The Prevalence of Caries and Tooth Wear in Cleft Children aged between 6 months and 6 years in the West of Scotland.

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Abstract

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Aim

To compare the prevalence of caries and tooth wear in the primary teeth of children with clefts with comparable national data and to determine whether there is an association between the occurrence of a cleft and socioeconomic deprivation in the West of Scotland.

Method

Ethical approval was granted from the West of Scotland Ethics Committee and the local Research and Development Office. Children attending the Oral Orthopaedic Prevention Clinic at Glasgow Dental Hospital and School were examined for caries according to British Association for the Study of Community Dentistry (BASCD) criteria (Pitts *et al.*, 1997) by two trained and calibrated examiners (KB and RW). A copy of the data collection criteria is appended (Appendix 1). Subjects were divided into five age groups: 0.5 – 1.49, 1.5 – 2.49, 2.5 – 3.49, 3.5 – 4.49 and 4.5 – 6.0 years. Mean dmft scores were compared to available national data (NDIP, 2008) for nursery and 5-year-old children in Scotland. Tooth wear scores for each age group were compared to available national data for the same age groups.

Results

Two hundred and nine subjects were examined: 45.9% were female, 54.1% male. Twenty-one (10%) had a recognised syndrome and were reported separately, leaving 188 subjects in the main study. Cleft Palate (CP) was the most commonly

occurring cleft in both syndromic and non-syndromic groups, followed in decreasing numbers by UCLP, BCLP, UCL and BCL.

The only age group of cleft children with a significantly higher level of caries than the general population was the 4.5 – 6.0 year-olds. Only 37.2% of the cleft subjects in this age group were caries free compared to the national figure of 57.7%. The mean dmft for the cleft group was 3.24 compared to 1.86 nationally. The other age groups had similar dmft and percentages of subjects caries free compared to the national data. The differences did not reach significance.

All the age groups in this study exhibited more tooth wear than their non-cleft peers. When looking specifically at wear extending into dentine, both the 1.5 – 2.49 and 3.5 – 4.49 age groups had significantly more wear when compared to the national data ($p \leq 0.05$).

Conclusions

By the age of 4.5 years, cleft children in the West of Scotland have significantly more caries than their non-cleft peers.

Children with a cleft aged 1.5 – 2.49 and 3.5 – 4.49 exhibit significantly more tooth wear into dentine on the upper primary incisors than their non-cleft peers.

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Author's Declaration

All data presented in this thesis is the original work of the author.

Karen Britton

Chapter 1 - Introduction

Orofacial clefts are the most commonly occurring craniofacial defect with an average prevalence of 1.2/1000 live births (Mossey & Little, 2002). Children born with an orofacial cleft have a high burden of care throughout their childhood. Most can expect at least one general anaesthetic for surgery, speech therapy, dental and orthodontic treatment and possible psychological problems associated with the appearance of their cleft.

Papers investigating the prevalence of caries in cleft children and adults have variously reported a higher prevalence of caries in cleft children, a lower prevalence or have concluded that there is no evidence to suggest a difference.

Two papers, Wong & King, 1998 and Hasslöf & Twetman, 2007, conducted systematic reviews of the literature. Hasslöf & Twetman, 2007 found potential problems with controls, caries diagnosis methods and a lack of data on “social class indicators”. Wong & King posed the question “How much do we really know about the oral health of children with CLP?” The authors reported on dental caries, oral hygiene and gingival disease. They concluded that there are many methodological difficulties in carrying out research in this field. These included small sample sizes, varying methods of presenting caries results, a lack of suitable control groups and a wide age range. They recommended that caries data should be presented by region and that this “would give a comprehensive picture of caries experience that is suitable for future comparisons”.

In this study I will present “a comprehensive picture” of caries and tooth wear experience in cleft children aged between 6 months and 6 years in the West of Scotland. I will present caries data in small age groups as mean dmft and dmfs, the percentage of subjects who are caries free, and caries by region (both anterior and posterior). It is hoped that this data will improve the evidence base in the West of Scotland and provide a database for comparison in any future studies.

Chapter 2 - Literature Review

2.1 What is a Cleft?

“Cleft” means split or separation. Clefts occur when there is failure of the embryonic prominences to fuse. This begins at around the sixth week of pregnancy when the maxillary prominence and the median nasal prominence fuse to form the upper lip and the premaxilla (Gorlin & Baylis, 2009; Sperber & Sperber, 2009).The secondary palate begins to form at the end of the sixth week. The maxillary prominences grow laterally to form the palatine shelves. These grow across and elevate displacing the tongue downwards, eventually meeting in the midline and fusing from front to back by week eleven (Gorlin & Baylis, 2009; Sperber & Sperber, 2009).

Clefts can be divided into those affecting the lip, the primary palate (premaxilla) and the secondary palate (hard and soft palate). Cleft lip and palate can occur separately or together and may be unilateral or bilateral.

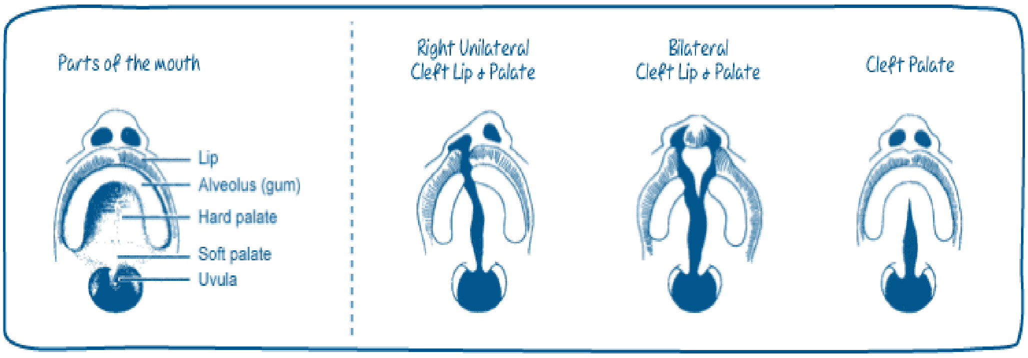

Figure 2-1 - Diagrammatic representation of various cleft types (www.clapa.com).

Cleft lip may be complete or incomplete i.e. extend into the nose or not. Cleft palate can also be subdivided according to whether the cleft is complete or “submucous” where there the muscle has not joined properly, but where the overlying mucosa is intact (Gorlin & Baylis, 2009; Mooney, 2009).

Figure 2-2 - Photographs of various cleft types (www.CLAPA.com).
Top left; Bilateral cleft lip and palate. Top right; Unilateral cleft lip and palate.
Bottom left; Unilateral cleft lip. Bottom right; Unilateral cleft lip and palate.

Clefts may occur as part of the phenotype of a syndrome (syndromic cleft) or may not be associated with a syndrome (non-syndromic cleft).

2.2 Epidemiology

There is great variation in the occurrence of cleft lip and palate both geographically and between different populations (Mossey & Little 2002). Clefts are particularly common in the Native American, Japanese and Chinese populations. In Europe clefts occur in around 1 in 1000 white births and Scotland has one of the highest cleft rates in Europe (www.eurocat.ulster.ac.uk/pubdata).

The prevalence of clefts was difficult to quantify accurately before the introduction of standardised national and international databases (Bixler, 1981; Sayeta *et al.*, 1989; Bellis & Wohlgemuth, 1999; Gregg *et al.*, 2008). Womersley and Stone (1987) reported a prevalence of facial clefts of 1.56 per 1000 total births in the West of Scotland between 1974 and 1985. A second West of Scotland study reported a total birth prevalence of 1.53 per 1000 in the period 1980 -1985 (Fitzpatrick *et al.*, 1994). Both studies reported a higher proportion of cleft palate cases compared to cleft lip with or without cleft palate.

Since 2000, Cleft Services in Scotland (CLEFTSiS) have gathered data for every child born in Scotland with a cleft. In 2008 ninety seven children were born with a cleft, of which 57 were referred for care to the Glasgow multi-disciplinary team (CLEFTSiS Annual Report, 2008), and in 2009 the respective numbers were 105 and 61 (CLEFTSiS Annual Report, 2009).

2.3 Aetiology

The exact causes of a cleft are still unknown. Some appear to be random, however there are many factors postulated to be associated with their development. These may be divided into genetic causes, including those associated with a syndrome, and environmental factors including drugs, smoking and alcohol consumption. In addition it seems likely that there are also relevant gene-environmental interactions involved in the aetiology of facial clefting (Shaw *et al.*, 1996; Romitti *et al.*, 1999; Shaw *et al.*, 1999; Hartsfield *et al.*, 2001).

2.3.1 Genetic Causes

Most cases of orofacial cleft are non-syndromic. However there are around 300 syndromes known to be associated with oral clefts, the most commonly occurring are: Van der Woude syndrome; Velocardiofacial syndrome; Trisomy 13 (Patau syndrome); Ectrodactyly-ectodermal dysplasia (EEC) syndrome; DiGeorge syndrome and Pierre Robin sequence (Gorlin, 2001). The details of all these syndromes may also be found on the Online Mendelian Inheritance in Man database on the NCBI website (2009).

There is strong evidence of family history being a significant risk factor for non-syndromic cleft lip and or palate, however these family aggregations do not seem consistent with a Mendelian form of inheritance and there may be interactions between several genes and environmental factors which are discussed in Section 2.3.2 below.

Animal and family based studies have provided much information regarding the inheritable nature of clefts. Several gene loci have been identified through animal and family based studies: MSX1, TGFA, TGFB3, IRF6, RARA, MTHRF, GABRB3, PAX9, PVRL1 and TBX2 (Murray, 1995; Wyszynski *et al.*, 1996; Murray, 2002; Marazita & Mooney, 2004; Krapels *et al.*, 2006; Weinberg *et al.*, 2006; Viera, 2008; Jugessur *et al.*, 2009). In their 2006 paper, Xu *et al.*, showed that in animal models ablation of TGFB2 results in a soft palate cleft and submucous cleft. MSX1, TGFR3 and PAX9 have been implicated in clefting with hypodontia (Slayton *et al.*, 2003), and MSX1, TGFA, PAX9 along with IRF6 are involved in addition with isolated hypodontia (Lidral & Reising, 2002; Viera *et al.*, 2004; Viera *et a*l., 2007). Figure 2-3, below, shows some of the genetic and environmental contributions to the aetiology of facial clefting.

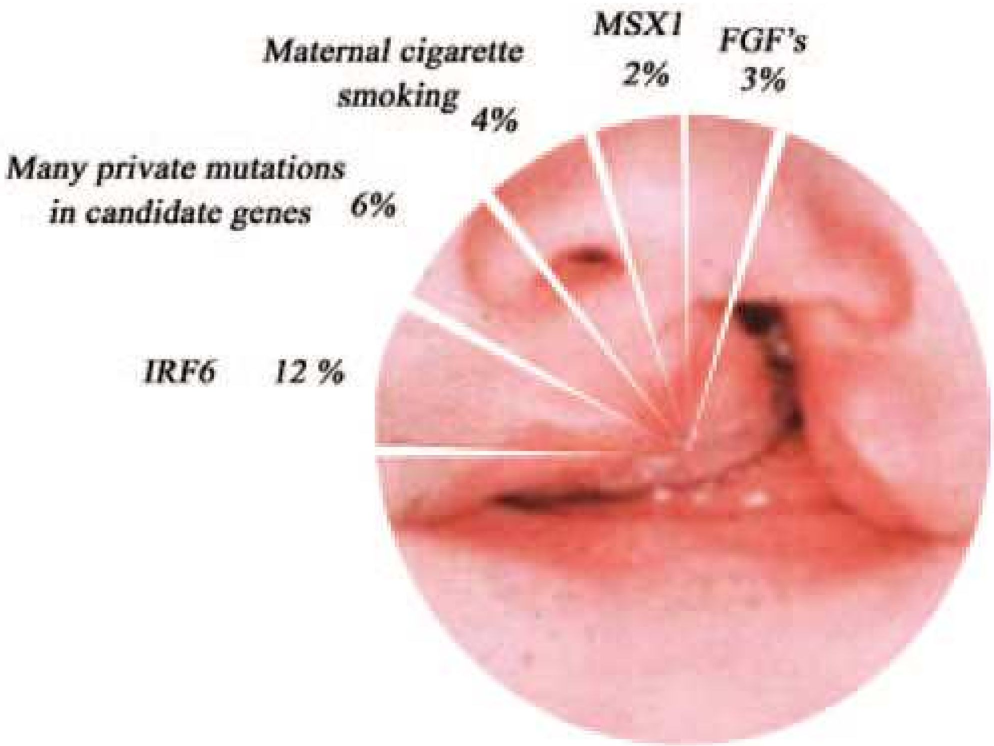

Figure 2-3 - Genetic Contributions to Cleft Lip and Palate (Viera *et al.*, 2008).

Menezes *et al.*, (2009) reported that people who are born with an orofacial cleft have a shorter lifespan and that this may be partly due to cancer. In this study the authors found that families with CLP had a higher rate of cancer, especially colon cancer, and that the gene AXIN2, which is associated with CLP can, when mutated, “increase the susceptibility to colon cancer”.

2.3.2 Environmental Factors

2.3.2.1 Deprivation

Two Scottish studies have looked at the link between socioeconomic status and the presence of a cleft.

Womersley and Stone (1987) collected data from the Glasgow Register of Congenital Malformations between 1974 and 1985. They subdivided the Greater Glasgow area into eight separate categories dependant on postcode and deprivation and looked at the prevalence of facial clefts within each category. Results showed that the highest numbers of clefts occurred in the two categories with the highest deprivation ranking i.e. “areas with local authority housing with young families, high unemployment and a preponderance of unskilled workers”. The greatest variance was for cleft palate alone and less variation was found for CL+/-P.

Clark *et al.*, in 2003 published a study investigating the association between socioeconomic status and orofacial clefts (OFC) in Scotland over the 10 year period 1989-1998. They found a “strong positive relationship whereby the prevalence of OFC at birth increased with increasing deprivation”. However unlike Womersley and Stone this was not statistically significant for CP but was for CL+/-P.

Both studies felt that the results reflected the greater likelihood of additional risk factors such as cigarette smoking during pregnancy in areas of higher socioeconomic deprivation.

Durning *et al.*, in 2007, published a paper investigating the relationship between orofacial clefts and material deprivation in Wales. The authors used patient data from a National Orofacial Cleft register and recorded all births with a cleft between 1982 and 2003. For all 831 babies born with a cleft within this timeframe, the cleft type and side were recorded and the Townsend score of material deprivation (Townsend, 1988) was calculated from the full postcode. The results showed a “statistically significant risk of 1.55 (CI 1.18 – 2.04) for orofacial clefts between the most and least deprived”. They also showed a significant rise in the rate of clefts per 100,000 live births with increasing deprivation for both CL/P and CP alone.

2.3.2.2 Folic acid

Insufficient intake of folic acid is widely understood to be associated with neural tube defects such as spina bifida (MRC Vitamin Study Research Group, 1991). The World Health Organisation (WHO) has recommended that women should take a daily dietary supplement of 400μg folic acid in the periconceptual period to reduce this risk (http://www.who.int/nutrition/publications/micronutrients/weekly_iron_folicacid.pdf).

The case for a causal relationship between folic acid deficiency and clefting of the lip and or palate is based on the fact that “neural crest cell proliferation and migration into the facial processes is folic acid dependent” (Loffredo *et al.*, 2001). Shaw *et al.*, in 1995 found that women who used multivitamins containing folic acid periconceptually “had a 25-50% reduction in risk for offspring with orofacial clefts compared to women who did not use such vitamins”. However, there are potential confounding factors such as the above mentioned socioeconomic status, smoking and alcohol intake and medications, making the study of a potential causal link with folic acid difficult. Several studies which have taken confounding factors into consideration have still found that vitamin supplementation has a protective effect for both CL+/-P and CP (Tolarová & Harris, 1995; Itikala *et al.*, 2001; Loffredo *et al.*, 2001; Bailey & Berry, 2005; Badovinac *et al.*, 2007) with a reduction in clefting of between 30% (Loffredo *et al.*, 2001) and 50% (Van Rooij *et al.*, 2004). However in their 2007 study in Norway, Wilcox *et al.*, found that an intake of 400μg or more of folic acid “provided no protection against CP alone” although it did reduce the risk of CL+/-P.

A population-based case-control study by Mossey *et al.*, (2007) looked at the association between pregnancy planning and orofacial clefts in 191 children born with non-syndromic orofacial cleft in the UK, including subjects from Greater Glasgow. The authors controlled for age, and socioeconomic status. Results showed “no association between CL+/-P or CP and total folate intake”.

However, planned pregnancies had a lower risk of orofacial clefts, and an unplanned pregnancy where the mother smoked in the first trimester nearly trebled the risk of an orofacial cleft when compared to a planned pregnancy where the mother did not smoke.

2.3.2.3 Alcohol

The risks of women drinking alcohol, a known human teratogen, during pregnancy has been acknowledged for some time (Martinez-Frias *et al.*, 2004). One of the most severe outcomes of heavy maternal drinking during pregnancy is Foetal Alcohol Syndrome, a combination of “specific craniofacial malformations, pre and post-natal growth retardation, and central nervous system disorders” (DeRoo *et al.,* 2008). However studies investigating the relationship between more moderate alcohol consumption during pregnancy and the risk of orofacial clefts have produced mixed results (Wyszynski & Beaty, 1996). Two studies found an increasing risk with increasing levels of alcohol consumed (Munger *et al.*, 1996; Romitti *et al.*, 1999). Werler *et al.*, (1991), Shaw & Lammer (1999) and DeRoo *et al.*, (2008) all reported greater risks of a cleft with more than 5 drinks per drinking occasion and DeRoo further commented that “maternal binge drinking may increase the risk of infant clefts”. However Lorente *et al.*, (2000) found no increasing risk according to dose for any cleft type, and Romitti *et al.*, in their 2007 study found no statistically significant risk associated with alcohol and any cleft phenotype, but did find that the odds ratio varied with the type of alcohol consumed, especially with CP (spirits>wine>beer) and that the risks further increased if there was no folic acid intake.

2.3.2.4 Smoking

Maternal smoking during pregnancy is associated with several pregnancy complications including pre-term delivery and low birth weight. It has also been shown to be associated with increased clefting compared to the offspring of mothers who did not smoke (Ericson *et al.*, 1979; Khoury *et al.*, 1987; Shaw *et al.*, 1996; Källén, 1997; Maestri *et al.*, 1997; Wyszynski & Tianxia, 2002).

