Original article

The Socio-demographic profile of children affected by Amelogenesis imperfecta and its impact on their families: a pilot study based on patients attending a Paedodontic clinic in Sri Lanka

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ABSTRACT

Aim: The appearance of teeth is fundamental to pleasing facial aesthetics and the health and well-being of a child. Amelogenesis Imperfecta (AI) is a disorder that affects the structure and appearance of the enamel of the teeth. The present pilot study aimed to assess the socio-demographic profile of children, affected by AI and its psychosocial impact on the families, compared to their non-affected siblings as perceived by mothers.

Method: Seventeen mothers with children affected by AI participated. The impact of the child's AI status on the family was assessed using a modified translated version of Family Impact Scale (FIS) which consisted of 27- items organized into 6 dimensions, namely parental/family impact, parental emotions, family conflict, impact on child in past/present context, perceived future impact on child and financial burden.

Results: Based on mothers respondents, the results revealed psychological impact as an overall score for 27 – items was 117.94 (±31.97) for affected families and the scores ranged from 62-174 which was significantly higher than the 0 score for controls. The highest median scores reported for *impact of AI on child's quality of life in past and present contexts* (median score= 39.0) and for *parental emotions* (median score=33.0). Mothers perceived a considerable impact of AI on child's quality of life in the future context as well (median score=21.0).

Conclusion: As emerged from the findings, there was a considerable perceived psychosocial impact on families reported by mothers having children with AI compared to their non-affected siblings. Thus it is important to provide necessary psycho-social support for those mothers.

Key words: Socio-demographic profile, Amelogenesis imperfecta, psychosocial impact

INTRODUCTION

Amelogenesis imperfecta (AI) is a rare inherited dental abnormality affecting the structure and clinical appearance of the enamel of both primary and permanent dentitions [1,2]. It could give rise to tooth disfiguration, sensitivity, tooth substance loss requiring life-long oral health care comprising of aesthetic dental treatment as well as oral hygiene improvement [3]. Hence, AI poses a challenge not only for affected children, but for their parents as well as oral health care system of the country.

The visible unaesthetic appearance of affected teeth with sensitivity by AI poses a significant psycho-social impact to affected children and adolescents such as negative self-perception, emotional disturbance, inhibited interactions, getting teased/ bullied, anxiety, low self-esteem, learning disabilities, low school achievement and social withdrawal [4,5,6,7]. Further, child's condition may have a negative impact on his or her family, predominantly the parents [8,9]. Therefore, AI could significantly influence the quality of life of affected children and their families, family impact assessment is important in this regard as highlighted by Locker et al., [7]. Despite, the great need for comprehensive assessment of the burden of AI in terms of occurrence, family impact and treatment; there is a dearth of information in respect to Sri Lankan patients. Such information is crucial for assessing the existing burden of AI in Sri Lankan context, to widen the existing epidemiological and clinical knowledge, as well as to plan efficient and effective oral health care services to the affected children and adolescents. Against this backdrop, present multi-component research study with a long-term follow up was aimed to address the remaining information gaps.

This manuscript is aimed to describe the psychosocial impact of having children affected by AI as perceived by mothers.

MATERIALS AND METHODS

All children with confirmed AI who presented to the Division of Pedodontics, Faculty of Dental Sciences, University of Peradeniya from January 2009 to December 2010 were included in the study. Hence, the non-probability consecutive sampling technique was used in this study. The condition was confirmed by clinic-pathological correlation. Accordingly, the sample consisted of 17 children affected by AI and 7 children from same families not affected by AI as the control group. The impact of the child's dental condition on the family was assessed by using: A pre-tested questionnaire which included the modified, translated version of Family Impact Scale (FIS) [7] for children with and without AI in the same family. This scale was used as there was no validated instrument to assess the impact of AI in Sri Lankan context. Content and Consensual validity of the translated modified scale was obtained by expert opinion.

Mothers were questioned on the perceived impact of child's disorder (AI) on the family and child's own quality of life over the past 6 months, as at present and in future context. Same questions were asked from the parents regarding siblings without AI (controls). However, the control group was less, as in some families all children were affected with AI.

