

How do children with amelogenesis imperfecta feel about their teeth?

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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 adolescents from their perspectives is not fully understood.

Aims. The aim of the study was to explore the impact of AI on children and adolescents through in-depth interviewing. The information derived from this was then used to construct a questionnaire to distribute to a larger cohort of AI patients.

Design. This research involved semistructured in-depth interviews with seven AI patients, and

common themes and concepts were then identified using framework analysis. A questionnaire was developed based on the themes and subthemes identified, and completed by 40 AI patients at various stages of treatment.

Results. Children and adolescents with AI exhibited concerns regarding aesthetics and function. 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 adolescents as a result of AI, including aesthetics, function, and psychosocial.

Background

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 manner¹. AI is a heterogeneous group of hereditary disorders of enamel formation and may be autosomal dominant, autosomal recessive, sex-linked, or sporadic².

A disturbance to any of the three steps required for enamel formation may result in a defective enamel structure, for example a decrease in the enamel matrix formation will result in hypoplastic AI. Disruption in the mineralization phase results in either hypomineralized (hypocalcified) AI (deficiency in the first phase of enamel mineralization) or

hypomineralized (hypomature) AI, caused by a deficiency in the final phase of enamel mineralization. Both hypoplastic and hypomineralized AI can coexist in the same individual or the same tooth². Classifications were traditionally based on phenotype, although it is now recommended that it is better to classify AI according to the mode of inheritance when known rather than the phenotype³.

The clinical appearance of AI can be remarkably different between types⁴. For example, hypoplastic AI is characterized by small crowns with thin enamel or enamel of normal thickness but with pits and grooves⁵. Hypocalcified AI presents as severely discoloured enamel, whereas the enamel in hypomature AI is usually of normal thickness with whitish opacities². The colour of the affected teeth ranges from normal to opaque white or yellow-brown⁶.

The aims of the management of AI are to improve aesthetics and function and reduce sensitivity of the affected dentition. Due to the sensitivity and pain encountered during tooth brushing, oral hygiene may be poor making it

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difficult to provide restorations⁷. When treating AI patients, it is important to have a multidisciplinary team where possible, this may include input from a paediatric dentist, orthodontist, periodontist, restorative dentist, and possibly a geneticist⁸. Due to the discolouration, sensitivity, and extensive treatment required, the psychosocial impact of AI should not be underestimated. A recent systematic review looking at associated dental and orofacial abnormalities in AI, suggested that further research is required into the quality of life and economic impacts associated with AI⁹.

It is surprising that the psychological impact of dental anomalies is a largely unresearched field. One study 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. Semistructured interviews were conducted with 21 patients (13 female and eight male) aged 10–15 years with different severities of DDE. After each interview, two photographs of the patient's teeth were taken, one with their teeth wet with saliva and another when their teeth were dry. The photographs were scored using the Thystrup and Fejerskov Index (TFI) and the Modified Developmental Defects of Enamel Index. The study concluded that variations in the impact of DDE were related to aspects of sense of self (the domains by which people judge their lives, or the 'looking glass' through which others judge them) rather than the extent of the defect directly¹⁰.

The psychosocial impact of AI on adult patients was explored with 59 family members, 30 with AI and 29 without AI, using a questionnaire to measure various psychosocial parameters such as self-image, self-esteem, social interaction anxiety, and self-perceived quality of life with regard to dental issues. The study concluded that adults with AI exhibited higher levels of social avoidance, distress, and self-consciousness about their teeth⁵. To date, there have been no studies looking at such impacts in children with AI.

Popular approaches to data collection in qualitative research are unstructured, or in-depth, interviews¹¹. Interviews are useful for exploring children's experiences in their own

words and can also inform questionnaires that are grounded in children's views.

Therefore, the aim of this study was to explore the impact of AI on children and adolescents, using semistructured in-depth interviews to identify themes that are important to AI patients. The second stage of the study was to use this data to develop a questionnaire to distribute to a larger cohort of AI patients.