When controlling for confounding factors Chung *et al.*, (2000) and Lorente *et al.*, (2000) found the odds ratio was 1.34 and 1.79 respectively for CL+/-CP, and

Källén (1997) found the odds ratio for maternal smoking and CL+/-CP was 1.16 (CI 1.02-1.32) and for isolated CP was 1.29 (CI 1.08-1.54). Lorente *et al.*, used data from a European multi-centre case-control study which included Glasgow. Wyszynski *et al*'s meta-analysis of 11 studies published from 1966 – 1996 gave an overall odds ratio of 1.29 (CI 1.18-1.42) for CL+/_P and 1.32 (CI 1.10-1.62) for CP (Wyszynski *et al.*, 1997). This was also found in a UK based study by Little *et al.*, (2004) which included data from Glasgow, who reported odds ratios of 1.9 (CI 1.3-3.1) for CL+/_P and 2.3 (CI 1.3-4.1) for CP only.

There appears to be a dose response in the first trimester (Khoury *et al.*, 1987; Chung *et al.*, 2000; Lorente *et al.*, 2000; Little *et al.*, 2004) whereby increasing numbers of cigarettes smoked resulted in an increasing risk of clefting in the foetus.

It is still unclear if exposure to maternal smoking acts in isolation to increase the risk of clefting but seems more likely that smoking interacts with various factors including genes which produce enzymes responsible for detoxifying cigarette smoke. Zieger *et al.*, (2005) found a gene-environment interaction between the infant genotype Taq1 site in TGFA and maternal smoking. Maestri *et al.*, (1997) reported interaction between maternal smoking and the transmission of markers near TGFA and TGFB3 as did Shaw *et al.*, (1996). However Beaty *et al.*, (1997) found no link between maternal smoking and TGFA genotype. Ramirez *et al.*, (2007) and Hartsfield *et al.*, (2001) reported no link between detoxification enzymes and an increased risk of clefting.

2.3.2.5 Drugs

2.3.2.5.1 Anti-epileptic medication

Special concerns have been voiced for women of child-bearing years with regard to contraception, pregnancy and teratogenicity that should be considered during counselling and selection of appropriate treatment. (Tatum *et al.*, 2004). Maternal use of anti-epileptic drugs (AED) results in an increased risk of major foetal malformations and minor foetal anomalies (Matalon *et al.*, 2002; Tatum *et al.*, 2004). Canger *et al.*, (1999) found an overall rate of malformations of 9.7%

compared to the Eurocat Study rate of 2.3% in the general population (Donati *et al.*, 1984 cited in Canger *et al.*, 1999).

Varying degrees of increased risk of facial clefts have been reported in children of AED treated mothers. Abrishamchian *et al.*, in 1994 conducted a population based case-control study and found that maternal epilepsy was associated with an increased risk of non syndromic CLP of 3.78 (CI 1.65-7.88) and for CP of 1.75 (CI 0.20-6.99). Hernandez-Diaz *et al.*, (2000) reported a relative risk of oral clefts of 2.5 (CI 1.5-4.2) after maternal exposure to anti-epileptic drugs. Friis, (1989) reported that the rate of facial clefts increased by a factor of 4.7.

Most studies agree that the greatest risk is in the use of polypharmacy i.e. the use of more than one drug to control maternal epilepsy (Abrishamchian *et al.*, 1994; Canger *et al.*, 1999; Tatum *et al.*, 2004). The ideal would be monotherapy and that “this is an important treatment goal” (Morrell, 1998).

2.3.2.5.2 Corticosteroids

Several studies have shown an association between maternal use of corticosteroids during the first trimester of pregnancy and the occurrence of an orofacial cleft in the infant (Rodriguez-Pinilla & Martinez-Frias, 1998; Carmichael & Shaw, 1999; Park-Wylie *et al.*, 2000; Edwards *et al.*, 2003; Källén, 2003; Carmichael *et al.*, 2007).The relative risks found varied widely between the studies and this may be due to the differences in study design and numbers.

Carmichael and Shaw’s 1999 population- based case control study included 662 cases of orofacial clefts. Corticosteroid use was shown to be associated with an increased risk of CL+/_P of 4.3 (CI 1.1-17.2) and CP alone of 5.3 (CI 1.1-26.5). Rodríguez-Pinilla & Martínez-Frías reported an increased risk of CL+/_P of 6.55 (CI 1.44-29.76) in their case control study of 1184 infants with non syndromic oral clefts.

Park-Wylie *et al.*, (2000) found an increased risk of oral cleft of 3.35 (CI 1.97-5.69) in 184 women exposed to prednisone in pregnancy.

Källén, in a 2003 study, investigated 1142 infants with orofacial clefts and the association between various drugs including anti-epileptics and glucocorticoids. There were only 32 incidences of orofacial cleft associated with exposure to glucocorticoids with a risk ratio of 1.44 (CI 0.98-2.03).

Carmichael *et al.*, (2007) interviewed the mothers of 1141 children with CLP and 628 children with CP and over 4000 controls. All cleft study subjects were part of the National Birth Defects study, a multi-state case-control study of over thirty birth defects in the USA. They reported a “moderately increased risk” of cleft associated with maternal use of corticosteroids during early pregnancy with odds ratios of 1.7 (CI 1.1 – 2.6) for children with CLP and 0.5 (CI 0.2 – 1.3) for CP alone.

2.3.2.6 Benzodiazepines

A meta-analysis of cohort and case-control studies in 1998 by Dolovich *et al.*, showed no association between exposure to benzodiazepines and oral cleft in the cohort studies, however the case-control studies did show an increased risk of oral cleft (1.79, CI 1.13 - 2.82).

2.3.2.7 Retinoids

Retinoic acid is an oxidised form of Vitamin A and is important in early embryogenesis. Synthetic retinoids are commonly used to treat severe cystic acne and keratinisation disorders e.g. severe psoriasis. The teratogenicity of retinoic acid has been shown in animal models and it is thought to interfere with the migration of neural crest cells and can result in malformations including micrognathia and cleft palate if taken during pregnancy (Lammer *et al.*, 1985; Chan *et al.*, 1996).

2.3.2.8 Folate antagonists

As mentioned already folic acid protects against neural tube defects and orofacial clefting. Folate antagonists can be dihydrofolate reductase inhibitors e.g. trimethoprim and methotrexate, or may affect other enzymes involved in the metabolism of folic acid or affect its absorption or hasten its degradation e.g. anti-epileptic drugs like carbamazepine, phenytoin and phenobarbital. Hernandez-Diaz *et al.*, (2000) found the relative risk of oral clefts in infants whose mothers took folic acid antagonists was 2.6 (CI 1.1-6.1).

2.4 Dental Caries

Caries is the breakdown of the hard tooth surface commonly known as tooth decay. The acidogenic theory of caries development is widely accepted (Tucker, 1932). This states that for caries to develop there must first be microorganisms, a suitable tooth surface on which they can metabolise carbohydrate from the diet, producing acid over time. This acid lowers the pH to a level below which the enamel will lose some of its mineral content. If this is allowed to continue unabated, the end result is breakdown of the tooth surface to form a cavity. The curve of the pH of plaque against time, the “Stephan Curve” (Figure 2-4) shows that this demineralisation can be halted, and indeed reversed, to allow the tooth enamel to remineralise if the pH of the plaque is allowed to return to 5.5 or above (ten Cate, 2009).

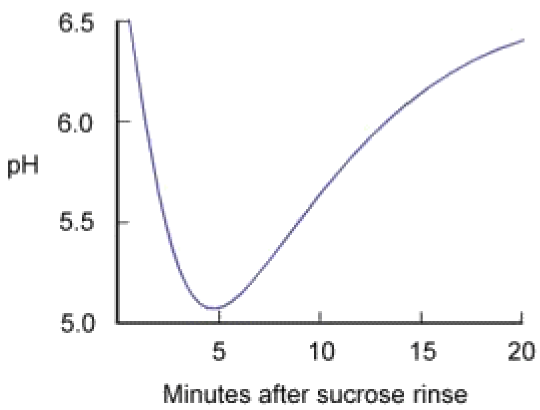

Figure 2-4 - The Stephan Curve.

Since caries is a multifactorial disease (Figure 2-5), it is therefore possible to prevent it by removing one or more of these factors. Tooth brushing will mechanically remove plaque and its component microorganisms, diet could be altered to minimise the number and frequency of acid attacks and fluoride toothpaste helps to remineralise the tooth surface (Marinho *et al.*, 2009).

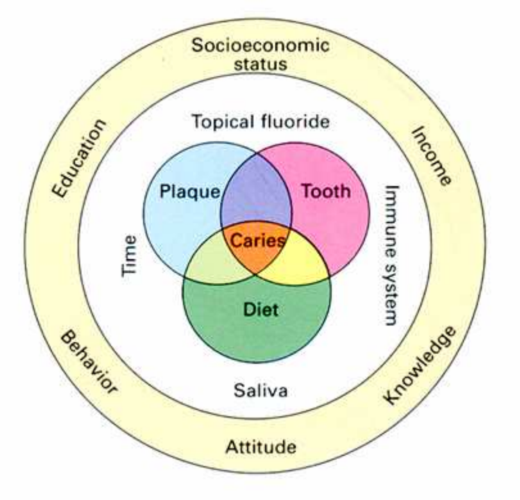

Figure 2-5 – Modified Keyes Diagram of factors determining caries development.

2.4.1 Caries Progression

Children who develop caries at an early age have an increased risk of developing further caries in the primary teeth (Johnsen, 1986; Greenwell *et al.*, 1990) and are likely to develop caries in the permanent dentition (Greenwell, 1990; Raadal & Espelid, 1992). The effect of caries in the primary dentition is cumulative until the age of 7 years, when it starts to decline due to exfoliation (Evans & Lo, 1992). The greatest risk factor for the development of further caries is whether there is early childhood caries (ECC or baby bottle caries) (Greenwell *et al.*, 1990; O'Sullivan & Tinanoff, 1993; Al-Shalan *et al.*, 1997).

In 1991 Grindefjord *et al.*, conducted a study investigating the prevalence of mutans streptococci in 1103 one-year-old children in Stockholm, Sweden. In a subsequent paper (Grindefjord *et al.*, 1995a) the authors investigated caries prevalence in relation to social and immigrant background, occurrence of mutans streptococci, dietary habits, oral hygiene and fluoride exposure. Seven hundred and eighty six of the original subjects were re-examined at 3 and 4 years of age. They reported that at the age of one year the highest risk factors for developing further caries by the time the child reached the age of 3.5 years were the:

- child's immigrant status
- mother's level of education

- presence of Strep. mutans
- amount of confectionary consumed.

If all the above risk factors were present, it resulted in an 87% probability that more caries would develop by 3.5 years.

In a further study, also published in 1995, (Grindefjord *et al.*,1995b), the authors reported on the proportion of the original study subjects who developed caries between the ages of 2.5 and 3.5 years. At baseline 11.3% of subjects had caries. Of these 92% developed new lesions within the year (of those 50% developed more than six new lesions). Whilst of those who were caries free at baseline, only 29% developed caries within the year.

Demers *et al.*, (1990) reviewed the predictors used to identify children at high risk of developing caries. They also concluded that a combination of factors provided a more effective prediction when used in screening. The best combination was of past caries experience together with the presence of the Streptococcus mutans bacteria and the socioeconomic status of the family.

Al-Shalan *et al.*, in their 1997 paper concluded that the presence of ECC at the first dental visit is a risk factor for future caries and that recalls and sealants are protective factors. This early identification of at risk children has been advocated for many years (Winter, 1988).

Gibson and Williams (1999) undertook further analysis of data from the National Diet and Nutrition Survey and investigated the “relative significance of dietary sugars, toothbrushing frequency and social class as predictors of caries experience” in children aged 1.5 to 4.5 years. They reported that “the strength of the association between social class and caries experience was twice that between toothbrushing and caries, and nearly three times that between sugar confectionary and caries”.

Harris *et al.*, in 2004, conducted a review of the literature on risk factors for dental caries and concluded that “a wide range of risk factors have been found to be significantly related to childhood caries" and that there was a lack of high quality longitudinal studies.

The above-mentioned clinical evidence is the reason why all children with a cleft in the West of Scotland are examined in detail so that children at an increased risk of developing caries can be offered effective preventive advice and treatment. It is also the reasoning behind dividing the subjects of this study into age groups to see when the caries starts, thus allowing more targeted prevention in the future to the younger age groups to reduce the burden of caries.

2.4.2 The Effect of Caries on Growth and Quality of Life

The proportion of children with sepsis increases markedly with caries experience (Pine *et al.*, 2006) and the resulting pain can be severe and impact negatively on daily life. Several studies have investigated the impact of caries on quality of life and the child's body weight and growth and have shown that children with early childhood caries weighed significantly less than their peers without caries. (Acs *et al.*, 1992; Ayhan *et al.*,1996; Sheiham, 2006; Cameron *et al.*, 2006). This latter cross-sectional study is of 165 children attending the Glasgow Dental Hospital and School for extractions under day stay general anaesthetic. The mean age was 5.7 years. Children with the worst decay (dmft 8-20) showed significantly lower body mass index.

Acs *et al.*, in 1992, conducted a retrospective review of records of children attending for sedation or general anaesthetic to manage early childhood caries (ECC) between 1987 and 1991. One hundred and fifteen children with matched controls were included. Their mean age was 3.2 years. The results showed that only 1.7% of the control subjects weighed less than 80% of their ideal weight for age, compared with 8.7% of the study subjects. Of those classified as having early childhood caries, 13.7% weighed less than 80% of their ideal. This was similar to the results of Ayhan *et al.*, in 1996 who found that of the children in their study with "rampant caries" 7.1% weighed less than 80% of their ideal weight compared to just 0.7 % of the matched control children. The fact that these children weighed less than 80% of their ideal weight means that they satisfy one of the diagnostic criteria for “failure to thrive” (Elice & Fields, 1990; Acs *et al.*, 1992).

Dental caries has been found to have a significant effect on a child’s general well being and quality of life, for example it can result in pain, disturbed eating and sleeping patterns and in missing school (Reisine, 1998; Shepherd *et al.*, 1999). However following complete dental rehabilitation of early childhood caries under sedation or general anaesthetic, there is a significant reduction in pain, better eating and sleeping patterns (Low *et al.*, 1999; Acs *et al.*, 2001; Filstrup *et al.*, 2003; Anderson *et al.*, 2004) and a “catch up” increase in growth velocity (Acs *et al.*, 1999). Acs *et al.*, in their 1999 paper found that a year and a half after complete dental rehabilitation there was no statistically significant difference between the weights of the study and control group children. Thomas & Primosch, (2002) could not confirm this “catch up” growth but did confirm the improvements in quality of life factors.

The above studies reinforce the already strongly held belief regarding the importance of early identification of high-risk children allowing the implementation of preventive programmes with advice on diet, oral hygiene and the use of sugar-free medication.

2.4.3 Caries and Deprivation

Numerous studies and reports have demonstrated a link between socioeconomic deprivation and health outcomes, including dental caries (James *et al.*, 1997; Pitts, 1998; Watt & Sheiham, 1999; Locker, 2000; Tickle *et al.*, 2000a; Tickle *et al.*, 2000b; Pine *et al.*, 2004; Downer *et al.*, 2005). Although the prevalence of caries in the UK has markedly reduced over the past two decades, children from areas of high socioeconomic deprivation continue to experience more caries than their peers from more affluent areas (Sweeney *et al.*, 1999; NDIP, 2008). 5-year-olds from more deprived areas are three times more likely to suffer from decayed and missing teeth than their more affluent peers (NDIP, 2003).

Sweeney *et al.*, (1999) in their Scottish study, reported a “striking association between increasing deprivation and increasing caries experience” and also that “children from the most deprived areas had significantly more untreated decay and missing teeth”. The authors reported a mean dmft for children in Depcat 1 (least deprived) areas of 1.48, which increased to 4.87 for those in Depcat 7 (most deprived) areas. Of the Depcat 1 children, 62.4% were caries free, compared to only 19.8% of the Depcat 7 children, and the Care Index (ft/dmft x 100) was 10.8% for Depcat 1 and 2.9% in Depcat 7.

The skewed nature of caries distribution in 5-year-olds can be found in the 2008 National Dental Inspection Programme (NDIP) of Scotland report which revealed that 11% of the population had 50% of teeth with established decay and that “all of the teeth with observed severe decay into the pulp were seen in just 3% of the children inspected”.

One of the stated goals for the WHO Global Goals for Oral Health (2020) is to “Reduce disparities in oral health between different socioeconomic groups within a country and inequalities in oral health across countries”.

2.4.4 Incidence / prevalence of caries in 0.5 – 6.5 year olds in Scotland and UK

Dental and oral health was adopted as one of the key priorities in "Health Education in Scotland - a National Policy Statement". Two dental targets were set for the year 2000: that 60% of 5-year-old school entrants should have no cavities, fillings, or extractions and that less than 10% of 45 to 54-year-olds should be without their own teeth. Following this the Scottish Forum for Public Health Medicine published a Scottish Needs Assessment Programme report in 1994 (SNAP, 1994; SNAP, 1998) into dental caries in children in response to the continuing high caries rate in Scotland and reported that there was “much to be done if the existing national target for 5-year-olds is to be met”.

Unfortunately, despite the improvements in general health over the past few decades and the trend towards a decrease in the amount of dental decay, many children in Scotland still have a significant amount of caries. At the age of five, upon entering Primary School, over 40% of Scottish children still have some dental decay (NDIP, 2008). Guidance for dental professionals regarding the prevention and treatment of dental caries in children has provided a framework within which to try to reduce the burden of caries and many policy documents have been produced (*Shaw*, 1997; Fayle *et* al., 2001; Rayner *et al., 2003;* EAPD, 2008). The Scottish Intercollegiate Guidelines Network (SIGN) published its SIGN 83 guidelines in 2005 and a recent draft document has been released for consultation in April 2009 by the Scottish Dental Clinical Effectiveness Programme to update the preventive and treatment advice available.

Epidemiological studies, which provide national oral health data, use standardised examination techniques and diagnostic criteria to allow direct comparisons between different studies. Epidemiological recording of caries occurs at the D_1 level, i.e. well established caries extending into dentine (SNAP update, 1998; NDIP 2008). Many clinicians would intervene and treat caries at a much earlier stage than this, at both D_1 level (very early decalcification of the enamel) and D_2 level (caries resulting in a breach of the enamel surface). It must therefore be remembered that a child recorded as having "no obvious caries" in an epidemiological study might in fact have caries requiring intervention in a clinical setting. Figure 2-6, below, shows the various stages of caries as discussed here.

