

An Investigation of The Impact of Amelogenesis Imperfecta (AI) on Children and Adolescents.

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Abstract

Background: *Amelogenesis Imperfecta (AI)* is an inherited dental condition affecting enamel, which can result in significant tooth discolouration and enamel breakdown, requiring lifelong dental care. The possible impact of this condition on children and young adults is not known.

Aims and Objectives: The aim of the study was to explore the impact of AI on children and young adults through in-depth interviewing and subsequent Framework Analysis. The information derived from this was then used to construct a questionnaire.

Methods: This research comprised of two parts, combining qualitative and quantitative methodology, in order to develop a questionnaire to distribute to a large cohort of AI patients. The first part involved semi-structured in-depth interviews with 7 AI patients and common themes and concepts were then identified using Framework Analysis. The second part of the study was the development of a questionnaire based on the themes and subthemes identified from part one of the research. This questionnaire was then distributed to 61 AI patients mixed between three cohorts of AI patients: pre, mid, and post-treatment.

Results: Children and adolescents with AI exhibited concerns regarding the aesthetics and function of their dentition. Patients also expressed a high level of concern regarding comments by other people and self consciousness associated with this. A small number of AI patients highlighted the effect of their dental treatment and health on their personal life.

Conclusion: The results indicate that there are marked impacts on children and young adults as a result of AI. These include aesthetics, function, and psycho-social aspects.

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DDent in Paediatric Dentistry

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List of abbreviations

ADAI	Autosomal Dominant Amelogenesis Imperfecta
AI	Amelogenesis Imperfecta
ARAI	Autosomal Recessive Amelogenesis Imperfecta
COHQOL	Child Oral Health Quality Of Life
CPQ	Child Perception Questionnaire
DD	Dentine Dysplasia
DDE	Developmental Defects of Enamel
DI	Dentinogenesis Imperfecta
EDH	Eastman Dental Hospital
IEE	Inner Enamel Epithelium
MIH	<i>Molar Incisor Hypomineralisation</i>
NatCen	National Centre for Social Research
OEE	Outer Enamel Epithelium
OHQOL	Oral Health Quality Of Life
OI	Osteogenesis Imperfecta
SSC	Stainless Steel Crown
TFI	<i>Thylstrup-Fejerskov Index</i>
UCLH	University College London Hospital

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1. REVIEW OF THE LITERATURE

Teeth are important in many ways; for function during mastication, since they are the first to initiate the process of digestion; to aid speech in conjunction with tongue, and as a component of facial appearance and aesthetics (Van der Geld *et al.*, 2007).

1.1 Dental Anomalies

Teeth are considered one of the specialized components of the craniofacial skeleton (Simmer, 2007). There are three main mineralized tissues that make up the tooth structure: enamel, dentine, and cementum, and each of these tissues are prone to development defects. Dental anomalies are “an aberration in which one or more teeth deviates from the normal in number, form, function, or position” (Mosby, 2007). Dental anomalies can range from missing, to discoloured or deformed teeth. Many are expressions of other, more complex disorders and dental anomalies may be caused by inherited genetic defects, result from spontaneous genetic mutations, or environmental factors (Nieminen, 2009).

1.1.1 Enamel formation

Enamel is the hardest tissue in the human body. It forms the outer layer of tooth structure and it consists of 96% with respect to weight of inorganic material, mainly calcium phosphate and well organised hydroxyapatite crystals, and 4% with respect to weight of organic material and water, and small quantities of sodium, chloride, and magnesium (Reyes-Gasga *and* Gutierrez, 2003).

Before studying any dental anomalies, it is important to understand the mechanisms and the stages of tooth development. The initiation of tooth formation starts around the 37th day of gestation (Singh, 2007). A thickening of the stratified squamous epithelium, also called the oral ectoderm, gives rise to the dental lamina which is the foundation for the tooth germ.

The tooth germ aggregates to form the tooth bud which will continue to proliferate and grow into the underlying mesenchyme. The deep surface of the bud bends into a dome to produce a cap shape. This marks two important steps, histodifferentiation and morphodifferentiation (Crawford *et al.*, 2007). At this stage the tooth germ consists of:

- a) Enamel organ (derived from ectoderm), this will later form the enamel.
- b) A dental papilla and the dental sac (from the mesenchyme) which will form the dentine, pulp, cementum, and periodontal ligament.

There are three steps required for normal enamel formation:

- 1- Enamel matrix formation,
- 2- Enamel matrix mineralization, and
- 3- Enamel maturation. (Nanci *et al.*, 2003)

Enamel formation starts from the process of histodifferentiation of the enamel organ which will produce four layers:

- a) The outer enamel epithelium (OEE) provides a protective barrier during enamel production
- b) The inner enamel epithelium (IEE) forms the enamel secreting cells, the ameloblasts.
- c) The stellate reticulum is found between the OEE and IEE.
- d) The stratum intermedium found between IEE and stellate reticulum (Nanci *et al.*, 2003).

Both the stellate reticulum and stratum intermedium have an important role in supporting the production of enamel by synthesizing glycosaminoglycans which will draw water into the enamel organ. The cells of the IEE differentiate into pre-ameloblasts and induce the dental papilla to differentiate into odontoblasts, which results in the start of dentinogenesis. The odontoblasts move to the centre of the tooth, leaving behind formed dentine. This induces the ameloblasts to move outward, leaving behind the enamel matrix. This is also known as the appositional stage (Nanci *et al.*, 2003).

The mineralization stage of the enamel matrix involves a two-step process. First the ameloblasts secrete an organic matrix that is mineralized to about 30% by weight. The rough endoplasmic reticulum in the ameloblast cell releases enamel proteins which is then partially mineralised by an enzyme called alkaline phosphatase. When the full thickness of enamel has been secreted by the ameloblasts, a continuous increase in mineral content begins. Water and proteins from the enamel matrix will be removed by the smooth-ended ameloblasts, and the transport of calcium and phosphate into the matrix will be achieved by ruffle-ended ameloblasts. This is also known as the calcification stage. The cells of the stratum intermedium also assist in the maturation stage.

The final stage is Maturation, which marks the end of calcification, of the enamel and dentine, just before the tooth is ready to erupt (Nanci, 2003).

The ameloblasts control the process of forming this organised structure, which involves a number of organic molecules (i.e. genes/proteins) including enamelin, amelogenin, ameloblastin, tuftelin, amelotin, dentine sialophosphoprotein, and enzymes such as kallikrein 4 and matrix metalloproteinase 20 (Crawford *et al.*, 2007). A mutation in the above can result in defective enamel, such as Autosomal Dominant Amelogenesis Imperfecta (ADAI) and Autosomal Recessive Amelogenesis Imperfecta (ARAI). Autosomal Dominant means the patient will be affected if only one parent has the defected gene, whereas in Autosomal Recessive, a patient needs to have two copies (from both parents) of the defected gene in order to be affected (Crawford *et al.*, 2007). The aetiology of AI will be discussed further in section 2.2.2.

1.1.2 Developmental disturbances affecting teeth

Developmental disturbances can affect the teeth in a number of different ways, and are categorized under five broad headings.

Disturbance in size:

Examples of this include microdontia; when one, or more, teeth are smaller than normal, or macrodontia; when one, or more teeth are larger in size than normal variation. The prevalence of microdontia and macrodontia in British school children is 2.5% and 1.1% respectively (Welbury, 2001).

Disturbance in number:

Hypodontia is the term used to describe the developmental absence of one or more primary or permanent teeth, excluding the third molars (Goodman *et al.*, 1994). It has a prevalence of 0.1-0.9% in the primary dentition and 3.5-6.5% in the permanent dentition (Brook, 1974). Supernumerary teeth are teeth in excess of the normal number. The most common site is the maxillary central incisors, followed by maxillary premolars (Sapp, 2004).

Disturbances in eruption:

Premature eruption of primary teeth, where they are present at birth, is termed natal teeth. If the primary teeth erupt during the first 30 days of life they are termed neonatal teeth. The lower primary mandibular central incisors are most commonly known to exhibit premature eruption (Massler *et al.*, 1950). In some cases the premature loss of primary teeth may lead to premature eruption of permanent teeth (Miyamoto *et al.*, 1976). Delayed eruption can be caused by local factors such as gingival fibromatosis or due to systemic conditions such as rickets, cleidocranial dysplasia, or cretinism (Sapp, 2004).

Disturbances in shape:

Abnormalities of tooth form have many varieties. Some of the examples found during childhood include; Gemination - this occurs as a result of incomplete division of the tooth germ. The tooth is single rooted with two crowns or one wide crown (Grover *et al.*, 1985). Fusion - occurs as a result of union between two separate tooth germs at any point between the crowns and the roots. It is differentiated from gemination by counting the teeth that are present. There will be one tooth missing from the arch segment if fusion exists (Soames *et al.*, 1997).

Dens Invaginatus; also known as dens in dente, is “a deep enamel-lined pit that extends for varying depths into the underlying dentine, often displacing the pulp chamber and sometimes altering the shape of the root” (Sapp, 2004). Hypercementosis is an increased deposition of cementum on the roots. It may be found in patients with Paget’s disease or hyperpituitarism. Roots tend to be bulbous and are often difficult to extract (Sapp, 2004). Taurodontism; which means “bull-like”, is a developmental defect most commonly seen in the permanent molars resulting in enlarged and elongated pulp chambers. It can be associated with Down syndrome, Amelogenesis Imperfecta, and hypodontia (Welbury, 2001).

Disturbances in tooth structure:

Developmental disturbance in tooth structure may be the result of defective formation of cementum, dentine, and enamel. These disturbances can be genetic / hereditary in origin, or acquired by systemic or environmental factors.

Cementum defects:

Hypophosphatasia is an autosomal recessive (AR) or autosomal dominant (AD) inherited condition affecting bone mineralization, due to a deficiency in serum alkaline phosphatase. It is usually detected in the neonatal and infantile periods. In the infantile form, the premature loss of primary teeth appears to be related to the absence of cementum (Mornet *et al.*, 2007).

Dentine defects:

The most common hereditary dentine disorders are Dentinogenesis Imperfecta (DI) and Dentine Dysplasia (DD) (Barron *et al.*, 2008). Both DI and DD are autosomal dominant conditions affecting the structure of the dentine in the primary and permanent teeth (Barron *et al.*, 2008). Shield's classification recognises three types of DI (I, II, and III) and two types of DD (I and II) (Shield, 1973).

DI type I is associated with Osteogenesis Imperfecta (OI) and affects both the primary and permanent teeth, although the permanent dentition is usually less affected. The colour ranges from bluish-grey to brown and yellow. Even though the enamel is normal, it can be easily chipped, exposing the underlying abnormal dentine, which results in attrition. On radiographic assessment the crowns have a bulbous shape and the roots are shorted thin. DI type I is a result of the mutation of one of two genes encoding type I collagen, COL1A1 AND COL1A2, affecting bone and connective tissues as well (Barron *et al.*, 2008).

DI type II is the most common type of hereditary dentine defects and is sometimes called hereditary opalescent dentine. It has an incidence of 1 in 8000 (Barron *et al.*, 2008) and has similar features to DI type I, but is not associated with OI.

DD type I (also called radicular DD) is the most common of the two types (prevalence of 1 in 100,000) and can affect both the primary and permanent dentitions. The colour, shape, form, and consistency of the teeth are usually unaffected. On radiographic assessment the roots look short, conical, or blunt with pulpal obliteration (Barron *et al.*, 2008).

DD type II (also called coronal DD) affects both dentitions with the primary teeth grey-bluish, brown, or yellow in colour. The roots are of normal shape and length with pulp stones found in the pulp chamber (Barron *et al.*, 2008).

Enamel defects:

Generalised developmental abnormalities of enamel may be attributed to genetics, systemic influences i.e. nutritional deficiencies or metabolic disorders, or may be idiopathic. They may also be caused by local factors such as trauma or infection (Welbury 2001).

Enamel hypoplasia is caused by deficient matrix (Webster, 2008) and can be local or generalized. An example of localised enamel hypoplasia is the Turner tooth which is caused by localized inflammation or trauma to the primary tooth during tooth development of the permanent successor. An example of generalized enamel hypoplasia is Hutchinson's incisors, resulting from congenital syphilis (Sapp, 1997).

Molar Incisor Hypomineralization (MIH) is defined as hypomineralization of systemic origin affecting one to four permanent first molars and frequently associated with affected incisors. The enamel can be soft, discoloured, and porous often leading to pain and sensitivity, with a prevalence ranging from 3.6 to 25% (Weerheijm *et al.*, 2004). Studies have suggested that if a child is unhealthy during the first 4 years of their life, they could be prone to MIH due to the disturbances during the enamel formation process, although the exact aetiology is still unknown (Beentjes *et al.*, 2002).

The commonest form of hereditary enamel defect is Amelogenesis Imperfecta.

1.2 Amelogenesis Imperfecta (AI)

Amelogenesis Imperfecta (AI) is an inherited dental condition affecting the structure and clinical appearance of the enamel of all, or nearly all, of the teeth in a more or less equal manner (Welbury, 2001). AI is a heterogeneous group of hereditary disorders of enamel formation and may be AD, AR, sex-linked, or sporadic (Crawford *et al.*, 2007).

1.2.1 Prevalence

There are varying reports regarding the prevalence of AI. One study in Michigan, USA showed a prevalence of one in 14000 (Witkop *et al.*, 1976). Another study in northern Sweden reported a prevalence of one in 718 (Backman *et al.*, 1986). In Turkey the reported prevalence was 43:10,000 (Atug-Atac *et al.*, 2007), and 10:10.000 in Argentina (Sedano *et al.*, 1975). Crawford *et al.* (2007) explained the wide variation in

prevalence rates were due to the different populations studied and the genetic differences for these populations.

1.2.2 Aetiology

It is thought that the changes in AI are due to a single gene mutation (Welbury, 2001). Each pattern of inheritance is associated with specific location on the inherited gene (Crawford, 2007).

Autosomal Dominant AI (ADAI) was thought to be linked to chromosome 4 (the enamelin gene ENAM), however, Mendoza *et al.* (2007) found a new locus AD on chromosome 8q24.3. Autosomal Recessive AI (ARAI) has been linked to consanguinity, with associated mutation of the matrix metalloproteinase 20 (MMP-20) and kallikrein 4 (KLK4) enzymes. It is seen more frequently in populations where intermarriage within the family is frequent (Crawford *et al.*, 2007). X-linked AI is thought to be associated with chromosome Xp22 which is the amelogenin gene AMLEX (Crawford *et al.*, 2007).

1.2.3 Classification

Any disturbances to any of the three steps required for enamel formation may result in a defective enamel structure. A decrease in the enamel matrix formation will result in a hypoplastic type of AI (deficiency in the amount of enamel). A disturbance in the mineralization phase results in a hypomineralized (hypocalcified) type AI (deficiency in the first phase of enamel mineralization) or hypomineralized (hypomature) type AI (deficiency in the second phase of enamel mineralization, which is the final growth and maturation of enamel) (Crawford *et al.*, 2007).

There are several classifications of AI in the literature (Alderred *et al.*, 2003; Hart *et al.*, 2002). Some were based on the phenotype only where the morphology and clinical structure are described; others considered the phenotype and the mode of inheritance (genotype). In 1945, Weinmann *et al.* introduced the first classification of AI (hypoplastic and hypocalcified) based on the phenotype. In 1956, Darling suggested five phenotypes based on clinical, radiographical, and histopathological findings. It was not until 1970, when Schulze based his classification of AI on phenotype and mode of inheritance, that the classification with three broad categories was developed: hypoplastic, hypocalcified, hypomaturational (Witkop and Rao, 1971). A fourth category was added to the Witkop classification, hypomaturational-hypoplasia with taurodontism (Winter and Brook, 1975).

In 1988, Witkop added 15 subtypes to Winter's four major categories, based on phenotype and mode of inheritance. A new classification, based on the molecular defect in addition to the phenotype and mode of inheritance, was introduced by Aldred and Crawford in 1995. Hart *et al.* (2002) proposed a molecular defect sub classification of the AMELX conditions. Finally, the most recent classification of AI in 2003 was based on mode of inheritance, phenotype, molecular defect, and biochemical result. It is recommended that it is better to classify AI according to the mode of inheritance when known rather than the phenotype (Aldred *et al.*, 2003).

The classification systems used for AI are shown in Table 1.1

Weinmann et al, 1945 [4]	Two types based on phenotype: hypoplastic and hypocalcified
Darling, 1956 [5]	<p>Five phenotypes based on clinical, microradiographic and histopathological findings.</p> <p>Hypoplastic: Group 1- generalised pitting. Group 2- vertical grooves. (now known to be x-linked AI) Group 3 – generalised hypoplasia.</p> <p>Hypocalcified: Type 4A – chalky, yellow, brown enamel. Type 4B – marked enamel discolouration and softness with post- eruptive loss of enamel. Type 5 – generalised or localised discolouration and chipping of enamel.</p>
Witkop, 1957 [6]	<p>Classification based primarily on phenotype. 5 types:</p> <ol style="list-style-type: none"> 1- Hypoplastic 2- Hypocalcification 3- Hypomaturation. 4- Pigmented hypomaturation 5- Local hypoplasia. <p>Added mode of inheritances as further means of delineating cases.</p>
Schulze, 1970[7]	Classification based primarily on phenotype and mode of inheritance.
Witkop and Rao, 1971[7]	<p>Classification based on phenotype and mode of inheritance. Three broad categories: hypoplastic, hypocalcified, hypomaturation,</p> <ul style="list-style-type: none"> • Hypoplastic. <ul style="list-style-type: none"> Autosomal dominant smooth hypoplastic –hypomaturation with taurodontism (subdivided into a and b according to author) Autosomal dominant smooth hypoplastic with eruption defect and resorption of teeth. Autosomal dominant rough hypoplastic. Autosomal dominant pitted hypoplastic. Autosomal local hypoplastic X- Linked dominant rough hypoplastic. • Hypocalcified

<p style="text-align: center;">Autosomal dominant hypocalcified.</p> <ul style="list-style-type: none"> • Hypomaturation <p style="text-align: center;">X- Linked recessive hypomaturation.</p> <p style="text-align: center;">Autosomal recessive pigmented hypomaturation.</p> <p style="text-align: center;">Autosomal dominant snow- capped teeth.</p> <p style="text-align: center;">White hypomature spots</p>
<p>Winter and Brook, 1975[9] Classification based primarily on phenotype. Four main categories: hypoplasia, hypocalcification, hypomaturation, hypomaturation-hypoplasia with taurodontism, with mode of inheritance as a secondary means of sub-classification.</p> <ul style="list-style-type: none"> • Hypoplasia <ul style="list-style-type: none"> Type I. Autosomal dominant thin and smooth hypoplasia with eruption defect and resorption of teeth. Type II. Autosomal dominant thin and rough hypoplasia Type III. Autosomal dominant randomly pitted hypoplasia. Type IV. Autosomal dominant localised hypoplasia. Type V. X- linked dominant rough hypoplasia. • Hypocalcification <ul style="list-style-type: none"> Autosomal dominant hypocalcification • Hypomaturation <ul style="list-style-type: none"> Type I. X- linked recessive hypomaturation Type II Autosomal recessive pigmented hypomaturation. Type III Snow-capped teeth. • Hypomaturation- hypoplasia with taurodontism. <ul style="list-style-type: none"> Type I. Autosomal dominant smooth hypomaturation with occasional hypoplastic pits and taurodontism. Type II. Autosomal dominant smooth hypomaturation with thin hypoplasia and taurodontism.
<p>Witkop and Sauk, 1976[2] Classification based on phenotype and mode of inheritance, similar to classification of Witkop and Rao(1971).</p>
<p>Sundell and Koch, 1985[10] Classification based on phenotype.</p>
<p>Witkop, 1988[11] Four major categories primarily on phenotype (hypoplastic, hypomaturation, hypomaturation-hypoplastic with taurodontism) subdivided into in to 15 subtypes by phenotype and secondarily by mode of inheritance.</p> <ul style="list-style-type: none"> Type I. Hypoplastic <ul style="list-style-type: none"> Type IA. Hypoplastic, pitted autosomal dominant. Type IB. Hypoplastic, local autosomal dominant. Type IC. Hypoplastic, local autosomal recessive. Type ID. Hypoplastic, smooth autosomal dominant. Type IE. Hypoplastic, smooth X-linked dominant. Type IF. Hypoplastic, rough autosomal dominant. Type IG. Enamel agenesis, autosomal recessive. Type II. Hypomaturation. <ul style="list-style-type: none"> Type IIA. Hypomaturation, pigmented autosomal recessive. Type IIB. Hypomaturation, X-linked recessive. Type IIC. Hypomaturation, snow-capped teeth, X-linked.

	<p>Type IID. Hypomaturation, snow-capped teeth, autosomal dominant.</p> <p>Type IIIA. Autosomal dominant.</p> <p>Type IIIB. Autosomal recessive.</p> <p>Type IV. Hypomaturation-hypoplastic with taurodontism.</p> <p>Type IVA. Hypomaturation-hypoplastic with taurodontism, autosomal dominant.</p> <p>Type IVB. Hypoplastic-hypomaturation with taurodontism, autosomal dominant.</p>
Aldred and Crawford, 1995 [12]	<p>Classification based on:</p> <p>Molecular defect (when known)</p> <p>Biochemical result (when known)</p> <p>Mode of inheritance</p> <p>Phenotype.</p>
Hart et al, 2002[13]	<p>Proposed a molecular defect sub classification of the AMELX</p> <p>Conditions:</p> <ol style="list-style-type: none"> 1.1 Genomic DNA sequence. 1.2 cDNA sequence. 1.3 Amino acid sequence. 1.4 Nucleotide and amino-acid sequences. 1.5 AMELX mutations described to date.
Aldred et al, 2003[1]	<p>Classification based on :</p> <p>Mode of inheritance.</p> <p>Phenotype- Clinical and Radiographic.</p> <p>Molecular defect (when known)</p> <p>Biochemical result (when known)</p>

Table 1.1 Classification systems applied to Amelogenesis Imperfecta
(From Crawford *et al. Orphanet Journal of Rare Diseases*, 2007. Reproduced with permission)

1.2.4 Clinical Features

The clinical appearance of AI can be remarkably different between types (Sapp, 2004). The colour of the affected teeth will range from normal to opaque white or yellow-brown in colour (Witkop *et al.*, 1976).

Hypoplastic AI is characterized by small crowns with thin enamel or enamel of normal thickness usually associated with pits and groves, due to deficiencies in the amount of enamel. (Coffield *et al.*, 2005). Examples of hypoplastic AI are shown in Figure 1.1(a-d).

ADAI mostly presents as the hypoplastic type, with thin smooth hard, or in some cases pitted, yellow to yellow-brown enamel with spacing between the teeth due to defect in the amount of enamel matrix produced . The enamel is hard and it is not subject to significant attrition. ARAI also usually presents as generalized hypoplastic pitted enamel (Hart *et al.*, 2002).

Hypocalcified AI occurs due to a defect in the initial crystallite formation. The enamel on the affected teeth is of normal thickness but can be easily removed and chipped because it is soft. The colour can vary between yellow and brown (Sapp, 2004), Figure 1.1(e and f).

Hypomature AI is characterized by poorly mineralized enamel of normal thickness due to a defect in the final growth and maturation of the enamel crystallites. The enamel is soft and can be easily chipped off and is white in colour (Coffield *et al.*, 2005). “Snow-capped” teeth are the mildest form of the hypomaturational type, affected teeth exhibit normal enamel with white opaque flecks on the incisal edges of the anterior teeth and the occlusal surfaces of the posterior teeth (Welbury, 2001) as in Figure 1.1 (g and h). Mutation of *KLK4* and *MMP-20* genes are associated with AR hypomaturational AI (Hart *et al.*, 2004).

In both hypocalcified and hypomature types of AI, sensitivity is a common problem due to the exposure of dentine as a result of the deficient or defective enamel (Witkop *et al.*, 1976).

The term dysmineralization was recommended to describe the probable spectrum of defects of enamel mineralization in AI (Crawford *et al.*, 2007). It is thought to have the same clinical features between affected members in the same family.

X-linked AI, affects males and females in a different way. Males show the trait fully and may have either hypoplastic or hypomatured enamel (Crawford *et al.*, 2007). In contrast, females may have vertical pitting and grooving of the enamel affecting the entire permanent dentition (Figure 1.1 a) due to the Lyonization effect. The enamel organ of females consists of ameloblasts that contain a genetically active X_m (maternal X) or X_p (paternal X) chromosome, One of the two copies of the X chromosome is inactivated and clones of pre-ameloblasts are randomly controlled by either normal or abnormal genes on the X-chromosome (Welbury, 2001). As the ameloblasts differentiate from the inner enamel epithelium, there are produced linear defects in enamel which are seen clinically as vertical stripes or bands of imperfect enamel (McLarty *et al.*, 1973).



Figure 1.1 Different clinical features of Amelogenesis Imperfecta. (a, b, c, d) showing various degrees of hypoplastic AI. (e and f) hypomineralised AI. (g and h) hypomature AI. (Crawford *et al.* *Orphanet Journal of Rare Diseases*, 2007. Reproduced with permission)

1.2.4.1 Associated dental features

Skeletal anterior open bite

Fifty percent of patients affected by AI have a skeletal anterior open bite where patients have difficulty biting together and biting into food (Aren *et al.*, 2003).

Patients who are homozygous i.e. have identical alleles of the ENAM mutation on chromosome 4, tend to have enamel pitting with an anterior open bite (Hart *et al.*, 2004).

Taurodontism is a condition found in molar teeth where the body of the tooth and pulp chamber are enlarged vertically at the expense of the roots (Seow, 1993). It appears to be another feature associated with AI, particularly where there is a mixture of hypomaturation and hypoplasia type AI.

Delayed eruption of the permanent teeth in patients with AI has been reported in the literature due to delayed dental maturity (Aren *et al.*, 2003).

Reduced crown size, due to tooth surface loss caused by attrition is another feature associated with AI (Collins *et al.*, 1999).

Dental hypersensitivity is one of the most common symptoms and has a great impact on patients' oral hygiene and daily life (Aren *et al.*, 2003). As a result, periodontal conditions such as calculus, gingivitis, gingival enlargement, and periodontitis can be found in AI patients (Poulsen *et al.*, 2008).

Other dental features associated with AI include; higher risk of caries, loss of vertical dimension, congenitally missing teeth, and root malformations (Gadhia *et al.*, 2012). Impacted teeth were also reported as a dental feature can be found in AI (Arkutu *et al.*, 2012).

1.2.4.2 Syndromes associated with AI

Nephrocalcinosis is a renal condition where there is an increase in the calcium content of the kidneys (Kirzioglu *et al.*, 2009). If left untreated it may lead to significant morbidity. There are a small number of cases described in the literature, mostly including siblings, where an association has been shown between AI and Nephrocalcinosis (MacGibbon,1972; Hall *et al.*, 1995). Hunter *et al.* (2007) recommended that all paediatric dentists should consider referring patients with hypoplastic AI for a renal ultrasound examination.

Cone rod dystrophy CRD is defined as "inherited retinal dystrophy that belong to the retinitis pigmentosa group, and more generally to pigmentary retinopathies" (Hamel, 2007). There are localized deposits in the cone rich macular area of the eye. Polok *et al.* (2008) suggested that there is a common aetiological factor, mutation of CNNM4 gene, between cone rod dystrophy CRD and AI. CNNM4 is considered an ion

transporter, especially for calcium and magnesium. A mutation to this gene may be implicated in defective enamel formation during the maturation stage.

1.2.5 Management of AI

The aims of the management of AI are to improve the aesthetics, function, and reduce sensitivity of the affected dentition. Some authors have suggested that infants born with enamel defects should be referred for early assessment before 1 year of age (Wright *et al.*, 2000). Treatment during childhood has been described as a temporary phase (Bouvier *et al.*, 1996). In the primary dentition, the primary molars can be protected by using preformed stainless steel crowns (SSC) and composite restorations can be used to restore anterior teeth. In some cases the use of inhalation sedation might be useful with anxious children (Sapir *et al.*, 2001). Other cases may require treatment under general anaesthesia due to the severity and complexity of the treatment required (Crawford *et al.*, 2007). Due to the sensitivity and pain encountered during tooth brushing, oral hygiene may be poor making it very difficult to provide restorations (Wright *et al.*, 2002).

In the permanent dentition, SSCs can be used to protect affected permanent molars from further attrition, with minimum tooth preparation needed by placing elastic separators between the molars and their adjacent teeth for one week to create a space for SSC. Composite veneers on anterior teeth have been recommended to be used especially with less hypomineralized teeth, where the enamel is reasonably hard and thin (Crawford *et al.*, 2007). Some authors advocated the use of bleaching to improve the aesthetics of affected teeth (Nathwani *et al.*, 2010). Microabrasion is a technique that uses a combination of erosion and abrasion to improve discolorations that are limited to the outer enamel layer, e.g. white to brown surface staining and idiopathic speckling (Welbury, 2001). It is the role of the paediatric dentist to deliver to the (adult) restorative dentist a patient who is motivated, with good oral care practices and with no treatment option compromised by previous treatment. (Crawford *et al.*, 2007).

When treating AI patients it is important to have a multidisciplinary team where possible. This may include input from a paediatric dentist, an orthodontist, a periodontist, a restorative dentist, and possibly a geneticist (Moretti *et al.*, 2007). From 6 years to late teens, interceptive measures from orthodontics regarding the extraction of the first adult molars with poor long term prognosis can help achieving excellent

outcomes (Arkutu *et al.*, 2012). The use of removable appliances (e.g. midlinescrew) for the correction of forms of malocclusion such as crossbites, with modification to incorporate aesthetic elements is recommended for orthodontic treatment of AI patients, in addition, AI patients with class III or II can benefit from growth modification by using functional appliances. (Arkutu *et al.*, 2012). As our knowledge of genetics increases, a discussion of likely risk and future inheritance with some of the affected families is important (Crawford *et al.*, 2007). Due to the discolouration, sensitivity and extensive treatment required, one should not underestimate the psychosocial impact on AI patients. This will be discussed further in the next section.

1.3 The psychosocial impact of dento-facial appearance

1.3.1 The psychosocial impact of facial appearance

The psychosocial impact of facial appearance can be seen in two main areas. The first is the response by others and society to appearance, as nowadays there is a greater emphasis on physical attractiveness. This can be seen in the media, where images of attractive faces are routinely used. The second effect is that on a person's own sense of well-being and self-esteem (Diener *et al.*, 1995). In a review of research into physical attractiveness, facial appearance was identified as being central to social interactions (Eagly *et al.*, 1991). Several studies have suggested that an attractive appearance can lead to positive self-concept and social well-being (Diener *et al.*, 1995; Kerosuo *et al.*, 1995). Albino *et al.* described appearance as 'the most consistent and compelling determinant of self and social perceptions and attributions, and it can have a significant impact on one's personal and social life' (Albino *et al.*, 1990).

There is a tendency to treat those with attractive faces in a positive way and, on the other hand, have negative attitudes and discriminatory behaviour towards those who may have a less attractive appearance (Langlois *et al.*, 2000). A person with, what is considered by society, an attractive facial appearance may be thought to be more socially skilled, intelligent, popular, and have a successful job, compared with less attractive individuals (Dion *et al.*, 1972). Less attractive subjects are often considered less trustworthy, intelligent, successful, well liked, and more aggressive and anti-social (Dion *et al.*, 1972; Baldwin *et al.*, 1980). People with facial disfigurements often complain of rejection and negative responses from others in society (Bull and Stevens, 1981; Rumsey *et al.*, 1982). A person's psychological well-being can be affected by perceptions and responses received from society.