The original English version of FIS was first translated into native Sinhala language and then back translated into English language by independent translators. It was pre-tested among maternal caregivers of children with Cleft Lip and Palate prior to corrective surgery who attended the Oral & Maxillo-Facial Unit, Provincial General Hospital Badulla, which provided a rural socio-cultural setting in Sri Lanka, similar to the study setting of the present study.

The original FIS of Locker et al., 2002 comprised of 14-items in 4 dimensions but the modified, translated FIS consisted of 27-items organized into 6 categories. Namely, parental/ family impact (5-items), parental emotions (4-items), family conflict (3-items), perceived impact on child at present and in the past (10-items), perceived future impact on child (3-items) and financial burden (1-item). Each item had a frequency score as, never = 0, once or twice = 1, sometimes = 2, often = 3 and all the time = 4 and severity score as never = 0, a little = 1, quite a bit = 1, very much = 2. Hence for each item the frequency score was multiplied by respective severity score to arrive at the final score. Accordingly the scores for each item irrespective of the category to which the respective item belonged to could range from 0-12. In the present analysis the total score was computed for each dimension rather than considering individual items to reflect the composite picture.

Data were entered using Excel spreadsheets and analyzed by SPSS-21 Statistical Software Package. Descriptive statistics were used to describe the results.

Ethics approval for the present study was obtained from the Faculty Research Committee, Faculty of Dental Sciences University of Peradeniya (RERC/2009/11/HERATH1).

RESULTS

Table 1: Socio-demographic Profile of children diagnosed with AI

Feature	Number (%)	
Age distribution (in years)		
1-5	1 (5.9)	
6-10	4 (23.5)	
11-15	11 (64.7)	
>15	1 (5.9)	
Gender distribution		
Male	7 (41)	
Female	10 (59)	
Race		
Sinhala	11 (64.7)	
Moor	4 (23.5)	
Tamil	2 (11.8)	
Parental education	Mother	Father
Primary	1 (5.9)	0 (0)
Secondary	1 (5.9)	3 (17.6)
GCE (O/L)	8 (47.1)	7 (41.2)
GCE (A/L)	5 (29.4)	5 (29.4)
Degree/diploma	2 (11.8)	2 (11.8)
Sub type of AI		
Type1 (hypoplastic)	3 (17.6)	
Type II (hypomaturation)	1 (5.9)	
Type III (hypocalcification)	5 (29.5)	
Type IV (hypomaturation-hypocalcification)	8 (47.0)	

The mean age of the sample was 11.29 (±3.38) years, ranging from 3 years to 16 years. Thus, majority of children with AI presented in the mixed dentition stage (Fig 1). With reference to level of education acquired by parents, mothers were less educated compared to fathers. Majority of mothers (64.8%) were housewives, looking after family and engaging in household chores. Among fathers, the majority (64.7%) were employed as skilled/unskilled workers, 23.5% as professionals and 11.8% as businessman. Overall, based on educational attainment and occupational status of parents, the children with AI belonged to low to middle socio-economic status.

Table 2: The Mean ± SD, Median and Range scores of dimensions of modified translated version of Family Impact Scale

Dimension	Mean Score	± SD	Median Score	Range
Parental/Family Activity	9.17	±6.26	8.0	2-24
Parental Emotion	33.59	±7.01	33.0	24-45
Family Conflict	4.41	±5.29	3.0	0-19
Perceived impact on Child's quality of life in the past & present	39.00	±18.03	39.0	10-73
Perceived Impact on Child's quality of Life in Future	21.00	±5.12	21.0	12-28
Financial Burden	10.76	±1.85	12.0	6-12

Based on maternal respondents, psychological impact of the condition can be presented as the mean overall score for 27-items of the scale as 117.94 (± 3.19) and the scores ranged from 62-174. The parental/family activity category scores ranged from 2-24 with the mean of 9.17 (±6.26), and the median score was 8.0. Parental emotion scores ranged from 24 to 45 with mean 33.59 (±7.01), and a median score of 33.0. The family conflict scores ranged from 0-19 with the mean of 4.41(±5.29), and a median score of 3.0. Moreover, impact scores of AI on child's quality of life in past and present context as perceived by parents ranged from 10-73 with the mean of 39 (±18.03) and a median score of 39.0. Furthermore, perceived future impact scores on child's quality of life ranged from 12-28 with the mean of 21 (±5.12) and a median score of 21.0. The single item perceived financial burden ranged from 6-12 with the mean of 10.76 (±1.85) and a median of 12.0 (Table, 2).