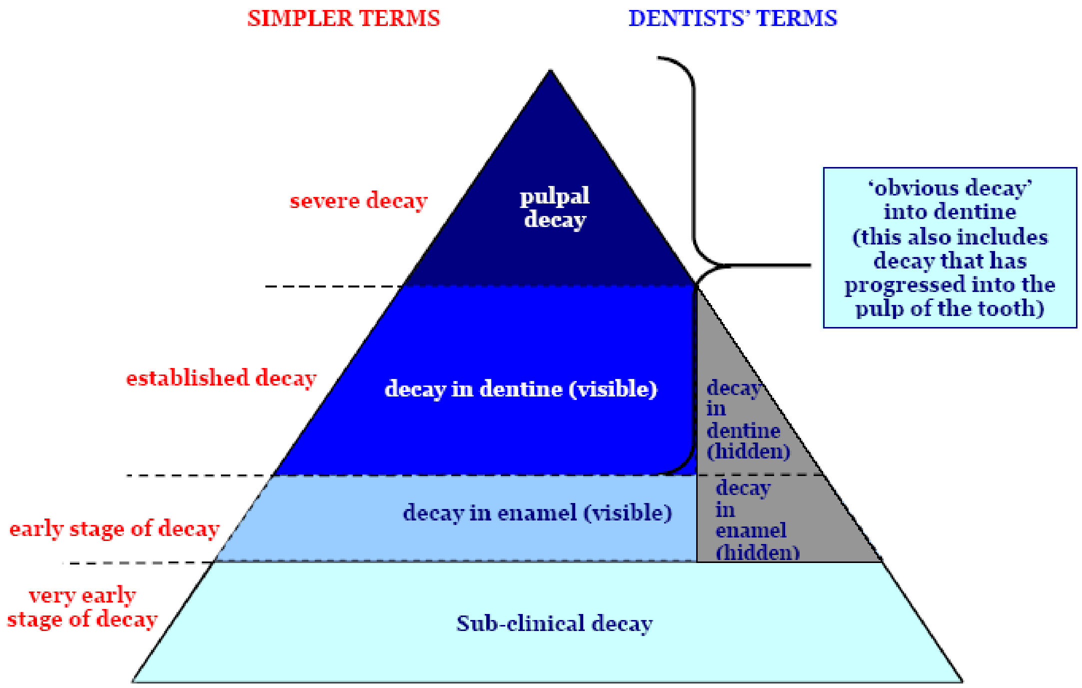

Figure 2-6 - Pyramid Diagram of Caries (NDIP, 2008).

The 1995 report of the National Diet and Nutrition Survey (Hinds & Gregory, 1995) examined 1658 children aged 1.5 – 4.5 years and included information on diet, caries, erosion and trauma. Four percent of 1.5 – 2.5 year olds had some caries with a mean dmft of 0.1, and this increased to 14% of 2.5 – 3.5 year olds with mean dmft of 0.5 and 30% of 3.5 – 4.5 year old children with a mean dmft of 1.3.

The report of the oral health survey of the National Diet and Nutrition Survey of 1997 (Walker, 2000) gives national UK data for 2672 young people aged 4 to 18 years. The mean incidence of caries in the 4 to 6-year-old age group was 37% and was highest in Scotland at 50% and lowest in London and the South East at 29%.

The most recent 2005/2006 British Association for the Study of Community Dentistry (BASCD) survey report of the dental health of 5-year-old children (Pitts *et al.*, 2007) examined 239,389 children across England, Wales, Scotland and the Isle of Man. Overall, 39.4% of children had caries at the D_3 level. The mean dmft was 1.57 but the mean dmft for children with dmft>0 was 3.99. The Care Index was 11%.

Dental disease in the West of Scotland is significantly higher than elsewhere in Scotland with only 35% of 5 year-old children in Greater Glasgow and Lanarkshire in 1999/2000 free from caries (SHBDEP, 1999/2000) and this rising to 41% for Greater Glasgow by 2004 (NDIP, 2004) compared to the national Scottish figure for 2004 of 50.7% caries free. However, possibly due to new national and local prevention programmes e.g. Childsmile (www.child-smile.org 2009) which includes nursery tooth brushing and twice yearly fluoride varnish applications, this has fallen significantly recently and the latest NDIP, 2008 figures show that in Greater Glasgow and Clyde 54.8% of 5-year-old children are caries free with the national average being 57.7% caries free at 5 years of age.

The majority of the dental disease is present in children from more deprived areas where 5-year-olds are three times more likely to suffer from decayed and missing teeth when compared to their peers from more affluent areas (NDIP, 2003). The most recent NDIP results (NDIP, 2008) show that this discrepancy continues with 73% of children from SIMD (Scottish Index of Multiple Deprivation) category 1 areas (least deprived) caries free, and only 42% of children in SIMD category 5 (most deprived). Further evidence of the skewed nature of caries distribution can be found in the NDIP figures showing that 42% of population had 100% of teeth with established decay, 11% of population had 50% of teeth with established decay, 4% of population had 25% of teeth with established decay (Taylor, 2004; NDIP, 2006).

The mean number of obviously decayed, missing and filled teeth for Scotland is currently 1.86 and for Greater Glasgow and Clyde is 2.07 (NDIP, 2008).

Until this year, apart from some local studies, there has been a paucity of data regarding caries nationally in the pre-school child in Scotland, however NDIP has recently introduced screening for nursery children.

In 1977, Sutcliffe investigated the interaction between oral hygiene, urban deprivation and caries rates of 3 and 4-year-old children in Edinburgh. The caries was recorded at the D_2 level using sharp probes. Of 1453 children, he found that 53% of the 3-year-olds and 37% of the 4-year-olds were caries free. The mean dmft was 1.9 in the 3-year-olds and 3.0 in the 4-year-olds. The study also revealed that one of the reasons why the children in areas of highest deprivation had more caries was poor oral hygiene. There was also a low Care Index (ft/dmft x 100) of 23 i.e. 23% of carious teeth had been restored.

Using the same criteria for deprivation and caries diagnosis as Sutcliffe, but using a blunt probe for examinations, Blinkhorn (1982) investigated the dietary habits of 203 nursery children in Edinburgh and recorded their dmft. Here the mean dmft in deprived areas was 2.10 for 3-year-olds and 4.47 for 4-year-olds. In non-deprived areas, the respective mean dmft values were 0.98 and 0.79. These higher caries rates in deprived areas were accounted for by the mothers being more likely to “give sweets after nursery, use sweets as a comforter, spend more money on sweets, and allow continued consumption of sweets throughout the day”.

Sweeney and Gelbier in their study published in 1999 investigated “the dental health status of pre-school children in a deprived urban community of Greater Glasgow” The results were to be used as a baseline prior to instigating a multiagency dental prevention programme in the area. Two hundred and forty eight children attending three nurseries were examined using the standard criteria of BASCD/SHBDEP (Pitts *et al.*, 1997). Thirty six percent of children aged three were caries free and their mean dmft was 3.14. For the children aged 4.5 – 5.0 years the respective figures were 14% and 6.14. It also reported that children from the most deprived postcode sector had a significantly higher dmft and more lesions that were unrestorable. The figures were also presented for different age groups. These are detailed in the following table (Table 2-1).

Age Group in Years	Dmft	% caries free
< 3.0	0.33	88.9
3.0 – 3.49	3.14	35.7
3.5 – 3.99	4.40	24.5
4.0 – 4.49	5.64	16.7
4.5 – 4.99	6.14	13.9
5.0 – 5.49	6.37	14.8

Table 2-1 - Sweeney & Gelbier, 1999.

A more recent study in Lanarkshire (Taylor, 2004) investigated caries in children of 1.5 – 4.5 years of age. This study found that at age 1.5 – 2.5 years 97% of children were caries free. At 2.5 – 3.5, this had dropped to 82%, and by 3.5 – 4.5 years of age had further reduced to only 50% of children being caries free. Children were more likely to have caries if they: had not had their teeth brushed before the age of one; brushed their own teeth; had not visited a dentist; used a teething aid. These figures, while still below the ideal, do demonstrate an improvement over time compared to the previous studies.

Blair *et al.*, in 2004 (Table 2-2) published their study investigating “nursery-based caries experience, before and after a community development-based oral health programme’s implementation”. This took place in an area of severe social deprivation in Glasgow and was intended to “develop and evaluate NHS-based strategies likely to improve dental health and reduce inequalities in pre-5-year-olds’ oral health”. Children were examined at baseline, two, and four years after the programme was initiated. The community-based health campaigns included nutrition advice (the promotion of breastfeeding, healthy snacks and a reduction in the consumption of caries inducing drinks), the provision of infant drinking cups, toothpaste and brushes and the introduction of toothbrushing in nurseries and playgroups.

Age Group in Years	dmft	% caries free at baseline	% caries free at 4 years	% reduction in mean dmft
3.0 – 3.99	3.14	38	51	46
4.0 – 4.99	5.9	17	40	37

Table 2-2 - Blair *et al*., 2004.

The above results, in Table 2-2, confirm that a preventive programme can significantly improve dental health in high caries risk children.

In 2009, NDIP published its report of the 2007 and 2008 survey of '3-year-olds' in NHS Greater Glasgow and Clyde, the first time that this age group of children have been included in the NDIP surveys. These were carried out by trained and calibrated examiners using the same criteria as the NDIP P1 dental inspections. The average age of the children inspected was 3.7 years (Range 2.8 – 4.5 years). 1890 children were examined in 2007 and 2643 children in 2008. Table 2-3 shows the mean dmft for 2007 and 2008. The report confirms the increasing dmft with increasing deprivation.

	2007	**2008**
Mean dmft	1.3	1.0
Mean dmft for children with dmft>0	4.1	4.0

Table 2-3 - Mean dmft for 5-year-olds in Scotland 2007and 2008.

In this study, I will present comparable data for children with a cleft in the West of Scotland in the various age groups regarding mean dmft, percentage caries free, and Care Index.

2.4.5 Incidence / prevalence of tooth wear in 0.5 – 6.5 year olds in Scotland and UK

Tooth wear has been a recognised problem for some time however this mostly involved the older population. It is now becoming apparent that it is also a problem in younger people, including children (Shaw & Sullivan, 2000; Linnet & Seow, 2001; Nunn *et al.*, 2003; O'Sullivan & Milosevic, 2008).

Tooth wear is usually multifactorial and is composed principally of three elements:

- Attrition – the result of tooth-to-tooth contact, during either normal function or parafunction e.g. grinding habits.

- Abrasion – the wear of the tooth surface by mechanical means other than tooth-to-tooth contact e.g. toothbrushing.

- Erosion – a process of wear by acid which does not come from bacterial metabolism e.g. acidic foods and drinks (extrinsic) or from reflux (intrinsic).

In most cases all three processes are likely to contribute to varying degrees. Exposure of dentine and pulpal involvement in primary teeth may occur more rapidly because of their thinner enamel.

A new Tooth Wear Index (TWI) was described by Smith and Knight in 1984. Prior to this, the existing indices required an accurate diagnosis of the aetiology of the damage and were used when only one of the conditions existed. The following table (Table 2-4) demonstrates the scoring used in the TWI.

SCORE	CHARACTERISTICS	EXTENT OF WEAR
0	Normal	None
1	Loss of enamel only	<1/3 tooth surface
2	Exposed dentine surface	1/3 – 2/3 tooth surface
3	Exposed pulp or secondary dentine	>2/3 tooth surface
4	Excluded	Excluded

Table 2-4 - Tooth Wear Index.

This index is useful for recording erosion at a population level however it is no substitute for study casts and photographs when monitoring “small changes” in erosion in individual patients (Shaw & O’Sullivan, 2000). In addition, Al-Malik *et al.*, (2001) concluded that “clinical assessment of erosion may underestimate the extent of the condition”.

There are no longitudinal studies investigating tooth wear, however, several national studies have been published which use the above mentioned diagnostic criteria and examination method, making them comparable. The only Scottish figures available were part of the national studies and are quoted below.

In two local studies, the prevalence of dental erosion increases with age and has been reported as 39% of 3-year-old children in East Cumbria (Jones & Nunn, 1995) and 47% in 5-year-old children in Ireland (Harding *et al.*, 2003).

Tooth wear was first assessed at a national level in the Children’s Dental Health in the United Kingdom survey of 1993 (O’Brien, 1994). This examined over 17,000 children aged between 5 and 15 years old, and found that 52% of 5-year-old children in the UK had palatal erosion of their primary incisors and 18% had buccal surface wear. Of the wear on the palatal surfaces, 24% involved the pulp of the tooth. Taking the results for Scotland alone, this jumped to 34% of 5-year-olds with pulpal involvement of the palatal surfaces of the primary incisor teeth.

The National Diet and Nutrition Survey of 1.5 – 4.5 year old children (Hinds & Gregory, 1995) examined the upper incisors of 1522 children for buccal erosion and 1496 for palatal erosion. 10% of children had buccal erosion and 19% had palatal erosion. The prevalence of palatal erosion into dentine was 3% for the 1.5 – 2.5 age group increasing to 6% and 13% respectively for the 2.5 – 3.5 and 3.5 – 4.5 year olds.

Of the 2672 children aged between 4 and 18 years who were examined in the National Diet and Nutrition Survey in 1997 (Walker, 2000) 65% of the 4 – 6 year olds had experienced erosion with 38% having erosion on the buccal surfaces and 58% on the palatal surfaces. Of those with palatal erosion, 19% had exposed dentine or pulpal involvement. The Scottish figures within this study were a prevalence of tooth wear of 71% in the 4 – 6 year old age group. Unfortunately, there was no detail on the Scottish figures for palatal erosion, which were quoted in the two previous studies.

The Children's Dental Health survey of 2003 (Lader *et al.*, 2003) examined 10,381 children aged 5, 8, 12 and 15 years. Of the 3265 5-year-olds examined, 20% had evidence of some erosion on the buccal surface of their upper primary incisors of which 3% extended into the dentine or pulp. The results for the palatal surfaces of these teeth were 53% of which 22% extended into dentine or pulp. The Scottish figures were not reported separately.

In this study, I will present tooth wear data for all age groups regarding the percentage with tooth wear and the percentage with wear on the palatal surfaces into dentine.

2.4.6 Caries in Cleft Children

Whereas there have been many studies investigating caries in the general child population, fewer studies have been carried out specifically for children with clefts.

Two review papers (Wong & King, 1998; Hasslof & Twetman, 2007) highlighted the problems associated with conducting caries studies in cleft children. Adequate sample and control group size and a narrow enough age range for valid statistical analysis being some of the issues raised. The recommendations are that data in genetic studies "should be presented for specific cleft types wherever possible" (Fraser, 1970) and that "syndrome and non-syndrome cases should be presented separately" (Bixler, 1981). This would give a more comprehensive picture of the caries experience for future comparison. However, neither of the review papers investigated the actual threshold for caries diagnosis used in individual studies.

2.4.6.1 Caries Diagnosis Threshold

Some studies record "sticky fissures", whilst others record at the caries into dentine level (D_3), and one paper does not include any information on the level at which caries was recorded.

Three papers, Bokhout *et al.*, 1996 and 1997 and Dahllöf *et al.*, 1989, used the caries diagnosis criteria described by Koch (Koch, 1967). Koch's paper assessed the effect of sodium fluoride in toothpaste and mouthwash on the incidence of caries in schoolchildren in Sweden in a 3-year randomised double-blind clinical trial. All children brushed their teeth just prior to examination and bitewing radiographs were available. The author recorded both obvious cavitation and a "sticky" pit or fissure, where a probe sticks in a fissure and requires a "definite pull" to remove it. This will result in a significant increase in caries reporting when compared to this study. Despite this, the higher caries prevalence found in children with a cleft in these studies, compared to the controls, is still valid.

Lin and Tsai in their 1999 paper use a modified version of the above criteria (Radike, 1972). This acknowledges that a sticky fissure alone is not an accurate indicator of caries (Slack *et al.*, 1958; Lussi, 1991; Penning *et al.*, 1992). In addition it requires the presence of an adjacent opacity, softening of the surrounding tooth surface or evidence of undermining of the enamel. The Lin and Tsai study investigated the prevalence of caries in 123 two-year-old cleft children. The subjects were divided into those who had been bottle-fed and those who had not. The dmfs score of the 48 bottle-fed children was 0.67 compared to 0.23 for the 75 non bottle-fed children.

All the other papers used the D_3 level for their caries diagnosis, whether that be the World Health Organisation (WHO) 1987 or 1997 guidance, or the UK National Survey guidance detailed by O'Brien in 1994 (Chapple & Nunn, 2001).

The World Health Organisation criteria records caries as present when a lesion has “an unmistakeable cavity, undermined enamel or a detectably softened floor or wall” i.e. at D_3 level (Paul & Brandt, 1998; Lucas *et al.*, 2000; Bian *et al.*, 2001; Ahluwalia *et al.*, 2004; Beseling & Dubois, 2004; Kirchberg *et al.*, 2004; Al-Wahani *et al.*, 2005; Stec-Slonicz *et al.*, 2007; Mutarai *et al.*, 2008).

Three papers used radiographs in addition to the clinical examination to assess the prevalence of caries (Lauterstein & Mendelsohn, 1964; Dahllöf *et al.*, 1989; Ahluwalia *et al.*, 2004). It is recognised that caries may be missed, especially caries on interproximal surfaces and early occlusal caries, if bitewing radiographs are not taken. Sköld *et al.*, (1997) reported that 1.8 more surfaces were assessed as carious when radiographs were employed. Anderson *et al.*, (2005) found that “between 30 and over 50% of these (approximal) surfaces show radiographic evidence of caries not detected from clinical examination”. Both these studies involved 5-year-old children. Thus the three previously mentioned cleft studies which utilised radiographs are likely to have a higher prevalence of caries compared to those which did not.

2.4.6.2 Prevalence of caries in cleft children

Johnsen and Dixon in 1984 (Cleveland, USA) looked at caries of the primary incisors only, using a sample of 64 children aged between 18 months and 4 years. The subjects had a variety of craniofacial anomalies including 41 with a cleft lip and or palate. Noting only frank cavities and not demineralisation, they found that 60% of those with BCLP, 20% of those with UCLP and 25% of patients with CP had carious primary incisors. However the numbers are very small and the results do not give a figure for dmfs/t or subdivide the results into smaller age groups.

Chapple and Nunn (2001) conducted a prevalence study of caries in children with clefts. The authors examined nineteen children from Newcastle, England aged 4 years, twenty-eight aged 8 years, and forty-four aged 12 years. As expected, caries prevalence increased with age, 63% of the 4-year-olds being caries free and only 34% of the 12-year-olds. The mean dmft for the 4-year-olds was 1.3 (CI 0.2-2.4) and for the 8-year-olds was 1.8 (CI 0.9-2.8). The DMFT for permanent teeth in 8-year-olds was 0.4 (CI 0.1-0.7) and in 12-year-olds 1.8 (CI 0.9-2.3).

Besseling and Dubois (2004) reported the prevalence of caries in cleft children in Southern Vietnam. This study examined 154 children with cleft lip and/or palate aged between 4 and 16 years. The results are presented by age group: 4-6 years (n=93), 11-13 years (n=33) and 14-16 years old (n=28). The dmft for the 4-6 year group was 11.2 +/- 6.45 and was 5.06 +/- 3.34 and 7.07 +/- 3.99 for the 11-13 year group and 14-16 year groups respectively.

