

ORIGINAL ARTICLE

Amelogenesis Imperfecta Restorations Survival Rate: A Retrospective Study

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ABSTRACT

Introduction: Amelogenesis imperfecta (AI) is a rare genetic disease affecting both dentitions. Factors such as age, socioeconomic status, dentition and AI type and severity should be taken into consideration in treatment planning. **Aim:** This retrospective study aimed to assess the survival rate of AI main restorative options and the effects of gender and dentition type. **Methods:** The study sample comprised 28 dental records of patients aged 5-17 years affected by AI (15 females, 13 males) and with anterior and/or posterior restoration in primary and/or permanent tooth/teeth. The fate of each restoration was classified into three categories: failed, withdrawn and censored and analyzed by the life table method of survival analysis. **Results:** Out of 233 restorations performed, the most frequently used restoration was preformed metal crowns (PMCs), followed by anterior composite, posterior composite, adhesive casting, Ketac fill and amalgam respectively. The two main restorations, PMCs and anterior composite were included in the statistical analysis. The survival rate of PMCs was significantly higher than anterior composite ($p < 0.001$). The anterior composite restorations survived significantly longer in males ($p < 0.05$). Females had significantly better survival rate of PMCs ($p < 0.05$). There was no statistically significant effect of the operator group of restoration survival. **Conclusion:** The anterior composite restorations survived significantly longer in males and females had significantly better survival rate of PMCs than males.

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INTRODUCTION

Some defects in dental enamel have been observed as inherited disorders primarily affecting enamel. Other defects in enamel have been associated with chromosomal aneuploidy, developmental defects and syndromes, metabolic and endocrine defects. Witkop defined Amelogenesis imperfecta (AI) as a hereditary defect of enamel affecting both the primary and permanent dentition (1). Several reasons for limiting the term AI to inherited defects primarily affecting only enamel were explained by Witkop (1). He mentioned that there are over 70 conditions in which intrinsic defects of enamel occur and any classification including all these disorders becomes unwieldy. The incidence of AI varies from 1 in 718 to 1 in 14,000 depending on the population studied (1,2). AI can be classified as hypoplastic, hypomaturation, hypocalcified, and hypomaturation-hypoplastic with taurodontism (1). In

addition, to dental sensitivity due to tooth wear, other clinical problems are associated with AI, such as delayed eruption and impaction of the teeth have been reported in the literature. Cases of anterior open bite also have been reported. Several case reports have stated that patients with AI seem to have a low caries activity (3-7). The increases susceptibility to dental caries might be due to changes in dental enamel that are related to amelogenin protein variation (1). Apparently amelogenin function can be affected by inflammation, and this can lead to changes in the structure of the dental enamel that later in life increases the risk of dental caries.

With the availability of various dental restorative materials and improved techniques, several studies have shown the use of glass ionomer cements, composite resin restorations and veneers, porcelain veneers, Preformed Metal Crowns (PMCs) or otherwise known as Stainless Steel Crowns (SSCs), laboratory fabricated crowns, and over dentures can be used to restore the affected primary and permanent dentitions (8-13). PMCs are one of the most effective and efficient restoration in restoring severely broken-down primary molars and young permanent molars in children (10, 11).

Treatment options of AI depend on the individual's specific diagnosis and phenotype. Unfortunately, because of limited studies and long-term follow up of restorative outcomes of patients especially children with AI, the available evidence is based on case reports. Therefore, there is a need of research on long-term prognosis and longevity of restorative outcomes of AI child dental patients. The aim of this retrospective study was to assess the survival rate of AI main restorative options performed in a post graduate paediatric dentistry department and to evaluate the effects of gender, operator, and dentition type.

MATERIALS AND METHODS

The study population consisted of dental records of 28 patients diagnosed with AI and treated during the period from 2001-2003 in the Division of Child Dental Health, Leeds Dental Institute, University of Leeds.

The selected dental records met the following criteria: accurate and legible; belonged to child dental patients aged 5-15 years and included one or more anterior and/or posterior restoration(s) in primary or permanent tooth/teeth. A special form for data collection was created to facilitate the collection and transference of information from each patient's record to a spreadsheet. The data sheet included data on both patient as well as restorations used, child's gender and the operator group (Postgraduate student/faculty). This study was approved by Research Ethics Committee, Leeds Healthcare/United Leeds Teaching Hospitals Trust No. 96/081.