Methods

Ethical approval was obtained and the project was registered with the Research and Development Directorate, and all research was undertaken in the Department of Paediatric Dentistry, Eastman Dental Hospital, UCLH Foundation Trust. The main researcher (MA) attended an interview skills course and also undertook face-to-face pilot interviews supervised by members of the research team, experienced in the field of qualitative research and in-depth interviewing. A topic guide was developed consisting of open-ended questions relating to AI.

Patient selection involved a purposive sampling approach to ensure a range of subject demographics. In qualitative research, it is often difficult to know exactly how many participants are required in advance, although small numbers of interviewees can give quality-rich data.

The inclusion criteria for the in-depth interviews 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) Patients able to speak English sufficiently well to be involved in an interview
- 6) No active restorative treatment yet undertaken

Patients were recruited for interview, 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. All patients were provided with an information sheet and verbal explanation. 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.

Face-to-face interviews were conducted away from the main clinical area to ensure privacy and to provide a less stressful environment, and a chaperone was present in all interviews. Parents were encouraged to remain outside the side surgery whilst their child was being interviewed, but they were given the opportunity to join the interview if they or their child requested. All interviews were recorded using a digital Dictaphone and then transcribed onto Microsoft Word, with all patients identified by a unique code number only to maintain patient confidentiality. Patients who participated in the study were told that if they found any question in the interview 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. The interviewer (MA) was not directly involved in the clinical care of the participants.

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 guide, they were also explored. The interviews started with general questions and then gradually moved on to more probing questions about aesthetics, sensitivity, function, and psychosocial aspects, such as effects on friendships and comments by other people. The interviews were transcribed immediately afterwards, and 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 process¹².

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 experiences¹¹. The first step involved identifying themes or

concepts by carrying out a thorough review of the data from the transcripts. The second step involved constructing a framework with a hierarchy of main themes, and each theme was subsequently divided into subthemes. An Excel spreadsheet was developed with a sheet for each theme, where the columns were subthemes and each row represented a patient. Individual patient quotes were then added to the appropriate cell.

Questionnaire development

The results from the in-depth interviews were used to develop the questionnaire. A combination of multichotomous and dichotomous responses was used, including 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, and there were 15 questions in total (Fig. 1). The wording of the questions and responses was in child-friendly language, to avoid ambiguity and to be easily understood. Leading questions, double negatives, loaded words, and hypothetical questions were avoided¹³. Several drafts of the questionnaire were developed and piloted amongst five child patients, not included in the study, but within the age range, to assess ease of use.

The questionnaire started with demographic questions, and the more personal and sensitive questions were gradually introduced in subsequent sections. The questions in the second section were mainly derived from the subthemes related to aesthetics and function and included questions regarding shape and colour of teeth, and pain or sensitivity from certain foods or drinks. The third section of the questionnaire was based on the psychosocial 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, for example a dedicated website or online support group.

Patients were also asked to complete a modified short version of the Child Percep-



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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 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

Fig. 1. Questionnaire for patients with amelogenesis imperfecta.

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	

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? _____

Fig. 1. (continued)

tion Questionnaire (CPQ₁₁₋₁₄), to validate part of the questionnaire¹⁴. The modified CPQ₁₁₋₁₄ contains 20 questions (the 16 questions included in the CPQ₁₁₋₁₄ and two additional questions relating to general oral

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 |

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:

Fig. 1. (continued)

health, and two from the regression specific model)¹⁵, and each question has five responses. The scores for each response are 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.

Patients attending the Eastman Dental Hospital from January to June 2012 were identi-



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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.

Fig. 1. (continued)

fied from clinic lists. Each patient, and their parent, was given information leaflets, and written consent was taken. Patients were asked to complete the questionnaire and leave it in the labelled collection box in the reception area. In cases where the patients could not complete the questionnaire at the same time as their appointment, a stamped addressed envelope was given so that the questionnaire could be completed at home and returned by mail. Each questionnaire was coded with an ID number.

Results

Interviews

A total of seven patients were interviewed, six of whom (85%) were female, and the average age was 14.2 years, with a range of 13–16 years. The gender, ethnicity, and pseudonyms for the patients are shown in Table 1. Interviews lasted between 13 and

20 min, with an average of 16 min for each interview. The main themes and subthemes are shown in Table 2.