Lages *et al.*, (2004) investigated the prevalence of caries in 78 Brazilian subjects aged between 1 and 32 years and found that the dmft for the 11 children in the 1-5 year age group was 2.91 +/- 3.99 and for the 31 children in the 6-12 year age group the dmft was 2.77 +/- 3.15.

Ankola *et al.*, (2005) assessed the primary dentition status and treatment needs of eighty-three Indian children with a cleft between 2 and 5-years-old. The dmft scores were presented for each year age (Table 2-5)

Age Group in years	dmft
2 years	6.57
3 years	3.95
4 years	4.05
5 years	6.27

Table 2-5 - Ankola et al, 2005.

2.4.6.3 Do Cleft children have a higher prevalence of caries than their non-cleft peers?

In most studies looking at caries in cleft children, the levels of caries were found to be higher in children with a cleft compared to the unaffected population (Dahllöf *et al.*, 1989; Bokhout *et al.*, 1997; Paul & Brandt, 1998; Chapple & Nunn, 2001; Hewson *et al.*, 2001; Ahluwalia *et al.*, 2004; Kirchberg *et al.*, 2004; Al-Wahadni *et al.*, 2005; Stec-Slonicz *et al.*, 2007; Mutarai *et al.*, 2008). This may be due to the irregularity of teeth around the cleft or to the lack of advice for parents on feeding and oral hygiene (Fitzgerald *et al.*, 1990). However, Lausterstein and Mendelsohn in their Chicago, USA study of 1964 found no greater amount of caries in children with a cleft compared the general population. They compared the dental caries experience of 285 cleft children with a mean age of 8.5 years, with 300 non-cleft children. The cleft group had a mean DMFT of 8.01 compared with 7.45 for the control group. However, they did find that cleft children living in fluoridated water areas “had an average of 2.01 less decayed teeth” than their cleft peers from non-fluoridated water areas. Likewise, Parapanisiou *et al.*, (2009) found no significant difference in “the prevalence of cavitated carious lesions between patients with cleft and without”. This, however, was a small study of only 41 cleft children with a similar number of matched controls. The mean age was 10.7 years with a range from 4 to 18 years. Caries scores were assessed by surface rather than by tooth after professional tooth cleaning and both bitewing and panoramic radiographs were available. The mean dmfs was 7.24 for cleft subjects and 8.38 for non-cleft subjects and the DMFS scores were respectively, 3.00 and 3.40. These figures were not broken down by age groups and so reflect a wide age range. The authors did find significantly higher plaque scores in the cleft subjects ($p = 0.0003$) as well as significantly more initial white spot lesions ($p = 0.000$).

The only study to date to find a decreased caries level in cleft children compared to their non-cleft peers, took place in Northern Ireland (Gregg *et al.*, 1999). Here the caries and restorative experience of 133 cleft children was recorded in a study assessing the value of specialist paediatric dental input into the cleft team in Belfast. Of the 72 five-year-old subjects, 50% were caries free and 42% had untreated caries. The national figures for 5-year-olds were 40% caries free and 70% with untreated caries. This shows a lower level of caries for cleft children compared to their non-cleft peers following specialist paediatric dental intervention including preventive advice and restorative care.

However many studies have disputed this and found an increase in the levels of caries in children with a cleft.

Dahllöf *et al.*, in 1989 (Stockholm, Sweden) had a sample size of 49 five and six-year-old cleft children, mean age 5.5 years. They did not separate the different cleft types and included three children with a syndrome. The results showed that 39% of the control group were caries free compared to only 22% of the cleft group and that the mean dfs was 7.0 for the cleft group compared to 3.9 for the control group.

Ishida *et al.*, in 1989 (cited in Wong & King 1998) looked at both the primary and permanent dentitions by dividing their sample into two age groups; 2-5 years old and 6-14 years old and again found an increased risk of caries in the cleft group.

Bokhout *et al.*, in 1996 showed an increased caries prevalence in 2.5-year-old Dutch children with clefts. Of the 76 children with clefts, 26% had caries with a mean dft of 0.59+/-1.35, whilst of the 75 control children, 5.3% had caries with a mean dft of 0.11+/-0.54. Half of the caries was present on the maxillary incisors.

In a second paper of 1997, Bokhout *et al.*, reported the results of a longitudinal study of 81 cleft children and 77 controls who were examined regularly from birth to 4 years of age. Caries developed in 30.9% of the cleft children compared to 6.5% of the controls. Caries occurred most commonly in the teeth adjacent to the cleft and molars of the cleft children.

Paul and Brandt (1998) investigated the oral health of 114 children attending the cleft clinics in Billericay, England. The age range was 3 to 18 years with a mean age of 8.0 years. Of the 41 subjects in the 3-5 year age group, 22% had evidence of caries with a mean dmfs of 1.9 (SD 6.5). When analysed by cleft type, 63.7% of those with CL, 61.4% of CP, and 45.8% of those with CLP were caries free.

Hewson *et al.*, (2001) investigated the caries experience of 90 cleft children in the west of Ireland and compared this to a control group of 100 non-cleft children. The age range was 18 months to nearly 17 years. Only 22% of the cleft group were caries free compared to 41% of the controls. The results were presented as a mean score for the primary and permanent teeth but were not subdivided into age groups. The cleft group had a mean dmft of 2.52 and a DMFT of 1.67 whereas the control figures were 0.93 and 2.07 respectively. The higher primary dmft score in the cleft group was statistically significant ($P<0.0001$) but not for the permanent scores.

Ahluwalia *et al.*, (2004) compared the caries rate, oral hygiene and the numbers of caries associated microorganisms present in 81 cleft palate children, aged between 6 and 16 years, with 61 controls in London, England. Results showed a statistically higher dmft and DMFT scores ($P<0.01$) in the cleft group compared to the controls. It also reported higher plaque scores, greater numbers of caries associated microorganisms and longer oral clearance times which may all be contributing factors to the greater levels of caries in the cleft group.

Kirchberg *et al.*, (2004) compared the caries experience of 623 children, aged between 6 and 16 years, with clefts in Leipzig, Germany with the results of the National Survey of 1998 of 47,646 children between 6 and 16 years carried out by the public health service in Germany. Caries was found “twice as often in children with clefts aged 6-7 years than in the control group” of the same age. Aged 6 years and 7 years the cleft dmft was 4.0 and 5.16 respectively, whilst the control dmft was 2.05 and 2.42 respectively. Likewise, in permanent teeth, “a significantly higher prevalence of caries was found in patients with a cleft aged 6-12 years”.

Al-Wahadni *et al.*, (2005) in their Jordanian study of 32 subjects with a cleft aged between 10 and 28 years divided their subjects by age in to two groups, 10-15 years (n=13) and 16-28 years (n=19) and compared the caries prevalence with 47 controls. In both age groups the DMFT was significantly greater for the cleft subjects than the controls.

Stec-Slonicz *et al.*, (2007) assessed the dental status of 37 Polish and 63 German subjects with cleft lip and palate aged between 3.5 – 18 years and compared the caries prevalence in the two groups with that of the corresponding local population. Both samples were divided into two age groups: 6-12 years and 13-18 years. However this does not account for the subjects in the 3.5 – 6.0 year age range for whom no data was reported. Caries prevalence in each group was higher than the general Polish and German population.

Mutarai *et al.*, (2008) compared the caries prevalence in 69 Southern Thai children aged between 18 and 36 months with that of 69 non-cleft controls. The cleft children had significantly higher dmft (mean dmft 9.190 than the controls (mean dmft 6.46).

Al-Dajani, (2009) in a study in Syria found an odds ratio of 2.52 for the association of dental caries with the presence of a CL+/-P. This study used a control group matching each of the 53 study subjects with one of their same sex siblings. By so doing the author concluded that “subjects with CL+/-P are susceptible to dental caries independently of socioeconomic status”.

2.4.6.4 Hypoplasia and developmental defects of enamel

Several papers have investigated hypoplasia and developmental enamel defects in cleft children (Ranta, 1986; Vichi & Franchi, 1995; Chapple & Nunn, 2001) using the Modified DDE Index (Clarkson & O’Mullane, 1988).

The precise aetiology remains unclear but is thought that, as teeth adjacent to the clefting site have different mineral structure antenatally, the same processes involved in clefting may play a role in enamel structural defects (Lagarde *et al.*, 1987; Malanczuk *et al.*, 1999). Others have postulated a link with surgical interventions and with feeding difficulties postnatally (Ranta, 1986).

Most authors (Vichi & Franchi, 1995; Malanczuk *et al.*, 1999; Chapple & Nunn, 2001) found developmental defects of enamel in both the primary and secondary dentitions. These same authors also found that dental abnormalities, including hypoplasia and hypodontia, occur more frequently in children with a cleft than in the unaffected population. Chapple and Nunn in their 2001 study demonstrated that hypoplasia was more likely to occur around the cleft site and found “a relationship between the severity of the cleft and the number of teeth with hypoplastic defects”.

2.4.6.5 Does Caries Prevalence Differ with the Type of Cleft?

Bian *et al.*, (2001) compared the caries experience of children with cleft lip only to those with cleft lip and palate. 104 Chinese children, aged between 3 and 6 years, were examined and their dmft scores determined. Seventy five percent of all the children had some caries experience. Twenty seven percent of the 3 to 4-year-olds were caries free, dropping to 23% of the 5 to 6-year-olds. Results showed that children in the CLP group had a higher prevalence of caries than those with cleft lip alone, the mean dmft being 4.1(SD +/_3.8) and 2.7 (SD +/-2.5) respectively.

Three studies have investigated the dental health of children with unilateral cleft lip and palate. The oral health status of Russian children with UCLP was evaluated by Turner *et al.*, (1998) in a visit to Moscow. 89 children aged between 5 and 9 years had a dental examination and their decayed, missing and filled teeth recorded. The missing component of the scores was not reported as the examiners were unsure whether the teeth were missing due to caries, exfoliation, or the cleft. Six percent of the subjects were caries free with a mean dft of 7.31(+/-1.70) and a mean DFT of 1.45 (+/_3.69). In the second study the caries prevalence of 60 UCLP children, aged between 3 and 15 years, attending a cleft clinic in London, England were compared to 60 matched non-cleft children attending a trauma clinic in the same hospital (Lucas *et al.*, 2000). There was no statistically significant difference between the mean dmfs or dmft of the two groups (cleft group 6.05; non-cleft group 6.97). Nor was there any significant difference in the plaque scores for the primary and permanent dentitions between the two groups. The final study (Stec-Slonicz *et al.*, 2007) found “no statistically significant difference in the frequency of caries” between the subjects in their study with UCLP and BCLP.

As mentioned previously, Johnsen and Dixon in 1984 found that 60% of children with BCLP, 20% of those with UCLP and 25% of patients with CP had carious primary incisors. Children with unilateral clefts had caries on the cleft side and “the pattern of caries tended to be unilateral”, and the children with fewer incisor teeth present had fewer carious lesions, suggesting that the caries may be due to crowding or the positioning of the incisors on the cleft side.

When analysed by cleft type, Paul and Brandt (1998) reported that caries was greatest in those with CLP, with only 45.8% caries free, followed by 61.4% of CP and 63.7% of the subjects in their study with CL were caries free.

Besseling and Dubois (2004) reported that when analysed by cleft type, the dmft in the 4 – 6 year age group of their study was highest for those with BCLP (13.50) followed by UCLP (12.14), CP (10.94) and CL (7.16) and that this was mirrored in the other age groups.

Kirchberg *et al.*, (2004) examined 623 children, aged between 6 and 16 years, with clefts in Leipzig Germany, and found the prevalence of caries was significantly higher in CLP children than those with CP alone in both primary and permanent teeth.

Ankola *et al*'s Indian study again found that the mean dmft was higher for clefts of the primary and secondary palate (5.63 +/- 5.45) and clefts of the secondary palate only (5.67 +/- 4.66) than of the primary palate alone (2.4 +/- 1.24).

Al-Wahadni *et al.*, (2005) found no significant difference in the caries experience of their UCLP when compared to the BCLP subjects.

However, Mutarai *et al.*, (2008) found that "cleft status was not an important factor for caries prevalence when oral health behaviours were controlled", suggesting that "frequent sugary food consumption and night-time feeding habit" were the variables significantly associated with dental caries.

2.4.6.6 The Importance of Caries Prevention in Children with a Cleft

The above studies detailing the greater prevalence of caries in cleft children compared to their non-cleft peers, highlights the importance of early regular preventive advice and treatment for all children with a cleft.

Three studies have been published which show the benefits, in terms of reduced caries prevalence, which can be achieved following the implementation of preventive programmes.

The earliest study is by Stephen and McFadyen published in 1977. This study was entitled "Three years of clinical caries prevention for cleft palate children". A preventive programme was initiated for children with a cleft in the West of Scotland. Patients were recruited on the first visit to the Oral Orthopaedic unit, usually two weeks after birth. A preventive programme of regular diet and oral hygiene advice, fluoride supplements in the form of drops, tablets or gel in individual applicator trays, and the placement of fissure sealants was instigated. Comparing the mean dmfs of the group having the preventive programme and controls of cleft children who had not, it resulted in a 98.7% reduction in dmfs.

In the study by Zschieschack and Grabowski (1999) in Rostock, Germany, the dmfs of 417 children aged 3 to 8 years old with CLP who had not received preventive treatment were compared to 258 CLP children who had. There was a caries reduction of greater than 50% in the group who had received regular dietary advice, oral hygiene instruction and topical and systemic fluoride supplements. In this study the major rise in caries prevalence was between the age of 3 and 4 years.

Cheng *et al.*, (2007) discussed strategies for the prevention of early childhood caries in children with CLP. Recommendations introduced at Royal Children's Hospital in Brisbane included:

- Early contact postnatally with a specialist in the cleft team for oral hygiene instruction and dietary and feeding advice.
- Post-surgical oral hygiene demonstration, especially around the cleft area.
- Regular dental visits with fissure sealants and fluoride supplements where required.

This recommendation for early specialist paediatric dental input has been validated by Gregg *et al.*, (1999) who achieved a lower caries rate in cleft children compared to their non-cleft peers in Belfast following the inclusion of a specialist paediatric dental input into the cleft team.

In this study, I will present caries data by age group. This will enable the targeted use of prevention to younger age groups in the future.

2.4.7 Tooth Wear in Cleft Children

The only study reporting the prevalence of tooth wear in children with a cleft is that by Chapple and Nunn published in 2001. Ninety-one children aged 4, 8 and 12 years were examined using standard indices under standard conditions of seating and lighting. The prevalence of caries, tooth wear, and developmental defects of enamel were reported. Only 19 four-year-old subjects were in the study and all exhibited erosion of enamel of the primary incisors and molar teeth. Unfortunately no more detail on the extent of the erosion, nor on the types of cleft the four-year-olds had, is given.

Chapter 3 – Aims

3.1 Aims

In children with a cleft lip and/or a cleft palate in the West of Scotland between the ages of 6 months and 6 years:

To determine;

1. The percentage of children who have no obvious caries by age group.

2. The mean dmfs and SD by age group.

3. The mean dt, mt, ft and dmft and SD by age group.

4. The relative proportion of caries in the anterior and posterior teeth.

5. The mean dmft for those children with dmft > 0 in each group.

6. Whether cleft children have more caries than their non-cleft peers.

7. The percentage of children with tooth wear extending into dentine on the buccal or palatal surfaces of the primary upper incisor teeth in each age group.

8. Whether there is any association between the occurrence of a cleft and socio-economic deprivation.

Chapter 4 – Method

4.1 Patient Identification and Recruitment

All children with a cleft, between the ages of 6 months and 6 years, in the West of Scotland are invited to attend the Oral Orthopaedic Prevention Clinic at Glasgow Dental Hospital and School. The children receive a dental examination and preventive dental advice is given to parents regarding toothbrushing and diet. All these children were eligible to take part in this study.

Ethical approval for this study was sought from the local Research and Development Office (Appendix 2) and the West of Scotland Ethics committee (Appendix 3). In the application to the West of Scotland Ethics committee we submitted on the basis that this research has no impact on patients, parents, or guardians and is essentially "routine clinical monitoring" and so anticipated that a patient information sheet and consent form may not be required, and that the study would be exempt from Site Specific Assessment. Ethical approval was granted by the West of Scotland Ethics committee on this basis, and approval was also granted by the local Research and Development Office.

4.2 Data Collection

Data was collected between 2007 and 2009 at the Oral Orthopaedic prevention clinic as part of the routine clinical monitoring of cleft children and involved a simple clinical dental examination under standard conditions using a light, dental mirror and CPITN probe. The data was then recorded on a standardised paper form used by CleftSiS, the managed clinical network for cleft services in Scotland, for their data collection; an example is appended (Appendix 1). As a dental examination is already part of the routine care for these children, no additional consent was required.

The data collected comprised:

- Name, Age, Sex, DoB

- Full postcode
- Side and type of cleft
- dmfs/dmft
- Tooth wear index for 52, 51, 61 and 62

This was thereafter entered, by the author, into a Microsoft Office Excel 2003 programme (© 1985-2003 Microsoft Corporation) on an encrypted laptop which enabled the calculation of the outcome measures as detailed in Chapter 3, Aims.

Each subject in the study was assigned a unique study number to ensure anonymity. Subjects were divided into the following age groups for analysis;

- 0.5 – 1.49 years
- 1.5 – 2.49 years
- 2.5 – 3.49 years
- 3.5 – 4.49 years
- 4.5 – 6.0 years.

4.2.1 Deprivation Categories

The full postcode was collected for each subject in this study to allow the degree of deprivation to be analysed using the Scottish Index of Multiple Deprivation (SIMD) system. It has been long acknowledged that there are great differences between the health of those living in areas of high deprivation and those in areas of low deprivation in lower social classes and those in upper social classes; the latter having significantly less mortality and morbidity.

Historically, the Carstairs Score and deprivation categories have been the most commonly used measures of deprivation in relation to health and disease within Scotland. These were first described in 1981 and revised in 1991 (Carstairs &

Morris 1991).The following table (Table 4-1) details the census information used to derive the Carstairs Score. The Carstairs Score is calculated by combining these variables for an area, usually a postcode sector, and appropriately weighting them.

Postcodes take a standard form; the area code followed in turn by the district, sector and unit codes e.g. G11 5AN, where G is the Area, 11 is the District, 5 is the Sector and AN is the Unit. The Carstairs analysis uses the postcode at Sector level. The deprivation scores for postcode sectors are restructured into seven deprivation categories giving Depcat Scores which range from Depcat 1 (the most affluent postcode sectors) to Depcat 7 (the most deprived postcode sectors).

CENSUS VARIABLE	DESCRIPTION
Overcrowding	Persons in private households living at a density of >1 person per room as a proportion of all persons in private households.
Male Unemployment	Proportion of economically active males who are seeking work.
Low Social Class	Proportion of all persons in private households with head of household in social class 4 or 5.
No car	Proportion of all persons in private households with no car.

