIDE SA 5001

Appendix 5

Raters' Questionnaire

Facial Appearance and Well-Being Following Treatment of People with Clefts

Panel Survey

Date: _____

Name: _____

(Your name is required for the purpose of matching and comparing the survey findings. It shall remain confidential and you will not be identified in any way in the analysis)

1254

1. With regards to the face, how attractive would you rate the following (Please indicate how attractive by marking on the line with a "/"):

i. Nose
Very unattractive Very attractive

---

ii. Lips
Very unattractive Very attractive

---

iii. Face
Very unattractive Very attractive

---

2. With regards to the face, do you think further surgery is required to change the appearance of the following (Please indicate how strongly you agree by marking on the line with a "/"):

i. Nose
Strongly disagree Strongly agree

---

ii. Lips
Strongly disagree Strongly agree

---

iii. Face
Strongly disagree Strongly agree