The majority of patients stated that it was their dentist who informed them about having 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:

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 [P1, 16 years]

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 [P2, 13 years]

The most common aesthetic issue raised by the interviewees was the colour of their teeth. Other important issues also highlighted by patients were the shape and size of their teeth, the appearance of their smile, and feelings about having photos or videos taken:

....if they were just a little more nice colour, not discoloured [P1, 16 years]

The colour, like when I say 'Sshhh' my front teeth show on there, they're yellow ones [P2, 13 years]

I don't like smiling with my teeth because I don't like them. If I had nice teeth maybe I would be able to smile more [P6, 13 years].

I don't like smiling with my teeth because I don't like them [P5, 15 years]

I will smile when all of my teeth are going to be white, nice shape. [P7, 13 years]

For others, sensitivity was also an issue:

Table 1. Demographic details of patients interviewed.

ID	Gender	Age (years)	Ethnicity
1	F	16	White
2	F	13	White
3	F	16	Asian
4	M	15	Asian
5	F	15	White
6	F	13	African
7	F	13	African

Table 2. Framework showing the main themes and subthemes identified from the interviews.

1. Background
1.1. Age
1.2. Gender
1.3. Ethnicity
1.4. Siblings
2. Amelogenesis Imperfecta (AI)
2.1. When did patient know about AI?
2.2. How did patient know about AI?
3. Aesthetics
3.1. Colour
3.2. Shape
3.3. Size
3.4. Photos/videos
3.5. Smile
4. Function
4.1. Pain/sensitivity
4.2. Avoiding certain foods or drinks
5. Psychosocial Aspects
5.1. Effects on friendships
5.2. Comments by people
5.3. Self-consciousness
5.4. Confidence
5.5. Teasing/name calling
5.6. Feeling different/isolated
5.7. Worries about future plans.
5.8. Effects on social interests
6. Health and Dental Health Concerns
6.1. Personal
6.2. Health related

It is the sensitivity more than the colour, the colour doesn't bother me, it's more the sensitivity. [P5, 15 years]

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 [P2, 13 years]

The majority of patients had experienced comments by other people about their teeth, and in some patients, this resulted in feelings of isolation or affected their confidence:

If someone sees it, they go 'Oh, don't you brush your teeth?' and stuff like that [P2, 13 years]

I'm not feeling well, I'm feeling a little bit bad, because they can see that my teeth look like dirty [P7, 13 years]

If the colour was fine, I'd feel a lot more confident [P2, 13 years]

The colour just knocks my confidence [P1, 16 years]

When all my friends are talking I'd want to join in but I don't want to show my teeth [P3, 16 years]

These findings suggested that AI can have a significant impact on children and adolescents and also demonstrated the benefits of using qualitative methods to investigate personal thoughts and opinions. The results of these interviews were used to develop a questionnaire to ascertain the views of a larger group of AI patients.

Questionnaire

The questionnaire was distributed to 61 AI patients, 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 returned the questionnaire by mail. The majority of patients were currently undergoing treatment (80%, $n = 32$), with the remaining eight patients in the pre-treatment phase, and the mean age was 13.2 years (range 10–16 years, SD 2.2 years). There were 19 females (47.5%, mean age 13) and 21 males (52.5%, mean age 14). The majority of participants 25 of 40 (62.5%) were White.

The first five questions were demographic or background questions. 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 of 40 (47.5%) ranked their confidence as (3). 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 (Fig. 2).

The next question assessed the importance of improving the colour, shape, and size of the teeth, the importance of reducing pain/sensitivity and the importance of improving the smile separately. Again categories 1 and 2 were combined (not important) and 4 and 5 (important) for ease of interpretation. There were no apparent differences in responses between stages of treatment or between genders with regard to the importance of treatment aims. Improving the colour of teeth was important for 90% of the respondents, with 74% stating that