Table 4-1 - Variables used to determine the Carstairs Scores.

More recently, a new Scottish Index of Multiple Deprivation (SIMD) has been introduced and is now used in the NDIP reports to analyse deprivation (www.Scotland.gov.uk/topics/statistics/SIMD). The latest version, SIMD 2006, combines information regarding current income, employment, health, education, skills and training, housing, geographic access and crime (Figure 4-1).

These seven domain scores are weighted to provide an overall score for each data zone. Unlike the Depcat system, SIMD uses the full postcode at unit level and is therefore a more accurate indicator of deprivation within a specific area. A second advantage of SIMD is that it will be updated after only three years compared to every ten for the Depcat system (Deprivation and Urban Rural Measurements in ISD, 2004).

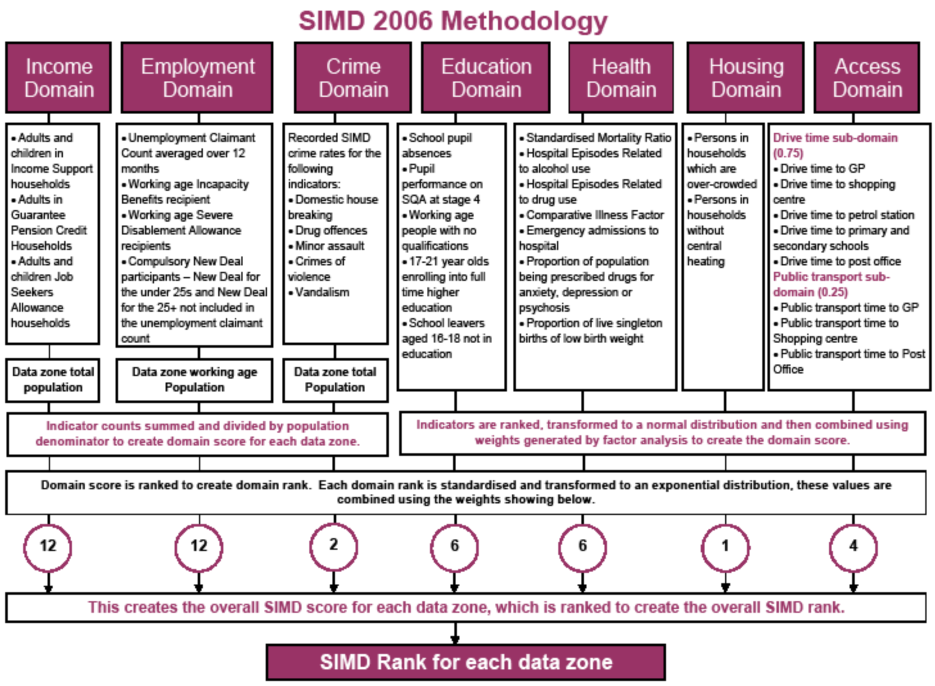

Figure 4-1 - SIMD 2006 Methodology
From www.Scotland.gov.uk/topics/statistics/SIMD.

4.2.2 Cleft Types

We recorded the type of cleft in each subject and also, if it was unilateral, the side affected.

4.2.3 Caries Data

After a full clinical dental examination, clinicians recorded the caries present in each subject as both dmfs and dmft according to BASCD criteria (Mitropoulos *et al.*, 1992; Pitts *et al.*, 1997). A copy of the coding system used is appended (Appendix 1).

Caries is recorded separately for the primary teeth using lower case letters, and the permanent teeth using capital letters. It may be noted either on a tooth basis or by each individual tooth surface. The number of decayed (d), missing (m), and filled (f) surfaces (s) or teeth (t) is totalled to give the dmfs/dmft for primary teeth. The same is done for the permanent teeth by totalling the numbers to give a final DMFS or DMFT score.

Whilst the dmft/DMFT scores give an accurate indication of caries experience at a population level, the surface level score, dmfs/DMFS gives far more detailed information about the caries experience of the individual. This is because it is possible to have three primary molar teeth affected by caries (dmft = 3) but for these three teeth to each have five surfaces affected by caries (dmfs = 15) and this is also true for the missing and filled components of the score.

The comparisons for this study population were the NDIP (National Dental Inspection Programme) figures for the appropriate age matched groups. NDIP is a yearly epidemiological monitoring programme of the dental health of Scottish children. It was established in 2002 following a review of the existing SHBDEP (Scottish Health Boards' Epidemiological Programme) scheme. On alternate years the children in Primary One (average age of 5 years) and Primary Seven (average age of 12 years) throughout Scotland are inspected by Community Dentists. The clinicians involved are calibrated to BASCD criteria to ensure the accuracy and reproducibility of the data collected and to allow true comparisons to be made between the different regions in Scotland. In 2007, for the first time, NDIP included dental inspections of pre-school children, and thus it is possible to use this new data as an age matched comparison group for our results in the younger age groups. The 2008 3-year-old NDIP subjects had an age range of 2.5 years to 4.5 years. The 2008 5-year-old NDIP subjects had a mean age of 5.4 years and an age range of 4.5 years to almost 6-years-old.

To ensure that direct comparisons are possible between the NDIP and the study data, the researcher and the other clinicians involved in the Oral Orthopaedic Prevention Clinic (Professor R. Welbury and Dr S. MacDonald) were NDIP trained and calibrated. This was undertaken on 5th and 6th March 2007 under the tutelage and supervision of Professor Lorna MacPherson, Professor of Dental Public Health and Dr Yvonne Blair, Dental Public Health, Glasgow Dental Hospital and School. The training component involved an illustrated lecture and discussion session on how to record the caries present in accordance with criteria set down by the British Association for the Study of Community Dentistry (Mitropoulos *et al.*, 1992; Pitts *et al.*, 1997). Thereafter a day of clinical training was held with the involvement of nursery children from Hamiltonhill Nursery in Possilpark, Glasgow. In the morning, the clinicians involved carried out inspections on each child and experienced NDIP dental nurses recorded their findings on the standardised data collection form. The data collected by each clinician was compared and, where there was any disagreement, the child involved was re-examined by the training clinician, and a discussion followed to review the case. During the afternoon, this was repeated, but without any opportunity for discussion of the cases, to determine whether any dentist lay out with the acceptable range of agreement (Landis & Koch, 1977; Pine *et al.*, 1997).

The paper records were entered into an encrypted laptop which was networked onto an NHS secure server and then transferred by NHS mail straight onto the secure server of the statistician who calculated the kappa scores and percentage agreement between the examiners. It was deemed on the day that all clinicians involved closely agreed and were therefore successfully calibrated. The agreement proportions were calculated by comparing each clinician with the median for the group (as is done with the NDIP calibration). The disagreement proportion was 1 out of 6 (i.e. one disagreement out of 6 calibration patients examined on the day). This disagreement was one clinician scoring a composite restoration in an occlusal surface where the other three did not. There was complete agreement on the dt component. The Kappa scores for all three clinicians involved in the Oral Orthopaedic Prevention Clinic was 1.0. Further detail of the calibration statistics can be seen in Appendix 4.

One of the main aims of this study was to statistically compare the study results to those of the National Dental Inspection Programme of Scotland. However, in order to compare the difference between the study mean dmft and the NDIP mean dmft and thereafter arrive at a p value, the figures required are the number of study subjects and the mean dmft figures with their standard deviations for both studies. I have been unable to obtain the standard deviation figures for the NDIP 3 and 5-year-old 2008 studies, which makes complete statistical analysis of the results impossible.

4.2.4 Tooth Wear Data

Tooth Wear scores were recorded for the four primary upper incisors according to the Tooth Wear Index (TWI) described by Smith and Knight (1984). The depth and extent of the wear was included. Where teeth were missing or caries was so extensive that assessment of wear was impossible, the tooth was recorded as “excluded”.

4.3 Statistical Analysis

This is a mainly descriptive study using means and proportions with standard deviation and confidence intervals where appropriate. The results were analysed using Minitab 15.1.0.0 (© 2006 Minitab Inc.) and SPSS 15.0 for Windows (© SPSS Inc., 1989-2006) and statistical significance was reported at a 5% level ($p \leq 0.05$).

Chapter 5 – Results

Two hundred and nine subjects were examined in the Oral Orthopaedic Prevention Clinic at Glasgow Dental Hospital and School over the 24 month period of March 2007 - December 2007, and January 2009 - April 2009.

Although in this study the numbers are relatively low, i.e. two hundred and nine subjects, this represents the majority of the available cleft population of the appropriate age range in the West of Scotland. Of the 289 subjects who would qualify for the study, nine died in infancy, forty live at a some distance from Glasgow and receive their preventive and continuing care nearer to home and one moved away from the area leaving 239 potential subjects. Therefore the study participation rate was 87.4%. Of the thirty potential subjects who did not participate, thirteen failed to attend at least one appointment during the study timeframe. The remaining seventeen were born in 2008 and barely six months old so that their initial appointment may have been just after the end of the study period.

The following table, Table 5-1, presents the main demographics of the study population by age group.

	0.5-1.49 Years		1.5-2.49 Years		2.5-3.49 Years		3.5-4.49 Years		4.5-6.0 Years	
Variable	**Number**	%	**Number**	%	**Number**	**%**	**Number**	**%**	**Number**	**%**
Female	21	45.65	21	46.67	22	61.11	9	29.03	23	45.10
Male	25	54.35	24	53.33	14	38.89	22	70.97	28	54.90
SIMD										
1	3	6.52	8	17.78	4	11.11	7	22.58	7	13.73
2	5	10.87	9	20.00	8	22.22	5	16.13	6	11.76
3	10	21.74	8	17.78	5	13.89	5	16.13	6	11.76
4	10	21.74	7	15.56	7	19.44	8	25.81	16	31.37
5	18	39.13	13	28.89	12	33.33	6	19.35	16	31.37
Cleft Types										
UCL	4	8.70	2	4.44	2	5.56	2	6.45	4	7.84
BCL	1	2.17	2	4.44	0	0	0	0	0	0
UCLP	12	26.09	9	20.00	9	25.00	12	38.71	12	23.53
BCLP	3	6.54	4	8.89	1	2.78	3	9.68	11	21.57
CP	22	47.83	23	51.11	21	58.33	10	32.26	19	37.25
Syndrome	4	8.70	5	11.11	3	8.33	4	12.90	5	9.80
Non-Syndrome	42	91.30	40	89.99	33	91.77	27	87.10	46	91.20

Table 5-1 - Demographics.

5.1 Gender ratio

Of the two hundred and nine subjects, 96 (45.9%) were female and 113 (54.1%) were male. The gender ratio within each age group may be seen in Table 5-1 above.

5.2 Cleft types

Cleft palate accounted for the majority of the clefts present, followed in decreasing numbers by UCLP, BCLP, UCL and BCL. Table 5-1 above, shows the proportion of each cleft type within each age group. The distribution of cleft types and the percentage of each cleft type within each gender are shown in and Figures 5-1 and 5-2.

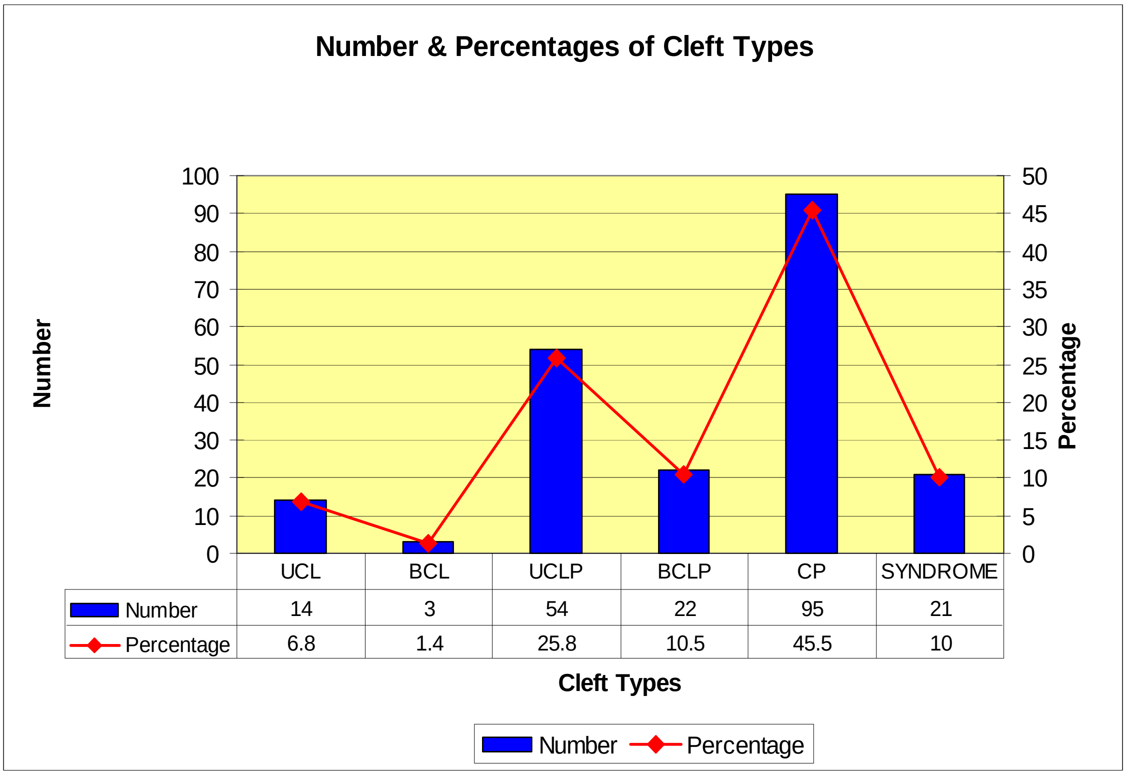

Figure 5-1 - Numbers and percentages of cleft types, n = 209.

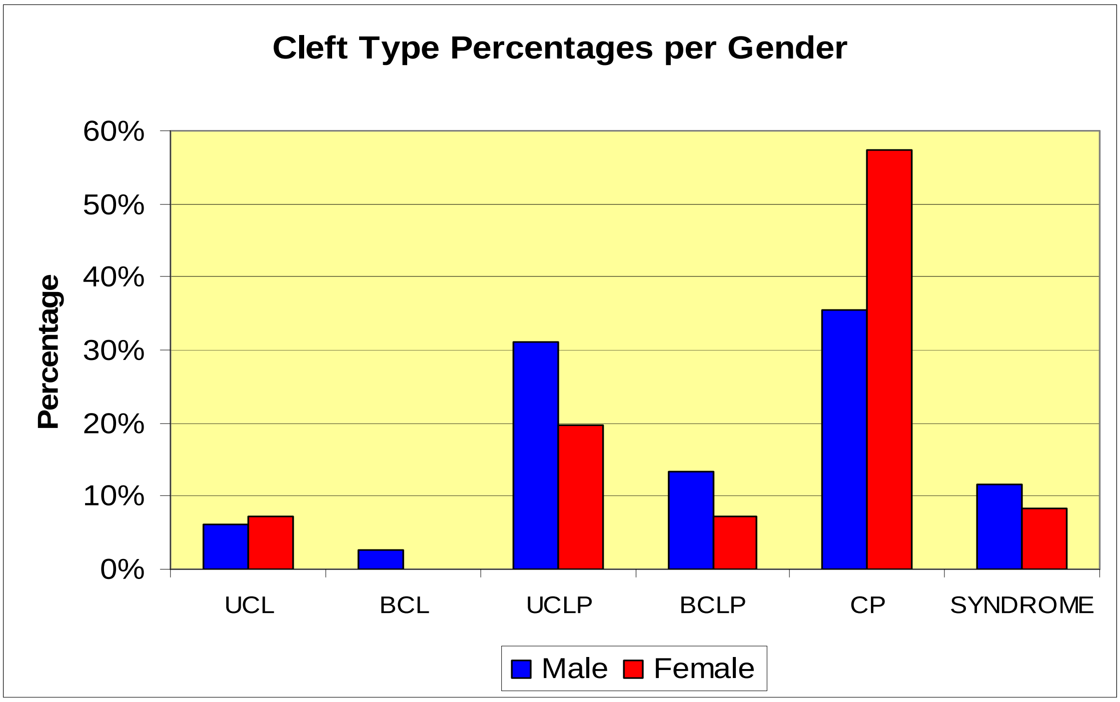

Figure 5-2 - Cleft type percentages by gender, n = 209.

5.3 Syndromes

21 of the two hundred and nine subjects had been diagnosed with a syndrome or an inherited chromosomal abnormality. 13 (61.9%) were male and 8 (38.1%) were female. Twelve had Pierre-Robin sequence, two had DiGeorge syndrome, and one case each of Kabuki, Sodium Valproate, Stickler, Goldenhaar and Van der Woude syndrome with one subject having both Kippel-Fiel and Sprengler syndromes.

Nineteen subjects (90.5%) had a cleft palate, one (4.8%) a UCLP, and one (4.8%) a BCLP.

80.9% of syndromic subjects had no obvious caries. The mean dmfs score was 4.76 and mean dmft was 1.43. The mean dmft for those with caries present was 7.5. Of the four subjects who had experienced caries, three were male and one female. One male subject had only one affected tooth with 2 carious surfaces, another male had three affected teeth with 5 affected surfaces, whilst the third male had 11 affected teeth and 45 affected surfaces. The only female subject with a syndrome had 15 carious teeth with 48 affected surfaces.

5.4 Percentage of subjects with no obvious decay

Table 5-2 below shows the percentage of subjects within each age group who had no obvious decay.

Age Group in Years	Numbers	Percentage with no obvious decay	95% CI for percentage
0.5 - 1.49	46	100	93.6, 100
1.5 - 2.49	45	91.1	78.7, 97.5
2.5 - 3.49	36	74.3	56.7, 87.5
3.5 - 4.49	31	67.7	48.6, 83.3
4.5 - 6.0	51	37.2	24.1, 51.9

Table 5-2 - Percentage with no obvious decay by age group, n = 209.

Table 5-3 below compares the percentage of study subjects who had no obvious decay with the 2008 NDIP results for the same age groups where available. 2008 3-year-old NDIP data set had an age range of 2.5 years to 4.5 years, and the 2008 NDIP 5-year-old data set had an age range of 4.5 years to almost 6-years-old.

Age Group in Years	Our Study	NDIP 3-year-olds (2008)	NDIP 5-year-olds (2008)
0.5 - 1.49	100		
1.5 - 2.49	91.1		
2.5 - 3.49	74.3	75.6	
3.5 - 4.49	67.7	75.6	
4.5 - 6.0	37.2		57.7*

Table 5-3 - Percentage with no obvious decay with NDIP comparators.
*** p = 0.004**

Table 5-4 and Figure 5-3 show the percentage of 5-year-old subjects with no obvious decay by SIMD quintile.