The fate of each restoration was classified into three categories (14): failed, withdrawn and censored. A restoration was considered to have failed if it was partially or completely lost, repaired or replaced due to caries or pulp pathology or extracted due to defective restoration. A restoration was withdrawn if the tooth was extracted due to orthodontic reason but was healthy, the restored tooth was exfoliated and the restoration was intact the visit before or the tooth was extracted due to any pulp pathology that did not appear to result from a defective restoration. If the restoration survived until the last date the patient attended the hospital, then it was designated "censored".

All data were entered into a personal computer via a spread sheet. The survival time of the restoration, which is an estimate of the number of months between the placement of the restoration and the time of critical event, was analyzed by the life table method of survival analysis.

RESULTS

Patient variables

The population sample consisted of 28 dental records of patients affected with amelogenesis imperfecta (15

females, 13 males) and attending the Department of Pediatric Dentistry, Division of Child Dental Health, Leeds Dental Institute, The University of Leeds. All data were recorded by the same investigator (AZ). Five out of 28 (18%) records were re-recorded for assessing the investigators reproducibility. All the transcribed information had 100% agreement, except for oral hygiene (93%), use of rubber dam (97%), date of restoration (93%), date of assessment of restoration (97%).

The age group of the patients at the time of study ranged between 5-17 years. It was difficult to assess the exact mode of inheritance as there was no sufficient information regarding family history. The most frequent type of AI was hypoplastic type (13 cases, 46.4%) followed by hypomaturation and hypomineralization types (6 cases, 21.4% each) and two cases (7%) hypoplastic hypomaturation with taurodontism. Five patients (17.9%) had dental anomalies associated with AI. Two had supernumerary teeth, two had taurodontism, and one case was associated with generalized root resorption.

Restoration Variables

Various treatment modalities had been identified and the survival rate was calculated using the survival life table. Out of 233 restorations performed, the most frequent restoration was PMCs, followed by anterior composite, posterior composite, adhesive coping, Ketac fill, and amalgam respectively. Two types of restorations (Anterior composite and PMCs) were included in the statistical analysis using the life table method of survival analysis. The rest were excluded from the analysis because they were very few in numbers and cannot give meaningful statistics.

The survival curve for both PMCs and anterior composite were described in Figure 1 which showed that as the restorations were followed up over time, the difference between the survival success rates increased. For both restorations, the observed difference in survival curves was found statistically significant when using log rank test (Log rank test $\chi^2=27$, $df =1$, $p<0.001$). The survival rate at 40 months was 80% for PMCS and 50% for anterior composite.

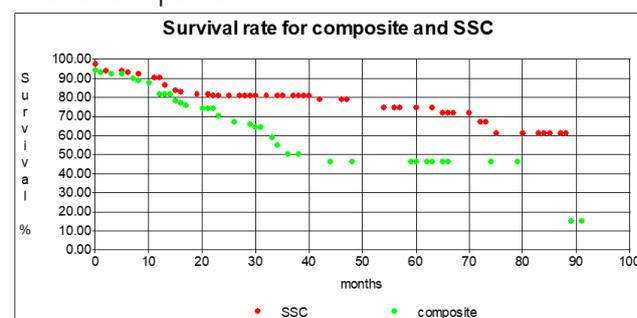


Figure 1: The survival rate for PMCs (SSCs) Vs composite restoration. Log rank test $\chi^2=27$, $df =1$, $p<0.001$. χ^2 -Chi Square; df - degree of freedom; p -Significance

Figure 2a describes the effect of gender in the survival curve for composite restoration. It can be noticed that the difference between the survival rates in females and males increased over time especially in males. The difference was statistically significant ($\chi^2= 4$, $df=1$, $p,0.05$). The survival rate at 30 months was almost the same for both genders (60%).

Figure 2b describes the effect of gender on the survival curve for PMCs restoration. It shows a difference in the survival rate in favour of females. Log rank test showed that this difference was statistically significant ($\chi^2= 6.41$, $df=1$, $p<0.05$). The survival rate at 60 months, however, was similar for both genders (approximately 75%).