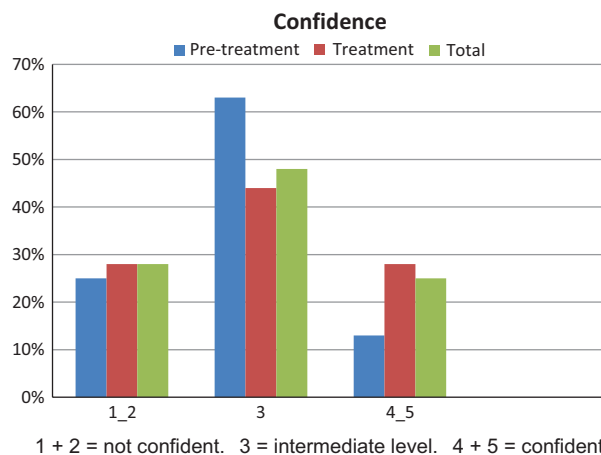


Fig. 2. Responses to question 6 for pre-treatment and treatment groups. (scale from 1 to 5).

improving the shape of the teeth was also important. The majority of patients wanted treatment to enhance their smile, with 77% of all respondents stating this was a reason for pursuing treatment. The majority of patients also wanted to have treatment to reduce pain and sensitivity, with a total of 74% saying this was important to them. The responses for this question are shown in Fig. 3, highlighting the importance of treatment aims to the respondents.

Question 8 asked whether there were any foods or drinks respondents avoided. Very few participants reported avoiding foods all of the time, with 'never' and 'occasionally' being the most common responses, both by gender and treatment phase. It is important to stress that the number of pre-treatment respondents was small ($n = 8$); therefore,

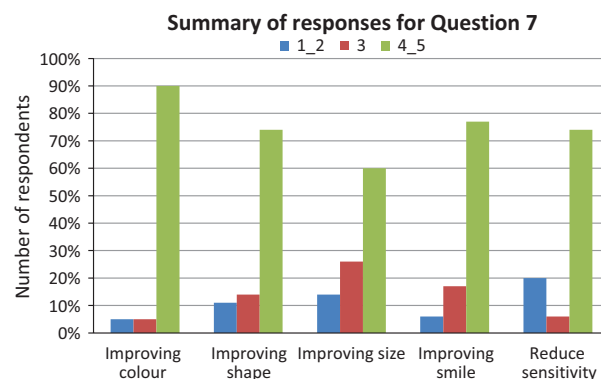


Fig. 3. Importance of treatment aims on a scale from 1 to 5 (1 being not important at all and 5 being very important).

these results need to be viewed with caution. Subsequent questions focused on psychosocial issues with 50% of respondents reporting being teased at least 'occasionally'. Amongst male respondents, 24% reported being teased 'often' in comparison with 11% of females. A number of respondents also reported that they avoided smiling, with 30% of respondents avoiding smiling 'occasionally' and 23% avoiding smiling 'often' or 'all of the time'. Females appeared to avoid smiling 'all of the time' more often than males (32% vs 14%).

Respondents were asked the single most important thing they wanted from treatment. The most common response was to improve the colour of their teeth (63%), with improving the smile (18%), and reducing sensitivity (10%) being the 2nd and 3rd choices. The final question, asked patients whether they thought it would be useful to have a website or 'support group' for patients with AI, and the majority (85%) answered yes, with only 15% answering no.

Child Perception Questionnaire

All 40 patients completed the CPQ₁₁₋₁₄ with no missing answers. The scores 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. The relationship between CPQ₁₁₋₁₄ score and level of confidence was explored, as part of the questionnaire validation process, with higher levels of confidence reported by the patients who had lower CPQ₁₁₋₁₄ scores (16). This indicated that those with better quality of life tended to show more confidence, possibly due to the lower impact of AI on their lives, whereas the respondents who were not confident tended to score higher on the CPQ₁₁₋₁₄ (38), suggesting a greater impact on quality of life.

Discussion

The aim of this study was to identify the main issues for AI patients and to construct a patient-centred questionnaire.

Interview findings

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. One of the main challenges, however, experienced by the researcher was to ensure the research/topic guide/etc. allowed young people to freely discuss their feelings and thoughts with a stranger. In addition, the clinical setting for the interviews, whilst necessary (as patients were attending for treatment), may have increased their anxiety and influenced their responses¹⁶.