SIMD Quintile	Percentage No Obvious Decay, This Study	Percentage No Obvious Decay, NDIP, 2008
Least deprived 1	50	73.1
2	50	67.7
3	50	58.6
4	26.7	52.2
Most Deprived 5	31.2	42.2

Table 5-4 - Percentage of 5-year-olds with no obvious decay by SIMD quintile, n = 51.

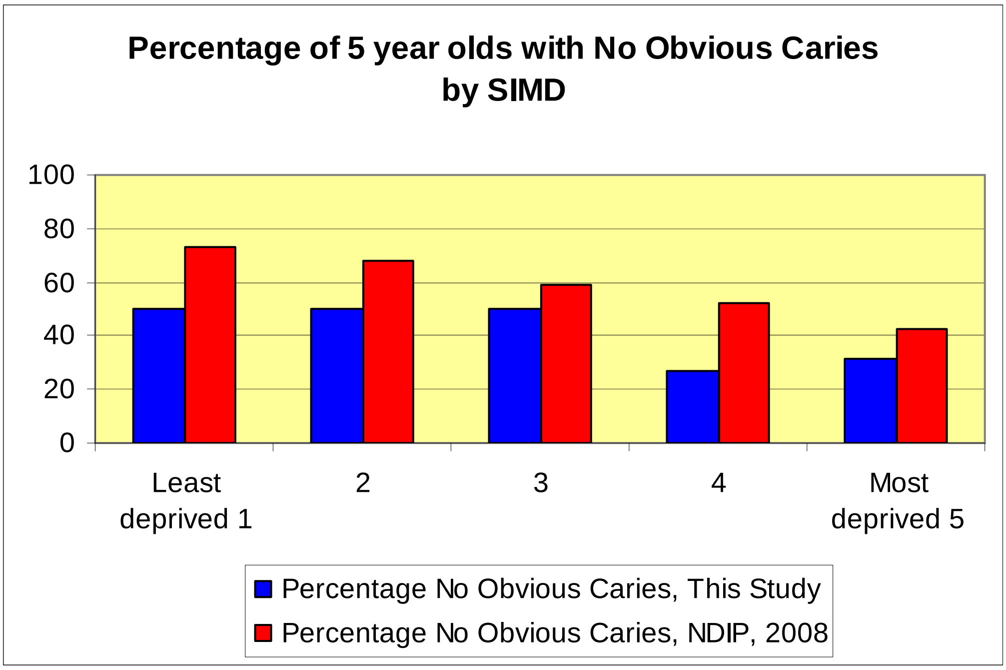

Figure 5-3 - Percentage of 5-year-olds with no obvious caries by SIMD quintile, n = 51.

5.5 Mean dmfs

Table 5-5 below shows the mean dmfs scores for each age group.

Age Group in Years	Mean dmfs	95% CI for mean
0.5 - 1.49	0	n/a
1.5 - 2.49	2.02	-0.73, 4.77
2.5 - 3.49	2.58	-0.24, 5.40
3.5 - 4.49	1.61	0.39, 2.84
4.5 - 6.0	8.67	4.06, 13.27

Table 5-5 - Mean dmfs by age group, n = 209.

5.6 Mean dt, mt, ft and dmft

Table 5-6 and Figure 5-4 below, show the mean dt, mt, ft and dmft figures for each age group.

	Means with 95% CI			
Age Group in Years	Dt	mt	ft	Dmft
0.5 - 1.49	0	0	0	0
1.5 - 2.49	0.49 (-0.14, 1.11)	0	0	0.49 (-0.13, 1.11)
2.5 - 3.49	1.0 (0.09, 1.90)	0	0	1.03 (0.10, 1.95)
3.5 - 4.49	0.77 (0.06, 1.48)	0.06 (-0.07, 0.19)	0.09 (-0.05, 0.24)	0.93 (0.21, 1.66)
4.5 - 6.0	1.57 (1.02, 2.12)	1.25 (0.32, 2.19)	0.35 (0.09, 0.61)	3.24 (0.21, 1.66)

Table 5-6 - Mean dt, ft, mt and dmft by age group, n = 209.

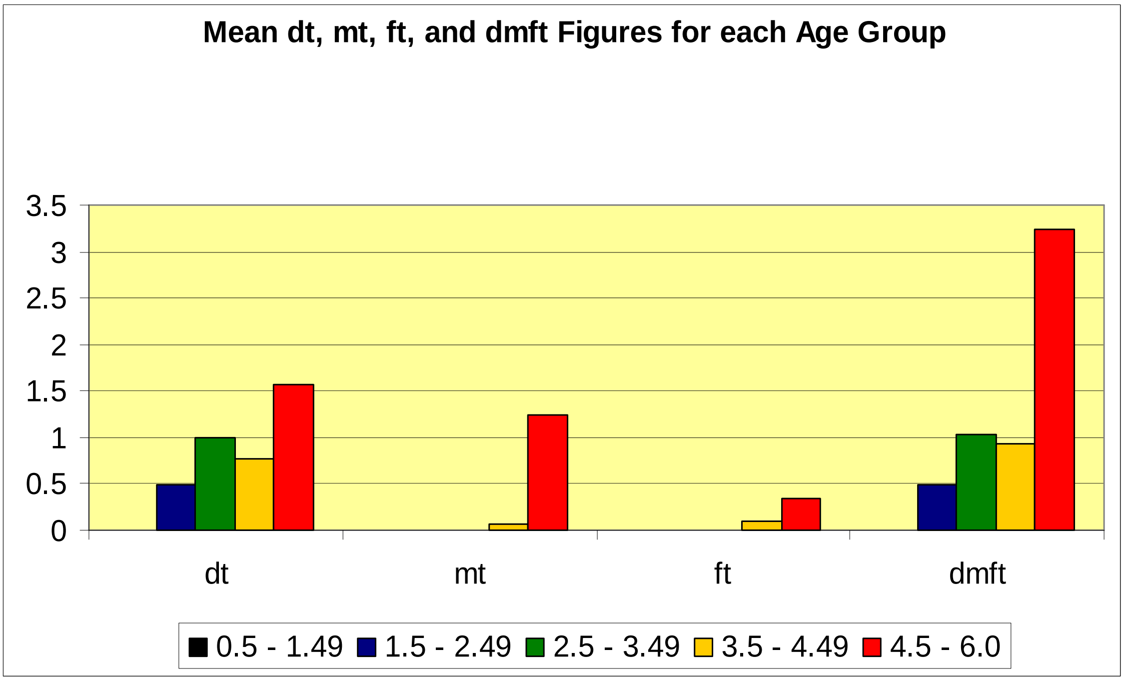

Figure 5-4 - Mean dt, mt, ft and dmft by age group, n = 209.

Table 5-7 below compares the study mean dmft with the NDIP, 2008 results for the same age groups where available. The NDIP SD values for the means were unavailable, so no p value could be calculated.

Age Group in Years	This Study	NDIP 3-year-olds (2008)	NDIP 5-year-olds (2008)
0.5 - 1.49	0		
1.5 - 2.49	0.49		
2.5 - 3.49	1.03	0.97	
3.5 - 4.49	0.94	0.97	
4.5 - 6.0	3.24		1.86

Table 5-7 - Mean study dmft and mean comparator dmft.

Table 5-8 and Figure 5-5 below show the mean dmft figures for the different cleft types.

	Mean dmft	Std. Deviation	95% Confidence Interval for Mean	
UCL	0.57	1.28	-0.17	1.31
BCL	0	0	0	0
UCLP	0.81	1.84	0.31	1.32
BCLP	2.77	5.98	0.12	5.43
CP	1.16	2.32	0.68	1.64
SYND	1.43	3.95	-0.37	3.23

Table 5-8 - Mean dmft for individual cleft types with SD and 95% CI, n = 209.

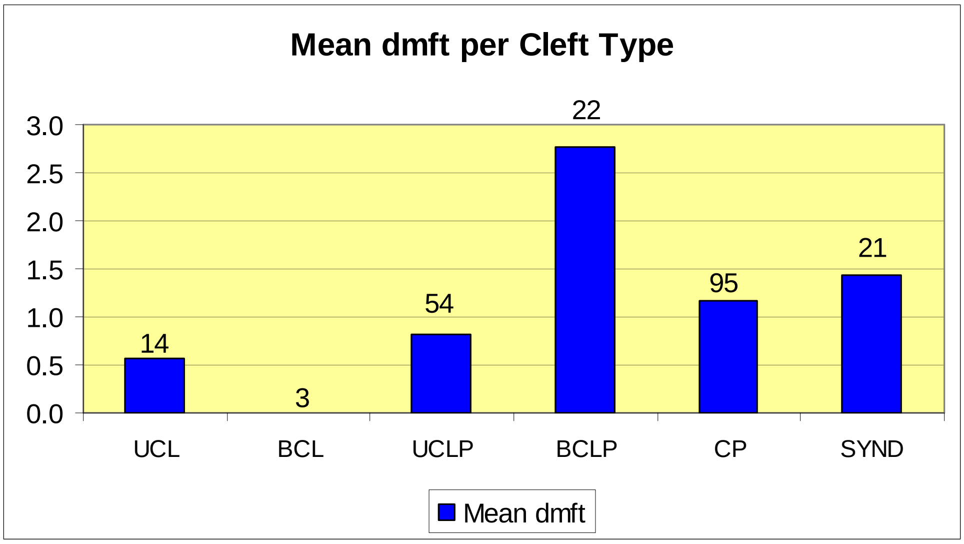

Figure 5-5 - Mean dmft by cleft type, n = 209.
Figures above columns indicate number of subjects with cleft type.

5.7 The distribution of caries between anterior and posterior teeth

Figures 5.6 and 5.7 demonstrate the proportion of caries in anterior and posterior teeth by tooth and surface detail.

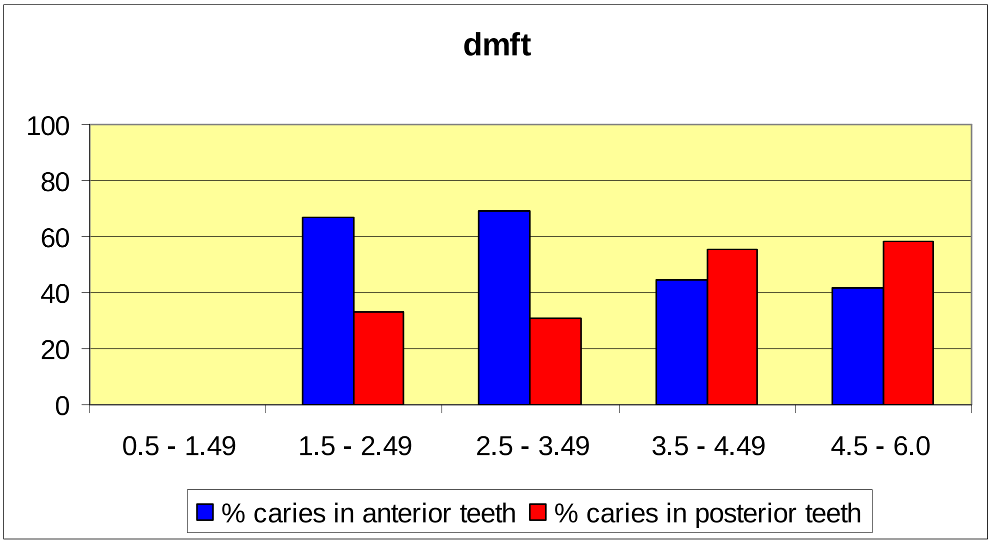

Figure 5-6 - Caries in anterior and posterior teeth (dmft) by age group, n = 209.

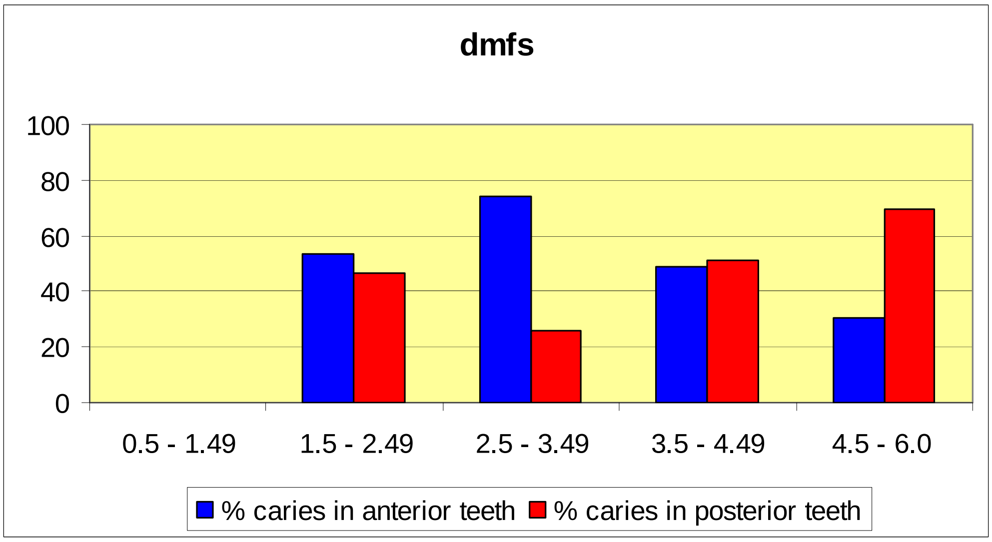

Figure 5-7 - Caries in anterior and posterior teeth (by surface) by age Group, n = 209.

5.8 Mean dmft where dmft > 0

Table 5-9 shows the mean dmft for those subjects who had a dmft score of greater than zero.

Age Group in Years	Mean dmft where dmft>0	SD
0.5 - 1.49	n/a	n/a
1.5 - 2.49	5.5	5.19
2.5 - 3.49	4.0	4.21
3.5 - 4.49	2.9	2.6
4.5 - 6.0	5.16	4.63

Table 5-9 - Mean dmft where dmft>0 by age group.

5.9 Care Index

Table 5-10 shows the Care Index for each age group (ft/dmft x100) with national comparisons where available.

Age Group in Years	Care Index	NDIP 5-yr-olds, 2008
0.5 - 1.49	n/a	
1.5 - 2.49	n/a	
2.5 - 3.49	n/a	
3.5 - 4.49	10.5	
4.5 - 6.0	10.9	9.68

Table 5-10 - Care Index by age group.

5.10 Percentage of subjects with tooth wear

Table 5-11 below, shows both the percentage of subjects with any sign of wear on their upper anterior primary incisors and the percentage of subjects where tooth wear extended into dentine.

Age Group in Years	Percentage with any tooth wear	Percentage with wear into dentine
0.5 - 1.49	2.1	2.17
1.5 - 2.49	36.0	8.89
2.5 - 3.49	45.7	8.33
3.5 - 4.49	63.3	32.26
4.5 - 6.0	80.0	27.45

Table 5-11 - Percentage of subjects with tooth wear, n = 209.

Table 5-12, below shows the comparison figures for tooth wear into dentine for the same age groups from the National Diet and Nutrition Survey of 1995 (Hinds & Gregory) and Children's Dental Health in the UK, 2003 (Lader *et* al., 2003).

Age Group in Years	Percentage tooth wear into dentine This Study	Percentage tooth wear into dentine Comparator Study
0.5 – 1.49	2.17	
1.5 – 2.49	8.89	3.0 * (Hinds & Gregory,1995)
2.5 – 3.49	8.33	6.0 (Hinds & Gregory,1995)
3.5 – 4.49	32.26	13.0 * (Hinds & Gregory,1995)
4.5 – 6.0	27.45	22.0 (Lader *et al*, 2003)

Table 5-12 - Comparison of tooth wear into dentine.
*** p ≤ 0.05**

5.11 SIMD

Table 5-13 and Figure 5-8 below show the number and percentage of subjects within each SIMD category.

	Number	Percentage
SIMD		.
Least Deprived 1	29	13.9
2	33	15.8
3	34	16.3
4	48	22.9
Most Deprived 5	65	31.1

Table 5-13 - Percentage of subjects within each SIMD quintile.

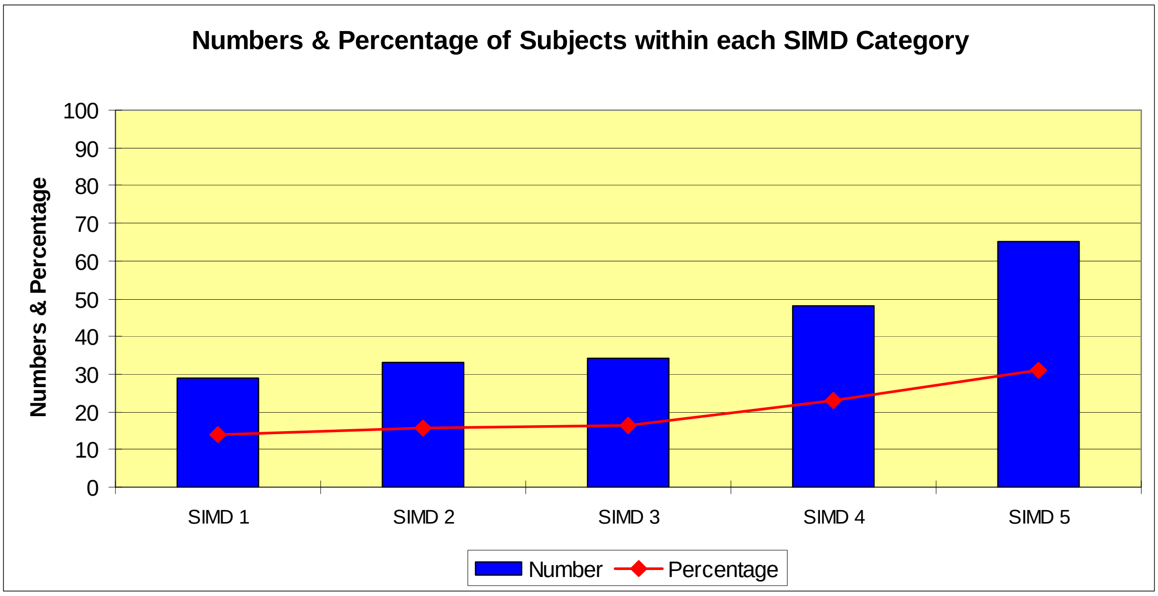

Figure 5-8 - Numbers and percentages of subjects within each SIMD quintile, n = 209.

Table 5-14 demonstrates the proportion of 5-year-olds within each SIMD quintile.

	5-year-olds, This Study		**5-year-olds, GGC**	
	Number	**Percentage**	**Number**	**Percentage**
SIMD		.		
Least Deprived 1	8	15.7	4381	19.5
2	6	11.8	3200	12.9
3	6	11.8	2907	11.7
4	15	29.4	3789	15.2
Most Deprived 5	16	31.3	10106	40.7

Table 5-14 - Percentage of 5-year-olds within each SIMD quintile.