1.3.1.1 The effect of appearance on social interactions

The psychosocial impact of facial appearance can be seen as early as infancy through the parent-child interaction. One study compared 144 mothers' attitudes and behaviours towards infants with different levels of attractiveness by using coloured photographs to rate the attractiveness on a 1 to 5 Likert-type scale, and then observing them feeding and playing with their baby in the hospital and at 3 months review. The study showed that mothers of attractive newborns were more affectionate and playful with their babies than were mothers of less attractive babies. The study also found that in the long term mothers of less attractive babies had a negative attitude, considering their child to be a burden on their lives (Langlois et al., 2000).

Teacher-student relationships can also be affected by facial attractiveness. In a study by Clifford and Walster (1973), 504 fifth-grade teachers were asked to predict 12 children's IQs, social relationships with classmates, and expected academic achievement based on photographs, report card information and attendance records. Six of the 12 photographs showed unattractive children and the other 6 showed attractive children. It was found that teachers expected attractive students to have higher IQs, attain higher academic results, and have better relationships with their classmates than unattractive students (Clifford and Walster, 1973).

Attractiveness can have a major influence on job success and employment decisions. Thirty students with identical qualifications were interviewed by thirty professional interviewers in a study. They found that attractive individuals were more likely to be employed than those who were less attractive (Dipboye *et al.*, 1975). It is true that attractiveness can play a major role in forming first impressions, but the long term qualities such as social skills and self-esteem are more important (Cunningham, 1999). The influence of facial attractiveness can have a negative effect on less attractive individuals. Less attractive people may have reduced self-confidence, self-esteem, and problems with their social behaviours and social skills (Cunningham, 1999).

1.3.2 The psychosocial impact of dental appearance

Dental aesthetics are part of the overall facial appearance and can have a great impact on how people perceive overall attractiveness. A study assessed facial aesthetics by evaluating various areas of the face. Seventy participants used visual analogue scales to judge the images, and it was found that the smile is the second facial feature, after

eyes, which people tend to view to assess another person's attractiveness, and that dental aesthetics can impact on overall facial appearance (Hassebrauck, 1998).

In recent years people have become more concerned and aware of their smile, and are keen to improve the appearance of their teeth (Kerosuo *et al.*, 1995). Dental appearance is considered to be very important socially (Shaw, 1981) and people with attractive teeth are thought to be more attractive, more intelligent, less likely to have aggressive behaviours, and teachers have higher expectations from them (Polak, 1975; Shaw, 1981; Lansdown, 1990; Kerosuo *et al.*, 1995).

One study investigated the social judgements made by children in relation to visible incisor tooth trauma. The 291 participants were year 7 and 10 school children (aged 11-12 years and 14-15 years respectively), who were asked to look at photographs of four different children's faces following trauma to their upper permanent incisors, both pre and post treatment. (The pre-treatment pictures were digitally modified to restore incisor aesthetics.) The participants were then asked to make a social judgment about the children in the pictures. The participants rated the children in the pictures using a four point Likert scale for three negative (rude, stupid, naughty) and six positive (clever, kind, honest, confident, careful, helpful) attributes. It was found that negative social judgments may be made on the basis of poor dental appearance, and that aesthetic dental treatment for children may yield important psychosocial benefits (Rodd *et al.* 2009)

Evidence for the impact of dento-facial appearance can also be found in the orthodontic literature. Using digital modifications, black and white photographs of an attractive boy and girl, and an unattractive boy and girl were given 5 different dental arrangements including normal incisors, prominent incisors, a missing lateral incisor, severely crowded incisors and a unilateral cleft lip. The photographs were then assessed by 42 children and 42 adults using visual analogue scales to record their impression of the child's social attractiveness. Children with a normal dental appearance were judged to be better looking, more intelligent, more desirable, and less likely to have aggressive behaviours than the less attractive children (Shaw, 1981).

A randomised, controlled trial investigating the psychosocial impact of early treatment of Class II division I malocclusion suggested that early treatment of this malocclusion can yield a great psychosocial benefit. Three different scales were used to assess the psychosocial impact of early treatment with Twin-block; the Piers-Harris children's self-

concept scale, the Childhood Experience questionnaire, and the Perception of the Benefits of Orthodontic Treatment Scale. It was concluded that early treatment resulted in a significant increase in self-concept and self-esteem and reduction in negative social experiences (O'Brien *et al.*, 2003).

A study investigating the effects of malocclusion and motivation for treatment in a risk-benefit appraisal of orthodontic treatment concluded that 'when personal dissatisfaction with dental appearance is felt in childhood, it might well remain for a lifetime' (Shaw *et al.*, 1991).

1.3.3 The psychosocial impact of dental anomalies

AI can result in significant tooth discolouration and require lifelong dental care. The possible psychological impact of this condition on children and young adults is not fully understood. A recent systematic review study looking at associated dental and orofacial abnormalities with AI, suggested that quality of life and economic issues need to be studied (Poulsen *et al* 2008).

It is surprising that the psychological impact of dental anomalies is a largely unresearched field. A study by Marshman *et al.* (2008) explored the impact of developmental defects of enamel (DDE) on young people, through their experiences of the condition and its meaning to their everyday life. Symbolic interactionism, where 'the self' is developed through interaction, was chosen as the theoretical framework to guide the study. Semi-structured interviews were conducted with 21 (13 female and 8 male) patients aged 10-15 years with different severities of DDE. All interviews were conducted in the participants' own homes and were recorded and transcribed. Each transcript was studied to assess the meaning of DDE for the participants using their own language and whether it had featured in their social interactions or had an impact on them. After each interview, two photographs of the patient's teeth were taken, one with their teeth wet with saliva, and another when their teeth are dry.

The photographs were scored using the Thystrup and Fejerskov Index (TFI) and the Modified Developmental Defects of Enamel Index. Recruitment continued until there were no new themes emerging from the interviews, indicating "saturation". The TFI scores ranged from 0 to 5 and the impact of DDE ranged from 'not bothered' to 'really quite bothered'. The study found that DDE had an impact on individuals who defined their sense of self by appearance and relied on other people's approval of their appearance. It was also found that age, gender, and severity of DDE had no links to

the degree of the impact. The study concluded that variations in the impact of DDE were related to aspects of sense of self rather than the extent of the defect directly (Marshman *et al.*, 2008).

Coffield *et al.* (2005) studied the psychosocial impact of AI on adult and adolescents patients. The age of 14 years or older was one of the inclusion criteria. Fifty nine family members, 30 with AI [mean age was 36.9 (-/+ 17.3) years, minimum age was 19.6 years] and 29 without AI [mean age was 46.6(-/+ 16.0) years] completed a questionnaire where psychometric scales were used to measure various psychosocial parameters such as self-image and self-esteem, social interaction anxiety, and self-perceived quality of life with regard to dental issues. More subjects with AI indicated having dental sensitivity (82.3%) and being teased about their teeth (93.3%) than subjects without AI. Subjects with AI were unhappier with the colour of their teeth (79.3%) than their unaffected counterparts (Figure 1.2).

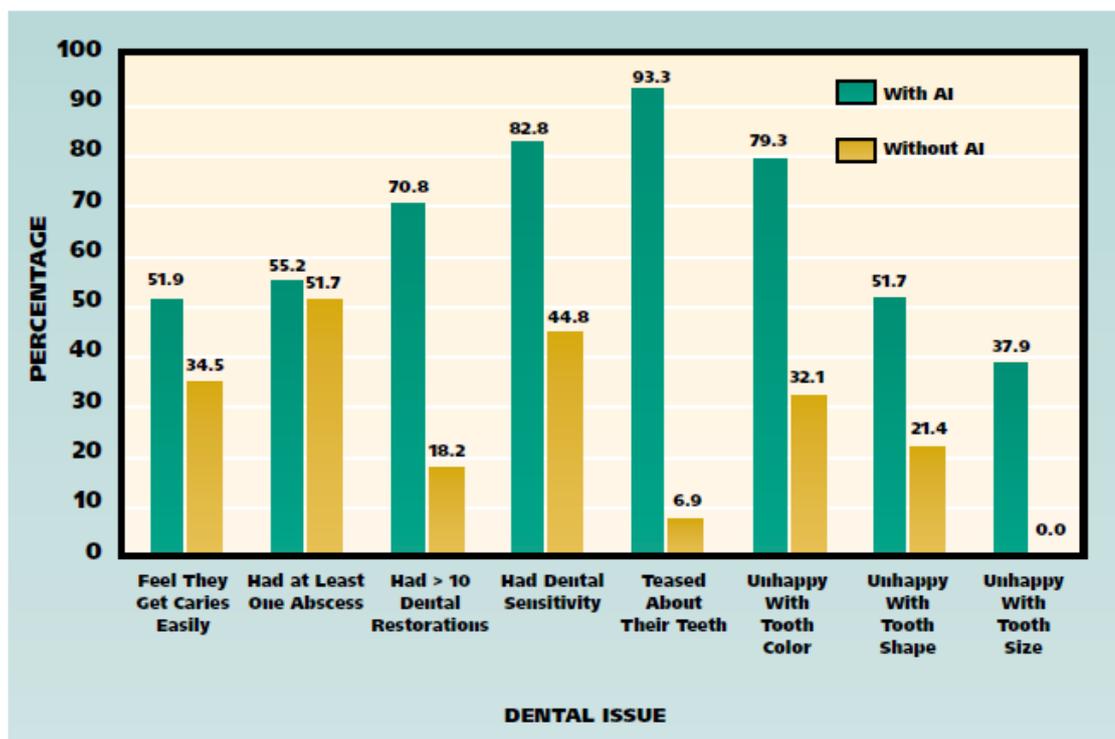


Figure 1.2 Self-reported dental experience of subjects with amelogenesis imperfecta compared with their family members who do not have the condition. (Coffield *et al.*, 2005).

The study concluded that patients with AI exhibited higher levels of social avoidance, distress, and self-consciousness about their teeth. This illustrates how a specific

developmental defect of teeth can influence a person's psychosocial well-being. To date there have been no studies looking at the impact of AI in children.

1.4 Qualitative Research

To investigate the possible impact of AI on children and young adults, qualitative research amongst this group of patients is required. Ritchie and Lewis (2006) provided a simple definition of qualitative research by highlighting the commonly agreed key elements which give qualitative research its unique character. They stated that qualitative research is 'directed at providing an in-depth and interpreted understanding of the social world of research participants, by learning about their social and material circumstances, their experiences, perspectives and histories'. Some researchers have defined qualitative research by highlighting the differences between qualitative and quantitative research, Strauss and Corbin, (1998) state: 'By the term 'qualitative research' we mean any type of research that produces findings not arrived at by statistical procedures or other means of quantification'.

Qualitative research has been through many developments since it was introduced earlier in the early twentieth century, and as a result many different schools have emerged (Ritchie and Lewis, 2006). Perhaps the most commonly known are (i) symbolic interactionism and grounded theory, (ii) phenomenology and ethnomethodology, and (iii) constructivism and critical theory. The theory of symbolic interactionism analyzes society by addressing the subjective meanings that people impose on objects, events, and behaviours. Phenomenology is more concerned about the studying of structures of consciousness as experienced from the first-person perspective. Ethnomethodology is the study of the everyday methods that people use for the construction of social order. Constructivism and critical theory involve creating meaning of the world through a series of individual filters placed over personal realities. The concept of grounded theory will be explained in more details in section 1.4.4. There are various methods or tools that are used to collect data in qualitative research, including in-depth interviews, observational methods, narratives, focus groups, and analysis of documents.

1.4.1 Sampling Strategies and Sample Size in Qualitative Research

When discussing sampling strategies for social research it is very important to distinguish between probability and non-probability samples (Greenfield, 1996). Probability sampling is generally considered a very rigorous approach to sampling and

it is more suitable for quantitative studies. Some of the most common types of probability sampling strategies, include simple random sampling, systematic random sampling, and stratified random sampling (Honigmann, 1982; Moser and Kalton, 1979).

When studying complex issues relating to human behaviour or phenomena, in qualitative research, non-probability sampling is preferred because the aim is to understand not the prevalence or distribution of certain phenomena, but rather the meaning of it from the affected subjects (Liamputtong and Ezzy, 2005). Therefore the main goal in qualitative research is to select samples that can provide rich information about certain features to study in depth, rather than increasing the sample size.

There are two main sampling strategies that have been developed for qualitative research, purposive sampling and theoretical sampling. The term purposive sampling is the most commonly used in the literature, but LeCompte and Preissle (1993) advocated that the term 'criterion based' is more appropriate because in this approach participants are selected because they have a certain feature that the researcher wishes to understand and explore (Ritchie and Lewis, 2006). Within purposive sampling there are different approaches for sample selection. Homogeneous sampling is when the participants for the study are selected because they all fall within a certain category or have the same culture (Holloway and Wheeler, 1996). Heterogeneous sampling is designed to select subjects who have certain phenomena but with a degree of variation (Robson, 2002).

Theoretical sampling is described by some authors as a particular kind of purposive sampling (Ritchie and Lewis, 2006). The researcher selects his or her subjects based on the potential amount of information that can be collected from them, in order to test certain theory. Glaser and Strauss (1967) defined theoretical sampling as 'the process of data collection for generating theory whereby the analyst jointly collects, codes, and analyses his data and decides what data to collect next and where to find them, in order to develop his theory as it emerges. This process of data collection is controlled by the emerging theory, whether substantive or formal. When there are no more new themes emerging from the collected data, the sampling process is terminated since a point of 'theoretical saturation' has been achieved (Coyne, 1997).

Selecting the appropriate sample size is a key point in any research study. In qualitative research, the sample sizes are usually small for three reasons (Ritchie and Lewis, 2006). Firstly, during data analysis there will be a point of saturation where no

more new themes arise and an increase in the sample size will not yield any new evidence. Secondly, the aim of qualitative research is not to answer questions about prevalence or incidence. There is therefore no need to have a sample size that is large enough to support statistically significant findings. Thirdly, qualitative research is highly intense, rich in details, and time consuming. Therefore, having a large sample size may be difficult and unmanageable (Ritchie and Lewis, 2006).

1.4.2 Data Collection Methods in Qualitative Research

Qualitative methods aim for validity through the use of certain tools in order to provide an in-depth and interpreted understanding of how people behave towards, or perceive, certain phenomena (Pope and Mays, 1995). Some of the main methods of data collection used in qualitative research are focus groups, in-depth interviews, and observation.

Focus Groups

Focus groups are also known as group discussions. The group participants interact with each other and share their views and experiences about a certain topic and, through their interaction, data are generated. Stewart and Shamdasi (1990) described focus groups as 'synergistic' and the idea is for the group to work together in order to generate data. One of the key features of the focus group is its spontaneity. The participants use their own language to draw their general framework of understanding. Diversity between focus group participants enriches the discussion, but there must be some common ground shared. It is the coordinator's role to encourage open discussion, include every participant in the conversation, and guide the flow of interactions. Some authors suggest the use of focus groups in order to encourage the least talkative subject to participate in the discussion (Trulsson *et al.*, 2002).

In-depth Interviews

One of the most frequently used methods of data collection in qualitative research is unstructured, or in-depth, interviews (Ritchie and Lewis, 2006). Pope and Mays (2006) believed that interviews were the most commonly used qualitative method in a health care setting. It may be described as a 'conversation with purpose' as described by Webb and Webb (1932) or a "two-way affair" (Liamputtong and Ezzy, 2005). Some of the key features of the in-depth interview are structure, flexibility, interactive nature,

and the using of probe questions. Before conducting any in-depth interviews, the researcher must design a topic guide to serve as an interview agenda (Burgess, 1984).

The researcher must maintain flexibility to allow the interviewees to respond and cover all topics in the order that suits them and be prepared to respond to relevant issues raised spontaneously by them. The interview must be interactive in nature and the researcher should ask an initial question in order to encourage the interviewee to talk freely. The initial response from the interviewee is usually at a 'surface' level. That is why the researcher should then probe further to obtain a more in-depth answer. Finally, the interview should allow the interviewee at some point to explore new thoughts. The interviews are usually face-to-face, and generally tape recorded in order for data to be captured in its natural form (Ritchie and Lewis, 2006).

1.4.3 Combining Qualitative and Quantitative Research

Despite the considerable potential for combining qualitative and quantitative research the debate as to whether the two should be, or can be, combined is still present in social research. The most recent views are that both research methods can complement each other, provided both methods and their data are clearly defined (Ritchie and Lewis, 2006). Some authors have suggested ways in which the methods can complement one another (Brannen, 1992; Bryman, 2001). One of the simple linkages that can be seen between the two methods is when qualitative research may precede statistical enquiry. This has been suggested to help devising areas of questioning for statistical study and to help discover and include comprehensible terms in a questionnaire (Pope and Mays, 2006). It is important to understand and appreciate that, despite the fact that both research methods can be combined or work together, each will offer different ways of knowing about the world, and that their evidence should not replicate each other (Ritchie and Lewis, 2006).

1.4.4 Data Analysis

Data analysis is challenging and considered the most complex stage of qualitative research (Thorne, 2000). There are many different approaches or strategies within qualitative analysis based on the nature of qualitative enquiry, primary aims, and focus (Ritchie and Lewis, 2006). Thematic analysis, content analysis, and constant comparative analysis are just few examples of different approaches or strategies within qualitative analysis. With thematic analysis, the task is to identify different themes or ideas by coding and indexing them after further exploration of each theme. It is very

important to create a manageable index which has a hierarchy of main and subthemes (Ritchie and Lewis, 2006). This process is central to building a thematic framework.

Content analysis involves the analysis of both the content and context of the collected data. Themes are identified and counted to determine the frequency each theme is encountered, then applying statistical methods to it.

Grounded theory was described by Glaser and Strauss (1967). It is designed to produce a theory from data collection about certain situation or phenomena. The idea is that concepts and themes emerge while the research is undertaken. It is not designed to test a hypothesis. Constant comparative analysis was a specially designed technique for use in the grounded theory approach (Glaser and Strauss, 1967). This technique is central to the grounded theory approach, where one piece of data is compared to all other data in order to explore and understand the variety between all pieces of data (Thorne, 2000). Data collection and analysis are both carried at the same time during this process until the researcher reaches the point of saturation where no more themes or concepts are arising. 'Coding' is considered the heart of constant comparison whereby each segment of speech is given a code representing a theme and each segment of speech are compared with the one before it and at the end of this process the researcher can create a theory or explanations about the data (Pope and Mays, 2006).

1.5 Questionnaire Development

Questionnaires are considered as a tool for data collection (De Vaus, 1996). Unfortunately, there is no widely accepted theory on how to design a questionnaire, with the result that medical professionals may not have the proper skills to design a questionnaire (Stone, 1993).

There are two types of questionnaire that are generally used, self-completion questionnaire and interview questionnaire; each one of them has its advantages and disadvantages. Some of the main advantages of self-completion questionnaire are the low cost, a large sample can be obtained, and it is generally familiar to most people. It can be completed at home or in the research setting (Williams, 2003). Disadvantages of self-completion questionnaire include the difficulty in ensuring that the participants answer all the questions themselves, and the low response rate especially if the questionnaire is posted to participants. There is always a possibility that the sample can be biased towards a better educated group when using the postal questionnaire (Black *et al.*, 1998).

In order to maximise the response rate some factors need to be considered while constructing a questionnaire (Edwards *et al.*, 2002). The questionnaire should be of appropriate length. Generally short and simple questionnaires will result in a higher response rate (Leung, 2001). The questions should be constructed to gain information related to the research topic. The questionnaire should have a simple and easy to understand title, with clear instructions on how to complete the questionnaire. It is important to guarantee confidentiality, preferably on the first page. When appropriate colours should be used and a personalised cover letter should be enclosed (Black *et al.*, 1998).

Other important factors include; the layout of the questionnaire, so that participants answer the questions correctly, and to facilitate the process of coding and analysis. The questions should be short, simple and specific starting with interesting and non-threatening issues in order to gain the participants attention and make them interested to finish the questionnaire. If the questionnaire is posted, then a stamped addressed envelope should be enclosed to insure ease of return. It is important to pay attention to all these details at the beginning of developing a questionnaire to improve the response rate (Walonick, 2004).

1.6 Psychometric Properties of Questionnaires

It is questionable whether the same measurements used to assess the quality and sustainability in quantitative evidence can be used or applied in qualitative investigations. Most of the debate is concerned with the concepts of validity and reliability.

1.6.1 Validity

To assess the validity of a questionnaire it is important to understand that there are different aspects of validity. Generally a questionnaire is thought to be valid if it measures what it is set out to measure (Black *et al.*, 1998). External validity is the ability to generalise the results to the general population, which is almost impossible in most qualitative research. However, internal validity is applicable in qualitative research and can be listed in three different types: (i) content validity measures the extent to which different indicators actually measure the variable aspects of the examined concept, (ii) face validity measures if a tool – e.g. a questionnaire – can be easily understood and if it is relevant to the research topic and (iii) criterion validity, where the assessment takes a form of comparing the new measure to a pre-existing valid ‘gold standard’. Construct validity is usually used when there is no gold standard to compare

to and it is defined as a theoretical measure of how meaningful a survey tool is, usually established by many investigators after many years of experience (Litwin, 2003). Some authors advocated that there is no right or wrong way of assessing the validity of a measure, since different situations may require different methods. (De Vaus, 1996).

1.6.2 Reliability

Reliability is usually understood to concern the replicability of research. It is set to assess the consistency and reproducibility of an instrument (Ritchie and Lewis, 2006). To assess the reliability of a self-completion questionnaire there are two aspects that need to be examined (Williams, 2003). First, to determine the internal consistency of a questionnaire the same questions should be asked in more than one way. The second aspect is called test-retest reliability, and it can be assessed by asking the participants to complete the same questionnaire on two separate occasions usually two to three weeks apart, and compare all responses at the end. Reliability in general is not recommended and considered to be inappropriate in qualitative research since there is no single reality to be achieved in the first place so the idea of replication is an artificial goal to pursue (Holstein Gubrium, 1997).

1.6.3 Readability

It is important that the questionnaire can be easily read and understood by the general public. Readability is set to assess this aspect of a questionnaire. During the pilot stage the participants can be asked to give their comments about the questionnaire at the end and assess their answers. There are some indices designed to assess readability like the Flesch Reading Ease Score (Flesch, 1948) and the Flesch-Kincaid Grade Level (Kincaid *et al.*, 1975) both measuring the average number of syllables in a word and the average number of words in a sentence.

1.6.4 Acceptability

This can be assessed during the pilot phase by looking into certain aspects of a questionnaire. For example, how much time was needed to complete the questionnaire, what did the participants think about answering the questions, did the wording of the questions convey the appropriate meaning and whether or not the questions were answered correctly (Williams, 2003).

1.7 Validation in Qualitative Research

There are different ways to validate or verify qualitative data. Generally they fall into two main categories, the first is concerned with internal validation and the second is concerned with external validation (Ritchie and Lewis, 2006).

Internal Validation

The constant comparative method - Content analysis will involve the analysis of both the content and context of the collected data. Themes will be identified and counted to determine the frequency each theme is encountered then applying statistical method to it.

Deviant case analysis – whenever there are deviant cases in the data they should not be ignored or forced into categories or classes. Instead they should be carefully reviewed and separately considered to help in further understanding and theory development (Ritchie and Lewis, 2006).

External Validation

Triangulation – in order to confirm and improve the research findings the step of triangulation should be applied. It will involve the use of different sources (e.g. interviews and questionnaires) and comparing their data together and looking for overall patterns (Patton, 2002).

Member or respondent validation – this will involve taking the research evidence collected from the data back to the participants who contributed to the data or to a group with the same experience or phenomena and assess the meaning that was assigned by the researcher, to see if it is confirmed by the participants (Ritchie and Lewis, 2006).

1.8 Generalising from Qualitative Research

Findings from qualitative studies can be generalised beyond the sample and context of the research. There are three different concepts that are involved with generalisation (i) representational generalisation mean where the findings from a sample size, can be generalised to the parent population that the sample was selected from, (ii) inferential generalisation asks whether the findings from the study can be generalised to different context or settings than the sampled one, and (iii) theoretical generalisation which is the most classic concept of generalisation and involves drawing theoretical statements from the findings of a study for general applications such as social structures and individual behaviours (Ritchie and Lewis, 2006).

1.9 Summary of the review of the literature

Amelogenesis Imperfecta (AI) can result in significant tooth discolouration and problems with sensitivity and function. The possible psychological impact of this condition on children and young adults is not fully understood. It is hoped that investigating this important aspect, using qualitative research methods will improve our understanding of the concerns of AI patients and providing better patient care for this group of patients.

Therefore the aim of this study is to explore the impact of AI on children and young adults through in-depth interviewing and a subsequent Framework Analysis. The information derived from this will then be used to construct a questionnaire.

2.0 Aims and Objectives

2.1 Aims

The aim of the study was to explore the impact of Amelogenesis Imperfecta (AI) on children and young adults.

2.2 Objectives

- To undertake semi-structured in-depth interviews with patients to identify themes that are important to AI patients.
- To undertake qualitative framework analysis based on the information from the interviews.
- To use the information from the framework analysis to develop a questionnaire, including the key themes and categories identified.
- To pilot and then distribute the questionnaire to AI patients in the Unit of Paediatric Dentistry at the Eastman Dental Hospital to assess the impact on children and young adults.

3.0 Subjects and Methods

3.1 Ethical Approval

Ethical approval for this research was granted in November 2010 by the Proportionate Review Sub-committee of the South East London REC 3 Research Ethical Committee (REC reference number: 10/H0808/156) (Appendix 1). Approval from the Research and Development Directorate of UCL Hospitals NHS Foundation Trust was also sought and granted.

3.2 Research Design

This research comprised of two parts, combining qualitative and quantitative methods, in order to develop a valid questionnaire to distribute to a large cohort of AI patients. Both parts of this research were undertaken in the Unit of Paediatric Dentistry, Eastman Dental Hospital. Part one involved the identification of common themes and concepts raised by AI patients in semi-structured, in-depth interviews. In part two a questionnaire based on the themes identified from part one of the research was developed and distributed to AI patients in the Unit of Paediatric Dentistry. This questionnaire aimed to investigate the impact of AI on children and young adults, attending the Eastman Dental Hospital.

4. Part One of the Study: In-depth Interviews & Framework Analysis

This part of the study aimed to obtain a deeper understanding of the main issues and concerns for patients with AI. The themes identified from this part were subsequently used to construct a valid patient-centred questionnaire.

4.1 Interviewing training

The main researcher (MA) attended an interview skills course at Aston University in April 2010. The course was intended to teach researchers who are involved in qualitative research how to undertake effective interviews. Following this, the main researcher (MA) and the research supervisors met to generate a topic guide consisting of open-ended questions relating to AI patients to be used in the interviews (Appendix 2). The questions were based on information gathered from the literature regarding AI and also from clinical experience. The questions were used as prompts during the interview but were not strictly adhered to. Four face-to-face pilot interviews were conducted with colleagues and supervised by senior research staff, experienced in the field of qualitative research and in-depth interviewing. The pilot interviews were tape recorded. Each pilot interview lasted between 25 and 30 minutes, followed by 15 to 20 minutes of feedback about the interview. This involved discussion and advice on how to use open ended questions, how to explore new ideas and responses in more detail, and how to handle sensitive issues if they were brought up during the interview process.

Patient selection involved a theoretical sampling approach to ensure a wide range of subjects and ideas. As discussed in Section 1.4.1, theoretical sampling is described by some authors as a particular kind of purposive sampling (Ritchie and Lewis, 2006). The researcher selects subjects based on the potential amount of information that can be collected from them, and when there are no more new themes emerging from the collected data, the sampling process is terminated since a point of 'theoretical saturation' has been achieved (Coyne, 1997). In qualitative research it is often difficult to know exactly how many participants are required in advance, although previous research indicated that approximately 15-20 patients would be sufficient (Pabari *et al.*, 2008).

4.2 Patient sample

The inclusion criteria were:

- 1- Male and female AI patients
- 2- Subjects of any ethnicity
- 3- Patients who were between 10 and 16 years of age
- 4- Patients with AI of a severity requiring restorative intervention
- 5- English speaking patients
- 6- No active restorative treatment yet undertaken

The age range of 10 to 16 years was chosen to ensure that the patients could comprehend the questions and discuss any issues or concerns they may have. English was the language chosen to avoid any miscommunications during translation and to allow a full discussion in the interviews. Patients who had undergone no active treatment were chosen in order to avoid any changes in the patients' thoughts and views about AI as a result of treatment they had received.

Patients were recruited from the Unit of Paediatric Dentistry at the Eastman Dental Hospital, UCLH Foundation Trust, from March to June 2011. Patients who satisfied the above criteria were approached by the research staff and asked if they would be willing to participate in the research. The main researcher (MA) attended the clinic every day to identify AI patients and asked colleagues and staff to identify any AI patients attending their clinic. The main researcher (MA) also attended the dental anomalies clinic to recruit any AI patients who satisfied the inclusion criteria. Each patient, and their parent/legal guardian, were provided with an information sheet outlining the details of the study (Appendices 3 and 4), and the purpose of the research was also explained verbally. If the patient and their parent/legal guardian agreed to participate, written informed consent was obtained from the parent/legal guardian and written assent from the patients (Appendices 5 and 6). Assurances were given regarding confidentiality; the information sheet included a statement which assured patients that their information would be recorded and coded in such a way that it was completely anonymous and that they would not be individually identified in the results.

4.3 Interviews

Face-to-face interviews were conducted in a side surgery to ensure privacy and to provide a less stressful environment than the main clinical area, and a chaperone was present in all the interviews. Parents were encouraged to remain outside the side surgery while their child was being interviewed, but they were given the opportunity to join the interview if their child requested. All interviews were recorded using a digital Dictaphone and then transcribed onto Microsoft Word, with all patients identified by code number only in order to maintain patient confidentiality. Patients who participated in the study were told that if they found any question in the interview or the questionnaire upsetting or of a sensitive nature they did not have to answer. They were also given the opportunity to stop the interview or withdraw from the study at any time if they wished.

All patients were asked open ended questions about themselves and their teeth using the topic guide but if the patient talked about issues which were not included in the interview schedule, they were also explored. The interviews started with general questions e.g. "Can you tell me a little bit about yourself?", and then gradually moved on to more probing questions about aesthetics, sensitivity, function and psycho social aspects, such as effects on friendships and comments by other people. The interview was transcribed immediately after the interview. The interview transcription and analysis were ongoing processes. The main researcher (MA) read through each interview transcript carefully to make sure that any emerging themes were identified and included in subsequent interviews. This technique allowed the researcher to establish when no more themes or ideas were arising and hence when to stop the interview process (Glaser and Strauss, 1967).

4.4 Framework analysis

A thematic analysis following the National Centre for Social Research (NatCen) approach was used to allow a detailed in-depth overview of the participants' personal experiences. This has been used in similar studies at the Eastman Dental Institute (Pabari *et al.*, 2008, Ryan *et al.*, 2009, Lee *et al.*, 2011). The first step involved identifying initial themes or concepts by carrying out a thorough review of the data from the transcripts. The second step involved constructing a framework index with a hierarchy of main themes and subthemes. An Excel spread sheet was developed with a sheet for each theme – columns were subthemes and each row represented a patient. Individual patient's quotes were then added to the appropriate cell.

The steps involved in framework analysis are shown in Figure 4.1 and the Excel spread sheet in Figure 4.2.