The reliability of the 27-item FIS scale was assessed by internal consistency, as shown by Cronbach's alpha, which reported a highly acceptable value of 0.83.

Interestingly, in the control group of siblings not affected by AI the impact scores for ll6 dimensions were reported as 0.

Figure 1: Two patients who presented with Hypoplastic type of AI

DISCUSSION

The present study aimed to explore the socio-demographic profile of children affected by AI and its psychosocial impact on the family compared to their siblings (normal children in the same family) not affected by AI as perceived by their mothers. However, the results should be interpreted cautiously as the study has inherent limitations such as small sample size and use of modified translated version of Family Impact Scale (MTFIS) of Locker, 2002[7], which needs further methodological assessment with validation using a large sample of parental caregivers of children affected by similar conditions. However, in the absence of any published studies on psychosocial impact on families having children affected by AI in Sri Lankan socio-cultural context, present study addresses an important information gap.

All dimensions and sub-dimensions of FIS were affected by having a child with AI as described my mothers. The most affected dimension based on frequency-severity score was mothers' perceived impact on child's quality of life in past and present context, parental emotion and mothers' perceived impact on child's quality of life in future context. The perceived negative impact was relatively low for financial burden, parental/family activity and family conflict dimensions compared to other dimensions (Table, 2). The highest median score reported for impact of AI on child's quality of life in past and present contexts as reported by mothers demonstrated the burden of the disorder (AI). Importantly the parents perceived considerable impact of AI on child's quality of life in future context as well (median score of 21.0). These findings corroborated with the considerably high scores on parental emotions dimension of FIS with a median score of 33.0. As the majority of children were adolescents (Figure 1), the findings reflected how mothers were worried about their children affected by AI going to achieve the life goals as young adults in future. Adolescence is a period of life for increased attention for appearance and grooming. Against this backdrop, Parekh et al., 2014[5] conducted a qualitative explorative study among children/ adolescents affected by AI. Findings revealed that there is marked aesthetic, functional and psychosocial impact of having AI among children and adolescents.

The visible unaesthetic appearance of AI affected teeth compounded by sensitivity, poses a significant psycho-social impact to affected children and adolescents, such as negative self-perception, emotional disturbance, inhibited interactions, being teased/bullied, anxiety, low self esteem, learning disabilities, low school achievement and social withdrawal [10,11].Coffield et al., 2005[10], reported that having AI, impacted on the psychosocial health of affected people, especially at younger ages such as, social avoidance and distress, higher levels of discomfort, dysfunction and high fear for negative evaluation. Moreover, there is evidence to support that oro-facial and dental anomalies affecting aesthetics of teeth could be an important cause for children and

adolescents being bullied at school and even at home environments [11].

The lowest median score (3.0) reported for family conflict dimension followed by family / parental activities dimension (median score of 8.0) could be probably attributed to cohesive nature of families in rural Sri Lanka, despite having a child with AI. Similarly, the relatively low impact scores (median score=12.0) for financial burden could be due to availability and accessibility to public dental care services free of charge at the point of delivery.

The high Cronbach's alpha value of the 27-item, modified, translated FIS provided evidence for suitability of this scale for group comparisons.

In conclusion, present study revealed that children affected by AI who attended the Division of Paedodontics belonged to low and middle socio-economic backgrounds. As reported by mothers, there was a considerable family impact due to AI status of the child especially for perceived impact of quality of life of the child in past, present and future as well as on parental emotions. There is a need for further studies in this regard with methodological refinement. Affected mothers and families should be provided necessary emotional support to overcome their suffering including comprehensive, long term restorative and preventive dental care provision. Paediatric Dentists should pay their attention, not only for providing successful restorative dental treatment and oral hygiene improvement of affected children but the psychosocial impact of their families as well.

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