Chapter 6 – Discussion

Both Wong & King, 1998 and Hasslof & Twetman, 2007, as part of their review papers on the incidence of caries in cleft children, discussed the problems associated with conducting this type of study. Small sample size, a wide age range, and lack of suitable controls were highlighted as potential downfalls.

In order to give a more accurate picture of the prevalence of caries, and to try to alleviate the potential problem of a wide age range, subjects were divided into yearly age bands from 6 months of age up to 6 years of age.

In scientific data, any controls should ideally be directly comparable to the study population in terms of age, gender, method of data collection (i.e. examination method and caries diagnosis threshold) and geographic location. Control studies should also have been conducted at the same time or within the same year. The controls chosen for this study were the NDIP Scottish national data. The 2008 figures for 3-years-olds and 5-year-olds were used as data was collected over the same timeframe and this ensured comparable data in terms of both age and geographic location.

The 3-year-old (nursery) NDIP survey included children from both the pre-school and ante-pre-school classes with an age range of 2.5 years to 4.5 years, and these figures have been used as the comparator figures for both our 2.5 – 3.49 and 3.5 – 4.49 age groups. As the NDIP nursery survey was carried out in the Greater Glasgow Health Board area, the resulting data is from the same geographic population as the study subjects.

The 5-year-old survey included children between the ages of 4.5 years and almost 6-years-old, and was used as the comparator for our 4.5 – 6.0 year age group. The NDIP survey was carried out throughout Scotland and the means include all the data nationwide, although where separate figures for some of the statistics are available by Health Board region, these will be quoted.

6.1 Gender ratio

Of the two hundred and nine subjects who were examined in this study, 45.9% were female and 54.1% were male. This is identical to the gender ratios reported by both Clark *et al.*, (2003) of 45% female and 55% male, and Womersley & Stone (1987) of 45.3% female and 54.6% male. Both these studies were conducted in Scotland. This shows that the gender ratio of orofacial cleft in Scotland has remained remarkably stable for the last 35 years.

6.2 Cleft types

We have endeavoured to present as much data as possible about the individual cleft types, however these have not been statistically analysed, because the statistical results would not be meaningful due to the small numbers involved.

Two of the other UK based cleft caries studies have reported the proportion of the various cleft types in their studies. Chapple & Nunn, (2001) combined the UCLP and BCLP subjects and, similarly to this study, found a majority presented with CP, followed in turn by U/BCLP and UCL. Ahluwalia *et* al., (2004) reported that the majority of their subjects presented with UCLP followed in turn by BCLP, CP and UCL. However one (Lucas *et al.*, 2000) focused solely on UCLP subjects and Hewson *et al.*, (2004) did not report which types of clefts their subjects had. None of the above studies reported findings regarding cleft types within both genders.

The figures looking at gender and cleft type are very similar to the results of Womersley & Stone (1987) and Clark *et al.*, (2003). Womersley & Stone also found that CP was the most commonly occurring orofacial cleft in Scotland between 1974 and 1985, followed in turn by CLP and CL. The same authors found that CP occurred more frequently in females (M:F = 1:1.6) and that CLP and CL alone occurred more commonly in males. Clark *et al*,. (2003) investigated the occurrence of facial clefts in Scotland between 1989 and 1998 and reported CP as the most commonly occurring type of cleft and that CP occurred in 56% of females and 44% of males. Although the authors of both these papers combined

CLP and CL alone, they found that these types occurred more commonly in males than females.

6.3 Syndromes

As mentioned in the results section, 21 (10%) of the subjects had been diagnosed with a syndrome or an inherited chromosomal abnormality.

Most of the papers investigating caries in cleft subjects either excluded all subjects with a syndrome from eligibility for their study (Lin & Tsai, 1999; Bian *et al.*, 2001; Besseling & Dubois, 2004; Al-Wahadni *et al.*, 2005; Mutarai *et al.*, 2008), or did not give any information regarding whether any of their subjects had a syndrome (Lauterstein & Mendelsohn, 1964; Johnsen & Dixon, 1984; Turner *et al.*, 1998; Lucas *et al.*, 2000; Hewson *et al.*, 2001; Ahluwalia *et al.*, 2004; Kirchberg *et al.*, 2004)

Two studies (Dahllöf *et al.*, 1989; Hasslof & Twetman, 2007) quoted the numbers of subjects with a syndrome included in their studies. Dahllöf *et al.*, 1989 included 3 syndromic subjects out of a total of 49 (6.1%), and Hasslof & Twetman, 2007 included 14 syndromic subjects in their total of 90 (15.5%). Our study result of 10% lies approximately midway between the two.

In line with recommendations from other papers, (Fraser, 1970; Bixler 1981) subjects with a syndrome were excluded from the main results of this study. However I will present some data separately in this section for the subjects with a syndrome, including: gender ratio, cleft types, and caries prevalence.

Of the 21 subjects with a syndrome, 13 (61.9%) were male and 8 (38.1%) were female. This mirrors the male bias toward the presence of a cleft found in non-syndrome subjects but is a higher percentage.

The mean dmfs, dmft and dmft >0 are substantially higher than that for the non-syndromic subjects. However this figure comprises only four subjects with caries, and spans a greater age range (2.5 – 6.0 years). As seen in the general non-cleft population, the caries present within this small group of syndromic cleft subjects is concentrated in a minority of the subjects.

6.4 Percentage of subjects with no obvious decay

All of the subjects in the 0.5 – 1.49 age group were caries free. No papers have been found detailing the caries prevalence of this age group in either the cleft or non-cleft populations. The percentage of subjects in this study remaining caries free reduces with each increasing age group until only 37.2% of the 4.5 – 6.0 years age group remained caries free.

In the 1.5 – 2.49 age group 91.1% of the subjects were caries free. NDIP does not include inspections for this young age group, however a study undertaken in Lanarkshire (Taylor, 2004) reported that 97% of non-cleft children in that region were caries free.

74.3% of the 2.5 – 3.49 age group were caries free. This is very similar to the figures from NDIP 2008 3-year-old survey which reported 75.6% of children had no obvious caries and the difference is not statistically significant (p = 0.844).

67.7% of the 3.5 – 4.49 age group were caries free. This is slightly lower than the NDIP 2008 figure of 75.6%, but is not statistically significant (p = 0.299).

20% fewer cleft children in the 4.5 – 6.0 year age group are caries free when compared to their non-cleft peers (p = 0.004). When considering the NDIP figures for the Greater Glasgow and Clyde (GGC) health board area alone, 27% fewer cleft children have no obvious caries (p = 0.01).

The follow-up question is then: “Why do the study subjects in the 4.5 – 6.0 year age group have more caries than their non-cleft peers?” Is this because of the presence of a cleft, or because there is a higher proportion of cleft subjects from more deprived areas?

When comparing the proportion of study 5-year-olds in each SIMD quintile to the proportion of 5-year-olds in the Greater Glasgow and Clyde area in each SIMD quintile, there is a similar trend towards increased numbers in the two most deprived quintiles. The numbers are too small to make statistical analysis reliable however, descriptively, it seems that there is not a higher level of deprivation within the cleft study group.

If the percentage of 5-year-olds who have no obvious caries is looked at by SIMD quintile, the cleft subjects in all SIMD quintiles have more caries than the NDIP comparators and there is not such a marked gradient between quintiles 1 and 5.

However, despite it being tempting to conclude that the caries is due to the presence of a cleft, without statistical evidence, no firm conclusions can be drawn as to what is the causal factor.

Other studies have reported the percentage of cleft children who were caries free. Chapple & Nunn (2001) reported that 63% of their 4-year-old cleft subjects in the Newcastle area were caries free, a figure which is slightly lower than in our 4-year-olds. Bokhout *et al.*, (1996) reported that 74% of the 2.5-year-old Dutch study subjects were caries free, again a similar figure to that found in this study. Hewson *et al.*, (2001) reported that 22% of their Irish cleft subjects were caries free. However this was a figure for the whole study population which ranged in age from 18 months to almost 17 years. Paul & Brandt (1998) also reported data for a wider age range in their study conducted in Billericay. 78% of the 41 subjects in their 3-5 year age group were caries free.

The only study reporting a significantly higher percentage of cleft children caries free was that of Gregg *et al.*, (1999). The authors reported that 50% of their 5-year-old subjects were caries free. This figure was achieved following specialist paediatric dental intervention for the cleft children, including preventive advice and restorative care.

6.5 Mean dmfs

Very few caries studies report figures for caries by tooth surface. The majority of both cleft and non-cleft studies report results for numbers of teeth affected by caries and quote mean dmft and mean dmft where >0.

Lauterstein & Mendelsohn (1964) examined 285 cleft children with a mean age of 8.5 years and found no significant difference in the permanent surface scores between cleft (mean DMFS 14.1) and non-cleft controls (13.3).

Lucas *et al.*, (2000) found no significant difference between their 60 cleft children, aged 3 – 15 years (mean dmfs 6.05) and the controls (6.97).

Unlike the two studies mentioned above, Paul & Brandt, (1998) divided their cleft subjects into smaller age bands. The authors reported a mean surface caries dmfs of 1.9 (SD 6.5) in their 41 study subjects aged 3 to 5 years. This figure is similar to our mean dmfs of 1.61 for the 3.5 – 4.49 age group. Our 4.5 – 6.0 age group had a greater mean dmfs (8.67) than Paul & Brandt whose subjects overlap in age with this group.

6.6 Mean dt, mt, ft and dmft

As previously discussed in the Literature Review, studies investigating caries in cleft children have been undertaken in many varied geographical locations over a long timeframe. These, together with the fact that many differing thresholds for the diagnosis of caries have been employed, makes valid statistical comparisons with many study results impossible. However where mean dmft values are reported in studies, these figures have been quoted and compared where appropriate.

6.6.1 Age group 0.5 – 1.49 years

None of the subjects in this age group had caries; all were caries free on examination. No other studies found have reported dmft for this age group of cleft children.

6.6.2 Age group 1.5 – 2.49 years

The mean dmft for this group was 0.49. The caries was present in only 4 (8.9%) of the 45 subjects in this group. Two of these subjects each had a dmft score of 1 and two each had a dmft score of 10.

Although Ankola et al., (2005), reported a much higher D_3 dmft of 6.57 for the 2-year-old cleft children in India, this must be viewed in relation to the generally higher prevalence of caries in India.

6.6.3 Age group 2.5 – 3.49 years

For the 25.7% of children in this age group in our study with caries, the mean dmft was 1.03.

Bokhout et al., (1996) had a very similar percentage of children aged 2.5 years with caries (26%), but a lower dft of 0.59. Ankola et al., (2005), reported a higher mean dmft of 3.95 for the 3-year-old children in their study.

When compared to the NDIP results for the 2008 inspections of nursery-aged children, the mean dmft figures are very similar (NDIP 0.97, our study 1.03) showing that there is no difference between children with a cleft and their non-cleft peers in the 2.5 – 3.49 age group.

6.6.4 Age group 3.5 – 4.49 years

Chapple & Nunn found a mean dmft of 1.3 for the 37% of the 4-year-old cleft children with caries in their 2001 study. Both these figures are slightly higher than our mean of 0.94 for the 32.3% of our 3.5 – 4.49 age group. This study took place in Newcastle, England, and although it used the D_3 threshold of diagnosis, it was carried out in 2001, since when there have been significant reductions in the incidence of caries in both England and Scotland. In addition a large proportion of the children in the Newcastle study will have been receiving fluoridated water.

When compared to the NDIP results for the 2008 inspections of nursery-aged children, the mean dmft figures are virtually identical (NDIP 0.97, our study 0.94) showing that there is little difference between children with a cleft and their non-cleft peers in the 3.5 – 4.49 age group.

6.6.5 Age group 4.5 – 6.0 years

The mean dmft for the 4.5 – 6.0 year age group was 3.24. This is a large increase in the prevalence of caries from a mean of 0.93 in the 3.5 – 4.49 year age group. It would be hoped that the recent extension of the Childsmile Programme in

Scotland into the Primary One classes in schools will help to reduce the mean dmft score in future.

When compared to the NDIP results for the 2008 inspections of 5-year-old children, the mean dmft in our study of 3.24 is greater than that of the general population at 1.86 (NDIP, 2008). Cleft children in this age group have an average of 1.38 more carious teeth than their non-cleft peers of the same age. This is certainly a clinically significant difference even although I have been unable to analyse the significance statistically. When compared to the NDIP, 2008 figures for the Greater Glasgow and Clyde area whose mean dmft for 5-year-olds was 2.07, the cleft study children had 1.17 more carious teeth.

The other studies discussed in the Literature Review section have either reported on older children aged above 6 years or have reported on a much wider age range. For example, Lages et al., (2004) reported the prevalence of caries in cleft subjects aged between 1 and 32 years. The authors divided the children into three groups 1-5 years, 6-12 years, and 13-18 years. Hewson et al., (2001) reported separate mean dmft for the primary teeth and DMFT for the permanent teeth, encompassing a potential age range of between around 6 months and around 11 years. These have too wide an age range to be compared to this study, so these studies and others with similar difficulties have not been included in the discussion section.

When looking at the individual components of the dmft score in this study, the mean dt score comprised the majority of the dmft score, followed in turn by the mt and ft elements. In other words, the subjects had more untreated decay and fewer teeth which had been treated by either extraction or restoration. For those teeth which had undergone treatment, more had been extracted than restored.

6.7 Cleft type and prevalence of caries

Subjects with BCLP had the highest mean dmft figure of 2.77. This was followed by CP (1.16), UCLP (0.81) and UCL (0.57).

Our sample sizes within each cleft type are too small to allow valid statistical analysis of these figures but generally these results show that subjects with more severe cleft types have a greater prevalence of caries than those with less extensive or severe clefts.

6.8 The proportion of caries present in anterior and posterior teeth

We were interested to see whether, as has been reported in previous studies, the caries in our study subjects was mainly on the anterior teeth around the cleft site, or whether it was distributed evenly between the anterior and posterior teeth.

Bokhout *et* al., (1996) reported that half the caries present in the 2.5-year-old cleft subjects affected the maxillary incisors. In his later (Bokhout *et* al., 1997) study reporting cleft subjects who were followed from birth to 4 years of age, the author reported that caries occurred most commonly in the teeth adjacent to the cleft. The study by Ahluwalia *et al.*, (2004) reported higher plaque scores, larger numbers of caries associated microorganisms and longer oral clearance times for cleft subjects compared to their controls.

Some authors (Johnsen & Dixon, 1984; Wong & King, 1998; Cheng *et* al., 2007) have suggested that crowding and rotation of teeth and scarring from surgery in the cleft area may make access for brushing more difficult and result in a higher prevalence of caries in the anterior teeth. Cheng *et* al., (2007) postulated that a lack of parental dietary counselling may also contribute to a greater caries risk.

With regard to both tooth and surface detail, the 1.5 – 2.49 and 2.5 – 3.49 age groups both presented with more caries in the anterior teeth. In the 3.5 – 4.49 age group the caries was distributed evenly between anterior and posterior teeth. By the 4.5 – 6.0 age group, there was more caries in the posterior teeth.

Unfortunately these results do not allow us to conclude that caries in the study subjects occurs more frequently near the cleft area. Perhaps a more plausible explanation involves eruption dates. It may be that the anterior teeth are more carious in the younger age groups because they have been erupted for a longer time and that by the 3.5 – 4.49 age group the posterior teeth have been exposed to the oral environment for long enough for caries to develop.

6.9 Mean dmft where dmft is greater than zero

In the youngest two age groups all the subjects were caries free and so this section was not applicable to them.

In 4.5 – 6.0 year age group the mean dmft for those subjects who did have caries was higher than the NDIP, (2008) results. The means were 5.16 in our study and 4.39 in NDIP. This mirrors the greater prevalence of caries that was found in the study population compared to the national population in Section 1.7. Although 0.77 more teeth with caries is a clinically significant difference, again as the NDIP standard deviation figures were unavailable, I have been unable to confirm this significance statistically.

The mean for the 2.5 – 3.49 age group was the same as the NDIP, 2008 results at 4.0. Only the mean for 3.5 – 4.49 age group was lower than the NDIP, 2008 results at 2.9 compared to 4.0.

6.10 Care Index

The Care Index is a measure of how much restorative treatment has been carried out. It is calculated, as previously mentioned, by the following formula:

$$ft \div dmft \times 100$$

It should perhaps, more accurately, be called the “Restorative Care Index” as it takes no account of the mt component of the dmft score (extractions due to caries being deemed a “failed treatment”).

The Care Index was not applicable for the youngest age group, as there was no caries present in this group.

For the 1.5 – 2.49 and 2.5 – 3.49 groups, the Care Index was zero. None of the subjects with caries had any of the affected surfaces either extracted or restored.

The 3.5 – 4.49 and 4.5 – 6.0 groups had a Care Index respectively of 10.5% and 10.9%, which is similar to that of the same age groups reported by NDIP, (2008) of 9.68%. This would be expected as the cleft children are, in the main, treated by dentists in general practice.

Of the ten subjects with caries in the 3.5 – 4.49 year age group, only one had been treated by extraction and two had restorations placed.

The 4.5 – 6.0 year age group had ten subjects who had a missing teeth component to their dmft score. This ranged from an mt score of only one up to fifteen. In comparison the ft component was present in ten subjects, but with a much lower range of only 1 to 4. The subjects with high dmfs/t scores were more likely to have a high ms and mt score. In other words, it was more likely that caries, if severe in extent, would be treated by extraction rather than restoration.

6.11 Percentage of subjects with tooth wear

There is only one paper with which to compare the tooth wear results of this study. Chapple & Nunn, (2001) reported that all of the 19 cleft subjects in their study aged 4 years had erosion of their primary molars and incisors. However, no detail is given as to what diagnostic criteria were used, so although their result of 100% is substantially higher than the 63.3% in this study, the results may not be directly comparable.

Of the national studies reporting tooth wear in children, most based their criteria on those used in the survey of children's dental health in 1993 (O'Brien, 1994). One, Harding *et al.*, 2003, used a modified version of that used by O'Brien.

6.11.1 0.5 – 1.49 year age group

There are no papers which quote figures for the prevalence of tooth wear in this age group. The study results will be used as a baseline for future comparisons in the cleft population of this age in the West of Scotland.

6.11.2 1.5 – 2.49 year age group

36% of the cleft subjects in this age group showed signs of tooth wear, with 8.9% exhibiting wear into dentine. This figure for wear into dentine is higher than that quoted by the National Diet and Nutrition Survey (Hinds & Gregory, 1995) in which study the figure for palatal erosion into dentine was 3% ($p = 0.05$).