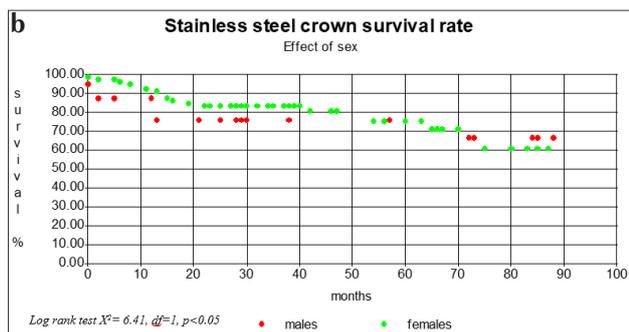
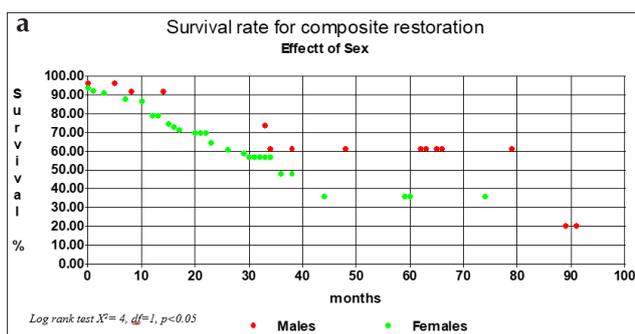


Figure 2: (a) The effect of sex on the survival rate for composite restoration (b) Effect of gender on survival rate for PMCs

The effect of operator group in the survival rate for composite restorations and PMCs are described in Figures 3 a,b. The operator group included teaching staff and postgraduates and not undergraduates because only few restorations were performed by undergraduates and could not be analyzed statistically. Visual comparison in these figures shows that there was small difference between the operator groups (teaching staff Vs postgraduates) for both types of restorations. This difference was not statistically significant ($\chi^2= 0.31$, $df=1$, $p = NS$) for composite and ($\chi^2= 0.00$, $df=1$, $p = NS$) for PMCs respectively.

The survival curve in Figure 4 describes the effect of dentition type (permanent Vs primary) on the survival rate for PMCs. Visual examination of this figure shows increased difference in the survival rate between the two dentition types. The difference was statistically significant using log rank test ($\chi^2= 27$, $df=1$, $p<0.001$).

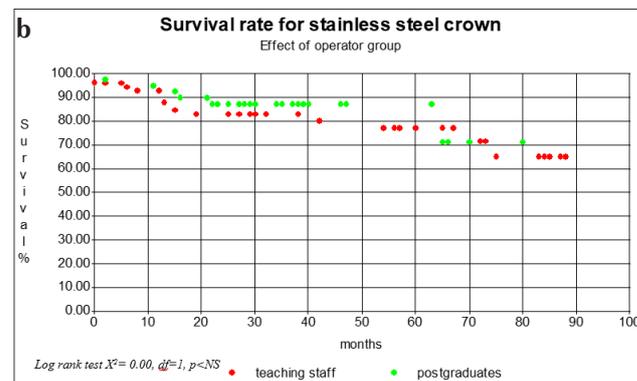
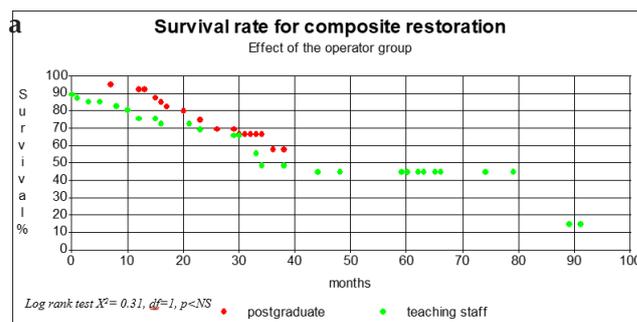


Figure 3: (a) The effect of operator group on the survival rate for composite restoration (b) Effect of operator group on survival rate for PMCs

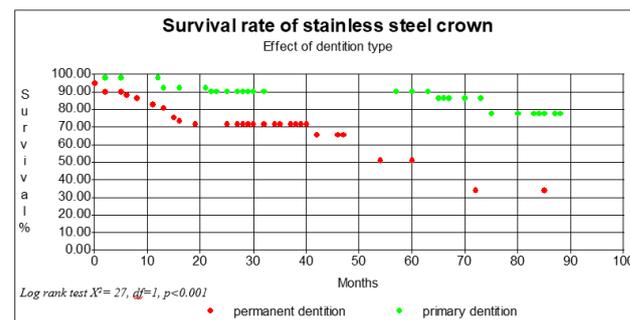


Figure 4: Effect of dentition type on survival rate for PMCs

DISCUSSION

In the present study the hypoplastic form of AI was the most frequent followed by hypomaturation and hypomineralization types. Other studies reported similar results with the hypoplastic form was the most frequent (2, 15-17). However, in a recent Norwegian study, Ohrvik and Hjortsju (18) with a smaller sample (15 children, mean age 17.3 years) reported one third (33.3%) were of hypocalcified type, 26.6% hypoplastic, 20% hypomaturation type and 20% not classified.