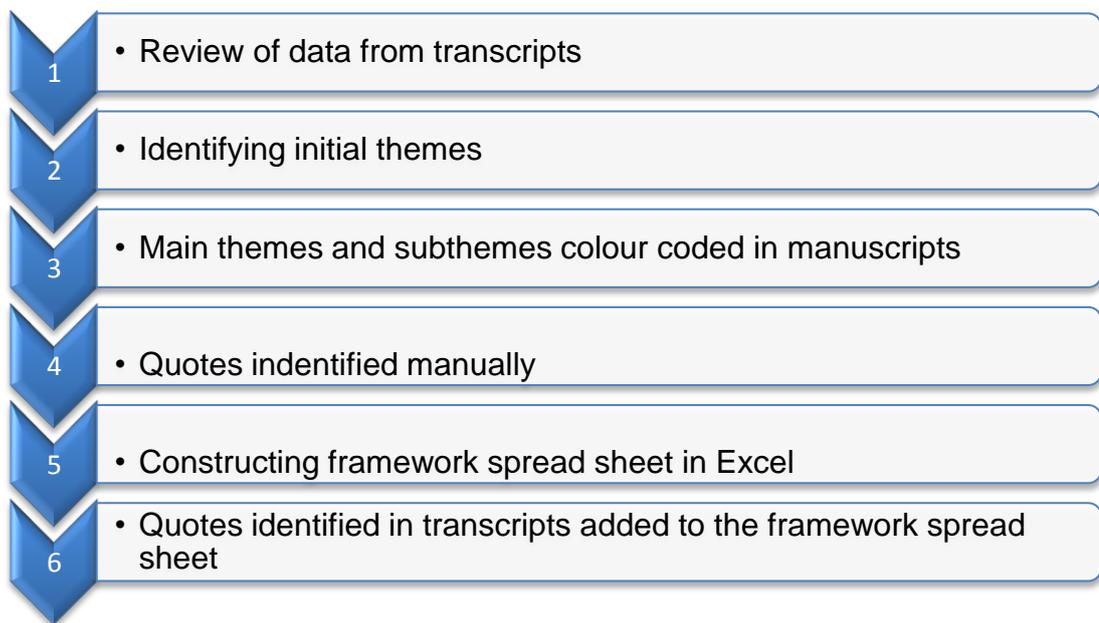


Figure 4.1 Flow chart showing the steps involved in the Framework analysis.

Order	Analytical Comments	3.1 Colour	3.2 Shape	3.3 Size	3.4 Photos/Vedios	3.5 Smiles
P1		It's the discolouring (55). Definitely the colour (59). It's not like completely white but it's more like a light yellow than mine (66). If they were just a little more nice colour, not discoloured (191)	Maybe chipped like they've broke off (136)		Whenever like someone takes a picture of me, I always close my mouth (45). I've had pictures and I've smiled, the whole picture is just completely ruined because you can just see this discolourant in your teeth (75). If no discolouration I could smile at the camera (193). When it's a picture, I always close my mouth, I always just not... I just don't like to see it in a picture (301).	I hate s...
P2		they are yellow (65). The colour, like when I say "Sshhh" my front teeth show on there, they're yellow ones (160).	and they're all different shapes really (107). if the shape was right, I would smile without looking like an idiot (239)		I don't like showing my teeth in pictures (72). I never show my teeth in photos (95).	I don't like showing my teeth in pictures (72). I never show my teeth in photos (95).
P3		it is not normal the colour (55). Ugly discoloured (66)	I don't like the shape (55). Misshaped (66).	they are small (71)		I don't like showing my teeth in pictures (72). I never show my teeth in photos (95).
P4		I don't want them to be like, you know, a really bad colour but they should be clean (178).	I think the shape (83).	my teeth are quite small (61). Yeah the size when I talk you can't see them (78). They are going smaller (250).		I don't like showing my teeth in pictures (72). I never show my teeth in photos (95).

Figure 4.2 Framework spread sheet showing quotes from transcripts in the appropriate cells according to the theme and subtheme. (the number in brackets refers to the line number from the transcript)

4.5 Results for Part One of the Study: In-depth interviews

Demographics

The demographic data for the patients interviewed in Part 1 of the study are shown in Table 4.1. Each patient was assigned an ID number. A total of 7 patients were interviewed, 6 of whom (85%) were female and the average age was 14.2 years, with a range of 13 to 16 years.

ID	Gender	Age (years)	Ethnicity
1	F	16	C
2	F	13	C
3	F	16	N
4	M	15	N
5	F	15	C
6	F	13	N
7	F	13	N

Table 4.1 Demographic details of patients interviewed

(M = Male, F = Female, C = Caucasian, N = Non-Caucasian)

One of the inclusion criteria for this part of the study was to identify AI patients who had not received any significant restorative input before. Within the time period, it was only possible to identify and approach 8 AI patients, as it was very difficult to find patients who never had any restorative treatment prior to their visit to the Unit of Paediatric Dentistry. One patient refused to participate due to the sensitivity of the subject.

The majority of patients interviewed 85.7 % (n=6) were diagnosed with Hypocalcified AI, and only one patient had Hypoplastic type of AI. Two patients indicated there is a family history where one of their parents had AI, the rest of the patients did not highlight any family history with AI.

4.5.1 Interviews – duration

Interviews lasted between 13 and 20 minutes, with an average of 16 minutes for each interview. The number of lines per interview ranged from 310 to 566 lines (average of 438 lines per interview) and in total, 5431 lines of transcripts (63 pages) were analysed manually.

4.5.2 Framework Analysis

The main themes and subthemes identified from the interviews are shown in Figure 4.3. Some patient quotes are included, with the 'P' number representing the ID of the patient who made the comment and the number in brackets referring to the line number from the transcript.

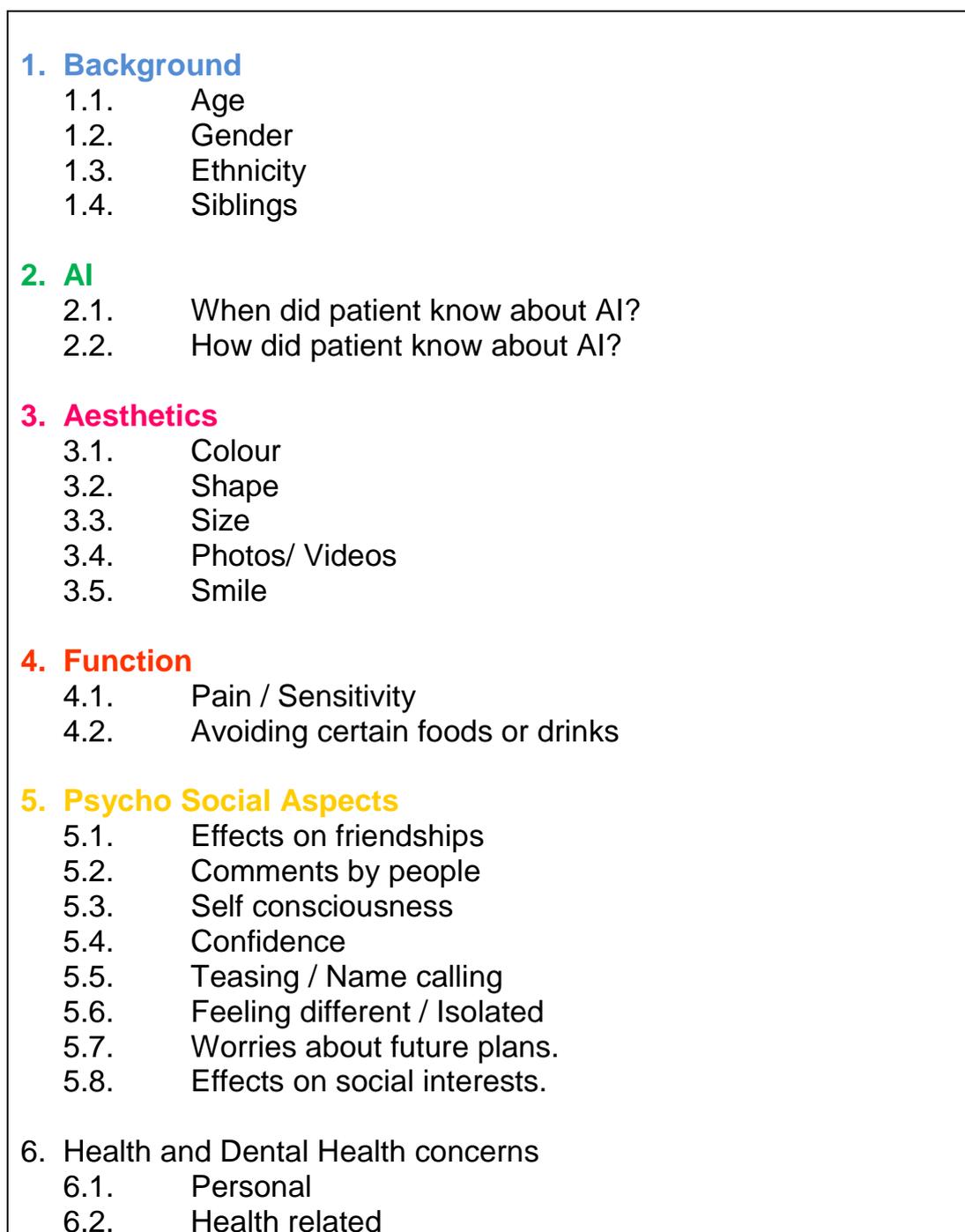


Figure 4.3 Framework showing the main themes and subthemes identified from the interviews.

Framework section 2: AI

The comments regarding AI were divided into 2 categories which were “When” and “How” the patient found out about AI. The majority of patients stated that it was their dentist who informed them about AI, with the exception of one patient who was informed by their General Medical Practitioner. A number of patients started to notice AI when they were in the mixed dentition stage.

(a) When did you found out about AI?

- [P2: 258] “I just didn’t really realise what it was till like a couple of years ago”
- [P7: 105] “I think I was about 10”
- [P3: 97] “Started to notice in primary school”
- [P1: 104] “I was younger, my first teeth were fine, then when my second set started to come through, they just started to change colour all of a sudden and over time”

(b) How did you find out about AI?

- [P4: 123] “I went to the dentist, she was talking about it”
- [P7: 113] “my doctor (GP) and my family told me”
- [P2: 253] “dentists, like muttered on about “Oh, yellow enamel” and I didn’t really know what it was and had to get mum to explain it to me”

Framework section 3: Aesthetics

The answers regarding aesthetics were coded into 5 categories which are listed below but the most common issue raised by the interviewees was the colour of their teeth. Other important issues also highlighted by patients were shape, size, smile, and feelings about having photos or videos taken.

(a) Colour

- [P1: 191] “.....if they were just a little more nice colour, not discoloured”
- [P3: 55] “it is not normal the colour”

- [P4: 178] "I don't want them to be like, you know, a really bad colour but they should be clean"
- [P2: 160] "The colour, like when I say "Sshhh" my front teeth show on there, they're yellow ones"
- [P7: 97] "It was okay but just the colour"

(b) Shape

- [P2: 107] "And they're all different shapes really"
- [P7: 306] "They were like triangle shape"
- [P6: 122] "They are quite round, I want them more square, normal like"

(c) Size

- [P3: 71] "They are small"
- [P4: 78] "Yeah the size, when I talk you can't see them"
- [P6: 40] "They are smaller"

(d) Photos/Videos

- [P1: 45] "Whenever like someone takes a picture of me, I always close my mouth"
- [P5: 323] "I don't like having my pictures taken"
- [P2: 72] "I don't like showing my teeth in pictures"
- [P6: 145] "like I went to a [photo] shoot and I didn't really want to smile with my teeth"

(e) Smile

- [P2: 84] "I can't really smile, to be honest, I can't get the facial expression right, I just don't like my teeth, why show them off"
- [P6: 108] "I don't like smiling with my teeth because I don't like them"
- [P1: 46] "I hate showing my teeth"
- [P3: 242] "I don't smile"
- [P7: 270] "I will smile when all of my teeth are going to be white, nice shape"

Framework section 4: Functional concerns

Questions regarding functional concerns were coded under 2 categories which were:

(a) Pain/Sensitivity

- [P5: 453] "It is the sensitivity more than the colour, the colour doesn't bother me, it's more the sensitivity."
- [P1: 171] "There is a bit of pain sometimes when brushing"
- [P4: 224] "If I eat cold things it starts hurting my teeth"
- [P6: 214] "With some cold stuff I do have sensitivity"

(b) Avoiding certain foods or drinks

- [P2: 227] "If there was no problem with sensitivity, I'd drink faster and bite down on ice lollies and not cringe when I think of it"
- [P6: 227] "Just I think really cold stuff"
- [P1: 153] "I couldn't lick a lolly properly because it was so cold to my teeth"
- [P5: 135] "It's fine, it's just hard food sometimes I struggle to bite"

Framework section 5: Psycho-Social Aspects

The psycho-social aspects were divided into 8 sub-themes which are described below. The majority of patients described comments by other people about their teeth, and one patient expressed worries about future plans.

(a) Effects on friendships

- [P1: 290] "I only keep to a small majority of friends"
- [P5: 261] "I tend not to always be out with my friends"

(b) Comments by people

- [P2: 115] "If someone sees it, they go "Oh, don't you brush your teeth?" and stuff like that"

- [P3: 223] "Oh my brothers and sisters say some comments"
- [P5: 278] "They will say, "Do you brush your teeth properly?" I always tell them"
- [P6: 130] "Sometimes when people mention things about them I don't like to then open it, my mouth again, or anything, I like to just keep my teeth hidden"

(c) Self consciousness

- [P1: 46] "I feel really self-conscious"
- [P5: 326] "I think that's me not the teeth but it could be part of the reason subconsciously, I don't know"
- [P7: 222] "I'm not feeling well, I'm feeling a little bit bad, because they can see that my teeth look like dirty"

(d) Confidence

- [P2: 233] "If the colour was fine, I'd feel a lot more confident"
- [P1: 281] "The colour just knocks my confidence"
- [P3: 233] "I'm still confident even though my teeth are like this"
- [P6: 189] "Would make me feel more... Courageous if they were't like that yellow"

(e) Teasing/Name calling

- [P1: 46] "I'm worried people are staring at me and laughing behind my back"

(f) Feeling different/ isolated

- [P1: 48] "I feel like I'm in my own world, sort of thing, not in my own world, but I just feel different and I don't want to be like that"
- [P3: 266] "Like when I'm outside with people and they have nice teeth I think about it more basically"
- [P2: 114] "It looks different to everyone else"

(g) Worries about future plans

[P6: 146] “In the future if it gets more, the modelling, then I wouldn’t really, or singing or whatever I do, I don’t really want to have bad teeth”

(h) Effects on social interests

[P4: 377] “Say it’s like a really cold day or something and I’m in the woods like cantering or something, sometimes I can feel the wind at the back of my teeth”

[P3: 389] “When all my friends are talking I’d want to join in but I don’t want to show my teeth”

Framework section 6: Dental Health

A small number of patients had some concerns regarding their dental health.

(a) Personal

[P1: 180] “Mum must have bought loads and loads of dental care from toothbrushes to toothpastes, and it just doesn’t work”

[P3: 192] “I don’t like brushing it because I don’t like looking at it, at my teeth

4.6 Discussion for Part One of the Study: In-depth interviews

4.6.1 Introduction

The importance of teeth should not be underestimated, since they are considered a main component of dento-facial appearance and have an important function during speech and mastication. It is well documented in the literature that dental appearance can have a great impact on patients (Hassebrauck, 1998). Whilst we know anecdotally that children may suffer teasing from their peers, the impact of AI, and the effect on quality of life is an under researched field. A recent systematic review recommended that the reduced quality of life and economic burden to patients with AI needs to be studied in depth (Poulsen *et al.*, 2008). To date the only research on the impact of AI was done by Coffield *et al.* (2005) who studied the psychosocial impact of AI on adult patients. The study indicated that patients with AI exhibited higher levels of social avoidance, distress, and self-consciousness about their teeth. This illustrates how a dental defect can influence psychosocial well-being. To date there have been no studies specifically looking at the impact of AI on children and young adults. It was therefore, the aim of this study to investigate and understand the impact of AI in children and young adults. This level of understanding may help the paediatric dentist, by designing a treatment plan addressing the patients' main concerns and also allowing appropriate management.

4.6.2 In-depth Interviews

The aim of this part of the study was to identify the main issues for AI patients and to construct a patient centred questionnaire. There are various methods that may be used to collect data in qualitative research and, for this study, in-depth interviews were chosen. One of the main advantages of in-depth interviews is that they allow the interviewees to be open and to share and discuss any thoughts or issues they might have, and focus on those issues that are important to them without being influenced by the interviewer's own ideas. However, one of the main challenges faced was interviewing teenagers, as the majority of the interviewees were shy at first and not willing initially to open up and talk about their feelings and thoughts. The researcher (MA) had to be sympathetic to this, ask more questions, allow patients time to consider their views, and encourage patients to expand on their answers. This had an impact on the length of the interviews; most of the interviews conducted with patients were shorter in time compared with the pilot interviews with colleagues. Other patients did feel confident enough to discuss concerns readily, for instance, when asked about

having photographs taken one patient expressed her concerns about future plans and careers. This issue had not been considered in the topic guide until the patient raised it.

In-depth interviews are laborious and time consuming in term of the transcription and analysis. It depends on the interviewing skills of the interviewer, and it is for this reason that the main researcher (MA) attended an interviewing skills course and practiced different possible interview scenarios with colleagues in order to be well prepared for interviews with patients. Some authors have argued that interviews lead to bias since the interviewer may lead the participant in their answers (Bowling, 1997). The main researcher (MA) avoided the use of leading questions during the interviews and followed the patient's thoughts and ideas to explore issues or concerns he or she might have.

Developing an interview style with open ended questions was not an easy task, the questions were carefully selected and worded to avoid simple yes or no answers and reduce bias (Black *et al.*, 1998). The pilot interviews were helpful in learning how to use open ended questions, explore new ideas and responses in more detail, and how to handle sensitive issues if they were brought up. Other possible confounders like malocclusion problems that might be associated with AI were discussed and decided best to be included in a separate study in liaison with the Orthodontics department to avoid any conflict or overlap of information regarding the impact of AI or malocclusion on patients.

Recruiting patients to participate in the interviews was considerably more difficult than originally anticipated. One patient's mother refused to allow her son to participate in this project due to the sensitivity regarding the subject. It was also hard to find AI patients who had not undergone any previous restorative treatment for their condition, since the majority of the AI patients in the Department of Paediatric Dentistry had undergone some form of treatment prior to their attendance, or were already patients in the Department. This meant that only 7 patients could be recruited within the allocated time interval. However, no new themes were identified at the last interview, therefore it was felt that the most relevant themes had been identified.

4.6.3 Framework Analysis

There are many different approaches or strategies within qualitative analysis, depending on the nature of the qualitative enquiry, primary aims, and focus. Thematic analysis, following the National Centre for Social Research (NatCen) approach, was used in this study. The NatCen approach aims mainly to organise data rather than

actually analysing it. This allowed the main researcher (MA) to manage the data from the interviews in an organised and well constructed manner by identifying initial themes or concepts after undertaking a thorough review of the transcripts, and then constructing a framework index incorporating main themes and subthemes. A framework chart was designed for organisation of the patient quotes. Framework analysis is a useful process, but it is time consuming. It requires manual labelling of data and comments which are then assigned to the correct theme and subtheme. One of the main difficulties faced during the framework phase was creating categories (themes and sub-themes) given the large volume of text that had to be analysed. This is a hard and challenging task to accomplish by one person; perhaps it might be useful in the future to create a team of specialist e.g. psychologist and orthodontics to assist in analysing the data from the interviews.

The semi-structured in-depth interviews with AI patients resulted in six main themes and these now will be discussed below in more detail.

4.6.3.1 Background

Most of the patients who were willing to be interviewed were females, and that might be because females in general were found to be more attentive conversationalists than males (Giles *et al.*, 1982). It could also be attributed to the fact that more regular dental attendance has been reported among females (25.75%) compared to males (16.6%) (Quteish Taani, 2002). It is hard to predict whether any gender differences could have had any effect. Both males and females appeared to share the same concerns and there were no differences in responses between the single male respondent and the females.

4.6.3.2 AI

The majority of patients started to notice their AI at a young age (around 6 years old), this is in agreement with previous research which suggested that children start noticing dental defects at a young age (Marshman *et al.*, 2008). When asked who told them first about AI, most patients answered that it was their dentist, with the exception of one patient who was informed by her general medical practitioner. The majority of patients were confused about the explanation given to them by their local dentist about their enamel condition. This may highlight the fact that dentists needs to be well informed regarding AI, and be able to explain the nature of this condition in a way that young children can understand. Further training at undergraduate or postgraduate level may

be required to teach dentist the skills required. It may also be that children find such information difficult to retain, hence it is very important to have good information sources such as information leaflets and specific internet websites about AI.

4.6.3.3 Aesthetics

All of the interviewees discussed concerns about the colour of their teeth and described teeth as being “yellow-brown” when asked about aesthetics. Adult AI patients were also more unhappy with the colour of their teeth (79.3%) than subjects without AI (32.1%) (Coffield *et al.*, 2005). The majority of patients were unhappy about the shape of their teeth and a small number were concerned about the size of their teeth too. The different issues raised regarding aesthetics were probably due to the fact that AI can present in different forms e.g. the hypoplastic form where teeth tend to be a different size and shape from normal teeth, or the hypocalcified type where teeth tend to have chalky, yellow, brown, soft enamel with post- eruptive loss. Several patients stated that they did not like being photographed or videoed because they did not want to show their teeth and others stated that they were not satisfied with their smile and would avoid smiling around other people. In a study by Porritt *et al.* (2010) assessing quality of life impact following childhood dento-alveolar trauma, the authors found that avoiding smiling or laughing when around other children had an impact on the child’s oral health related quality of life. Another study investigating children’s experiences of enamel defects also found that a number of children were reluctant to smile (Rodd *et al.*, 2009).

There are clinical implications regarding the importance of dental aesthetics in AI patients, as their level of concerns may lead to high expectations of having better aesthetics following treatment. It is important for the clinician to manage the patient’s expectations by understanding their motivation and thereby hopefully achieving optimum levels of satisfaction with treatment outcome (Abdel-Kader, 2006). Clinicians must discuss the objectives and limitations of treatment at the outset in order to achieve fully informed consent, set realistic expectations and hopefully to avoid any dissatisfaction with the outcome of care (Cunningham *et al.*, 1996). Aesthetic dental treatment for children may yield important psychosocial benefits (Rodd *et al.* 2009), but this does rely on patients being appropriately prepared and having realistic expectations.

4.6.3.4 Function

A large number of patients said that they had experienced pain and sensitivity from their teeth. In addition some patients reported avoiding certain foods or drinks (hard food and fizzy drinks) due to dental sensitivity. The issue of dental sensitivity was found to be a major factor in the impact of AI on adult patients as stated by Coffield *et al.* (2005), with 82.3% indicating sensitivity. Patients affected by Dentinogenesis Imperfecta were also found to benefit from early treatment to improve function and minimise nutritional deficits and psychosocial distress (Barron *et al.*, 2008).

4.6.3.5 Psycho-Social Aspects

The majority of patients said they had received some comments from other people about their teeth. A person's psychological well-being can be affected by perceptions and responses received from society (Bull and Stevens, 1981; Rumsey *et al.*, 1982). A small number of patients expressed feeling different or being isolated. Other issues that arose during the interviews were effects on friendship and on social interests. This is in accordance with the findings of Dion *et al.* (1972) where unattractive people were selected as friends less frequently and were considered more anti-social. This may lead to attractive children being treated more positively than unattractive children as found by Langlois *et al.* (2000), and children with AI may be similarly affected.

Teasing and name calling were discussed in the interviews. This was also highlighted with adult AI patients, with 93.3% of subjects reporting being teased about their teeth (Coffield *et al.*, 2005). This was also found with respect to psychosocial effects of malocclusion, where children with malocclusions were found to be more susceptible to teasing by their peers (Shaw *et al.* 1980). Marshman *et al.* (2008) found that some young people had experienced teasing and name calling due to developmental enamel defects of their teeth. Teasing history is considered an influencing factor for seeking treatment in orthodontics (Shaw *et al.*, 1980; DiBiase and Sandler, 2001). Clinicians should be sensitive to such issues in children, which may require support or referral to counselling services.

The impact of facial appearance can be seen in a person's own sense of well-being and self-esteem (Diener *et al.*, 1995). Less attractive people may have reduced self-confidence, self-esteem, and problems with their social behaviours and social skills

(Cunningham, 1999). Self consciousness and confidence were mentioned by a number of patients as one of the reasons they wished to undergo treatment; this is in accordance with the findings of McKiernan *et al.* (1992) where improved confidence in interpersonal relationships was seen as being the most important benefit of treatment after improving aesthetics in adult patients. The use of wax mock ups, or digital images of restored teeth may help the clinician explain the possible treatment options, and may help in achieving realistic expectations to avoid and dissatisfactions with the outcome of care.

An interesting issue raised by one patient was worries about their future plans and career. Dipboye *et al.* (1975) found that attractive individuals were more likely to be employed than those who were less attractive and McKiernan *et al.* (1992) found that improved career prospects were one of the main benefits anticipated by adult orthodontic patients.

4.6.3.6 Health and Dental Health Concerns

The final theme was dental health concerns. Some interviewees questioned the effectiveness of their brushing techniques and oral hygiene because they noticed that no matter how much effort they put in to brushing their teeth, they still could not improve the colour. Adult subjects with AI also reported feeling that they get “cavities” easier than others (Coffield *et al.*, 2005). Anagnostopoulos *et al.* (2011) found that stronger self-efficacy beliefs and greater perceived severity of oral diseases were related to increased tooth brushing frequency. These findings suggest that the majority of patients with AI are not aware that the colour of their teeth is not as a result of poor brushing technique but is a result of the condition. It is the clinicians’ responsibility to reinforce oral hygiene instructions, and help maintain positive attitude toward dental care whilst reassuring the patient that this is not their fault.

This also highlighted that AI needs to be explained more clearly to patients, and that further information may be required for patients and parents with AI.

5. Part Two of the Study: Questionnaire Development and Distribution

5.1 Materials and methods

The findings from the first part of the study suggested that AI can have a significant impact on children and adolescents and also demonstrate how beneficial it is to use qualitative methods to investigate personal thoughts and opinions. The results of these interviews allowed the development of a questionnaire to ascertain the views of more AI patients.

5.1.1 Questionnaire Development

The steps involved in designing a questionnaire as proposed by Williams (2003) were followed. The first step was defining the study population. The questionnaire was intended to be distributed to all AI patients attending the Eastman Dental Hospital, Unit of Paediatric Dentistry from January – June 2012.

The second step was formulating the questions and responses for the questionnaire. A combination of multichotomous and dichotomous responses were used and consisted of multiple choices, Likert scales, and simple yes or no answers. The questions and their responses were carefully designed based on the themes and subthemes identified from the interviews. The wording of the questions and responses was in child friendly language, in order to avoid ambiguity and be easily understood. Leading questions, double negatives, loaded words, and hypothetical questions were avoided (Black *et al.*, 1998).

It was important to ensure the wording of the questions was right before circulating the questionnaire, as this has a significant influence on the responses given (Larsen *et al.*, 1987). Some of the questions were given “Other” as an optional response followed by space to write any information, and there was additional space for any further comments or any unexpected responses at the end of the questionnaire. This was in order to give the participants a chance to express any issues or concerns that did not come up during the interviewing process. The questions with a Likert scale responses were grouped together in order to avoid any confusion for the patients and make it easy for them to answer the same format of questions altogether. The patients were clearly informed to choose only one answer for each question.

The third step was designing the layout of the questionnaire. The front page of the questionnaire included the title and a short introduction highlighting the purpose of the study. A guarantee of confidentiality was also given on the front page. A combination of brightly coloured fonts and pictures were used throughout the questionnaire to make it more attractive and child friendly. All instructions were in bold in order to assist the patients when answering the questions. Williams (2003) suggested that the questionnaire should be divided into sections in order to make it less intimidating for the patients and this recommendation was followed. The questionnaire consisted of fifteen items divided into three sections.

The questionnaire started with demographic questions and the more personal and sensitive questions were gradually introduced in the second and third sections. The questions in the second section were mainly derived from the subthemes related to aesthetics and function and included questions regarding shape, colour of teeth, and pain or sensitivity from certain foods or drinks. The third section of the questionnaire was developed based on the psycho-social aspects identified from the interviews and included questions about self consciousness, confidence, and comments by other people. A final question assessed whether there was a need for further information regarding AI, e.g. a dedicated website or online support group. The questionnaire was printed on good quality paper with Arial 14 font (Appendix 7).

The readability of the questionnaire was assessed in Microsoft Word using the Flesch Reading Ease Score and Flesch-Kincaid grade level. The reading age of the questionnaire was 8-9 years old, and was therefore considered acceptable for the age group under investigation in this study.

The patients were also asked to complete the Child Perception Questionnaire (CPQ11-14). The CPQ11-14 was developed by Jokovic *et al.* (2002) at the University of Toronto as a measure of Oral Health-Related Quality of Life (OHRQoL) for children. It is designed to assess a child's perceptions of the impact of oral disorders on physical and psycho-social functioning. The long version of the CPQ11-14 consisted of 37 questions, whilst the short version consisted of 20 questions. The CPQ11-14 covers subject areas such as oral symptoms, functional limitations, emotional limitations, and social well-being. The short version of the CPQ11-14 has been tested and validated (Foster *et al.*, 2008). The CPQ11-14 was distributed alongside the newly developed questionnaire in keeping with the recommendation to use generic and condition

specific oral health measures in studies of this type (Appendix 8). Both the AI questionnaire and the CPQ 11-14 were sent to the ethical committee once the AI questionnaire had been developed. This was in keeping with the original ethics approval which had asked for the questionnaire to be sent prior to use.

As described by Williams (2003), a pre-pilot was undertaken as an information-gathering exercise in which colleagues were asked to identify any possible modifications needed before distributing the pilot questionnaire to patients. Initially, the research group read the questionnaire and made amendments until all of the questions were considered to be acceptable to respondents. Seven versions of the questionnaire were developed and edited before piloting on six colleagues and three patients. Only three patients were included in the piloting phase in order to ensure more patients would be available for the next phase. The average time needed to complete the questionnaire was 1 minute and 47 seconds for colleagues and 2 minutes and 30 seconds for patients. The wording of the responses for question 8 was modified following the pilot in order to allow the patient to list any food or drinks that might be avoided either occasionally, often, or all of the time. The CPQ11-14 was continued at the end of the AI questionnaire in order to ensure the continuation of answering all questions and to avoid any confusion for the patients on which questionnaire should be answered first.

5.1.2 Questionnaire Distribution

This study was investigating opinions rather than generating a score for each individual patient. This meant a hypothesis was not being tested and sample size was less of an issue. Therefore, the aim was to identify as many AI patients as possible attending the Unit of Paediatric Dentistry at the Eastman Dental Hospital for their regular appointment from January to June 2012. The patients were identified by asking colleagues and staff about any AI patients attending their clinic every day, and also from the Dental Anomalies clinic. Each patient, and their parent, were given information leaflets (Appendices 3 and 4) and a full explanation about the project. If they agreed to participate, the consent form (Appendices 5 and 6) was signed and the patient was asked to complete the questionnaire and leave it in the collection box labelled "AI Questionnaire" sited in the reception area. In cases where the patients could not complete the questionnaire at the same time as their appointment for any reason, they were given a stamped addressed envelope, including the information leaflets and

consent forms, and asked to complete the questionnaire at home and return it by mail. Each questionnaire was coded with an ID number and the patients were divided in three groups: pre-treatment, mid-treatment, and post-treatment.