6.11.3 2.5 – 3.49 year age group

Almost half of the subjects in this age group (45.7%) were diagnosed with tooth wear, with 8.3% showing wear into dentine. Again, this figure is higher than that quoted in the National Diet and Nutrition Survey (Hinds & Gregory, 1995) where palatal erosion into dentine occurred in 6% of the study's 2.5 – 3.5 yr olds. Although higher, the difference between the two studies is less than in the 1.5 – 2.49 age group and did not reach significance.

6.11.4 3.5 – 4.49 year age group

The percentage of subjects in this age group showing any signs of tooth wear increased to 63.3%, considerably higher than that reported by Jones & Nunn, 1995 of 39% of 3-year-old children in their study in East Cumbria. 32.3 % of the subjects in this study demonstrated wear into dentine, almost double the figure reported by the National Diet and Nutrition Survey (Hinds & Gregory, 1995) of 13% for 3.5 – 4.5 year olds. This difference was statistically significant ($p = 0.006$).

6.11.5 4.5 – 6.0 year age group

80% of subjects in this age group showed signs of tooth wear, of whom 27.5% had wear into dentine. These figures are higher than those quoted by O'Brien, (1994), Hinds & Gregory, (1995) and Walker, (2000). The percentage of wear into dentine in this age group was lower than in the 3.5 – 4.49 age group. The difference may be due to exfoliation of the primary incisors which occurs at around this time.

O'Brien, (1994) reported that 52% of 5-year-old children in the UK had palatal erosion of their primary incisors and 18% had buccal surface wear. Of the wear on the palatal surfaces, 24% involved the pulp of the tooth. Taking the results for Scotland alone, this jumped to 34% of 5-year-olds with pulpal involvement of the palatal surfaces of the primary incisor teeth. Hinds & Gregory, (1995) reported that 65% of the 4 – 6 year olds had experienced erosion. The Scottish figures within this study were a prevalence of tooth wear of 71% in the 4 – 6 year age group. Unfortunately, there was no detail on the Scottish figures for erosion into dentine, which was quoted in the two previous studies. Walker, (2000) reported 19% had exposed dentine or pulpal involvement.

6.12 Deprivation and the presence of a cleft

The SIMD measure of deprivation divides the population into five equal "quintiles", and as such, one would expect 20% of the population to be represented in each quintile. In this study there is a linear trend of increasing proportions of subjects within each category from the least deprived (13.9%) to the most deprived quintile (31.1%) and there are less than the expected 20% within SIMD category 1 (least deprived) whilst there are more than the expected 20% in SIMD category 5 (most deprived). It would seem an obvious assumption that this reflects an association between deprivation and the presence of a cleft. However as there are well documented large pockets of severe deprivation in the study area, the results may only reflect the higher levels of deprivation within the Greater Glasgow area.

Figures for the proportion of children in the Greater Glasgow and Clyde area within each SIMD quintile were available for 5-year-olds and when comparing these to the study population, the figures show a broadly similar distribution. Approximately 60% of both the study and general population of 5-year-olds are present in the two most deprived SIMD quintiles. This makes it more likely that the results are due to the higher levels of deprivation found in the Greater Glasgow and Clyde area and not to an association between deprivation and the presence of a cleft in the West of Scotland.

Chapter 7 – Principal Findings

7.1 1. The percentage of subjects with no obvious caries

Age Group in Years	Percentage with no obvious decay
0.5 - 1.49	100
1.5 - 2.49	91.1
2.5 - 3.49	74.3
3.5 - 4.49	67.7
4.5 - 6.0	37.2

7.2 2. The mean dmfs by age group

Age Group in Years	Mean dmfs
0.5 - 1.49	0
1.5 - 2.49	2.02
2.5 - 3.49	2.58
3.5 - 4.49	1.61
4.5 - 6.0	8.67

7.3 3. The mean dt, mt, ft and dmft by age group

	Means			
Age Group in Years	dt	mt	ft	dmft
0.5 - 1.49	0	0	0	0
1.5 - 2.49	0.49	0	0	0.49
2.5 - 3.49	1	0	0	1.03
3.5 - 4.49	0.77	0.06	0.09	0.93
4.5 - 6.0	1.57	1.25	0.35	3.24

7.4 4. The relative proportion of caries in anterior and posterior teeth

Age Group in Years	% caries in anterior teeth (dmft)	% caries in posterior teeth (dmft)
0.5 - 1.49	n/a	n/a
1.5 - 2.49	66.7	33.3
2.5 - 3.49	69.4	30.6
3.5 - 4.49	44.4	55.6
4.5 - 6.0	41.8	58.2

Age Group in Years	% caries in anterior teeth (dmfs)	% caries in posterior teeth (dmfs)
0.5 - 1.49	n/a	n/a
1.5 - 2.49	53.5	46.5
2.5 - 3.49	74.2	25.8
3.5 - 4.49	48.8	51.2
4.5 - 6.0	30.6	69.4

7.5 5. The mean dmft for those children with dmft > 0 in each age group

Age Group in Years	Mean dmft where dmft>0
0.5 - 1.49	n/a
1.5 - 2.49	5.5
2.5 - 3.49	4.0
3.5 - 4.49	2.9
4.5 - 6.0	5.2

7.6 6. Whether cleft children have more caries than their non-cleft peers

There was no difference between the mean dmft scores for the cleft subjects and their non-cleft peers in the age groups below 4.5 years of age.

The 4.5 – 6.0 year age group had more caries than their non-cleft peers (mean dmft respectively of 3.24 and 1.86).

7.7 7. The percentage of children with tooth wear extending into dentine on the buccal or palatal surfaces of the primary upper incisor teeth in each age group

Age Group in Years	Percentage with wear into Dentine
0.5 - 1.49	2.2
1.5 - 2.49	8.9
2.5 - 3.49	8.3
3.5 - 4.49	32.3
4.5 - .0	27.5

7.8 8. Is there any correlation between deprivation and the presence of a cleft?

	Number	**Percentage**
SIMD		.
1	29	13.9
2	33	15.8
3	34	16.3
4	48	22.9
5	65	31.1

Chapter 8 – Recommendations

Young children with a cleft in the West of Scotland have a considerable burden of dental decay in addition to the surgical and potential orthodontic and speech therapy care during their childhood.

Dental decay can result in considerable pain, abscess, difficulty eating and sleeping and absence from school. In Scotland dental disease requiring tooth extraction is the most common reason for children to be admitted to hospital for general anaesthetic. However, dental caries is a preventable disease and parents, with the encouragement and support of dental professionals, can help to reduce the risk of their child developing tooth decay by brushing their child's teeth with the correct strength of fluoride toothpaste twice daily and by following a healthy, low sugar diet. Dental professionals can also assist by giving dietary advice, prescribing fluoride supplements, regularly applying fluoride varnish (a highly concentrated topical fluoride), and where these are insufficient to stop caries developing, by restoring the caries as early as possible.

All of the above are already provided at the Oral Orthopaedic Prevention Clinic at the Glasgow Dental Hospital and School however, as has been shown by this study, cleft children in the West of Scotland still have a higher prevalence of caries than their non-cleft peers.

In order to reduce this additional burden of dental disease, I would recommend:

- Targeting prevention at the 2.5 – 3.49 age group.
- Provision of fluoride tablets at appropriate doses for all children over the age of 3 years (Stephen & MacFadyen, 1977).
- Recruitment of a dedicated Dental Therapist to the service.
- Provision of a dedicated Dental Therapist treatment clinic incorporating additional education and advice regarding prevention of dental caries.

Targeting additional prevention to the age groups below which the greatest increase in caries incidence occurs, should greatly reduce the caries incidence.

A new Dental Therapy prevention and treatment clinic would help to improve the dental health of children with a cleft and, if not eliminate dental caries altogether (a laudable if difficult aim), would at least improve the restorative Care Index for our patients and reduce morbidity associated with dental decay.

Appendices

Appendix 1

Data collection form

Caries Diagnostic Codes and Criteria

- The examination is principally visual. Instruments or gauze may be used to clean a surface.
- Use of air to dry the tooth permitted.

Examination

Tooth	**Codes**
Code	Criteria
6	Extracted due to caries
7	Extracted for orthodontic reasons
U	Unerupted
T	Missing due to trauma

Primary molars are not recorded as code 6 after age 10

Surface codes

Code	Criteria
G	Sound
E	Enamel caries
1	Arrested dentine caries
2V	Dentine caries (uncavitated)
2C	Dentine caries (cavitated)
3	Unrestorable caries (pulpal)
4	Filled and decay (2v, 2c, 3)
F	Filled and sound
R	Filled needs replacing but not carious
$	Sealed surface
N	Obvious sealant restoration
C	Crown/advanced restoration
9	Excluded
T	Unrestored fracture
X	Restored fracture
Y	Restored fracture needs replacing

Basic Periodontal Examination

For teeth 16,11,26,36,31,46

0 healthy
1 bleeding on probing
2 plaque retention factor
3 shallow pocket
4 deep pocket
* furcation / recession

False pockets in children <11 years

Toothwear

On the buccal and lingual surfaces of primary and permanent maxillary incisor teeth will be assessed for loss of surface enamel characteristics, and / or exposure of dentine or pulp.

Assess the **depth** and **area** of loss of tooth tissue for each surface using the following criteria:

Depth

Code 0	Normal
Code 1	Enamel only - loss of surface characterisation
Code 2	Enamel and dentine - loss of enamel, exposing dentine
Code 3	Enamel into pulp - loss of enamel and dentine resulting in pulpal exposure
Code 9	Excluded

Area

For affected surfaces

Code 0	Normal
Code 1	Less the 1/3 of surface involved
Code 2	1/3 to 2/3 of surface involved
Code 3	Greater than 2/3 of surface involved
Code 9	Excluded

Developmental Defects of Enamel

SCOTS is a modification of DDE which includes symmetry and the subjects assessment.

How it works:

Teeth examined wet
4 upper incisors examined (For CleftSiS purposes all teeth will be coded but only upper incisors for extent and symmetry)

The components are:

Subject
Examiner asks the child "Do you think you have any marks on your upper front teeth which won't brush off?"

- Record yes or no

If "yes" ask the following:

- I never think about them
- I think about them but they don't bother me
- I think they might bother me when I get older
- They bother me from time to time
- They bother me a little
- They bother me a lot
- I get very upset by them

Examination

Opacities present

0 Normal
1 Demarcated opacity
2 Diffuse opacity
3 Hypoplasia
4 Demarcated & diffuse (1 & 2)
5 Demarcated & hypoplastic (1 & 3)
6 Diffuse & hypoplastic (2 & 3)
7 Other eg tetracycline staining
8 Demarcated, diffuse and hypoplastic (1 & 2 & 3)
9 Excluded eg missing

All the teeth.

Minimum extent of worse opacity

< 1/3	less than 1/3 of tooth surface area affected
>1/3<2/3	Between 1/3 and 2/3 " of tooth surface area affected
>2/3	more than 2/3 of tooth surface area affected

Symmetry of opacities

0 Defects not symmetrical
1 One type of defect symmetrical
2 More than one type of defect symmetrical

Only the perm. incisors.
21|12

Appendix 2

Research and Development Approval Letter

North Glasgow University Hospitals NHS Trust

NHS
Greater Glasgow

West Research Office
Administration Building
Ground Floor Room 9
Western Infirmary
Glasgow G11 6NT
Tel: 0141 211 6281

Our Ref JG/LR
Enquiries to Judith Godden
Direct Line 0141 211 1817
e-mail: *Judith.Godden@northglasgow.scot.nhs.uk*

19th Jan 2007

Professor Richard Welbury
University of Glasgow Dental School
378 Sauchiehall St
Glasgow
G2 3JZ

Dear Professor Welbury

REC Ref: 06/S0709/143 **R&D Ref:** WN06DN008
Title: An investigation into the incidence of Caries and Tooth Wear in Cleft children between the ages of 6 months and 6 years in the West of Scotland

We are pleased to inform you that, based on the information provided, this project has been granted overall Management Approval and may now proceed. This includes Finance, Pharmacy and a favourable Research Ethics Committee opinion.

Under Research Governance, we are required to hold a sponsor file containing the following documents: Protocol, Amendments and Ethics approval. While the study is ongoing you are responsible for updating us with all study amendments.

Further management approval will be required for amendments that increase patient numbers, increase or change the test procedures or bring about a change in pharmacy requirements. Please contact the R&D office if you wish to discuss any future amendments.

Thank you for your current and future collaboration.

Yours sincerely

Dr Judith Godden
Academic Research Co-ordinator

Appendix 3

West of Scotland Ethics Committee Approval letter

North Glasgow University Hospitals
Division

NHS
Greater
Glasgow

West Glasgow Ethics Committee 2
Western Infirmary
Dumbarton Road
Glasgow
G11 6NT

Telephone: 0141 211 6238
Facsimile: 0141 211 1920

19 December 2006

Prof Richard Welbury
Professor of Paediatric Dentistry
University of Glasgow Dental School
378 Sauchiehall Street
Glasgow G2 3JZ

Dear Prof Welbury

Full title of study:	**An investigation into the incidence of Caries and Tooth Wear in Cleft children between the ages of 6 months and 6 years in the West of Scotland.**
REC reference number:	**06/S0709/143**

The Research Ethics Committee reviewed the above application at the meeting held on 19 December 2006. Thank you for attending to discuss the study.

Ethical opinion

The committee agreed that there were no ethical issues with this study.

The members of the Committee present gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation.

Ethical review of research sites

The Committee agreed that all sites in this study should be exempt from site-specific assessment (SSA). There is no need to complete Part C of the application form or to inform Local Research Ethics Committees (LRECs) about the research. The favourable opinion for the study applies to all sites involved in the research.

Conditions of approval

The favourable opinion is given provided that you comply with the conditions set out in the attached document. You are advised to study the conditions carefully.

Approved documents

The documents reviewed and approved at the meeting were:

Document	*Version*	*Date*
Application	5.2	29 November 2006
Investigator CV		29 November 2006
Protocol		29 November 2006
Covering Letter		29 November 2006
Compensation Arrangements		29 November 2006

Research governance approval

You should arrange for the R&D Department at all relevant NHS care organisations to be notified that the research will be taking place, and provide a copy of the REC application, the protocol and this letter.

All researchers and research collaborators who will be participating in the research at a NHS site must obtain final research governance approval before commencing any research procedures. Where a substantive contract is not held with the care organisation, it may be necessary for an honorary contract to be issued before approval for the research can be given.

Membership of the Committee

The members of the Ethics Committee who were present at the meeting are listed on the attached sheet.

Dr M T Hosey declared an interest

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

06/S0709/143 **Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project

Yours sincerely

Andrea H Torrie
Ethics Manager – West Glasgow LRECs

Email: andrea.torrie@northglasgow.scot.nhs.uk

Enclosures: *List of names and professions of members who were present at the meeting and those who submitted written comments*
Standard approval conditions SL-AC2

Copy to: *Dr Judith Godden*
NHS Greater Glasgow and Clyde Primary Care Division
West Research Office, Ground Floor, Room 9,
Western Infirmary

West Glasgow Ethics Committee 2

Attendance at Committee meeting on 19 December 2006

Committee Members:

Name	*Profession*	*Present?*	*Notes*
Dr Sue Langridge	***Chair***		
Dr R Lindsay	***(until 9.55 a.m.)***		
Dr R Soutar			
Rev R Currie			
Sister C Donald			
Mr J McHugh			
Dr M T Hosey			
Dr A Crighton			
Dr A Shaukat			
Mrs A H Torrie			

Also in attendance:

Name	*Position (or reason for attending)*
Ms S Jenner	

Appendix 4

Calibration Information

ID	dmftx_BRIT	dmftx_WELB	dmftx_MACD	dmftx_BLAIR	dmftx_MED
Desson	1	1	1	1	1
Dolan	0	0	0	1	0
Flannigan	0	0	0	0	0
Johnson	1	1	1	1	1
Mckay	1	1	1	1	1
Moffat	0	0	0	0	0
N = 6					

ID	dt_BRIT	dt_WELB	dt_MACD	dt_BLAIR	dt_MED
Desson	4	4	5	4	4
Dolan	0	0	0	0	0
Flannigan	0	0	0	0	0
Johnson	2	2	2	3	2
Mckay	2	1	1	1	1
Moffat	0	0	0	0	0
N = 6					

ID	mt_BRIT	mt_WELB	mt_MACD	mt_BLAIR	mt_MED
Desson	0	0	0	0	0
Dolan	0	0	0	0	0
Flannigan	0	0	0	0	0
Johnson	0	0	0	0	0
Mckay	0	0	0	0	0
Moffat	0	0	0	0	0
N = 6					

ID	ft_BRIT	ft_WELB	ft_MACD	ft_BLAIR	ft_MED
Desson	0	0	0	0	0
Dolan	0	0	0	1	0
Flannigan	0	0	0	0	0
Johnson	0	0	0	0	0
Mckay	0	0	0	0	0
Moffat	0	0	0	0	0
N = 6					

Table of dmftx_BRIT by dmftx_MED			
dmftx_BRIT	**dmftx_MED**		
Frequency	**0**	**>=1**	**Total**
0	3	0	3
>=1	0	3	3
Total	3	3	6

McNemar's Test	
Statistic (S)	.
DF	1
Pr > S	.
NOTE: There are no discordant pairs.	

Simple Kappa Coefficient	
Kappa	1.0000
ASE	0.0000
95% Lower Conf Limit	1.0000
95% Upper Conf Limit	1.0000

Table of dmftx_WELB by dmftx_MED			
dmftx_WELB	**dmftx_MED**		
Frequency	**0**	**>=1**	**Total**
0	3	0	3
>=1	0	3	3
Total	3	3	6

McNemar's Test	
Statistic (S)	.
DF	1
Pr > S	.
NOTE: There are no discordant pairs.	

Simple Kappa Coefficient	
Kappa	1.0000
ASE	0.0000
95% Lower Conf Limit	1.0000
95% Upper Conf Limit	1.0000

Table of dmftx_MACD by dmftx_MED			
dmftx_MACD	**dmftx_MED**		
Frequency	**0**	**>=1**	**Total**
0	3	0	3
>=1	0	3	3
Total	3	3	6

McNemar's Test	
Statistic (S)	.
DF	1
Pr > S	.
NOTE: There are no discordant pairs.	

Simple Kappa Coefficient	
Kappa	1.0000
ASE	0.0000
95% Lower Conf Limit	1.0000
95% Upper Conf Limit	1.0000

Table of dmftx_BLAIR by dmftx_MED			
dmftx_BLAIR	**dmftx_MED**		
Frequency	**0**	**>=1**	**Total**
0	2	0	2
>=1	1	3	4
Total	3	3	6

McNemar's Test	
Statistic (S)	1.0000
DF	1
Pr > S	0.3173

Simple Kappa Coefficient	
Kappa	0.6667
ASE	0.2869
95% Lower Conf Limit	0.1044
95% Upper Conf Limit	1.0000

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