Taurodontism associated with Amelogenesis imperfecta has been reported by a few studies in the literature. For example, a study by Crawford (19) reported two separate families exhibited hypoplastic hypomaturation with taurodontism in their teeth. Likewise in the present study two cases were identified as having hypoplastic hypomineralization with taurodontism.

The survival life table method was utilized in this study. It divides the time into intervals and calculates survival at each interval. The main two restorations' (anterior composite and PMCs) scatter graphs showed that as the restorations were followed up over time the difference in the survival rates between the two restorations increased. This result was not expected with this statistically significant difference because of the usually less occlusal and leverage forces on the anterior teeth. From the patient's records, it was not possible to establish the exact reason for restoration failures and re-restoring the teeth. One of the reasons which might have contributed the lower survival rate of anterior composite veneers is the fact that composite veneers placed on anterior permanent teeth need re-restoring because of the eruption of teeth of young children over time. In a recent study by Mazzetti et al. in 2022 (20) the ceramic veneers had superior longevity than direct composite veneers in both success and survival analysis. However, composite veneers are considered the best available option in young dental patients. Collares et al (21) concluded that there was a difference in longevity of restorations for different teeth in the arch, with restorations in central incisors being the most susceptible to failure and replacement. They added that the risk for restoration failure increased in children below 12 years. They had a 17% higher risk for failure when compared with 18 - 25-year-olds.

There was a significant difference in the composite veneer restorations longevity between the females and males ($p < 0.05$) in the present study. This difference might be attributed to the fact that, females are more concerned about their aesthetics and because the composite veneers tend to discolor over time, females tend to visit the dentist and demand replacing the restoration to improve their aesthetics. Another potential factor which might explain the lower survival rate for composite veneers in females and males as mentioned above is the fact that most of the restorations were placed on anterior young permanent teeth which were not fully erupted and over time these teeth tend to erupt. The eruption of such teeth will expose the affected enamel which would concern the aesthetics and warranting replacing their restorations.

Ohrvik and Hjortsju (18) concluded in a retrospective study that prefabricated composite veneers should be avoided when possible, and direct composite restoration may be used as interim restorations in AI patients. However, all ceramic enamel-dentin bonded restorations are considered as first treatment option for young and old patients.

The survival rate of PMCs varied significantly between males and females with restorations placed in females survived longer. The reason was not clear but may be attributed to more occlusal forces in males that may be responsible for restoration dislodgement.

There was no statistically significant effect of the operator groups on the survival rate of both restorations. Because of the nature and the difficulty of AI cases treatment, most of the restorations in this study were performed by postgraduates. Collares et al (21) reported that anterior composite restorations placed by general dental practitioners showed an adequate clinical performance, with a significant difference in outcome among operators.

PMCs are considered one of the most cost-effective and efficient restoration in restoring severely broken-down primary molars and young permanent molars in children (10, 11, 22). Comparable results were reported in a prospective study (Zagdown et al. (14) on the longevity and success rates of PMCs and cast adhesive copings for the restoration of first permanent molars affected by either amelogenesis imperfecta or severe enamel hypoplasia. However, adhesive cast copings preserved more tooth tissue but were more technique sensitive and cost more than PMCs. The exact reason for lower survival rate of PMCs in the permanent molars in the present study was unknown but might be due to heavier occlusal forces in permanent dentition and/or the difference in anatomy in primary and permanent dentition. Recently, Chaipattanawan et al in 2022 (23) recommended PMCs for use as interim restorations in children's permanent molars, but their longevity may be constrained, requiring replacement over time.

One of the main potential limitations of this study is its retrospective design where the results are based on the accuracy of the recorded data. Nevertheless, the adopted strict inclusion criteria helped to achieve the aims of the study. As the available studies are mainly case reports, further retrospective and prospective research studies are needed to evaluate the clinical success and satisfaction of AI children and their parents utilizing different types of bonded restorations.

CONCLUSION

Within the limitations of this retrospective study. Hypoplastic form of AI was the most frequent. The anterior composite restorations survived significantly longer in males than females and females had significantly better survival rate of preformed metal crowns than males. Dental practitioners, therefore, should take in consideration the gender of the patient and dentition type when selecting the restorative material.

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