Due to initial difficulties with patient recruitment, AI patients not attending EDH within the time period were also identified and posted the questionnaire. A list of AI patients was identified from previous audits within the department. Members of staff were all asked to provide a list of their own AI patients. Each patient was mailed an envelope containing the questionnaire, consent forms, information leaflets, a stamped addressed envelope to return the completed questionnaire and consent form, and an explanatory cover letter (Appendix 10). All of the questionnaires and consent forms were coded accordingly. The parents/patients were asked to keep one copy of their consent form for their own records. A mailing list was created that included the patient's ID, date the questionnaire was sent, the date the questionnaire was received, and the stage of treatment the patient was in. If the patient did not reply within 2 weeks a reminder letter was sent.

5.2 Results for Questionnaire

5.2.1 Demographics

The questionnaire was distributed to 61 AI patients at the Unit of Paediatric Dentistry, Eastman Dental Hospital and the response rate was 66% (40/61). Of the 40 respondents, 25 patients completed their questionnaire whilst attending their regular dental appointment at the Unit of Paediatric Dentistry, and 15 (out of 33) returned questionnaires by mail.

Originally the patients were divided into three groups; pre, during and post treatment. Due to the limited number of responses from the post-treatment patient (1 respondent) it was decided to combine the 'during' and 'post-treatment' groups into one category (Treatment). The number of respondents in the two groups is shown in (Table 5.1)

Stage of Treatment	%	n
Pre-treatment	20	8
Treatment	80	32
Total	100	40

Table 5.1 Number of respondents by treatment stage.

The mean age was 13.2 years (range 10 to 16 years, SD 2.2 years). There were 19/40 (47.5%, mean age 13) females and 21/40 (52.5%, mean age 14) males.

The majority of participants 25/40 (62.5%) were Caucasians. The distribution of ethnic groups is illustrated in (Table 5.2).

Ethnic group	%	n
White	62.5	25
Asian	25.0	10
Black	10.0	4
Other	2.5	1

Table 5.2 Ethnicity of respondents

Q4. Do you remember noticing anything different about your teeth before your dentist sent you to this hospital?	Yes %(n)	No %(n)
	64.0% (25)	36.0%(14)

Table 5.3 Responses to Question 4. Total sample n=39

One patient did not answer Questions 4 hence the sample size was reduced to 39. There did not appear to be any difference between phase of treatment and gender.

Q5. Who first said it would be a good idea for you to have treatment for your teeth?	%(n)
Self	5.0% (2)
Mum/Dad	37.5% (15)
Dentist	55.0% (22)
GMP	2.5% (1)

Table 5.4 Responses to Question 5. Total sample n= 40

The most common response to question number 5 was a suggestion from the patient's dentist (55% of respondents).

Question 6 asked participants to assess their own level of confidence on a scale from (1) to (5), 1 being not at all confident and 5 being very confident and 19/40 (47.5%) ranked their confidence as (3). When examined between genders, there did not appear to be a difference in the responses between males and females. For ease of interpretation of the answers, categories 1 and 2 (not confident) and 4 and 5 (confident) were combined. The numbers of respondents scoring not confident or confident were similar in the treatment group (Figure 5.1).

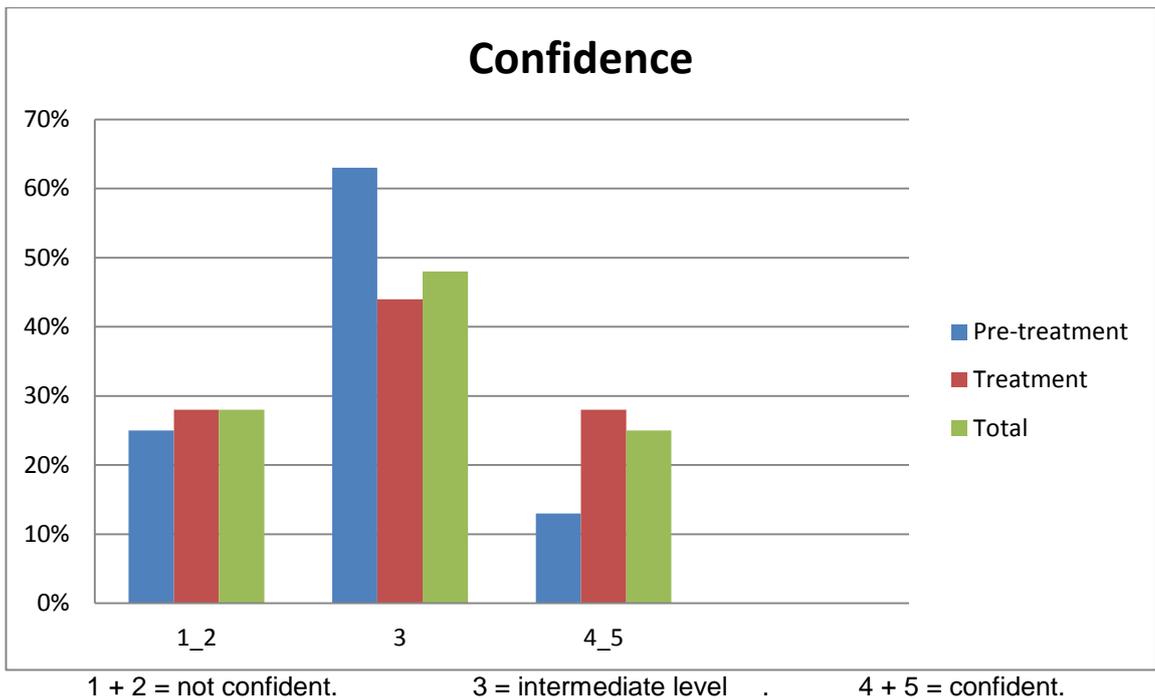


Figure 5.1 Responses to question 6 for Pre-treatment and Treatment groups. (scale from 1 to 5)

Question 7 assessed the importance of improving the colour, shape and size of the teeth the importance of reducing pain/sensitivity and improving the smile separately.

The results for improving the colour were examined by gender (Table 5.5) and between phases of treatment (Figure 5.2). Again categories 1 and 2 were combined (not important) and 4 and 5 (important) to ease interpretation.

		Importance of colour % (n)			Total % (n)
		Not important 1-2	3	Important 4-5	
Gender	Female	5.0% (1)	11.0% (2)	84.0% (16)	47.5% (19)
	Male	5.0% (1)	0.0% (0)	95.0% (20)	52.5% (21)
Total		5.0% (2)	5.0% (2)	90.0% (36)	100% (40)

Table 5.5 Importance of improving colour of teeth for female and male respondents.

The most common response was to improve the colour of teeth, with (90%) of the respondents stating that it was important.

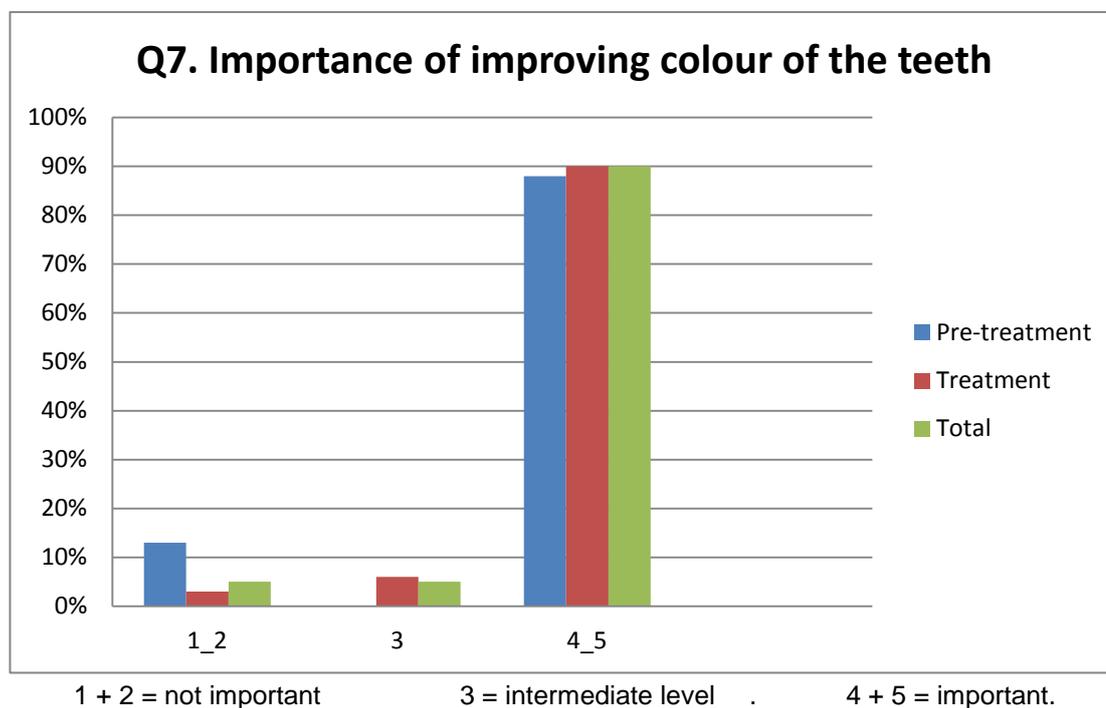


Figure 5.2 Importance of improving colour of the teeth for pre-treatment and treatment groups.

The total number of respondents who answered the rest of question 7 was 35, as 5 patients did not complete the question fully. Therefore the responses about the importance of improving shape, size, smile, and sensitivity was restricted to n=35 subjects.

There were no apparent differences in responses between stage of treatment (Table 5.6) and gender (Table 5.7) with regard to the importance of improving the shape of the teeth. The majority (74%) agreed that it was very important with only (11%) saying it was not important.

		Importance of treatment to improve the shape of the teeth % (n)			Total % (n)
		Not important		Important	
		1-2	3	4-5	
Phase	Pre-treatment	33.0% (2)	(0)	67.0% (4)	15.0% (6)
	Treatment	7.0% (2)	17.0% (5)	76.0% (22)	72.5% (29)
Total		11.0% (4)	14.0% (5)	74.0% (26)	87.5% (35)

Table 5.6 Importance of treatment to improve the shape of the teeth for pre-treatment and treatment groups.

	Importance of treatment to improve the shape of the teeth % (n)			Total % (n)
	Not important		Important	
	1-2	3	4-5	
Gender Female	12.0% (2)	12.0% (2)	76.0% (13)	42.5% (17)
Male	11.0% (2)	17.0% (3)	72.0% (13)	45.0% (18)
Total	11.0% (4)	14.0% (5)	74.0% (26)	87.5% (35)

Table 5.7 Importance of treatment to improve the shape of the teeth for female and male respondents.

In terms of the importance of treatment to improve the size of the teeth, the majority of patients (60%) indicated it was important and there appeared to be agreement between females (71%), as shown in table 5.9, and patients in the treatment group (69%) on this issue (table 5.10).

	Importance of treatment to improve size of teeth % (n)			Total % (n)
	Not important		Important	
	1-2	3	4-5	
Phase Pre-treatment	33.0% (2)	50.0% (3)	17.0% (1)	15.0% (6)
Treatment	10.0% (3)	21.0% (6)	69.0% (20)	72.5% (29)
Total	14.0% (5)	26.0% (9)	60.0% (21)	87.5% (35)

Table 5.8 Importance of treatment to correct size of the teeth for pre-treatment and treatment groups.

	Importance of treatment to improve size of teeth % (n)			Total % (n)
	Not important		Important	
	1-2	3	4-5	
Gender Female	12.0% (2)	18.0% (3)	71.0% (12)	42.5% (17)
Male	17.0% (3)	33.0% (6)	50.0% (9)	45.0% (18)
Total	14.0% (5)	26.0% (9)	60.0% (21)	87.5% (35)

Table 5.9 Importance of treatment to improve size of teeth for female and male respondents.

	Importance of improving smile % (n)			Total % (n)
	Not important		Important	
	1-2	3	4-5	
Phase Pre-treatment	17.0% (1)	17.0% (1)	67.0% (4)	15.0% (6)
Treatment	3.0% (1)	17.0% (5)	79.0% (23)	72.5% (29)
Total	6.0% (2)	17.0% (6)	77.0% (27)	87.5% (35)

Table 5.10 Importance of improving the smile for pre-treatment and treatment groups.

The majority of patients wanted treatment to enhance their smile and saw this as being important, with 77% of all respondents stated this was a reason for pursuing treatment (table 5.11).

	Importance of improving smile % (n)			Total % (n)
	Not important		Important	
	1-2	3	4-5	
Gender Female	6.0% (1)	24.0% (4)	71.0% (12)	42.5% (17)
Male	6.0% (1)	11.0% (2)	83.0% (15)	45.0% (18)
Total	6.0% (2)	17.0% (6)	77.0% (27)	87.5% (35)

Table 5.11 Importance of improving the smile for female and male respondents.

The majority of patients also wanted to have treatment to reduce pain and sensitivity, a total of 74% said this was important to them (tables 5.12 & 5.13).

	Importance of reducing pain/sensitivity % (n)			Total % (n)
	Not important		Important	
	1-2	3	4-5	
Phase Pre-treatment	50.0% (3)	17.0% (1)	33.0% (2)	15.0% (6)
Treatment	14.0% (4)	3.0% (1)	83.0% (24)	72.5% (29)
Total	20.0% (7)	6.0% (2)	74.0% (26)	87.5% (35)

Table 5.12 Importance of reducing pain/sensitivity for pre-treatment and treatment groups.

		Importance of reducing pain/sensitivity % (n)			Total % (n)
		Not important		Important	
		1-2	3	4-5	
Gender	Female	18.0% (3)	6.0% (1)	76.0% (13)	42.5% (17)
	Male	22.0% (4)	6.0% (1)	72.0% (13)	45.0% (18)
Total		20.0% (7)	6.0% (2)	74.0% (26)	87.5% (35)

Table 5.13 Importance of reducing pain/sensitivity for female and male respondents.

The summary of responses for question 7 is shown in Figure 5.3, highlighting that all treatment aims were considered important by the majority of respondents.

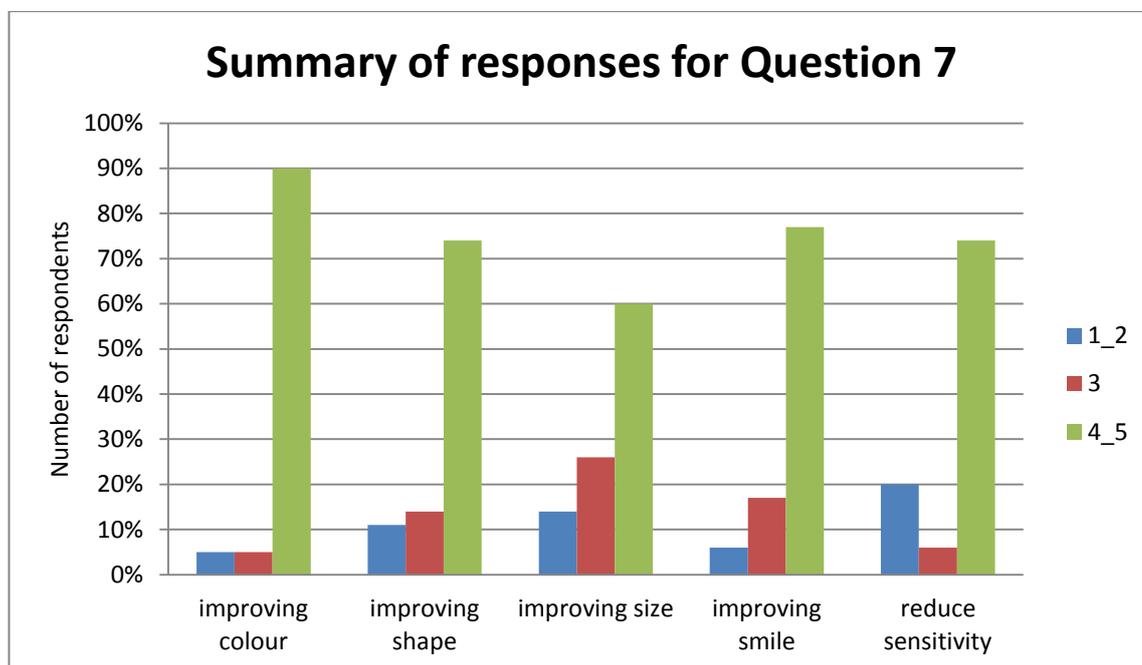


Figure 5.3 Importance of treatment aims on a scale from 1-5 (1 being not important at all and 5 being very important).

Question 8 asked respondents if there were any foods or drinks they avoided. Very few participants reported avoiding foods all of the time, with never and occasionally being the most common responses, both by gender (Table 5.14) and treatment phase (Table 5.15)

		Avoid food or drinks % (n)				Total % (n)
		Never	Occasionally	Often	All of the time	
Gender	Female	42.0% (8)	42.0% (8)	11.0% (2)	5.0% (1)	47.5% (19)
	Male	43.0% (9)	19.0% (4)	33.0% (7)	5.0% (1)	52.5% (21)
Total		43.0% (17)	30.0% (12)	23.0% (9)	5.0% (2)	100% (40)

Table 5.14 Responses to question 8 for female and male respondents.

		Avoid food or drinks % (n)				Total % (n)
		Never	Occasionally	Often	All of the time	
Phase	Pre-treatment	50.0% (4)	25.0% (2)	13.0% (1)	13.0% (1)	20.0% (8)
	Treatment	40.0% (13)	31.0% (10)	25.0% (8)	3.0% (1)	80.0% (32)
Total		43.0% (17)	30.0% (12)	23.0% (9)	5.0% (2)	100% (40)

Table 5.15 Responses to question 8 for pre-treatment and treatment groups.

Question 9 asked respondents about their experiences of teasing and 50% of respondents reported being teased at least “occasionally” (Table 5.17). Amongst male respondents, 24% reported being teased “often” in comparison with 11% of females (Table 5.17).

		Teased % (n)				Total % (n)
		Never	Occasionally	Often	All of the time	
Phase	Pre-treatment	38.0% (3)	25.0% (2)	38.0% (3)	0.0% (0)	20.0% (8)
	Treatment	22.0% (7)	56.0% (18)	13.0% (4)	9.0% (3)	80.0% (32)
Total		25.0% (10)	50.0% (20)	18.0% (7)	8.0% (3)	100% (40)

Table 5.16 Responses to question 9 for pre-treatment and treatment groups.

		Teased % (n)				Total % (n)
		Never	Occasionally	Often	All of the time	
Gender	Female	16.0% (3)	58.0% (11)	11.0% (2)	16.0 % (3)	47.5% (19)
	Male	33.0% (7)	43.0% (9)	24.0% (5)	0.0% (0)	52.5% (21)
Total		25.0% (10)	50.0% (20)	18.0% (7)	8.0% (3)	100% (40)

Table 5.17 Responses to question 9 for female and male respondents.

Question 10 asked patients about avoiding smiling. The most common response for question 10 was that the respondents “occasionally” avoided smiling (30%), although 23% avoided smiling “often” or “all of the time”. Females appeared to avoid smiling “all of the time” more often (32%) than males (14%) as shown in Table 5.18. The treatment group had similar responses for “never”, “occasionally”, “often” and “all the time” as shown in table 5.19.

		Avoid smiling % (n)				Total % (n)
		Never	Occasionally	Often	All of the time	
Gender	Female	32.0% (6)	21.0% (4)	16.0% (3)	32.0% (6)	47.5% (19)
	Male	19.0% (4)	38.0% (8)	29.0% (6)	14.0% (3)	52.2% (21)
Total		25.0% (10)	30.0% (12)	23.0% (9)	23.0% (9)	100% (40)

Table 5.18 Responses to question 10 for female and male respondents.

		Avoid smiling % (n)				Total % (n)
		Never	Occasionally	Often	All of the time	
Phase	Pre-treatment	38.0% (3)	38.0% (3)	13.0% (1)	13.0% (1)	20.0% (8)
	Treatment	22.0% (7)	28.0% (9)	25.0% (8)	25.0% (8)	80.0% (32)
Total		25.0% (10)	30.0% (12)	23.0% (9)	23.0% (9)	100% (40)

Table 5.19 Responses to question 10 for pre-treatment and treatment groups.

Question 11 asked participants about their tendency to “feel different” to their peers. Almost half of patients (45%) indicated that they never felt different or “left out”, although (30%) felt occasionally left out and (20%) often felt different. Females indicated feeling “often” left out more often than males (32% of 10%), but in contrast males felt left out “all of the time” more often than females (10% of 0%) as shown in Table 5.20. Fewer respondents in the pre-treatment group felt different, than in the

treatment group (table 5.21), although the small numbers involved meant this could not be tested for significance.

		Feel different % (n)				Total % (n)
		Never	Occasionally	Often	All of the time	
Gender	Female	32.0% (6)	37.0% (7)	32.0% (6)	0.0% (0)	47.5% (19)
	Male	57.0% (12)	24.0% (5)	10.0% (2)	10.0% (2)	52.2% (21)
Total		45.0% (18)	30.0% (12)	20.0% (8)	5.0% (2)	100% (40)

Table 5.20 Responses to question 11 for female and male respondents.

		Feel different % (n)				Total % (n)
		Never	Occasionally	Often	All of the time	
Phase	Pre-treatment	50.0 % (4)	38.0% (3)	0.0% (0)	13.0 % (1)	20.0% (8)
	Treatment	44.0% (14)	28.0% (9)	25.0% (8)	3.0 % (1)	80.0% (32)
Total		45.0% (18)	30.0% (12)	20.0% (8)	5.0 % (2)	100% (40)

Table 5.21 Responses to question 11 for pre-treatment and treatment groups.

Question 12 asked respondents if they worried about future plans. The majority of patients (55%) indicated that they never worried that their teeth may affect their future plans. Although (25%) admitted to occasionally worries (Table 5.22). There were no obvious differences between males and females (table 5.23).

		Worry about future plans % (n)				Total % (n)
		Never	Occasionally	Often	All of the time	
Phase	Pre-treatment	63.0% (5)	25.0% (2)	0.0% (0)	13.0% (1)	20.0% (8)
	Treatment	53.0% (17)	25.0% (8)	9.0% (3)	13.0% (4)	80.0% (32)
Total		55.0% (22)	25.0% (10)	8.0% (3)	13.0% (5)	100% (40)

Table 5.22 Responses to question 12 for pre-treatment and treatment groups.

		Worry about future plans % (n)				Total % (n)
		Never	Occasionally	Often	All of the time	
Gender	Female	58.0% (11)	21.0% (4)	5.0% (1)	16.0% (3)	47.5% (19)
	Male	52.0% (11)	29.0% (6)	10.0% (2)	10.0% (2)	52.2% (21)
Total		55.0% (22)	25.0% (10)	8.0% (3)	13.0% (5)	100% (40)

Table 5.23 Responses to question 12 for female and male respondents.

Question 13 asked if their dental problems had any effect on sports and hobbies. The most common response to question 13 was that AI never affected hobbies (80%) and there did not appear to be any obvious differences between gender or phase of treatment (Tables 5.24 & 5.25).

		Affect hobbies % (n)			Total % (n)
		Never	Occasionally	Often	
Gender	Female	79.0% (15)	11.0% (2)	11.0% (2)	47.5% (19)
	Male	81.0% (17)	10.0% (2)	10.0% (2)	52.2% (21)
Total		80.0% (32)	10.0% (4)	10.0% (4)	100% (40)

Table 5.24 Responses to question 13 for female and male respondents.

		Affect hobbies % (n)			Total % (n)
		Never	Occasionally	Often	
Phase	Pre-treatment	88.0% (7)	0.0% (0)	13.0% (1)	20.0%(8)
	Treatment	78.0% (25)	13.0% (4)	9.0% (3)	80.0% (32)
Total		80.0% (32)	10.0% (4)	10.0% (4)	100% (40)

Table 5.25 Responses to question 13 for pre-treatment and treatment groups.

Question 14 asked respondents the single most important thing they wanted from treatment. The most common response for question 14 was to improve the colour (63%), with improving the smile (18%), and reducing sensitivity (10%) being 2nd and 3rd choices (Figure 5.4). When examined between genders, more males selected to improve the colour than females (86% of 37%), while more females chose to improve the smile than males (32% of 5%) as shown in Figure 5.5 and table 5.26.

		What is the most important thing you would like from the treatment of your teeth % (n)					Total % (n)
		improve the colour	improve the size	improve smile	reduce sensitivity	feel better about myself	
Gender	Female	37.0% (7)	5.0% (1)	32.0% (6)	16.0% (3)	11.0% (2)	47.5%(19)
	Male	86.0% (18)	5.0% (1)	5.0% (1)	5.0% (1)	0.0% (0)	52.2% (21)
Total		63.0% (25)	5.0% (2)	18.0% (7)	10.0% (4)	5.0% (2)	100% (40)

Table 5.26 Responses to question 14 for female and male respondents.

		What is the most important thing you would like from the treatment of your teeth % (n)					Total % (n)
		improve the colour	improve the size	improve smile	reduce sensitivity	feel better about myself	
Phase	Pre-treatment	75.0% (6)	13.0% (1)	0.0%(0)	0.0%(0)	13.0% (1)	20.0% (8)
	Treatment	59.0% (19)	3.0% (1)	22.0% (7)	13.0% (4)	3.0% (1)	80.0%(32)
Total		63.0% (25)	5.0% (2)	18.0% (7)	10.0% (4)	5.0% (2)	100%(40)

Table 5.27 Responses to question 14 for pre-treatment and treatment groups.

Interestingly, improving the colour and size of the teeth were selected most frequently in the pre-treatment group (75% and 13% respectively), with improving the smile and reducing sensitivity not being selected. In the treatment group, improving the colour and smile plus reducing sensitivity were all selected, as shown in (Table 5.27).

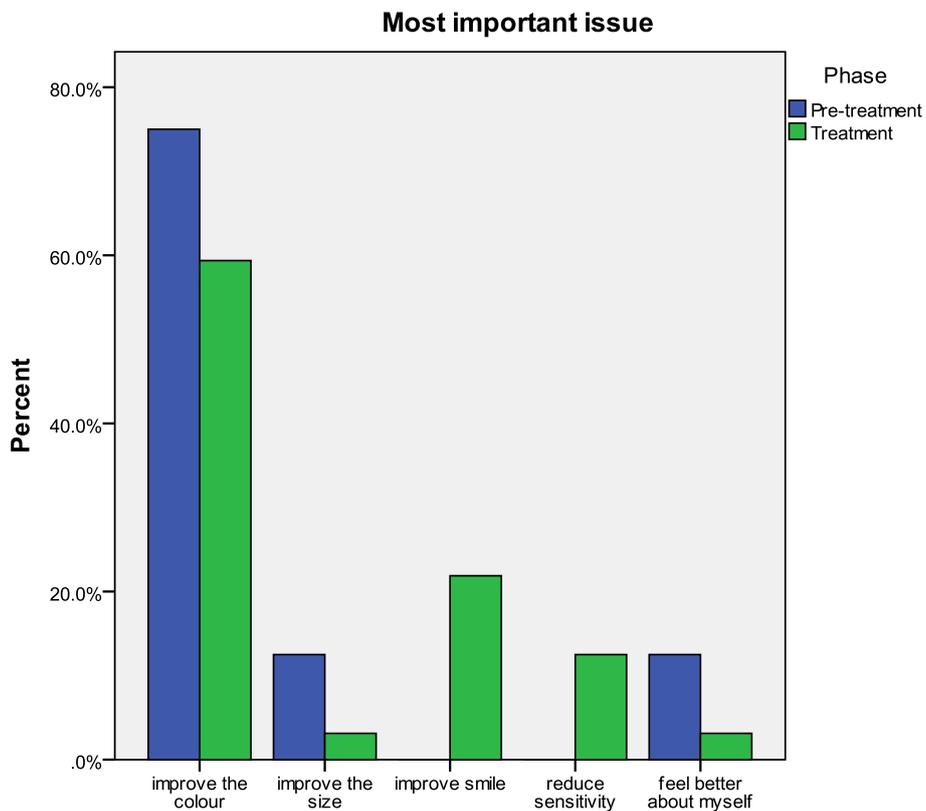


Figure 5.4 Responses to question 14 regarding the single most important aim of treatment between treatment groups.

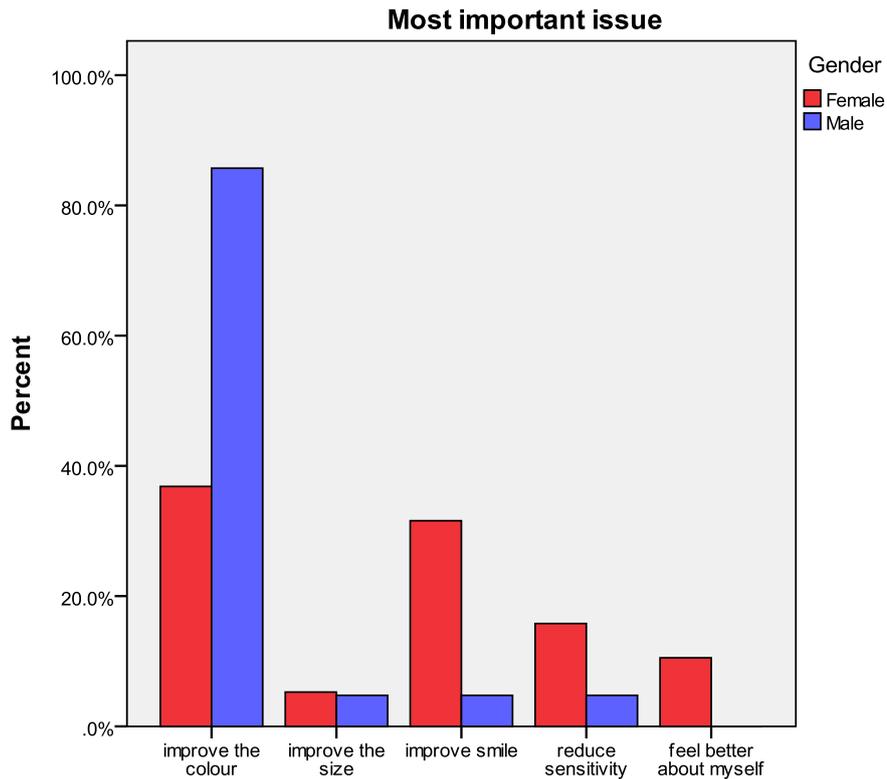


Figure 5.5 Responses to question 14 regarding the single most important aim of treatment between genders.

The final question, asked patients if they thought it would be useful to have a website or “support group” for patients with AI, (85%) answered yes and (15%) answered no.

Child Perception Questionnaire (CPQ)

The Child Perception Questionnaire (CPQ) contained 20 questions and each question had five responses. The scores for each response was 0= ‘Never’, 1= ‘Once or twice’, 2= ‘Sometimes’, 3= ‘Often’, and 4= ‘Every day or almost every day’. The maximum score for each question was 4, therefore the maximum possible total score for the CPQ was 80. A high score on the CPQ indicated a greater impact on the patient’s life. All 40 patients completed the CPQ with no missing answers.

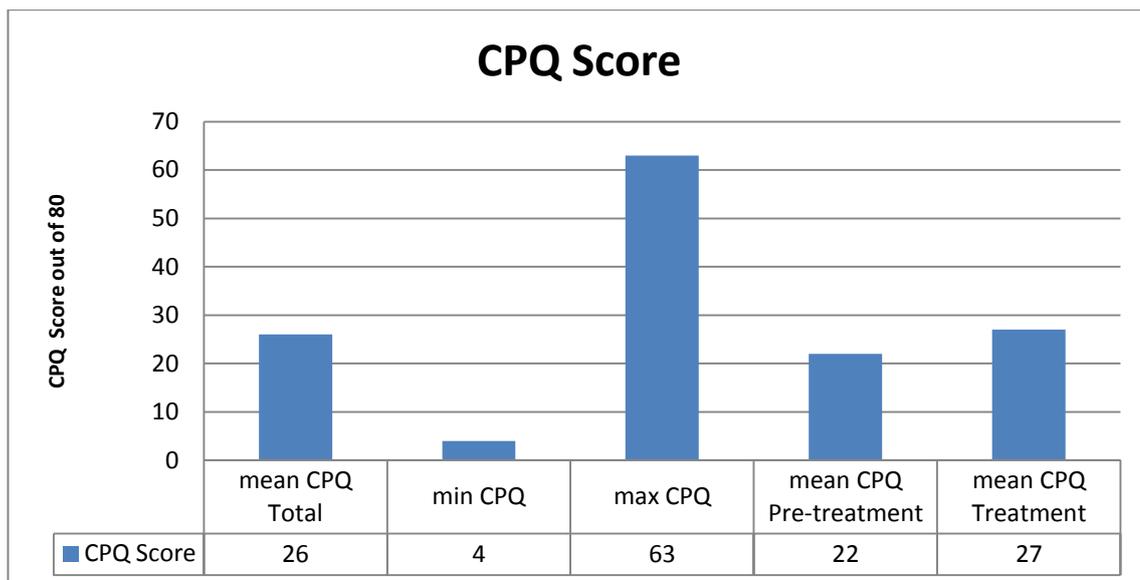


Figure 5.6 Total CPQ scores

The scores of the CPQ ranged from 4 to 63 with similar distributions between the Pre-treatment and Treatment groups. When examined between genders, females had a slightly higher mean score than males (females 28, males 24), as shown in Figure 5.6. The data was not subjected to further statistical analysis due to the small numbers involved.

However, the relationship between CPQ score and level of confidence was explored (Table 5.28). The higher level of confidence reported by the patients matched with the lower CPQ score (16), indicating a lower impact on quality of life, whereas the respondents who were not confident scored higher CPQ score (38), suggesting a greater impact on quality of life.

Confidence level	Not confident		Confident
	1+2	3	4+5
Mean CPQ score (+SD)	38 (8)	24 (14)	16 (10)

Table 5.28 Comparison between the mean CPQ score (+ standard deviation), with the level of confidence reported by respondents.

The mean value score for each CPQ question are listed in Table 5.29.

CPQ questions	Mean score value (Min=0 Max=4)
1- Would you say the health of your teeth, lips, jaws and mouth is	2.68
2- How much does the condition of your teeth, lips, jaws or mouth affect your life overall?	1.80
In the past 3 months, how often have you had:	
3- Sores in your mouth?	1.25
4- Bad Breath?	1.25
5- Food stuck in between your teeth?	1.95
6- Difficulty biting or chewing food like apples, corn on the cob or steak?	1.65
7- Difficult to drink or eat hot or cold foods?	1.25
8- Difficulty saying any words?	0.23
9- Trouble sleeping?	0.38
10- Pain in your teeth, lips, jaws or mouth?	1.45
11- Taken longer than others to eat a meal?	1.13
12- Felt irritable or frustrated?	1.00
13- Felt shy or embarrassed?	1.58
14- Been upset?	1.08
15- Been concerned what other people think about your teeth, lips, mouth or jaws?	1.83
16- Avoided smiling or laughing when around other children?	1.30
17- Not wanted to speak or read out loud in class?	0.95
18- Other children teased you or called you names?	0.83
19- Had other children ask you questions about your teeth, lips, jaws or mouth?	1.30
20- Argued with other children or your family?	1.02

Table 5.29 Mean score value for each CPQ question.

5.4 Discussion

5.4.1 Questionnaire Development

When designing the questionnaire the researcher used a combination of brightly coloured fonts and pictures throughout the questionnaire to make it more attractive and child friendly. The colour choices were carefully selected to avoid any colours that might be challenging for people who are colour blind.

The short version of the CPQ11-14 was used as it is quick and easy for children to complete. Initially the CPQ11-14 and the AI questionnaire were given as separate documents during the piloting stage, but in order to avoid any confusion and to make sure that the participants answered both questionnaires at the same time the CPQ11-14 was attached at the end of the AI questionnaire and the participants were given instructions to turn to the next page and complete the CPQ11-14. This ensured the answering of both questionnaires as one unit and made the process of data collection easier.

5.4.2 Response rate and sample size

A response rate of 66% (40/61) was achieved for the second part of this study. Williams (2003) considered a response rate of 75% to be “extremely good”. In a similar study looking at the psychosocial impact of AI on adults and adolescents patients the response rate was 44.1% (Coffield *et al.*, 2005). In another study looking at the children’s perspectives in the management of visible enamel defect via developed questionnaire the response rate was 72% (Rodd *et al.*, 2010). Whilst this did not represent all of the AI patients in the Department, a fair cross section of patients was achieved. Of the 25 questionnaires distributed to patients in the clinic as they attended their booked appointments, all of the questionnaires were completed and returned on the same day (100%). However, only 14 of the 33 mailed questionnaires were returned (42%). These findings indicate that the probability of the patients completing and returning a questionnaire is much higher when patients are approached personally and asked to complete the questionnaire.

It was very difficult to identify AI patients within the department since a complete database for all AI patients who attend the Department did not exist. The number of patient in the pre-treatment group (n=8) was small because the lower age for this project was 10 years, so it was difficult to find patients who had never had any restorative treatment by that age as most of the patients who were referred to the Eastman Dental Hospital had some treatment before their referral. The post treatment

group (n=1) was also small due to the nature of AI, it was difficult to identify a group of patients who had completed active treatment. In addition, most of the post-treatment patients were either discharged from the department once the treatment was completed or they were transferred to adult services. Therefore, as many AI patients as possible were recruited to participate in this study within the time constraints, and it was decided to have only two groups: pre-treatment and treatment.

5.4.3 Results

Demographics

There were a similar number of males and females in this study (47% females and 52.5% males). This differs from research in other areas, such as Orthodontics, where there is often a predominance of female participants (Khan and Harrocks, 1991). However, when Coffield *et al.* (2005) studied the psychological impact of AI on adult patients they found a similar gender distribution among AI patients (43% females and 56% males). The similar female to male ratio suggests that there are no differences between genders when it comes to seeking treatment for developmental enamel defects.

Question 4 (Table 5.3)

The majority of patients (64%) stated that they had noticed something different about their teeth before they were referred to the EDH for treatment, which suggests that this type of dental anomaly cannot be over looked and children can identify such dental defects. This is in accordance with the findings of Rodd *et al.* (2010) who found that children could assess the colour of their teeth before and after treatment. This finding is also comparable with that of Burden *et al.* (1995), who found that children were aware of their own dental aesthetics irrespective of gender or social background.

Question 5 (Table 5.4)

Approximately half of the patients (55%) stated that it was their general dental practitioner who had initially suggested treatment for their teeth. This is in accordance with the findings from the first part of the study where the majority of AI patients interviewed indicated that it was their dentist who told them about their AI. This highlights the fact that dentists needs to be well informed regarding AI, and be able to explain the nature of this condition in a way that young children understand. Other

people who suggested treatment were the patient's parents (37.5%), the patients themselves (5%) and the GMP (2.5%)

Question 6 (Figure 5.1)

The mean confidence score for AI patients was 3 on a scale of 1 – 5 (47.5%). Matell and Jacoby (1972) found that on a five point Likert scale, an average of 20% of respondents chose the mid-point (3) but the percentages in this study were much higher. There were no obvious differences found between males and females with respect to confidence. It was not considered appropriate to statistically analyse these findings due to the small numbers involved. However, more post treatment respondents selected 4 or 5 as a response (28%) than pre-treatment (12.5%). This does suggest there may be an improvement in confidence when treatment starts, and this is worthy of further study. According to Rodd *et al.* (2010) children who received dental treatment in the last 12 months reported being more confident following treatment of visible enamel defects. This suggests that children need to be followed for a reasonable period after their treatment to assess their level of confidence. Coffield *et al.* (2005) found self-esteem scores were similar for patients with and without AI, but self esteem and confidence are different psychological concepts so should not be directly compared.

Question 7 (Tables 5.5-5.12 and Figures 5.2 and 5.3)

Improving the colour of teeth was the most important overall reason to have treatment for AI (90%) which was in agreement with the findings of the in-depth interviews, where all interviewed patients highlighted improving the colour of their teeth as an important expectation from their treatment. Similarly Coffield *et al.* (2005) reported that the majority of AI patients were unhappy with the colour of their teeth (79%). Improving the size of the teeth was also seen as important, but by fewer of the respondents; (60%) said this was important. Improving the smile was again important (77%), and a similar percentage (74%) saw a reduction in sensitivity as important. In recent years, people have become more concerned about dental aesthetics and more aware of their smile, and are keen to improve the appearance of their teeth (Kerosuo *et al.*, 1995). Dental appearance is considered to be very important socially (Shaw, 1981). Clinicians must appreciate the importance of dental aesthetics particularly with young AI patients, since it is well documented that appearance is the most valued characteristic among young

dental patients (Prokhorov *et al.*, 1993). The desire to improve the smile and appearance have been considered motivating factors for seeking dental treatment (McKiernan *et al.*, 1992).

Question 8 (Table 5.13 and 5.14)

Just under half (43%) of the patients reported that they did not avoid any food or drinks because of their teeth, although (30%) reported occasionally avoiding food or drinks, and (23%) often. When examined closely between genders and phases of treatment, more females (42%) than males (14%) indicated that they occasionally avoided food or drinks, but more males reported often avoiding food or drinks (33% males and 11% females). It is difficult to draw conclusions between pre-treatment and treatment groups as there were only 8 patients in the pre-treatment group. However, this is an important area to look at in future studies. The possibility of needing to avoid certain food or drinks in both groups is difficult for patients and something which should be discussed with them when embarking on treatment, as treatment may not solve this completely. The issue of pain and sensitivity was well highlighted by AI patients during the interview stage. In addition, more adult AI patients reported pain and sensitivity compared to unaffected individuals (Coffield *et al.*, 2005).

Question 9 (Table 5.16 and 5.17)

Responses for question 9 showed that 50% of AI patients were occasionally teased about their teeth. Males were teased "often" more frequently than females (24% cf 11%), but females were teased "all of the time" more frequently than males (16% cf 0%). Patients in the pre-treatment group were "often" teased about their teeth (38%) more frequently than patients in the treatment group (13%). The majority of interviewees in the first part of this study reported a history of teasing and were distressed by this. A teasing history has also been reported to affect adult AI patients (93.3%) as found by Coffield *et al.* (2005). This was also found with respect to children with malocclusions, who were found to be more susceptible to teasing by their peers (Shaw *et al.*, 1980). Marshman *et al.* (2008) found that some young people had experienced teasing and name calling about their developmental enamel defects and a teasing history is considered an influencing factor for seeking treatment in orthodontics (Shaw *et al.*, 1980; DiBiase and Sandler, 2001) Pabari *et al.* (2008) found that 45.9% of adult orthodontics patients seeking treatment had a history of teasing or negative

comments about their dental appearance. It has been suggested that treating patients who have history of teasing about their appearance can yield a great benefit to their psychosocial well-being (Helm *et al.*, 1985). However, this is not something which can ever be guaranteed. The findings do also illustrate the importance of highlighting this issue with patients and offering support and counselling should it be required.

Question 10 (Table 5.18 and 5.19)

The percentage of patients who indicated that they occasionally avoided smiling because of their teeth was 30%, and approximately equal numbers replied never (25%) or often (23%). When examined between genders, females were more likely to avoid smiling “all of the time” than males (32% cf 14%). Whereas males reported avoiding smiling “often” more frequently than females (29% cf 16%). There was no major difference in the distribution of responses for this question between the two stages of treatment. This issue was highlighted during the interview stage of this study where patients mentioned avoiding smiling, particularly when having photos or videos taken. This is in accordance with the findings of Marshman *et al.* (2008), where subjects expressed feelings of self-consciousness and not wanting to show their teeth. In another study, children with untreated enamel defects were also found to be reluctant to smile (Rodd *et al.*, 2010). When assessing the quality of life following childhood dental injuries the issue of avoiding smiling or laughing when amongst peers was raised by children who had sustained dental trauma (Porritt *et al.*, 2010).

Question 11 (Table 5.20 and 5.21)

Almost half of patients (45%) indicated that they never felt different, isolated or “left out”, 32% of females indicated that they often feel different compared with 10% of males. However, males felt different “all of the time” more frequently than females (10% cf 0%). This is in accordance with the findings from adult AI patients where they demonstrated high levels of social avoidance and distress (Coffield *et al.*, 2005). Another study looking at Chinese patients with clefts found that they exhibited higher levels of social avoidance and distress than unaffected subjects (Berk *et al.*, 2001). This tendency to feel “different” may mean that AI patients are more distressed and anxious in social interactions and this may result in a threat to social interactions in adulthood.

Question 12 (Table 5.22 and 5.23)

The majority of patients (55%) indicated that they were not worried about future plans and career opportunities. However, a quarter of the patients (25%) stated that they occasionally worried that their teeth may affect their future plans. McKiernan *et al.* (1992) found that adult orthodontic patients saw improved career prospects as one of the main benefits of treatment. Dipboye *et al.* (1975) found that attractive individuals were more likely to be employed than those who were less attractive. Therefore AI children and adolescents may be right in their concerns regarding future plans and this may lead to them having higher expectations of the outcomes of treatment. This is something which should be discussed at the outset of treatment.

Question 13 (Table 5.24 and 5.25)

The most common response to question 13 was that AI did not affect sports and hobbies (80%) with no major differences between gender or phase of treatment. This question should perhaps be clarified in future studies if this questionnaire was used, it would be useful to investigate what type hobbies AI patients may feel they have to avoid. This would be a useful amendment for future work.

Question 14 (Table 5.26 and 5.27)

The most commonly perceived benefits of treatment were to improve the colour of the teeth (63%), to improve the smile (18%), and to reduce sensitivity (10%). When examined between genders, more females (32%) chose improving the smile than males (5%). This was also noted from the first part of this study during the interviewing stage. Coffield *et al.* (2005) found that adults with AI were more unhappy with the colour of their teeth (79.3%) than subjects without AI (32.1%). The clinical implication regarding the importance of dental aesthetics in AI patients is that high levels of concerns may lead to higher expectations of better aesthetics following treatment, and clinicians should clearly discuss the limitations of treatment.

It is important for the clinician to appreciate that appearance is very important among children and young adults, and to manage the patient's expectations by understanding the motivation and thereby hopefully achieving better levels of satisfaction with treatment outcome (Abdel-Kader, 2006). Clinicians must discuss the objectives and limitations of treatment at the outset in order to achieve fully informed consent, set

realistic expectations and hopefully to avoid any dissatisfaction with the outcome of care (Cunningham *et al.*, 1996). Aesthetic dental treatment for children may yield important psychosocial benefits (Rodd *et al.*, 2010), but this does rely on patients being appropriately prepared and having realistic expectations.

Question 15

Eighty five percent of patients answered 'yes' when asked about having a website or support group for patients with the same dental condition. This highlighted the wish for resources to gain more information about AI. Other ways to provide information include using information leaflets, articles in magazine, use of social networking, videos and DVDs.

The CPQ (Table 28 and Figure 5.6)

The CPQ had originally been included in the study to aid validation of the questionnaire developed from the interviews. Unfortunately due to the small number of patients recruited in the pre-treatment group this was not feasible.

The CPQ is designed to assess the impact of oral conditions on children and was therefore appropriate for the research topic posed in this study. It is considered useful for comparisons between populations but unfortunately has limited ability to measure the effects of a certain condition (Cunningham *et al.*, 2000). The responses to the questionnaire were analysed in terms of the mean scores of the CPQ. There was a relatively small difference in the mean score for the pre-treatment group and treatment group, however this may be a reflection of the small sample size. There was however a noticeable relation between the high score of CPQ and the low level of confidence, which suggests that AI patients with low confidence may experience a great psychosocial impact.

6.0 Summary

A questionnaire was developed and was intended to be distributed to all AI patients attending the Unit of Paediatric Dentistry, Eastman Dental Hospital. All the questions and their responses in the questionnaire were based on the themes and subthemes identified from the in-depth interviews from the first part of the study. The patients were identified by asking colleagues and staff about any AI patients attending their clinic every day, and also from the Dental Anomalies clinic. Forty patients out of sixty one completed the questionnaire. The patients were divided into two groups; pre-treatment and treatment. The majority of patients (64%) reported noticing something different about their teeth prior to their referral to the Paediatric Department. The confidence level of AI patients seems to be mostly in the intermediate level between “not confident” and “confident”. Improving the colour of the teeth was the most common response by AI patients, with (90%) of the respondents stating that was important. Enhancing the smile was seen as being important and (77%) of all respondents stated this was a reason for pursuing treatment. Interestingly, the desire to reduce sensitivity and pain came third after the importance of improving the colour of the teeth and smile, with 74% stating this as an important issue. A small number of patients (20%) reported often feeling different or “left out”. A quarter of the patients (25%) stated that they occasionally worried that their teeth may affect their future plans. It seemed that AI did not affect sports and hobbies of patients, although this area needs further investigation in the future. Improving the colour of the teeth was the most commonly perceived benefits of treatment (63%) followed by improving the smile (18%), and to reduce sensitivity (10%). There was an overwhelming desire (85%) by patients to have a website or a support group for AI patients to gain more information about AI.

7.0 Conclusion for Part One and Part Two of the study

A total of 47 patients were included in this study and the following findings were noted:

- 1) In part one and two, children and young adults were found to have impacts due to their AI, in terms of concerns regarding aesthetics, function, and psychosocial issues.
- 2) In part two, the small number of pre-treatment patients makes such group comparisons inappropriate. However the following conclusions may be drawn:
 - (i) Improving the colour of the teeth, improving the smile and reducing dental sensitivity were all perceived to be important aims of treatment.
 - (ii) The single most important aim was improvement in colour.
- 3) In both parts one and two of the study, the issue of teasing was evident. This can have long term consequences for patients and appropriate ways of offering support and counselling in such situations should be available.
- 4) Patients said they would value additional information about AI, such as from a website or support group.

This study has an implication in understanding the impact of Amelogenesis Imperfecta on children and young adults. As clinicians it is important to understand and manage the patient's expectations and discuss the objectives and limitations of treatment to achieve fully informed consent and avoid any dissatisfaction with the outcome of care.

Indeed, the impact of Amelogenesis Imperfecta on children and young adults will influence the assessment of the effectiveness of treatment, and should include multiple factors such as normative, psychological and social factors.

Suggestions for Future Research

- 1- Repeat the study on a larger scale with a multi-centre study to assess whether the findings for this cohort of patients are comparable with other AI patients and to allow such group comparisons.
- 2- To assess the validity (mainly internal validity) and the reliability of the designed questionnaire via repeating the same questionnaire on a large cohort of AI patients.
- 3- To undertake a longitudinal study to investigate effects of treatment on AI patients.
- 4- It would also be useful to compare the impact of AI on patients with different types of AI, but this would need to be a multi-centre study to allow adequate numbers to be recruited.
- 5- It would be valuable to further investigate the impact of AI on patients who never had any treatment for their teeth; however it would be difficult to obtain adequate sample size.
- 6- The other ways to provide information including websites or online support groups, using information leaflets, articles in magazine, use of social networking, videos and DVDs needs to be explored.
- 7- It would be beneficial to investigate the impact of other developmental dental defects e.g. *Dentinogenesis Imperfecta*, and compare it to the findings of this study.

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Appendices

Appendix 1: Ethical Approval

South East London REC 3

(formerly King's College Hospital Research Ethics Committee)
1st Floor Camberwell Building
King's College Hospital
94 Denmark Hill
London
SE5 9RS
Telephone: 020 3311 7227
Facsimile: 020 3311 7280

Miss Susan Parekh
Clinical Lecturer
UCL Eastman Dental Institute
Unit of Paediatric Dentistry
UCL Eastman Dental Institute
256 Gray's Inn Road, London
WC1X 8LD

28 October 2010

Dear Miss Parekh

Full title of study: **How do children with Amelogenesis Imperfecta (AI) feel about the appearance of their teeth?**
REC reference number: **10/H0808/156**

Thank you for your application for ethical review, which was received on 22 October 2010. I can confirm that the application is valid and will be reviewed by the Committee at the meeting on 03 November 2010.

One of the REC members is appointed as the lead reviewer for each application reviewed by the sub-committee. The lead reviewer for your application is Nora Donaldson.

Please note that the lead reviewer may wish to contact you by phone or email between November 1st and 3rd to clarify any points that might be raised by members and assist the sub-committee in reaching a decision.

If you will not be available between these dates, you are welcome to nominate another key investigator or a representative of the study sponsor who would be able to respond to the lead reviewer's queries on your behalf. If this is your preferred option, please identify this person to us and ensure we have their contact details.

You are not required to attend a meeting of the sub-committee.

Please do not send any further documentation or revised documentation prior to the review unless requested.

Documents received

The documents to be reviewed are as follows:

Document	Version	Date
Investigator CV		20 September 2010
Protocol	1	22 September 2010
Student's CV		22 September 2010
REC application	45983/159656/1/210	23 October 2010
Participant Information Sheet: Parents	1	22 September 2010
Participant Information Sheet: Patient	1	22 September 2010
Participant Consent Form: Parents	1	22 September 2010
Participant Consent Form: Patient	1	22 September 2010
Key Investigator's CV 1		22 September 2010
Key Investigator's CV 2		22 September 2010
Key Investigator's CV 3		
Referees or other scientific critique report		27 October 2010

No changes may be made to the application before the meeting. If you envisage that changes might be required, we would advise you to withdraw the application and re-submit it.

Notification of the Committee's decision

We aim to notify the outcome of the sub-committee review to you in writing within 10 working days from the date of receipt of a valid application.

If the sub-committee is unable to give an opinion because the application raises material ethical issues requiring further discussion at a full meeting of a Research Ethics Committee, your application will be referred for review to the next available meeting. We will contact you to explain the arrangements for further review and check they are convenient for you. You will be notified of the final decision within 60 days of the date on which we originally received your application. If the first available meeting date offered to you is not suitable, you may request review by another REC. In this case the 60 day clock would be stopped and restarted from the closing date for applications submitted to that REC.

R&D approval

All researchers and local research collaborators who intend to participate in this study at sites in the National Health Service (NHS) or Health and Social Care (HSC) in Northern

Ireland should apply to the R&D office for the relevant care organisation. A copy of the Site-Specific Information (SSI) Form should be included with the application for R&D approval.

You should advise researchers and local collaborators accordingly.

The R&D approval process may take place at the same time as the ethical review.

Final

R&D approval will not be confirmed until after a favourable ethical opinion has been given

by this Committee.

Guidance on applying for R&D approval is available at

<http://www.rdforum.nhs.uk/rdform>.

There is no requirement for separate Site-Specific Assessment as part of the ethical review

of this research. The SSI Form should not be submitted to local RECs.

Communication with other bodies

All correspondence from the REC about the application will be copied to the research sponsor. It will be your responsibility to ensure that other investigators, research collaborators and NHS care organisation(s) involved in the study are kept informed of the progress of the review, as necessary.

10/H0808/156

Please quote this number on all correspondence

Yours sincerely

Atul Patel
Committee Co-ordinator

Email: atul.patel@imperial.nhs.uk

Copy to:

Sponsor Phillip Diamond
R&D, Ground Floor, Rosenheim Wing
25 Grafton Way
London
WC1E 6DB

Student Mr Mohammad Almehteb
Unit of Paediatric Dentistry
UCL Eastman Dental Institute
256 Gray's Inn Road, London
WC1X 8LD

Appendix 2: Topic guide for in-depth interviews

Introduction

- Introduce self
- Introduce study: who/what it's for, what is it about
- Key points:
 - purpose of interview
 - length of interview
 - voluntary nature and right to withdraw
 - will not affect care
 - reasons for recording the interview
- Confidentiality, and how findings will be reported
- Any questions?

1. Background and personal circumstances

- Q: What are their personal circumstances at present? Siblings etc.?
- Age, activities
- What they like to do in their spare time

2. Feelings about AI

- What do you know about the condition of your teeth?
- Does anyone else in your family have teeth like yours?
- Is there anything you do or don't like about your teeth?
- How do you feel about your teeth generally?
- Is there anything you do/ don't do because of your teeth?
- How do you feel about your teeth?
- If there is anything you want to change about your teeth what would it be?
- Have any of your friends, family or other people ever made good or bad comments about your teeth? If so what?
- Do you know whether you need to have any treatment for your teeth?
- Do they expect other people to notice the difference? Who? How will they feel if these things don't change/happen?
- How do you feel if these things don't change or happen?
- Is there anything else you want to add?

Contact details

**Miss Susan Parekh, Dr Susan
Cunningham, Miss Amanda O'Donnell,
Mr Mohammad Almhateb**

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Website: www.uclh.nhs.uk

UCL Hospitals cannot accept responsibility
for information

provided by external organisations.

Appendix 3: Patient's Information Leaflet



If you need a large print,
audio or translated copy of
this document, please
contact us on 0207 915
1269. We will try our best to
meet your needs.

If you wish to discuss this study with a member
of the research team or an independent expert
who is not part of the research team, please
ask Miss Susan Parekh

Thank you for taking the time to read this
leaflet.

Publication date:

Date last reviewed:

Version number:

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University College London Hospitals 
NHS Foundation Trust

**How do children with
Amelogenesis Imperfecta (AI)
feel about the appearance of
their teeth?**

Patient Information Leaflet



Version:

Date:11-02-2011

Invitation

We are doing a research project and we would like you to take part in it. This leaflet tells you about the project and why we are doing it, so that you can decide if you want to take part. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information.

What is the purpose of the study?

Our project is to help find out how you really feel about yourself and your teeth.

Why have I been invited?

You have been invited to take part because you have been diagnosed with Amelogenesis Imperfecta (AI), and offered treatment to correct your teeth/smile.

Do I have to take part?

No. It is up to you to decide. If you do decide to participate we will ask the parents to sign a consent form, and the patient will sign an assent form. If you change your mind, you are free to withdraw at any time, without giving a reason. The standard of care you receive will not be affected in any way.

What will happen to me if I take part?

We will ask you some questions about you and how do you feel about having AI. This should take about 1-1.5 hours depending on how much you want to say. There are

no right or wrong answers; we are just interested in your ideas. Some of these ideas will then be used to develop a questionnaire. You will not need to do anything else. We would prefer to interview you without your mum/dad being there, however, if you would like them with you then that is perfectly OK.

What are the possible disadvantages or risks of taking part?

There are no risks anticipated. None of your answers will affect your treatment in any way.

What are the possible benefits?

The information from this study will hopefully be used to help us understand better what makes you and other patients feel the way you do about yourselves and your teeth. We hope to set up a support group for patients with AI using research from this study.

What will happen with the results?

We hope to publish the results of the study on completion. All confidential information will be coded and you will not be identifiable in any way.

Will my taking part in the study remain confidential?

Yes. All information that is collected about you during the research will remain strictly confidential and will be seen only by the investigators named on this sheet. The safety and security of the data will be the responsibility of the principal investigator (Miss Susan Parekh). The data held about you will include the results of the interview and also

your age and gender (male or female). This information will be recorded in such a way that it is completely anonymous and you cannot be individually identified in anyway.

Who has reviewed the study?

All research in the NHS is looked at by independent group, called a Research Ethics Committee to protect your safety, rights, wellbeing and dignity. This study has been reviewed by [South East London] Research Ethical Proportionate Review Sub-Committee. If you would like to see a summary of the findings from the study when it is completed, please tell Miss Parekh or any of the other dentists you see.

Version: 2

Date:11-02-2011

Contact details

**Miss Susan Parekh, Dr Susan
Cunningham, Miss Amanda O'Donnell,
Mr Mohammad Almehteb**

Tel: 020 7915 1269

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Website: www.uclh.nhs.uk

UCL Hospitals cannot accept responsibility
for information

provided by external organisations.

Appendix 4: Parent's Information Leaflet



If you need a large print,
audio or translated copy of
this document, please
contact us on 0207 915
1269. We will try our best to
meet your needs.

If you wish to discuss this study with a member
of the research team or an independent expert
who is not part of the research team, please
ask Miss Susan Parekh

Thank you for taking the time to read this
leaflet.

Publication date:

Date last reviewed:

Version number:

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University College London Hospitals 
NHS Foundation Trust

How do children with Amelogenesis Imperfecta (AI) feel about the appearance of their teeth?

Parent's Information Leaflet



Version: 2

Date: 11-02-2011

Invitation

Your child is being invited to take part in a research study. Before you make a decision, it is important you know why the research is being done and what it would involve from you. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information.

What is the purpose of the study?

Certain dental conditions can be inherited, and cause significant problems, such as discolouration and sensitivity. Amelogenesis Imperfecta (AI) is one of these conditions, and can require lifelong dental care. The possible impact of this condition on children and young adults is not known, as it is a largely unresearched field. We are interested in finding out about the possible impacts of AI on children and young adults, both pre and post-treatment.

Why has my child been chosen?

We are asking all patients who have been diagnosed with AI, and are appropriate for our study to participate.

Does my child have to take part?

No. It is up to you and your child to decide. If you do decide to participate we will ask the parents to sign a consent form, and the patient will sign an assent form. If

you change your mind, you are free to withdraw at any time, without giving a reason. The standard of care you receive will not be affected in any way.

What will happen to my child if he/she takes part?

We will ask your child some questions about how he/she feels about having AI and about their appearance of their teeth. This should only take about 1-1.5 hours depending on how much your child wants to say. There are no right or wrong answers; we are just interested in your child's ideas. Some of these ideas will then be used to develop a questionnaire for a larger study of this condition. You will not need to do anything else. We would prefer to interview your child by themselves, however, if your child would like you with him/her then that is perfectly OK.

What are the possible disadvantages or risks of taking part?

There are no risks anticipated. None of your child's answers will affect his/her treatment in any way.

What are the possible benefits?

We cannot promise the study will help you directly but the information we get from this study will help improve treatment of other patients. We also hope to set up a support group for patients with AI using the research from this study.

What will happen with the results?

We hope to publish the results of the study on completion. All confidential information will be coded and you will not be identifiable in any way.

Will my taking part in the study remain confidential?

Yes. All information that is collected about your child during the research will remain strictly confidential and will be seen only by the investigators named on this sheet. The safety and security of the data will be the responsibility of the principal investigator (Miss Susan Parekh). The data held about your child will include the results of the interview and also your gender (male or female). This information will be recorded in such a way that it is completely anonymous and your child cannot be individually identified in anyway.

Who has reviewed the study?

All research in the NHS is looked at by independent group, called a Research Ethics Committee to protect your safety, rights, wellbeing and dignity. This study has been reviewed by [South East London] Research Ethical Proportionate Review Sub-Committee. If you would like to see a summary of the findings from the study when it is completed, please tell Miss Parekh or any of the other dentists you see.

Version: 2

Date: 11-02-2011

Appendix 5: Parents' Consent

University College London Hospitals



NHS Foundation Trust

Eastman Dental Hospital
256 Gray's Inn Road
London
WC1X 8LD
Tel: 020 7915 1000
Fax: 020 7915 1012
Web: www.uclh.nhs.uk

Centre Number:

Patient Identification Number for this study:

Form version: 1 Date: 22-9-2010

REC Ref No: 10/H0808/156

CONSENT FORM FOR PARENTS / GUARDIANS

Title of project **How do children with Amelogenesis Imperfecta (AI) feel about the appearance of their teeth?**

Name of Principal investigators:

Mr Mohammad Almehteb; Miss Susan Parekh; Professor Susan
Cunningham; Miss Amanda O'Donnell

Please tick box

1. I confirm that I have read and understood the information sheet (Version 1 dated 22-9-2010) for the above study and have had the opportunity to ask questions.

2. I confirm that I have had sufficient time to consider whether or not I wish my child to be included in the study

3. I understand that my child's participation is voluntary and that they are free to withdraw at any time, without giving any reason, without his/her medical care or legal rights being affected.

4. I understand that relevant sections of my child's medical notes and data collected during the study may be looked at by individuals from [UCL Eastman Dental Institute], from regulatory authorities or from the NHS Trust, where it is relevant to my child taking part in research. I give permission for these individuals to have access to my child's records.

5. I agree for my child to take part in the above study.

—

Name of patient

Date

Signature

_____	_____	_____
Name of Person taking consent	Date	Signature
Miss Susan Parekh	020-7915-1269	
Researcher (to be contacted if there are any problems)		

Comments or concerns during the study

If you have any comments or concerns you may discuss these with the investigator. If you wish to go further and complain about any aspect of the way you have been approached or treated during the course of the study, you should write or get in touch with the Complaints Manager, UCL Hospitals. Please quote the reference number at the top this consent form.

- 1 form for patient;
- 1 to be kept as part of the study documentation,
- 1 to be kept with hospital notes

Appendix 6: Questionnaire



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UNIT OF PAEDIATRIC DENTISTRY

Questionnaire for Patients with Amelogenesis Imperfecta (AI)

Please would you help us by filling in this questionnaire to find out how you really feel about yourself and your teeth.

There are no right or wrong answers; we just want to know your thoughts.

Guarantee of Confidentiality

All information you give will remain private and no one will know your name.

Filling in this questionnaire will not affect your treatment in any way.

Please turn over

Please remember:

- Do not write your name on the paper.
- This is not a test and there are no right or wrong answers.

Section 1: About You

1. How old are you? _____ years.

2. Are you

Boy

Girl

3. Which of these groups do you think you belong to **(Tick one box only)**

White

Asian

Black

Mixed

Other group

Section 2: About Your Teeth

4. Do you remember noticing anything different about your teeth before your dentist sent you to this hospital?

Yes

No

Please turn over

5. Who first said it would be a good idea for you to have treatment for your teeth?

(Tick **ONE** box only)

You

Mum or Dad

Other family members

Your Dentist

Your Doctor

Other

If other, please say who

6. On a scale of 1 to 5, how confident do you feel because of your teeth?

(Please choose only ONE answer and place a circle around it ○)

Not at all confident

Very confident

1

2

3

4

5

Please turn over

7. On a scale of 1 to 5, how important is it/was it that you had treatment for your teeth

(Please choose ONE answer only and place a circle around it ○)

	Not important at all			very important	
To improve the colour of your teeth	1	2	3	4	5
To improve the shape of your teeth	1	2	3	4	5
To improve the size of your teeth	1	2	3	4	5
To improve your smile	1	2	3	4	5
To reduce pain/sensitivity	1	2	3	4	5
Other reasons (please tell us)					

8. Do you avoid any food or drinks because of your teeth?

(Please choose only ONE answer and place a circle around it ○)

Never	Occasionally	Often	All of the time
1	2	3	4

If your answer is “Occasionally”, “Often”, or “All of the time”, then please tell us which food or drinks?

Please turn over

Section 3: About Your Feelings

(For each of the following 5 questions, please circle ONE answer only)

9. Have you ever been teased about your teeth or have other people ever made hurtful remarks about your teeth?

Never	Occasionally	Often	All of the time
1	2	3	4

10. Do you ever avoid smiling because of your teeth?

Never	Occasionally	Often	All of the time
1	2	3	4

11. Do you ever feel “different” or “left out”?

Never	Occasionally	Often	All of the time
1	2	3	4

12. Do you ever worry that your teeth may affect your future plans?

Never	Occasionally	Often	All of the time
1	2	3	4

13. Do your teeth ever affect things you do in your spare time or hobbies (for example, sports or music)?

Never	Occasionally	Often	All of the time
1	2	3	4

Please turn over

14. What is the most important thing you would like from the treatment of your teeth?

(Tick ONE box only)

Improve the colour of my teeth

Improve the shape of my teeth

Improve the size of my teeth

Improve my smile

Reduce sensitivity from my teeth

To feel better about myself

If the most important thing for you is not listed above, please write it below: _____

Please turn over



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(Now, this question is for You and your family)

15. Do you think it is useful to have a website or “support group” for patients with the same dental problem you have (Amelogenesis Imperfecta)?

Yes

No

You have now finished

Thank you for helping us by doing this questionnaire.

If you have anything else to tell us, please write it below

If you have any questions, please contact Dr Parekh on 020 3456 1269

Appendix 7: Child Perception Questionnaire (CPQ)

SHORT-FORM CHILD PERCEPTIONS QUESTIONNAIRE (CPQ11-14)

These next few questions are about how you feel about your teeth. There are no "right" or "wrong" answers- please answer as best you can. Please tick the box which applies to you.

1. Would you say the health of your teeth, lips, jaws and mouth is:

Excellent Very Good Good Fair Poor

2. How much does the condition of your teeth, lips, jaws or mouth affect your life overall?

Not at all Very little Some A lot Very much

In the past 3 months, how often have you had:

3. Sores in your mouth?

Never Once or twice Sometimes Often Every day or almost every day

4. Bad Breath?

Never Once or twice Sometimes Often Every day or almost every day

5. Food stuck in between your teeth?

Never Once or twice Sometimes Often Every day or almost every day

6. Difficulty biting or chewing food like apples, corn on the cob or steak?

Never Once or twice Sometimes Often Every day or almost every day

In the past 3 months, how often have you had:

7. Difficult to drink or eat hot or cold foods?

Never Once or twice Sometimes Often Every day or almost every day

8. Difficulty saying any words?

Never Once or twice Sometimes Often Every day or almost every day

9. Trouble sleeping?

Never Once or twice Sometimes Often Every day or almost every day

10. Pain in your teeth, lips, jaws or mouth?

Never Once or twice Sometimes Often Every day or almost every day

11. Taken longer than others to eat a meal?

Never Once or twice Sometimes Often Every day or almost every day

12. Felt irritable or frustrated?

Never Once or twice Sometimes Often Every day or almost every day

13. Felt shy or embarrassed?

Never Once or twice Sometimes Often Every day or almost every day

14. Been upset?

Never Once or twice Sometimes Often Every day or almost every day

15. Been concerned what other people think about your teeth, lips, mouth or jaws?

Never Once or twice Sometimes Often Every day or almost every day

In the past 3 months, how often have you had:

16. Avoided smiling or laughing when around other children?

Never Once or twice Sometimes Often Every day or almost every day

17. Not wanted to speak or read out loud in class?

Never Once or twice Sometimes Often Every day or almost every day

18. Other children teased you or called you names?

Never Once or twice Sometimes Often Every day or almost every day

19. Had other children ask you questions about your teeth, lips, jaws or mouth?

Never Once or twice Sometimes Often Every day or almost every day

20. Argued with other children or your family?

Never Once or twice Sometimes Often Every day or almost every day

Appendix 8: Explanatory Letter

Dear Parents

We are contacting you because your child is / was a patient at the Department of Paediatric Dentistry, Eastman Dental Hospital for treatment of a condition associated with their teeth, called Amelogenesis Imperfecta (AI).

AI can cause significant problems, such as discolouration and sensitivity, but the possible impact on children and young adults is not known, due to limited research in this area.

We would like to invite your child to participate in a research study to assess the possible impacts of AI on children and young adults.

In this envelope you will find:

- A questionnaire
- Consent Form for Parents (3 copies)
- Consent Form for Patient (3 copies)
- Parent's Information Leaflet
- Patient's Information Leaflet
- A stamped envelope

We would like your child to complete the attached questionnaire about how he/she feels about having AI and about their appearance of their teeth. There are no right or wrong answers; we are just interested in your child's ideas. The questionnaire should not take more than 5 minutes to complete.

For more information about the research study, please read the **"Parent's & Patient's Information Leaflet"** attached in this packet.

If you agree for your child to participate in this research study, kindly do the following steps:

- 1- Sign and date the three copies of the consent forms please keep one copy for yourself and put the other two in the stamped return envelope.
- 2- Ask your child to complete the questionnaire.
- 3- When finished, put the completed questionnaire with the two copies of Consent Forms in the stamped envelope and return to us.

The information we get from this study will hopefully help improve treatment of many children with AI. Finally, we would like to thank you and your child for taking the time to help us in this research project. Your thoughts and support are greatly appreciated. If you would like any further information, please use the contact number on the information sheet.

Yours Sincerely,



Mohammad Almehteb

DDent Paediatric Dentistry Postgraduate



Dr Susan Parekh

Clinical Lecturer/Honorary consultant

**An audit of safeguarding children
knowledge in postgraduate students at the
Eastman Dental Hospital, UCLH**

&

**The efficacy of safeguarding children
training**

(Audit Report)

2012

Mohammad Almehteb

Eastman Dental Institute/Hospital

University College London/Hospital

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ABBREVIATIONS

AAPD	American Academy of Pediatric Dentistry
DoH	Department of Health
EDI/H	Eastman Dental Institute/Hospital
GDP	General Dental Practitioner
NICE	National Institute for Health and Clinical Excellence
RCSEng	Royal College of Surgeons of England
UCLH	University College London Hospital

ABSTRACT

Background: Safeguarding children is everyone's responsibility & even if children attend with parents / grandparents all healthcare professionals have a crucial role in identifying child welfare concerns and features of possible abuse or neglect. The dental team has an ethical responsibility to find out about and follow local procedures for child protection and to follow them if a child is or might be at risk of abuse or neglect. Studies exist looking at practitioners' knowledge about safeguarding children but postgraduate students have not been looked at previously. At the Eastman Dental Hospital and Institute there are a lot of postgraduate students and their knowledge base is important to explore.

Aim: The aim of this audit project was to assess the knowledge and awareness of the postgraduates at the Eastman Dental Hospital regarding safeguarding children and to look at the efficacy of safeguarding children training with questions being asked pre and post training

Standard: The four standards set for this audit were that 100% of all postgraduate students should correctly answer key questions identified with the Trust safeguarding Lead Paediatrician, that when the first year questionnaires were repeated – results were the same or better, and thirdly the second and third year responses be the same or better than the second first year responses. Also, all postgraduates – second, third and the repeat first years should have had appropriate safeguarding children training.

Method: A questionnaire was completed by 1st, 2nd, and 3rd year postgraduates at the Eastman Dental Hospital. The time scale for this audit was from October 2011 to august 2012. The first year postgraduates had to complete the same questionnaire twice - first before they had any induction or training about safeguarding children at the Eastman Dental Hospital, and the second time was after they have had "Level I" safeguarding children training. The second and third year postgraduates had to complete the questionnaire only once.

Results: Overall, data was collected for 51 students in the first year, 20 students in the second year, and 20 students in the third year. The first year postgraduate students showed better knowledge about the safeguarding children than the second and third years. The information retention regarding safeguarding children seemed to be reduced with postgraduates as they advanced through years of their course.

Outcome: Postgraduate students showed some deficiencies in knowledge as our gold standards were not all met. Targeted training may help this in the future. Training will be developed and a re-audit will take place in December 2012.

AUDIT PERSONNEL

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BACKGROUND

Safeguarding children is everyone's responsibility and even if children or young adults attend with parents / grandparents all health care professionals need awareness of signs and symptoms of maltreatment. There are many types of abuse with different features including neglect, physical, sexual and emotional abuse, and fabricated or induced illness. Members of the dental team are in a position where they may observe the signs of child abuse or neglect or hear something that causes them concern about a child. The dental team has an ethical responsibility to find out about and follow local procedures for child protection and to follow them if a child is or might be at risk of abuse or neglect (Standards for dental professionals, GDC 2005). There is also a responsibility to ensure that children are not at risk from members of the profession.

RECOGNIZING ABUSE AND NEGLECT

In Scotland, the "Non-organic failure to thrive" is recognised as a fifth category in child abuse. Table 1 below gives more details and examples for each type of child abuse.

Professionals need to be aware of and be sensitive to different family patterns and life style. However, child abuse cannot be condoned for cultural or religious reasons.

There are risk factors that make a child more vulnerable for abuse. These factors are parental factors, child factors, and family / social factors. Parental factors include history of abusive childhood experiences, learning difficulties and little or no ante-natal care. Child factors are things like not attending school and a child with disability and lastly family / social factors include drug and alcohol abuse and weak supportive networks.

Category	Definition⁵	Examples of signs the dental team may observe
Physical Abuse	Hitting, shaking, throwing, poisoning, burning or scalding, drowning, suffocating or otherwise causing physical harm to a child; fabricating or inducing illness (formerly known as Munchausen by proxy syndrome).	Bruises; abrasions and lacerations; burns; bite marks; bone fractures; intra-oral injuries. See also Figures 3 and 4.
Emotional Abuse	Persistent emotional maltreatment such as to cause severe and persistent adverse effects on the child's emotional development; conveying to children that they are worthless or unloved; imposing inappropriate expectations; preventing normal social interaction; seeing or hearing the ill-treatment of another (eg domestic violence).	Interaction with parent, eg parent using threatening or abusive language; poor growth; developmental delay; social immaturity; aggression; indiscriminate friendliness; challenging behaviour.
Sexual Abuse	Forcing or enticing a child to take part in sexual activities, whether or not the child is aware of what is happening; either involving physical contact or non-contact activities (eg involving children in looking at or producing pornographic material).	Direct allegation (disclosure); sexually transmitted oral infections; pregnancy; trauma including oral trauma; emotional and behavioural changes.
Neglect	Persistent failure to meet a child's basic physical and/or psychological needs, likely to result in the serious impairment of the child's health or development; failing to provide adequate food, clothing, shelter; failing to protect from danger; failure to ensure access to appropriate medical care.	Failure to thrive; frequent injuries; inappropriate clothing; ingrained dirt; developmental delay; behavioural changes, eg withdrawn or attention-seeking; untreated dental disease with repeated episodes of dental pain.

Table 1. Recognizing abuse and neglect. (Courtesy of: Safeguarding children in dental practice, Dental Update, 2007)

SIGNS OF ABUSE AND NEGLECT

Signs of physical abuse can frequently be seen in the head and neck region during dental examination, but can also be seen in other parts of the child's body like wrists, back, and ankles.

Bruising is the most common physical abuse injury that can be seen in a child. It can be presented in different patterns that could suggest possibility of abuse. Bite marks may also be noticed by dentists during general dental examination which may appear as circular or oval patterns of abrasions and lacerations. Lacerations, abrasions and scars are also considered signs of abuse. Injuries to the upper labial fraenum may be considered an indicator of abuse. However, a torn labial fraenum should be assessed in the context of the child's medical and social history, stage of development, and the explanations given about the cause. Other oral injuries that can be considered as a sign of abuse are lip laceration and teeth fracture with unsuitable explanations. Burn injuries seen on any area that should not be expected to come into contact with a hot object (for example, cigarettes, iron) in an accident for example soles of feet, the back of a hand or buttock, can be considered a sign of abuse. General fractures (one or more) in the absence of a predisposing medical condition (eg Osteogenesis Imperfecta) can be considered a sign of abuse.

Other injuries that may not be possible to observe during dental visit like spinal injuries, intracranial injuries, and visceral injuries can be a sign of abuse.

Emotional abuse is the persistent emotional maltreatment causing severe and persistent adverse effect on the child's emotional development. It may involve conveying to the child that he or she is worthless or unloved. Signs of emotional abuse can be seen in different behaviour and emotional states, for example fearful, withdrawn, low self-esteem, aggressive, habitual body rocking, and over-friendliness to strangers including healthcare professionals. It can also be seen between parent-child interactions where there is negativity or hostility towards a child or a parent refuses to allow a child to speak to the dentist on their own.

Sexual abuse involves forcing a child to take part in sexual activities, whether or not the child is aware of what is happening. The activities may involve physical contact like rape, or non-contact activities like involving a child in looking at pornographic material or watching sexual activities or encouraging children to behave in sexually

inappropriate ways. Signs of sexual abuse include sexually transmitted infections, pregnancy and emotional and behavioural changes.

Neglect is the persistent failure to meet the child's basic physical and/or psychological needs, likely to result in the serious impairment of the child's health or development. It can be seen as early as during pregnancy as a result of maternal substance abuse. After birth, neglect can include failure to provide adequate food, clothing and shelter and failure to access appropriate medical / dental care and treatment with dental caries and neglect being increasingly discussed. As a result of neglect a child may present with untreated dental diseases with repeated episodes of dental pain.

ROLE OF THE DENTAL TEAM

Every staff member, whether dentist, dental nurse, receptionist or practice manager - has an important role in recognizing signs of abuse and neglect. This is clearly stated in the publication of the department of education and skills by HM government (2006) "*All those who come in contact with children and families in their everyday work, including practitioners who do not have a specific role in relation to child protection, have a duty to safeguard and promote the welfare of children*".

Because abuse or neglect may present in a number of different ways like signs and symptoms and observation of child behaviour or parent-child interactions, the dental team is considered in to be in a good position to recognise abuse and neglect. As health care professionals it is important to remember that our first duty is to the child and no child should be left untreated or in pain because of underlying concerns about abuse.

The first step to be taken if there are any concerns about child abuse should always be to discuss this with an appropriate colleague like an experienced dentist etc where possible. A referral to the local social services may be made. If however a discussion has not taken place and you have concerns ***no action is never an option***. Protocols vary but usually referrals should be made in writing or verbal if urgent within 48 hours stating the facts of the case and reason for concerns along with any action plan already in place. It is considered good practice to explain any concerns to the child and parents and inform them about the intention to refer them to social services and seek their consent. Research shows that being open and

honest from the start results in better outcomes for children. However there are certain exceptions and reasonable judgment must be made in each case. Discussion of concerns should be avoided in certain circumstances for example where the discussion may put the child at greater risk or where parents / carers are being violent or abusive.

PROFESSIONAL RESPONSIBILITY

There are number of guidelines set-up to help healthcare professionals in managing safeguarding children concerns.

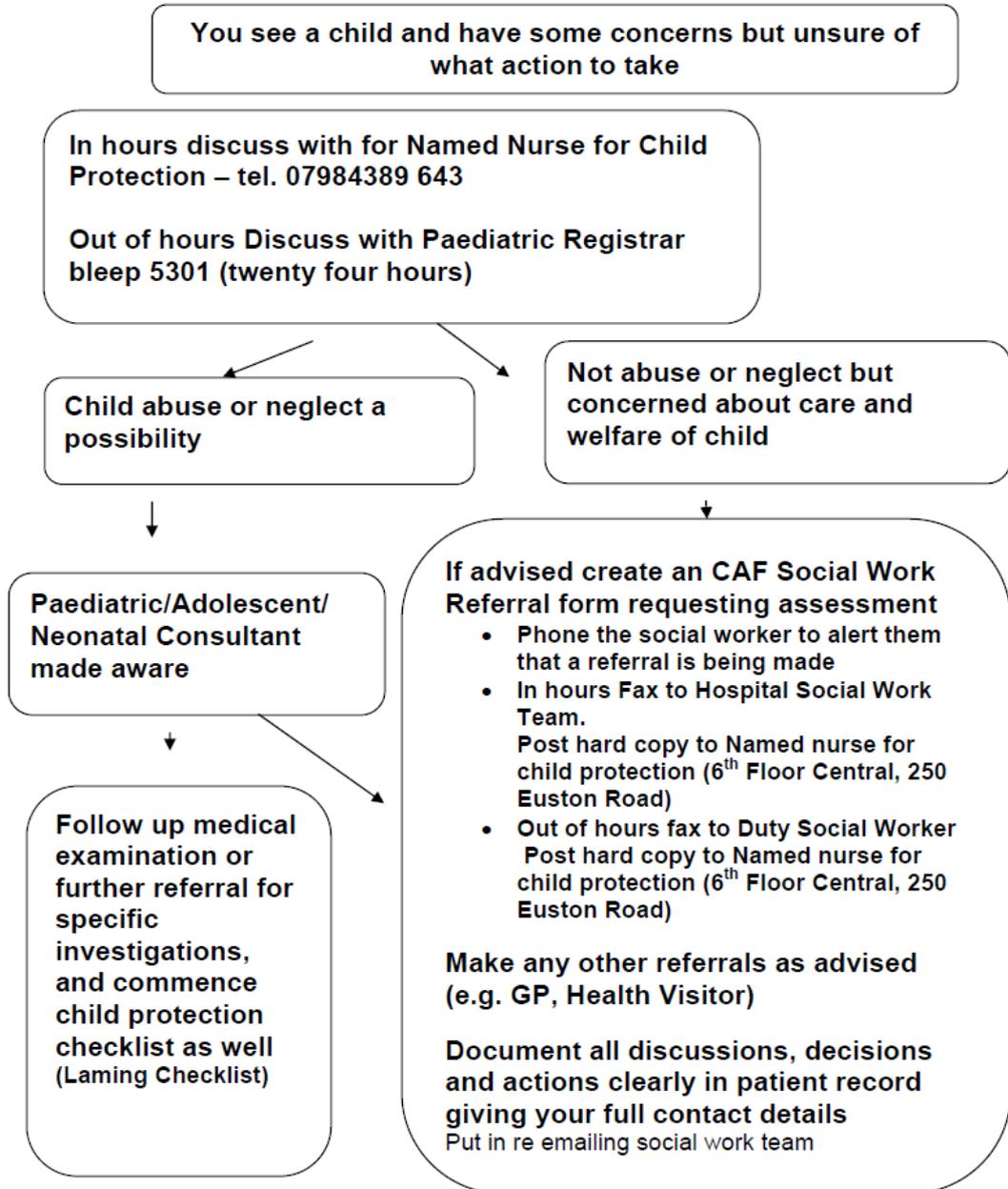
At the University College of London Hospital (UCLH), NHS Foundation Trust, the local policy and procedures for child safeguarding and protection is set up – most recent version being May 2012. The aim of the UCLH policy is to provide guidance to health care professionals within the trust to know what to do if they have any concerns about child's safety or well being. It is designed for all those who come in contacts with children and their families in their every day work.

They can provide a framework to ensure appropriate information sharing. The policy advises staff to be open and honest with the family from the beginning about why/what, how and with whom information will, or could be shared, and seek their agreement, unless it is unsafe or inappropriate to do so as mentioned previously. If in doubt it is good practice to seek advice from a senior colleague. A referral to social care may be made including informing the social work team by phone and completing and sending a common assessment framework (CAF) form either electronically (eCAF) or by hand.

A follow up examination is required for further referral or specific investigations. All discussions, decisions and actions must be clearly documented in patient record.

The policy indicates that all staff should be aware of some of the universal health services like the need for all children to be registered with a GP and if they are not a help should be provided to do this. Safeguarding children should be done in a multi-agency way and multidisciplinary meetings held to discuss any concerns or issues about safeguarding children.

23.0 DECISION TREE



UCLH - 2012
 Published Date: May 2012
 Review by Date: May 2014
 Policy/procedure only current on date printed, refer to Insight for definitive version.

Figure 1. Summary of UCLH child safeguarding and protection policy and procedures.

At the Eastman Dental Hospital - one of the Specialist Hospitals of UCLH - all staff members are required to have Level 1 safeguarding children training delivered by

the trust as part of their mandatory training programme. This has to be done annually. Non-clinical staff is required to have Level 1 safeguarding children training only. This is usually done as an e-learning exercise. All clinical staff requires a minimum of Level 2 training, and all other staff members who come in to regular contact with children on a daily basis like paediatrics, neonatology, obstetrics, and accident and emergency must have Level 3 training. The latter is a day course of training with multi-agency and multidisciplinary elements and the training has to be updated every 3 years.

The NICE guideline '**When to suspect child maltreatment**' – July 2009, provides a summary of the alerting features that healthcare professional might come across that suggest a child might be being maltreated.

If you encounter an alerting feature described in this guidance it is good practice to follow the process outlined below.

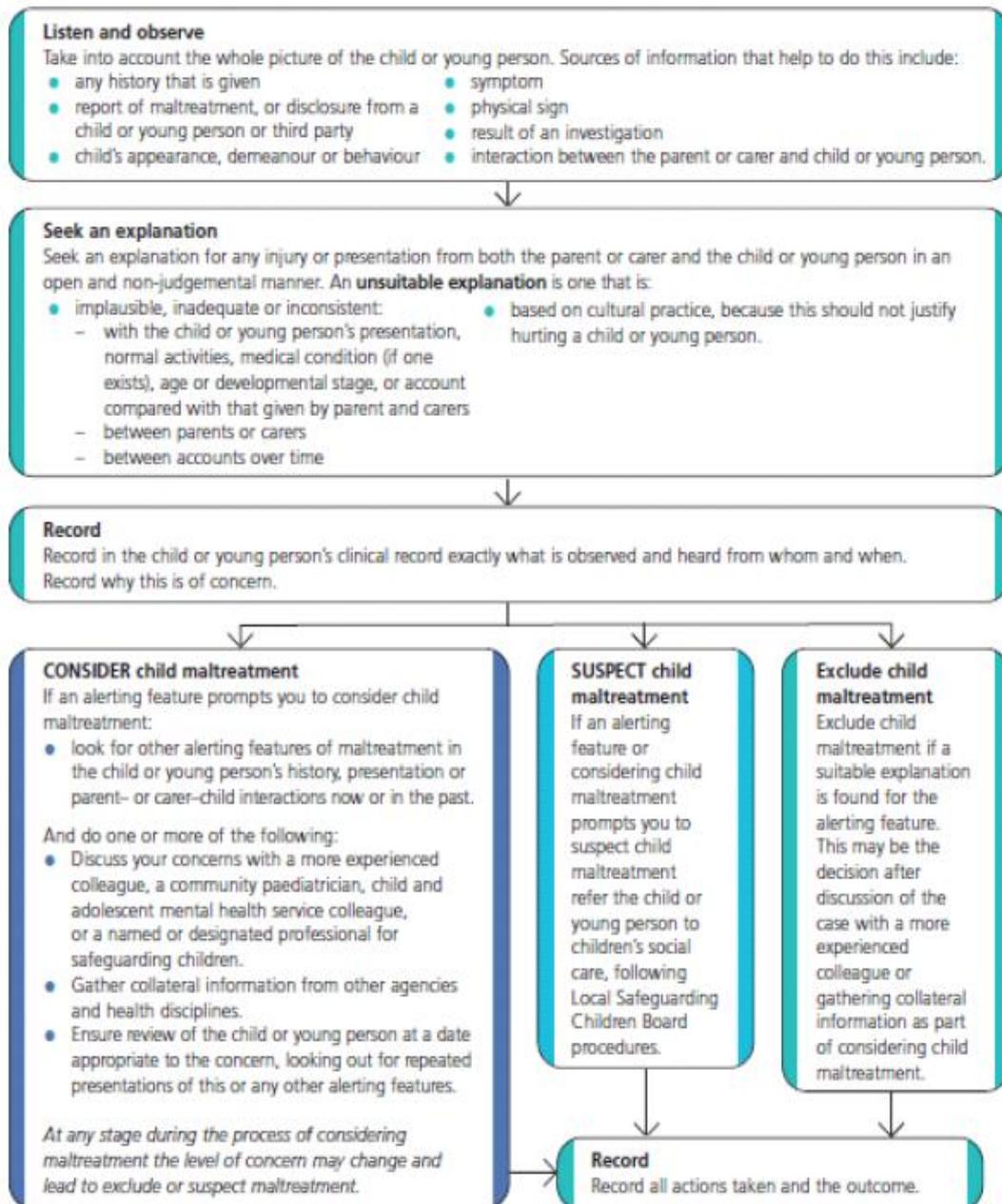


Figure 2. Flow chart for using the NICE guideline 'When to suspect child maltreatment'

The purpose is to raise awareness and help healthcare professionals who are not specialists in child protection to identify children who may be being maltreated. It does not give healthcare professionals recommendations on how to diagnose, confirm or disprove child maltreatment. The alerting features described in the guideline are similar what mentioned earlier include physical, emotional signs. Because some alerting features are more likely to indicate child maltreatment than

others, the guideline advises healthcare professionals to either 'consider' or 'suspect' child maltreatment as possible explanation for their observation. The guideline advice 'considering' child maltreatment when the healthcare professional thinks it might be one reason for the alerting feature, but they are not sure. The guideline advises healthcare professional to 'suspect' child maltreatment if they see one of the alerting features that is more likely to mean that a child has been maltreated, but these are not proof of it. With regards to dentistry, the NICE guideline mentions in physical Features (*incl. oral injury*), and in suspected neglect/abuse – that the oral cavity should be examined.

Other guidelines are included in the references list but include HM Government (2010): 'Working Together to Safeguard Children', and British Society of Paediatric Dentistry's: a policy document on dental neglect in children (2010). This latter document defined dental neglect can be defined as the persistent failure to meet a child's basic oral health needs, likely to result in the serious impairment of a child's oral or general health or development. Talked about three tiers of neglect and putting systems in place to safeguard children.

In summary, the dentist's role is to recognise the possibility of child abuse and provide essential emergency dental treatment if required. A dentist should be familiar with child protection policy and procedure and know who to access if having any concerns (Local Safeguarding Children's Board LSCB) and how to carry out referral to the social work team. It is also important that dentists access training to provide them with knowledge skills and confidence to deal with child protection concerns or referral and seek advice or support. Still – often health care professionals are fearful of raising a concern but it must be remembered that the majority of referrals are *because a child is in need of help & support* – a section 17 and not in need of protection – a section 47). Information may help to complete the picture of a child's life as in the majority of safeguarding children cases with devastating outcomes –information has not been shared.

At the Eastman Dental Hospital there are a large number of Postgraduate students treating patients. The academic and NHS staff work closely with children and their families in different departments. Studies exist looking at dental practitioners' knowledge about safeguarding children but postgraduate students have not been looked at previously. One study was done to assess child protection training and experience among dental professional with an interest in paediatric dentistry

showed there is a significant gap between recognising signs of abuse and responding effectively where only 67% of the respondents managed to identify signs of abuse and only 29% had ever made a child protection referral (Harris *et al.*, 2009).

This audit was discussed with the Safeguarding lead for Children who was actively involved in designing the data collection form and deciding on information to be asked and its importance.

AIMS AND OBJECTIVES

The aims of this audit were to,

1. Assess the knowledge and awareness of all of the postgraduate students at the Eastman Dental Hospital regarding safeguarding children
2. To look at the overall efficacy of safeguarding children training in the first year students only as audit questions asked before, and after training

Standard:

The gold standards set for this audit were that,

1. 100% of all postgraduate students should correctly answer key questions on the questionnaire (*3 of them and 2 scenarios*) as identified with the Trust safeguarding Lead Paediatrician as below, the acceptable answers in bold.

Q19- *If you suspected but were not certain of a safeguarding issue, would you, (only tick one)*

Take action anyway

Take no action

Discuss with a senior colleague if possible or co-worker

Q20- *As a clinician, if you were treating an adult patient, are you responsible for safeguarding their children or grand-children?*

Yes

No

Q21- *Safeguarding children is the responsibility of:*

(only tick one)

- Clinicians*
- Other dental team members*
- Every one**
- Admin staff*

What would you do in the following 2 scenarios?

1. *A 5 year old child who has multiple carious teeth misses 2 appointments for treatment under general anaesthesia. Would you..... (only tick one)*

- Do nothing*
- Discuss your concerns with a more experienced colleague**
- Refer to social services**

2. *A child with dental trauma attends your practice. Their account of the trauma changed three times and the stories do not fit the injury. The child interaction with the parents is unusual. The presentation is delayed (2 weeks) Would you (only tick one)*

- Do nothing*
- Discuss your concerns with a more experienced colleague**
- Refer to social services**

- 2. When the first year questionnaires were repeated – results were the same or better
- 3. Second and third year responses same or better than second first year responses
- 4. All postgraduates should have had some form of safeguarding training in the repeat first year, second and third year groups

Method: This was a retrospective process audit. Data was collected on a questionnaire – see Appendix A –and was completed by 1st, 2nd, and 3rd year postgraduates at the Eastman Dental Hospital. The time scale for this audit was from October 2011 to August 2012. The first year postgraduates had to complete the same questionnaire twice - first before they had any induction or training about safeguarding children at the Eastman Dental Hospital, and the second time was after they have had “Level I” safeguarding children training (6/10/11 and then 30/5/12). The second and third year postgraduates had to complete the questionnaire only once.

The preliminary data were recorded on data collection sheet (appendix A) and later transferred to an Excel spreadsheet. The data were examined and presented by simple graphs (pie and bar charts) and analysed by descriptive statistics.

RESULTS

1 - Interpreting Questionnaire for 1st year students (First attempt)

The total number of post graduate students participating in the first questionnaire was 51 with gender 26 males and 25 females.

The first year postgraduates graduated from different countries with the majority being UK graduates (25%) and the results are listed in Table 2

Country of graduation	Frequency	Percent
Egypt	3	5.8
Germany	1	1.9
Greece	6	11.5
Hong Kong	1	1.9
India	3	5.8
Ireland	3	5.8
Italy	2	3.8
Jordan	1	1.9
KSA	1	1.9
Libya	1	1.9
Malaysia	6	11.5
New Zealand	1	1.9
Pakistan	1	1.9
Portugal	1	1.9
Spain	2	3.8
Thailand	1	1.9
UAE	3	3.8
UK	13	25.0
Total	51	100.0

Table 2. Distribution of countries that 1st year postgraduates graduated from.

The mean year of graduation from the dental school for the first year postgraduates was (2005), range 1996 – 2010, and the result are shown in Table 3.

Year of Graduation	Frequency	Percent
1996	1	2.0
1997	1	2.0
1998	1	2.0
1999	1	2.0
2000	2	3.9
2001	1	2.0
2002	6	11.8
2003	2	3.9
2004	2	3.9
2005	7	13.7
2006	6	11.8
2007	1	2.0
2008	9	17.6
2009	8	15.7
2010	3	5.9
Total	51	100.0

Table 3. Distribution of graduation year for the 1st year postgraduates.

The highest number of postgraduate students were from the endodontic department (13). Only one student was from the special care unit. Other departments included paediatric dentistry with 5 students, orthodontics with 10 students, periodontics with 2 students, oral surgery with 7 students, OMS with 4 students and prosthodontics with 8 students. The participants ranged between 23 and 40 years of age.

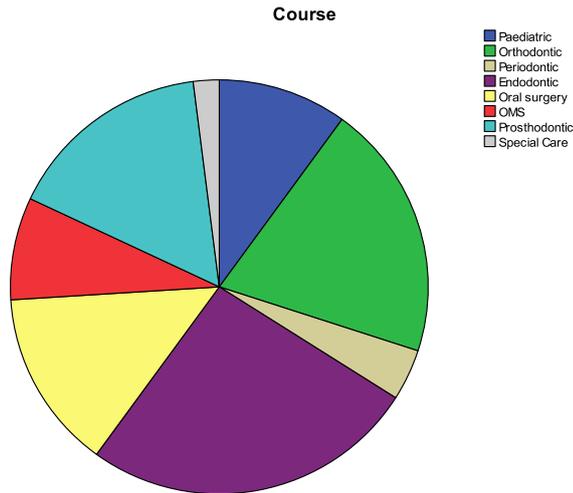


Figure 3. Distribution of 1st year postgraduate students from each department at the EDH

When asked whether they had provided dental treatment for children before, all 51 students indicated that they had. Where treatment had been provided is shown below. Two participants did not indicate where they had provided the treatment.

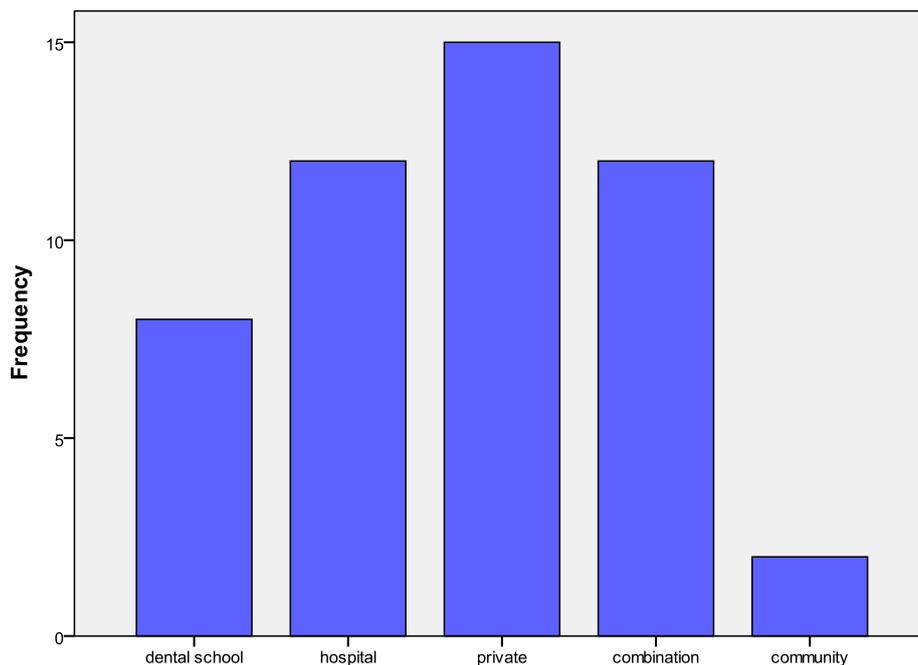


Figure 4. Places where 1st year postgraduate student provided dental treatment for children before their enrolment in the EDH

When asked whether they had taken a safeguarding children course before the majority had not as seen in Figure 5 below.

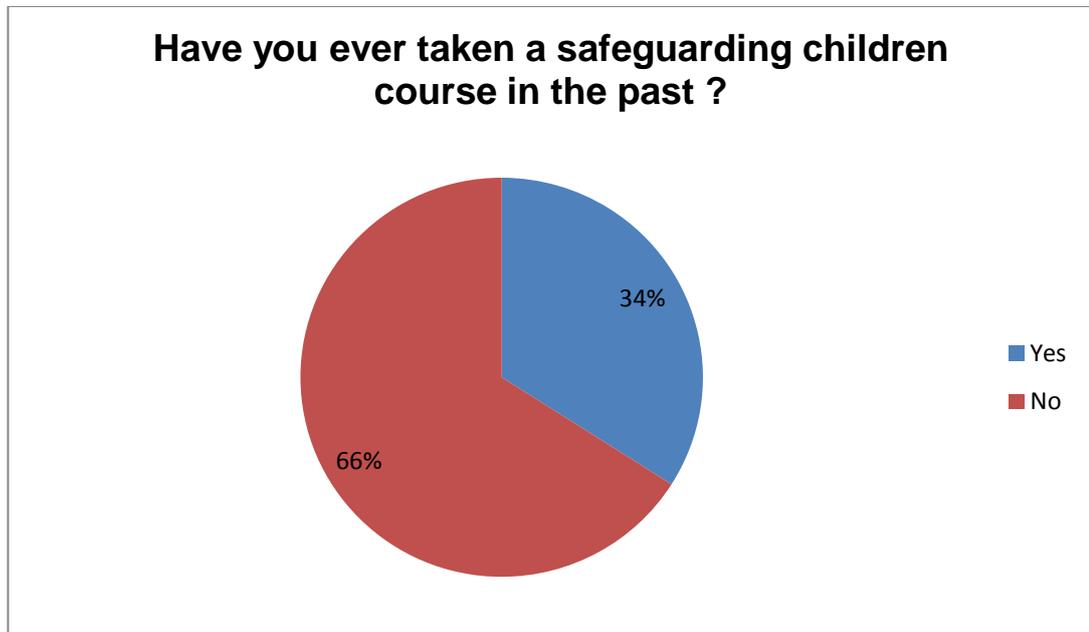


Figure 5. Distribution of 1st year postgraduate students who had safeguarding children course in the past

From those who had taken a course on safeguarding children, 10 students had been trained a year ago, 3 students 2 years ago, 2 students 3 years ago and 2 students five years ago. Only 3 students indicated the level of training in safeguarding children with two students saying it was level 2 while 1 student indicated level 1 training.

When asked whether they felt confident regarding safeguarding children and the actions that should be taken on a scale of 1 (not confident) to 5 (very confident) – the majority put a score of 3 as below.

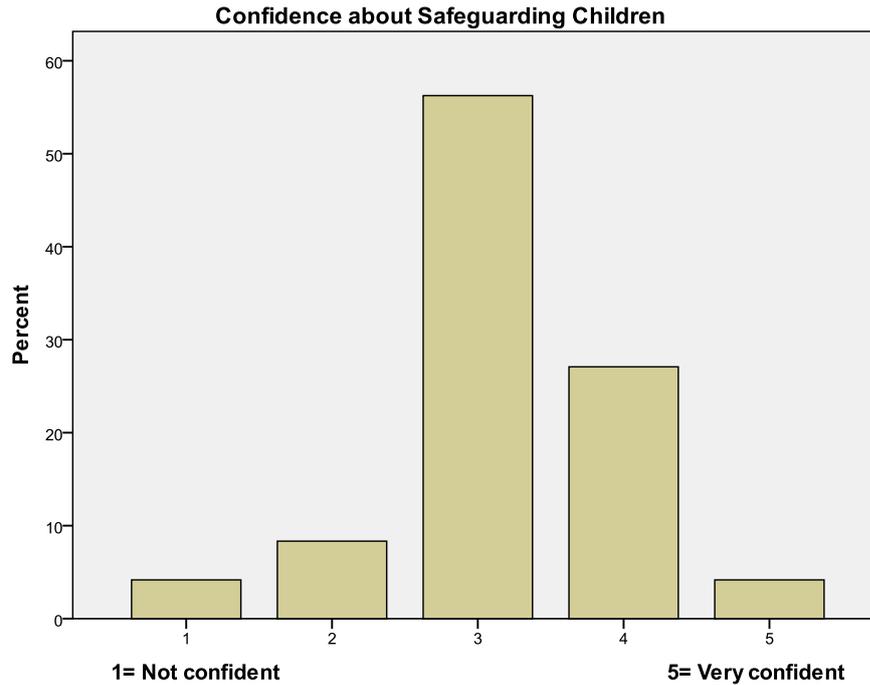


Figure 6. Confidence level of 1st year postgraduate students about safeguarding children

Asked whether they were aware of documents or guidelines on safeguarding, 16 students indicated that they were aware while 31 students were not. Four students did not give any answer.

On the issue of being aware of NICE guidelines as attributed to safeguarding, only 9 students indicated awareness of their existence, 41 students had no idea and one student did not answer the question.

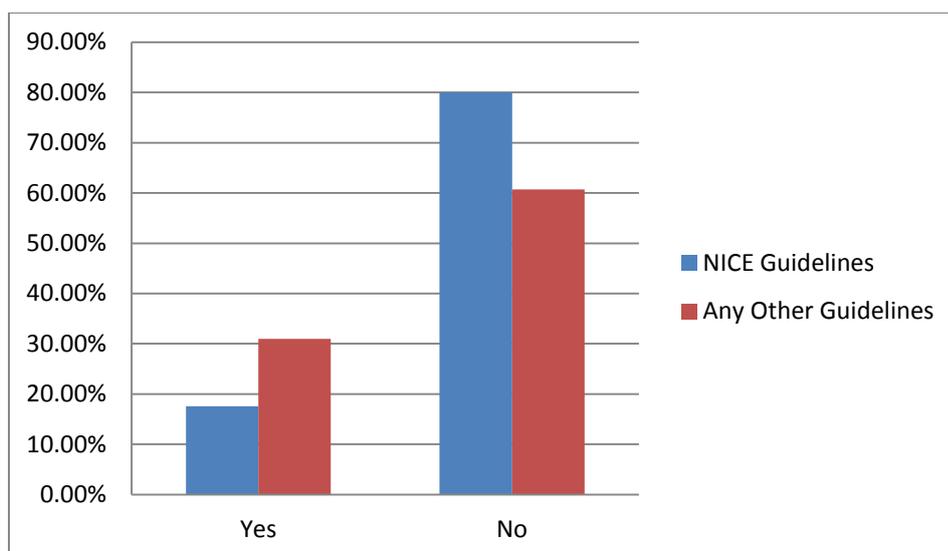


Figure 7. Awareness of the 1st year postgraduate students of the NICE guidelines or any other documents or guidelines regarding safeguarding children.

The participants were required to indicate whether they *had received* any documents or guidelines on safeguarding children in their previous workplace. Twenty one students indicated that they had received some however 29 said they had not. When asked whether the participants could name any Oro-Facial signs of abuse (e.g. bruises, bites, burn, etc), 12 (24.5%) students only named one sign while 19 (38.8%) students managed to name only two signs, and other results are included in the table below. Two students did not answer the question.

Number of Oro-Facial signs of abuse given by students	Number of students (%)
1	12 (24.5%)
2	19 (38.8%)
3	14 (28.6%)
4	2 (4%)
5	2 (4%)

Table 4. The number of Oro-Facial signs of abuse identified by the 1st year postgraduate students

Regarding types of abuse, the results are as shown in figure 8.

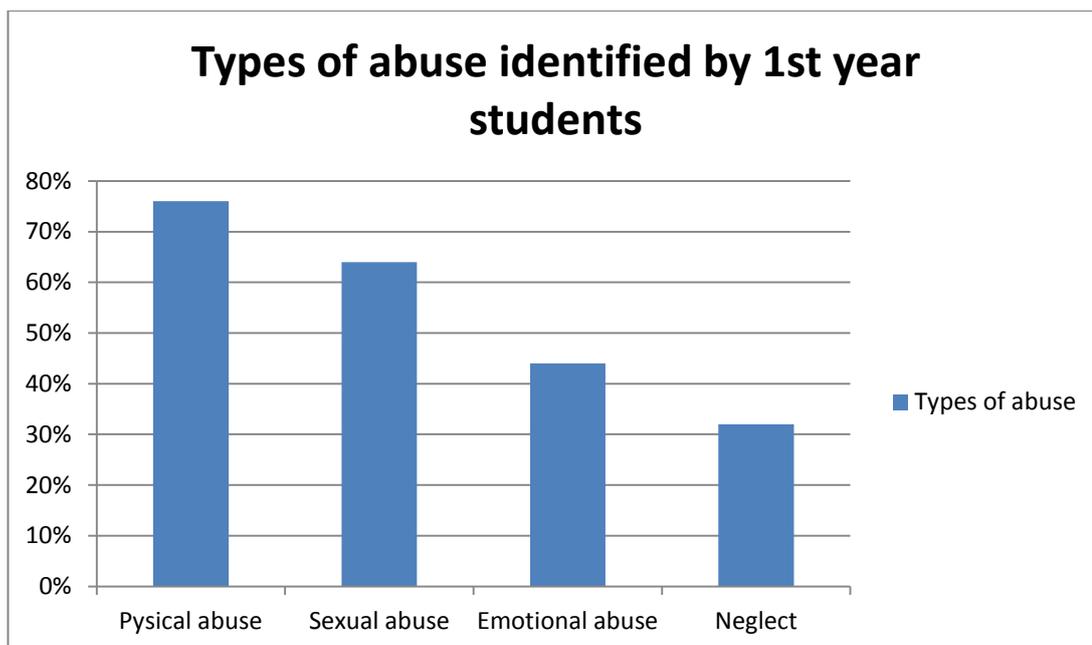


Figure 8. Types of abuse the 1st year postgraduate students can identify

Participants were asked what they would do if they suspected that there were safeguarding issues, albeit uncertain. Three students said they would take action, 47 students indicated that they would discuss with colleagues.

If you suspected but were not certain of a safeguarding issue, what would you do?	% (n)
Take action	6% (3)
Discuss with colleague	94% (47)
Take no action	0% (0)

Table 5. Responses of what to do if suspect safeguarding issue as given by 1st year postgraduate students

When asked whether they were responsible for safeguarding the children or grandchildren of an adult patient as a clinician, 25 (49%) students answered yes. Twenty two (43%) students said they would not be responsible. Four students did not answer this question.

The questionnaire sought to establish from participants who was responsible for safeguarding children. One participant indicated that it was clinician’s responsibility, 2 students said it was the responsibility of other team members, 1 student said it was administration staffs’ responsibility while the majority (46 students) indicated that it was everyone’s responsibility. This can be seen clearly below in figure 9.

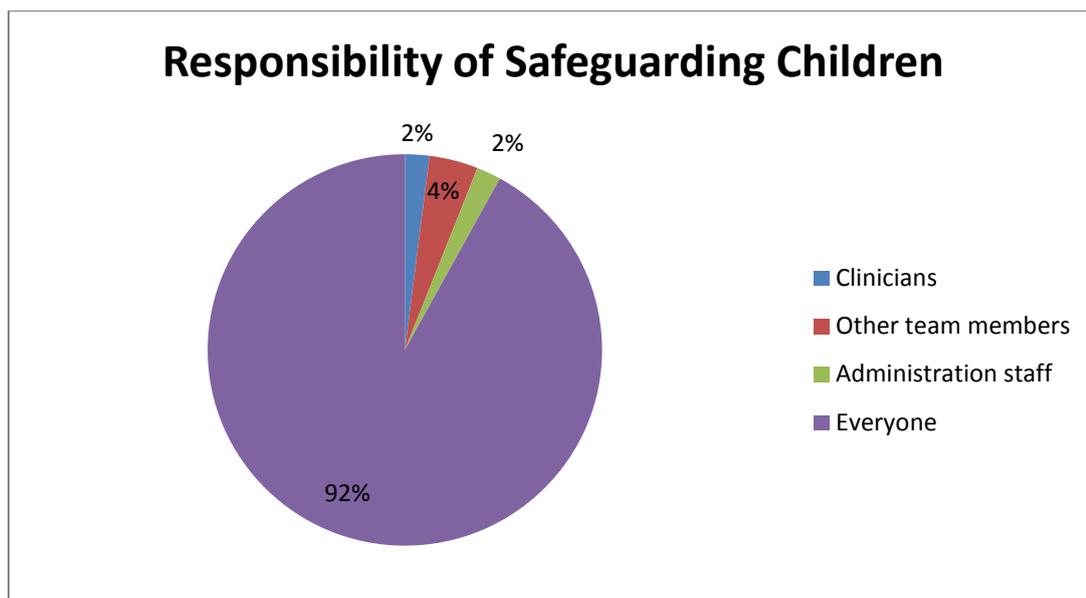


Figure 9. The responsibility of safeguarding children relays on whom as thought by 1st year postgraduate students

The participants were given two scenarios where a safeguarding issue might be involved and were asked to give their reaction if they faced such incidents.

A 5 year old child who has multiple carious teeth misses 2 appointments for treatment under general anaesthesia.	% (n)
Refer to social services	12% (6)
Discuss with colleague	88% (44)
Do nothing	0% (0)

Table 6. Responses to scenario number one by 1st year postgraduate students

<p style="text-align: center;">A child with dental trauma attends your practice. Their account of the trauma changed three times and the stories do not fit the injury. The child interaction with the parents is unusual. The presentation is delayed (2 weeks)</p>	<p style="text-align: center;">% (n)</p>
<p style="text-align: center;">Refer to social services</p>	<p style="text-align: center;">56% (28)</p>
<p style="text-align: center;">Discuss with colleague</p>	<p style="text-align: center;">44% (22)</p>
<p style="text-align: center;">Do nothing</p>	<p style="text-align: center;">0% (0)</p>

Table 7. Responses to scenario number two by 1st year postgraduate students

One student did not give an answer in the first and second

2 - Interpreting Questionnaire for 1st year students (Second attempt)

After a number of months on their courses – the same first year participants who had now had their safeguarding children training, were asked to repeat / complete the same questionnaire they had before.

The number of students was fifty one. Some results remained unchanged i.e. their ages and countries of origin, ration of males to female etc. Information was also the same regarding the course taken, providing dental treatment for children before and in the same places.

When asked whether they had participated in safeguarding children course in the past, 32 students said they had unlike in the previous survey where only 17 students had indicated that they had taken the course. This means that an additional 15 students had taken the training, but 18 students indicated that they had not taken the course.

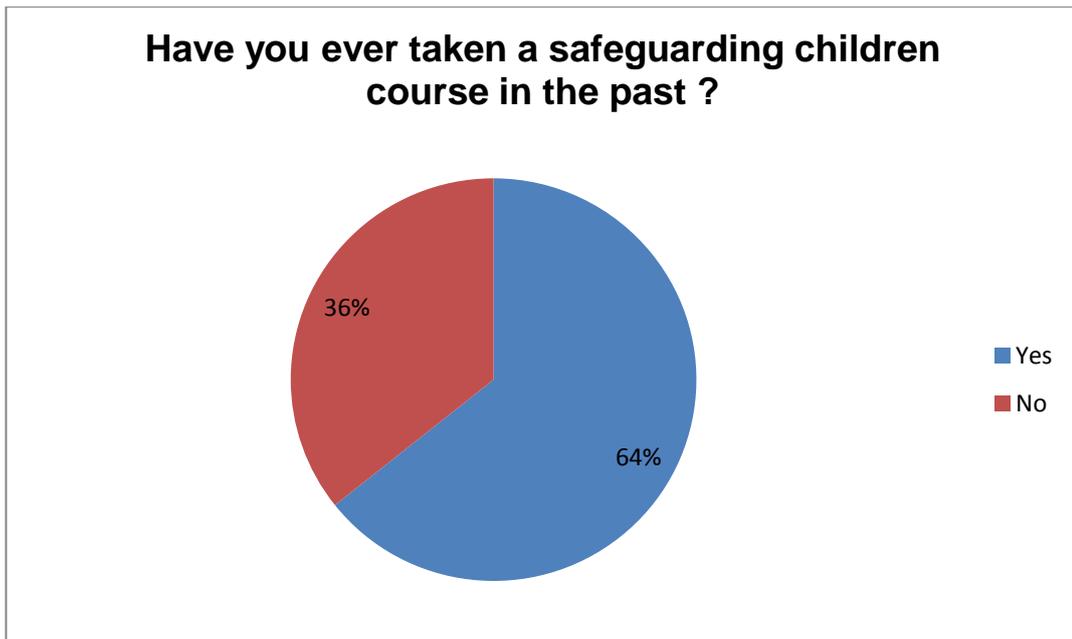


Figure 10. Distribution of 1st year postgraduate students who indicated they had safeguarding children course in the past (2nd attempt after training)

Regarding the confidence level about safeguarding children and the actions to be taken, there was an overwhelming level of confidence amongst the students. All of them were confident after the training. Thirty three students were not sure whether they were confident or not compared to previous 27. Thirteen indicated they were relatively confident while 2 students said they were very confident.

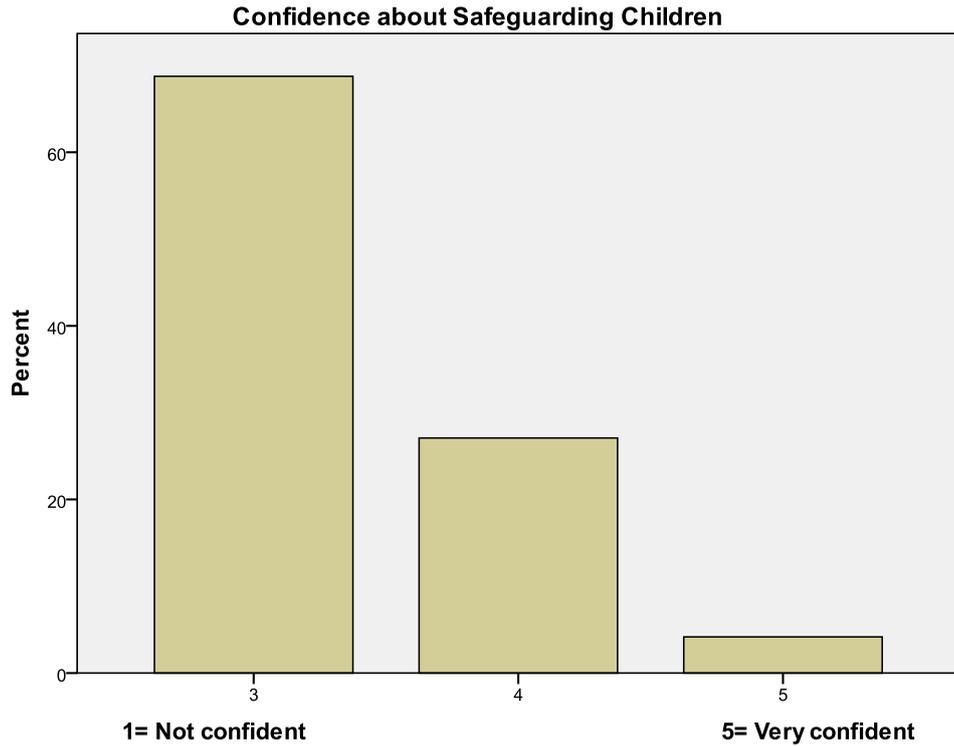


Figure 11. Confidence level of 1st year postgraduate students about safeguarding children post training

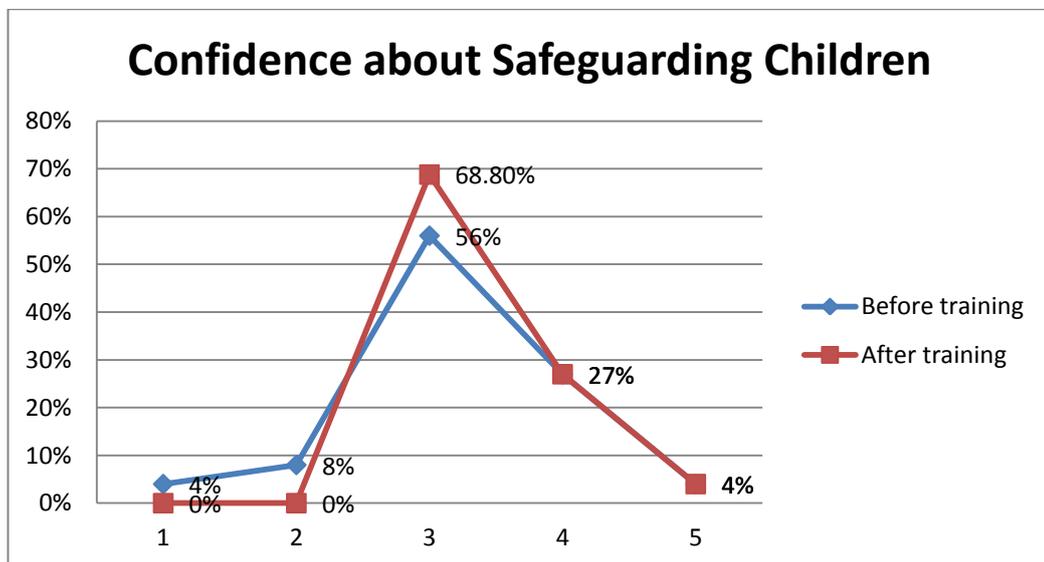


Figure 12. Comparison of level of confidence about safeguarding children for the 1st year postgraduate students pre and post training

With regards to awareness of the existence of documents or guidelines on safeguarding children. Forty three students indicated that they were now aware of the documents and guidelines. Only 5 students indicated that they were not aware.

Regarding the existence of any NICE guidelines in relation to safeguarding children, 44 students indicated that they were aware while 6 students answered negatively. In regard to receiving any documents or guidelines in the previous workplace, the results remained the same which were, twenty one students indicated that they had received - 29 said they had not.

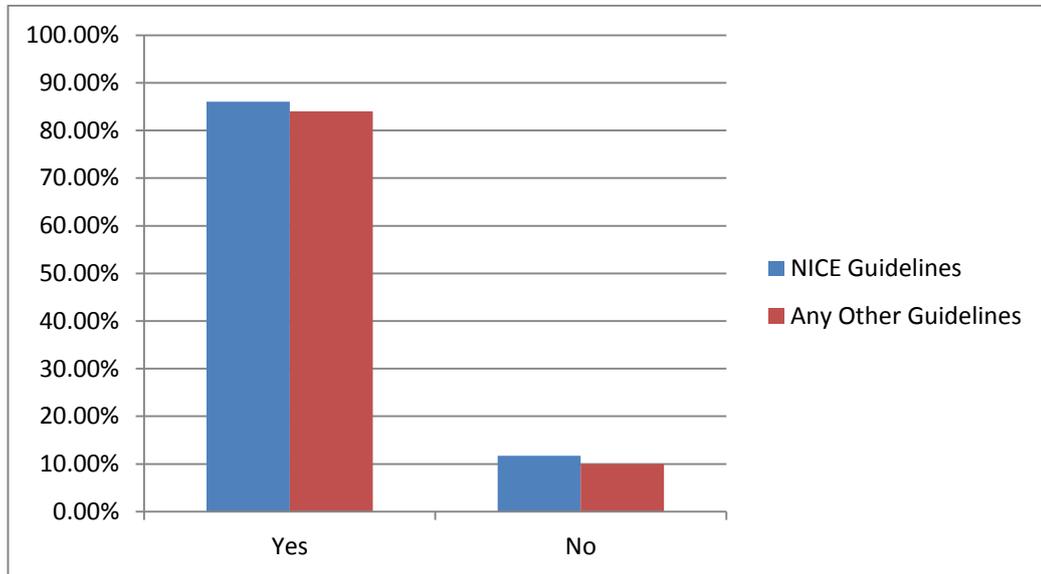


Figure 13. Comparison of awareness of the NICE guidelines or any other documents or guidelines by 1st year postgraduate students (2nd attempt after training)

When looking at types of abuse, 39 students identified emotional abuse. With regard to identification of sexual abuse, 47 students identified it while 4 could not. Forty seven students identified physical abuse and 27 students were able to list neglect as a type of abuse while 23 could not.

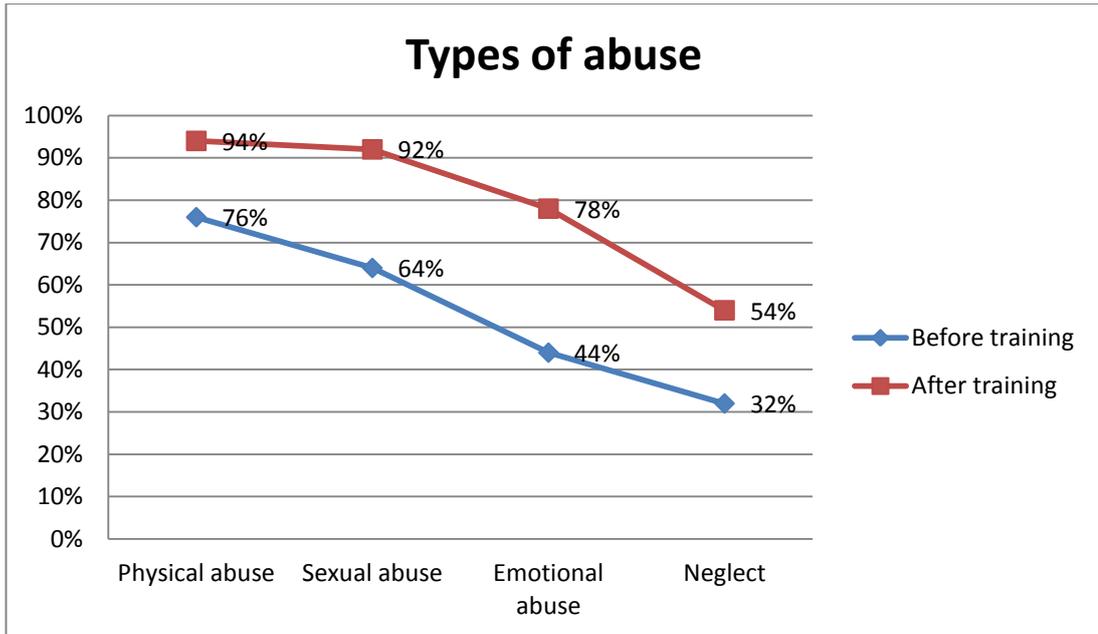


Figure 14. Types of abuse identified by 1st year postgraduate students before and after their training in safeguarding children

The number of Oro-Facial signs of abuse identified by the 1st year postgraduate students after training are shown in Table 8.

Number of Oro-Facial signs of abuse given by students	Number of students (%)
1	3 (6%)
2	12 (24.5%)
3	25 (51%)
4	7 (14%)
5	2 (4%)

Table 8. The number of Oro-Facial signs of abuse identified by the 1st year postgraduate students after training

Number of students who opted to take action upon suspecting safeguarding issues was 2. Forty eight students would discuss such as situation with a senior colleague.

If you suspected but were not certain of a safeguarding issue, what would you do?	% (n) Before training	% (n) After training
Take action	6% (3)	4% (2)
Discuss with colleague	94% (47)	96% (48)
Take no action	0% (0)	0% (0)

Table 9. Responses of the 1st year postgraduate student to suspicion of safeguarding issue.

None of the students felt that safeguarding children is the responsibility of administration staff or other team members. Forty nine (98%) students indicated that it was the responsibility of everyone. Only one participant said it was the clinician's responsibility. Students who felt they should discuss a situation where they suspect safeguarding issues with a senior colleague to increase to 48 (94%) compared to previous survey where 44 (86%) students would discuss the situation with a colleague.

When asked whether they were responsible for safeguarding the children or grandchildren of an adult patient as a clinician, 41 (87%) said yes compared to 25 (49%) students before training, and only 6 (12%) students compared to 22 (43%) students said they would not be responsible.

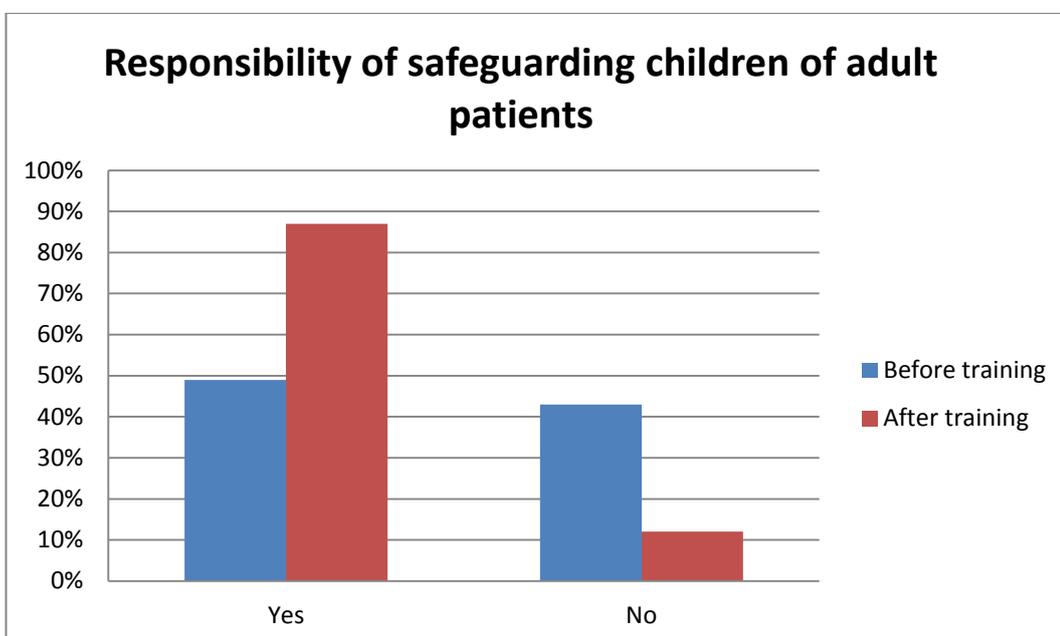


Figure 15. Distribution of 1st year postgraduate students who thought as clinicians they were responsible of safeguarding children of their adult patients.

When faced with the same two scenarios given before where safeguarding issue might be involved, the responses of the first year students changed. For the first scenario, the number of students who would discuss the case with a colleague increased from 44 (88%) to 48 (96%) and the number of students elected to refer to social services decreased from 6 (12%) to 2 (4%). The number of participants who would discuss with a senior colleague for the second scenario increased to 34 (68%) students compared to 22 (44%) students who would discuss the same situation with a colleague. Those who would refer to social services decreased from 28 (56%) to 16 (32%) students. Only one student did not give an answer to both scenarios.

A 5 year old child who has multiple carious teeth misses 2 appointments for treatment under general anaesthesia.	% (n) Before training	% (n) After training
Refer to social services	12% (6)	4% (2)
Discuss with colleague	88% (44)	96% (48)
Do nothing	0% (0)	0% (0)

Table 10. Responses to the first scenario by 1st year postgraduate students

A child with dental trauma attends your practice. Their account of the trauma changed three times and the stories do not fit the injury. The child interaction with the parents is unusual. The presentation is delayed (2 weeks)	% (n) Before training	% (n) After training
Refer to social services	56% (28)	32% (16)
Discuss with colleague	44% (22)	68% (34)
Do nothing	0% (0)	0% (0)

Table 11. Responses to the second scenario by 1st year postgraduate students

3 - Interpreting Questionnaire for 2nd year students

When the same questionnaire was presented to 2nd year postgraduate students, the results were different. The total number of participants was 20. The age range varied and was between 27 and 34 years. The number of males was 9 and 11 females. More UK graduates presented in the second year postgraduates (45%). The number of courses taken was five: Paediatric (4 students), Orthodontic (7 students), Periodontic (5 students), Oral surgery (1 student) and Prosthodontic (3 students).

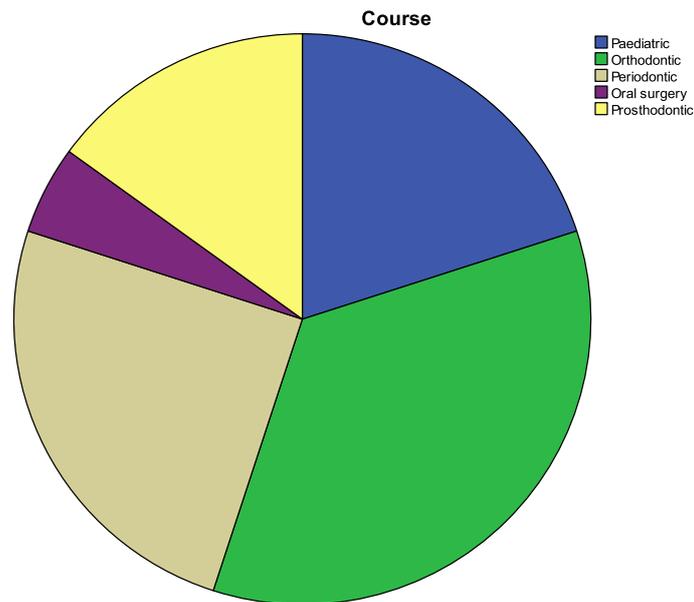


Figure 16 Distribution of 2nd year postgraduate students from each department at the EDH

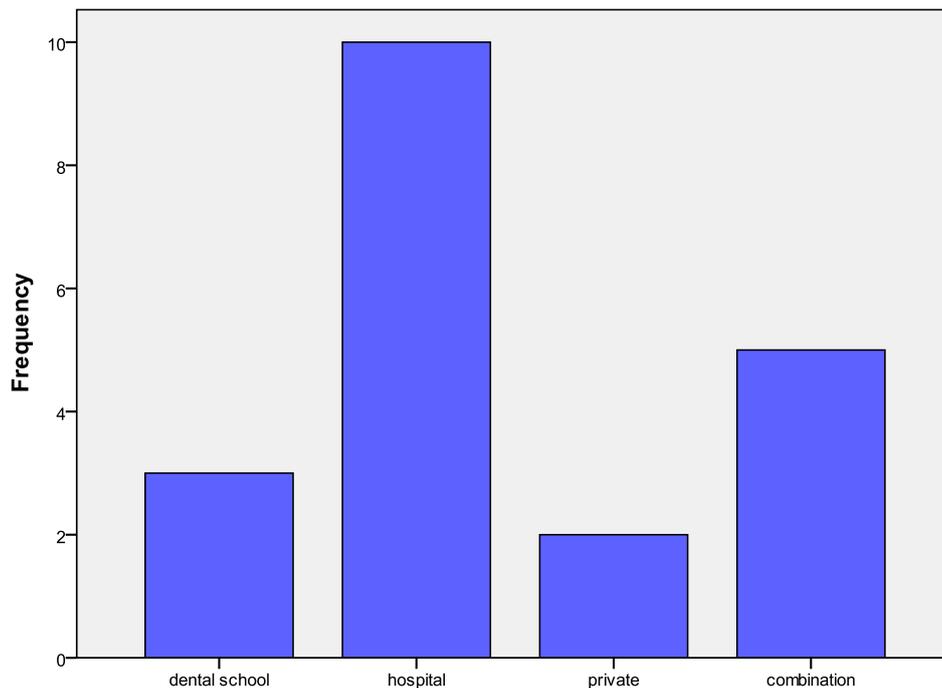


Figure 17. Places where 2nd year postgraduate student provided dental treatment for children before

All the students had provided dental treatment to children before. Three students had provided treatment at dental school, 10 in hospitals, 2 in private and 5 in both hospitals in private. Eleven students felt that they would treat children in the institution while 9 felt otherwise.

A bigger percentage (65%) of the students indicated they had not taken safeguarding course before compared to (36%) for 1st year students. In addition to the questions, students were asked whether they felt anxious about safeguarding children. Five students said they felt anxious while 14 students felt not anxious about safeguarding children and one student did not answer the question.

Eight students indicated they were not aware of any guidelines on safeguarding children. Eight students (40%) indicated that they were aware of NICE guidelines.

On whether they had received any guidelines about safeguarding children in previous workplace, 9 students (45%) indicated they had, while 11 students (55%) said they had not.

Seven students (35%) were being able to identify emotional abuse. Twelve students (60%) said they would identify sexual abuse. Fourteen students (70%) indicated

they would identify physical abuse. Sixty percent of 2nd years would identify any type of neglect. The number of Oro-Facial signs of abuse given by the 2nd year postgraduate students is show in Table 11 below. Four students (20%) did not give any Oro-Facial signs of abuse.

Number of Oro-Facial signs of abuse given by students	Number of students (%)
1	5 (25%)
2	8 (40%)
3	1 (5%)
4	2 (10%)
5	0 (0%)

Table 12. The number of Oro-Facial signs of abuse identified by the 2nd year postgraduate students.

Twenty percent of students would take action if they suspected safeguarding issues. Ten percent would take no action While 70 percent would discuss the situation with a senior colleague.

In answering whether they would be responsible for an adult patient’s children, 12 students (63%) answered yes while (37%) answered no. Seventeen (85%) students indicated that it is the responsibility of everyone to safeguard children, 2 students (10%) mentioned clinicians and 1 student (5%) mentioned other dental team members.

Regarding the scenario where the student suspected safeguarding issues, the results are shown in the tables below.

A 5 year old child who has multiple carious teeth misses 2 appointments for treatment under general anaesthesia.	% (n)
Refer to social services	10% (2)
Discuss with colleague	85% (17)
Do nothing	5% (1)

Table 13. Responses to scenario number one by 2nd year postgraduate students

A child with dental trauma attends your practice. Their account of the trauma changed three times and the stories do not fit the injury. The child interaction with the parents is unusual. The presentation is delayed (2 weeks)	% (n)
Refer to social services	30% (6)
Discuss with colleague	65% (13)
Do nothing	5% (1)

Table 14. Responses to scenario number two by 2nd year postgraduate students

4 - Interpreting Questionnaire for 3rd year students

When the same questionnaire was presented to 3rd year postgraduate students, the number of students participated in the questionnaire were 20 with 10 males and 10 females. Their mean age was 30 years. Their mean year of graduation was 2004 (range 2001 - 2007), and the UK graduates consisted (65%) of the group. The number of courses taken was five and the results are shown in the chart below.

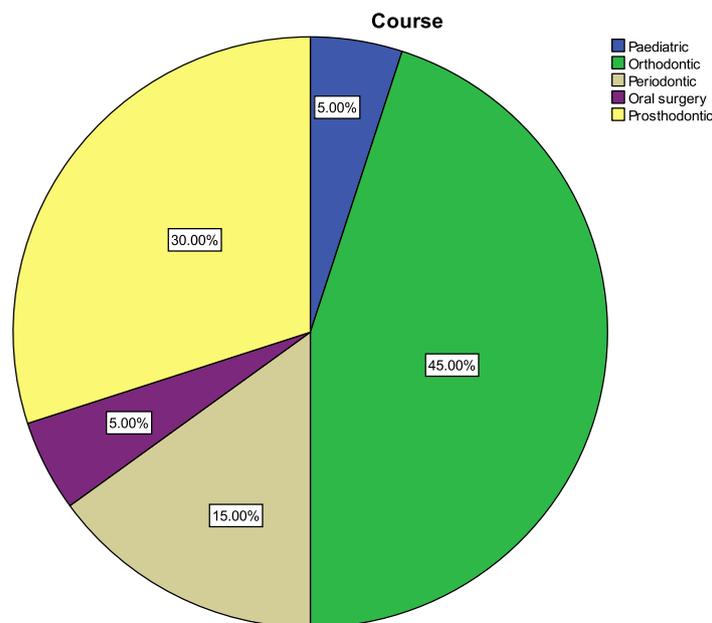


Figure 18. Distribution of 3rd year postgraduate students from each department at the EDH

All the students had provided dental treatment to children. Four students provided treatment in hospitals, 2 students in private and 14 students (70%) in both hospitals and private.

Half of the students (50%) expected to treat children in the institution, and half of the students (50%) indicated taken safeguarding children course in the past, while the other half did not believe they had any training in safeguarding children in the past. When asked if they were anxious about safeguarding children, 11 (55%) students said they feel anxious and 9 (45%) were not anxious.

Only 31 percent (6 students) were aware of documents on safeguarding of children, and the majority of students were not aware of the NICE guidelines regarding safeguarding children.

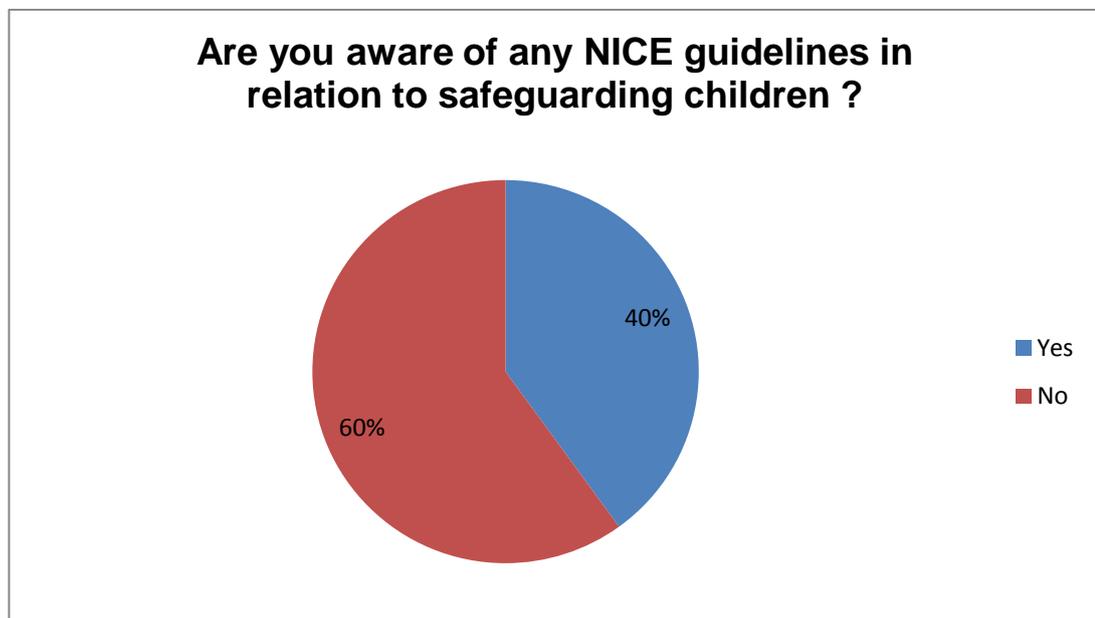


Figure 19. Awareness of NICE guidelines among 3rd year postgraduate students.

Forty percent (8 students) had previously received guidelines in former workplace.

Seven students (35%) could identify emotional abuse, 45 percent (9 students) could identify sexual abuse, 18 students (90%) indicated they would identify physical abuse, and 40 percent (8 students) could identify neglect as a type of abuse.

The numbers of Oro-Facial signs of abuse given by the 3rd year students are shown in Table below. Four students could not answer the question.

Number of Oro-Facial signs of abuse given by students	Number of students (%)
1	5 (25%)
2	5 (25%)
3	4 (20%)
4	1 (5%)
7	1 (5%)

Table 15. The number of Oro-Facial signs of abuse identified by the 3rd year postgraduate students.

The majority of students (75%) said they will discuss with senior colleague if they suspected but were not certain of safeguarding issue. Only two students (10%) chose to take no action and 3 students (15%) would take action.

In answering whether they would be responsible for an adult patient's children, 15 students (75%) answered yes, while 4 students (20%) said no. Eighteen (90%) students indicated that it is the responsibility of everyone to safeguard children, 1 students (5%) mentioned clinicians and 1 student (5%) mentioned other dental team members.

The 3rd year postgraduate students were given the same two scenarios give to the 1st and 2nd year students where a safeguarding issue might be involved and were asked to give their reaction if they faced such incidents. The results are listed in tables below.

A 5 year old child who has multiple carious teeth misses 2 appointments for treatment under general anaesthesia.	% (n)
Refer to social services	5% (1)
Discuss with colleague	90% (18)
Do nothing	5% (1)

Table 16. Responses to scenario number one by 3rd year postgraduate students

<p>A child with dental trauma attends your practice. Their account of the trauma changed three times and the stories do not fit the injury. The child interaction with the parents is unusual. The presentation is delayed (2 weeks)</p>	<p>% (n)</p>
<p>Refer to social services</p>	<p>35% (7)</p>
<p>Discuss with colleague</p>	<p>65% (13)</p>
<p>Do nothing</p>	<p>0% (0)</p>

Table 17. Responses to scenario number two by 3rd year postgraduate students

DISCUSSION

1- First attempt for 1st year postgraduate students

The results of the first year postgraduates before and after training were different. Initially in the questionnaire responses, although the majority had been relatively recently qualified from universities, they may not have had a lot of experience associated with identifying children who needed safeguarding or teaching on the subject. Also, practice varies according to country and culture.

There were some inconsistencies between the answers given by 1st year students in their first attempt. For example, when the participants were required to indicate whether they had received any documents or guidelines on safeguarding children in their previous workplace, 21 students indicated that they had received and 29 said they had not. This is inconsistent as in a previous question, only 16 students had indicated that they were aware of the guidelines yet 21 had received documents or guidelines.

Even though (98%) of the 1st year postgraduate initially elected to take action on both scenarios presented to them regarding safeguarding, either to discuss with colleague or refer to social services, one student (2%) did not give an answer to both scenarios which mean that not all students (100%) managed to give the correct answer as set by our standards for this audit.

2- Second attempt for 1st year postgraduate students

Even though they all had training in safeguarding children as part of their common core course in their first year at the EDH, when asked whether they had participated in safeguarding children course in the past, 32 students said they had, unlike in the previous survey where only 17 students had indicated that they had taken the course. This means that an additional 15 students had taken the training, but 18 students indicated that they had not taken the course.

Forty three students indicated that they were now aware of the documents and guidelines compared to 16 students in the previous survey.

The training seems to have increased the awareness regarding the existence of any NICE guidelines in relation to safeguarding children. After being trained, 44 students

indicated that they were aware while 6 students answered in the negative. This is in comparison to 9 and 41 students who had answered yes and no previously respectively. The training seems to have increased students' ability to identify emotional abuse as a type, 39 students could now identify, up from 22 students. Eleven students could not identify emotional abuse now compared to 28 who previously could not.

With regard to identification of sexual abuse, 47 students could now identify while 4 said they could not. This is in contrast to previous survey in which 32 students could identify and 18 students could not. Forty seven students could identify physical abuse but 3 students still could not identify. This is an increase of 9 students who could identify. Twenty seven students could now identify neglect, an increase of 11 students from previous survey.

There was a significant increase in students who could name Oro-Facial signs of abuse. The training decreased the number of those who could only list one sign of Oro-Facial abuse (from 12 to 3). More students can now identify at least 3 signs (from 14 to 25). However the number of students who could identify 5 signs of Oro-Facial signs of abuse remained the same (5).

The training helped increase the number of students who felt they were responsible for an adult patient's children or grandchildren from 25 to 41. There has been an increase in the number of students (8%) who now believe that safeguarding children is the responsibility of every one. The increase may be attributed to students' understanding of safeguarding children as the responsibility of not only health staff and professionals, but everyone. The understanding also led to students who felt they should discuss a situation where they suspect safeguarding issues with a senior colleague to increase from (86%) to (94%). The training had helped increase the confidence level of 6 students.

When faced with the same two scenarios where safeguarding issue might be involved, before and after training, more students elected to discuss the issue with a senior colleague after training than before, but fewer students chose to refer the case to social services.

3- Second year postgraduates' responses

When presented the same questionnaire to the 2nd year postgraduate students the results were different again and there were variations in the responses given. Due

to the variations between the length of each programme, the number of students were less than the first year students since there were some students who are enrolled in a one year program and others enrolled in two or three years programme.

Although they had safeguarding training as part of their introductory course in their first year, interestingly more students in the 2nd year (65%) believe they did not have safeguarding course before than the 1st year students (36%) after training. Five students indicated they were anxious about safeguarding children. This is significant and should be picked up and investigated during training. When assessing their awareness of any documents or guidelines regarding safeguarding children, more students in the 2nd year (40%) were not aware of any guidelines or documents compared to and (10%) in the 1st year students after training. This highlights the importance and the need of emphasising guidelines and protocols training during the induction course. Similarly when asked about their awareness of the NICE guidelines, the number of students who were not aware of the NICE guidelines increase from 1st to 2nd year (12%, 40%) respectively.

Fewer students in the 2nd year were able to list all four types of abuse compared to the 1st year students. Almost similar numbers of students across all three groups were able to list the same number of Oro-Facial signs of abuse, with 4 students in the 2nd year could not answer the question which is unacceptable and can be due to lack of knowledge in the subject.

There was an overwhelming difference in the reaction to suspicion of child maltreatment where (96%) of the 1st year students decided they will discuss the case with a senior colleague compared to (70%) of the 2nd year students. When asked about their responsibility toward safeguarding adult patient's children more student's in the 1st year students (96%) answered yes compared to the 2nd year students (63%). Ten percent indicated they would take no action if they suspected safeguarding issues which is unacceptable response in these situations. In both scenarios (5%) of the students selected "do nothing" as an answer which highlight the need for more teaching.

4- Third year postgraduates' responses

These were similar to the 2nd year postgraduates as discussed but more students in the 3rd year (50%) believed they had not had a safeguarding course before than the

1st year students (36%) after training. This could be a result of information overload accumulated over the three years. More students in the 3rd year (55%) felt anxious about safeguarding children than in the 2nd year students (25%). This percentage could be possibly lowered if anxiety about safeguarding children was assessed during the appraisal process and support / education provided. When assessing their awareness of any documents or guidelines regarding safeguarding children, more students in the 3rd year (68%) were not aware of any guidelines or documents compared to the 2nd year (40%) and (10%) in the 1st year students after training. Perhaps this is because training or protocols are not visible on the clinic. The number of students who were not aware of the NICE guidelines increases from 2nd to 3rd year (40% to 60%). Fewer students in the 3rd years were able to list all four types of abuse compared to the 1st year students. However, when compared with the 2nd year students, more 3rd year students managed to identify physical abuse (90%) than (70%) of 2nd year students. Similar to the 2nd year, 4 students did not identify any Oro-Facial signs of abuse which considered a poor response. Two students (10%) said they will take no action if they suspected child maltreatment which is bad and can be contributed to fear or lack of understanding to protocols and procedures. Four students (20%) thought they were not responsible of safeguarding children's of their adult patients, and it might be due to focusing their attention on children patients rather than all contacts. Only one student decided to "do nothing" in scenario 1 while in scenario 2 all students chose to take action. This is perhaps because trauma is more obvious than dental caries or neglect.

As stated at the start the gold standards set for this audit were that,

1. 100% of all postgraduate students should correctly answer key questions on the questionnaire (*3 of them and 2 scenarios*) as identified with the Trust safeguarding Lead Paediatrician:
 - With regard to the response if suspected safeguarding issue, only (96%) of the 1st year, (70%) of the 2nd year, and (75%) of the 3rd year students decided to take action which is the correct answer.
 - When asked about their responsibility to safeguard the children of adult patients, (87%) of the 1st year, (63%) of the

2nd year, and (73%) of the 3rd year gave the correct answer which is **yes**.

- Eighty nine percent of the 1st year, (85%) of the 2nd year, and (90%) of the 3rd year believed that safeguarding children is the responsibility of every one.
- For the first scenario, only (96%) of the 1st year, (95%) of the 2nd and 3rd year students gave the correct answer by taking action, although (5%) of the 2nd and 3rd year students decided to do nothing.
- For the second scenario, (100%) of the 1st year, (95%) of the 2nd year, and (100%) of the 3rd year gave the correct answer by taking action in such situation.

A 100% correct response was not achieved for all the key questions, which high light the need for more training and assessment about safeguarding children.

2. When the first year questionnaires were repeated – results were the same or better.
 - The result indicated there was an improvement in the responses given by the 1st year students, which high lights the positive effect of training.
3. Second and third year responses same or better than second first year responses.
 - The responses of the 2nd and 3rd year students were not better than the second 1st year responses. This could be due to lack of continues training and assessment about safeguarding children.
4. All postgraduates should have had some form of safeguarding training in the repeat first year, second and third year groups.
 - Not all postgraduates indicated they had some form of safeguarding training in the past. Some students still could not remember if they had safeguarding training as part of their induction course, which could be due to the increased academic overload. This high light the emphasis to continue

expressing the importance of safeguarding children during training.

CONCLUSION

Not all our audit standards were reached and a lack of knowledge about safeguarding children has been identified in postgraduate students at EDH, UCLH. This could be due to shortage of training and education about this subject. The data suggests that training in safeguarding children helped 1st year students to be more aware, and as a result more competent, with safeguarding children. The first year postgraduate students showed more knowledge about the subject than the second and third years. The information retention regarding safeguarding children seems to be reduced with postgraduates as they advance through years of their course. Targeted training may help this in the future.

AUDIT OUTCOMES / ACTION

The results of this audit will be presented locally to the paediatric department staff at a meeting in September, 2012. Presentation to the Trust safeguarding committee will be later in the year.

To liaise with the Trust safeguarding Lead Nurse and Paediatrician and the Clinical Director and Dean of EDH / EDI respectively to look at developing targeted training in safeguarding children for all postgraduate students at EDH.

Remind all areas in the hospital – NHS and academic appointments that safeguarding children must always be a regular part of clinical governance and fed back to health and safety and quality assurance committees.

There will be a re-audit of safeguarding knowledge of all postgraduate students at the Eastman Dental Hospital in December 2012.

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I would like to thank all the postgraduate students at the Eastman Dental Institution for their participation in this project.

APPENDICES

APPENDIX A

Safeguarding Knowledge, Data Collected 6-10-11

1- Name: _____

(purely for audit recording – will not be disclosed)

2- Age in years _____

3- Gender: Male Female

4- Country the Dental degree awarded from? : _____

5- In what year? : _____

6- Course taking now at the Eastman? (eg. Ortho) : _____

7- Length of course? : _____ year

8- Full time

9- Part time If yes, rest of time Hospital Private practice
General NHS Practice Hospital No other work Combination

10- Have you provided dental treatment for children before?

Yes No

If yes, When? : Last 12 months 1-2 years ago 2 - 4 years ago

>4 years ago

Where? : Dental school Hospital Private practice
Combination

11- Do you expect to treat children as part of your EDI Course?:

Yes No

12- Have you ever taken a safeguarding children course in the past?

Yes

No

If yes approximately how long ago? : _____ year

If known, "level" of the course: _____

13- On a scale of 1 to 5 how confident are you about safeguarding children and actions to be taken?

Not confident very

confident

1

2

3

4

5

14- Are you aware of any guidelines or documents on safeguarding children? :

No Yes , Name if known:

15- Are you aware of any NICE guidelines in relation to safeguarding?

Yes No

16- Where you worked previously, did you receive any guidelines or documents about safeguarding children?

Yes No

17- Can you list 4 types of abuse? :

- 1.
- 2.
- 3.
- 4.

18- Can you name any Oro-Facial signs of abuse? :

19- If you suspected but were not certain of a safeguarding issue, would you,

(only tick one)

- Take action anyway
- Take no action
- Discuss with a senior colleague if possible or co-worker

20- As a clinician, if you were treating an adult patient, are you responsible for safeguarding their children or grand-children?

Yes No

21- Safeguarding children is the responsibility of:

(only tick one)

- Clinicians
- Other dental team members
- Every one
- Admin staff

What would you do in the following 2 scenarios?

- 3. A 5 year old child who has multiple carious teeth misses 2 appointments for treatment under general anaesthesia.**

Would you..... (only tick one)

- Do nothing
- Discuss your concerns with a more experienced colleague
- Refer to social services

- 4. A child with dental trauma attends your practice. Their account of the trauma changed three times and the stories do not fit the injury. The child interaction with the parents is unusual. The presentation is delayed (2 weeks)**

Would you (only tick one)

- Do nothing
- Discuss your concerns with a more experienced colleague
- Refer to social services

Thank you very much for completing this questionnaire

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