Recruiting patients to participate in the interviews was also considerably more difficult than originally anticipated, as it was hard to find AI patients who had not undergone any previous restorative treatment for their condition. This meant that only seven patients could be recruited within the allocated time interval. When conditions are relatively rare and a single centre study is being undertaken, this can reduce the number of patients available, as found in a study looking at adolescents' perspectives of living with Treacher Collins syndrome, where only six patients were recruited. There is the potential, however, to conduct good qualitative research with small sample sizes due to the richness of data produced¹⁷. No new themes, however, were identified at the last interview; therefore, it was felt that the most relevant themes had probably been identified.

Most of the patients who were willing to be interviewed were females and that might be because females in general have been found to be more attentive conversationalists than males¹⁸. It could also be attributed to the fact that more regular dental attendance has been reported amongst females (25.8%) compared with males (16.6%)¹⁹. 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 male respondent and the females.

The majority of patients stated they noticed their AI at a young age (around 6 years old), which is in agreement with previous research, suggesting that children start noticing dental defects relatively early¹⁰. The majority of patients were confused about the explanation given to them by their local dentist about their enamel condition, which highlights the importance of general dentists being well informed regarding AI, to be able to explain the nature of this condition in a way that young children and parents can understand.

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 reported as being more unhappy with the colour of their teeth (79%) than subjects without AI (32%)⁵. Another study investigating children's experiences of enamel defects also found that a number of children were reluctant to smile¹⁰.

There are clinical implications regarding the importance of dental aesthetics in AI patients, as their 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 outcome²⁰. 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 dissatisfaction with the outcome of care²¹. Aesthetic dental treatment for children may yield important psychosocial benefits²², but this does rely on patients being appropriately prepared and having realistic expectations.

Teasing and name calling were discussed in the interviews, and this was also highlighted in a study with adult AI patients, where 93.3% of subjects reporting being teased about their teeth⁵. A previous study also found that some young people had experienced teasing and name calling due to developmental enamel defects¹⁰. Clinicians should be sensitive to such issues in children, which may require support or referral to counselling services in some cases.

Questionnaire findings

Improving the colour of teeth was the most important overall reason for undergoing treatment for AI (90%) and the findings of the in-depth interviews, and questionnaires were in agreement. Improving the size of the teeth was also seen as important, but by fewer respondents (60%). Improving the smile was considered important by 77% of respondents, and a similar percentage (74%) saw a reduction in sensitivity as important. It has been shown that the smile is the second facial feature, after eyes, which people view to assess another person's attractiveness, and dental aesthetics is also known to impact on overall facial appearance²³. Clinicians must appreciate the importance of dental aesthetics particularly with young AI patients, as it is well documented that appearance is the most valued characteristic amongst young dental patients²⁴. The desire to improve the smile and appearance is considered motivating factors for seeking dental treatment²⁵.

A limitation of this study was that the questionnaire was not subject to test-retest reliability to determine the validity. A further study is needed to ascertain reliability (by asking a subsection of the original sample to redo the questionnaire), before the questionnaire can be used by other researchers. It is hoped that once the questionnaire has been validated, it can be used in other centres to increase the sample size and allow statistical testing between genders. Using multiple centres would be beneficial to increase the sample and identify further what aspects of AI affect OHRQOL.

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, and this is an area of research that needs to be explored further.

Conclusion

Patient's own opinions of appearance are important, and this study highlights the need for further patient-centred research into dental anomalies. Children and adolescents were found to have impacts due to their AI, in

terms of concerns regarding aesthetics, function, and psychosocial issues. Improving the colour of the teeth, improving the smile, and reducing dental sensitivity were all perceived to be important aims of treatment, with improvement in colour being the single most important aim. The issue of teasing was also evident. This can have long-term consequences for patients and appropriate ways of offering support and counselling in such situations should be available.

Why this paper is important to paediatric dentists:

- Discusses the impacts of quality of life in AI children and adolescents
- Highlights the concerns regarding aesthetics for this group
- Highlights the concerns regarding sensitivity of teeth

Conflict of interest

The authors declare no conflict of interest.

References

- 1 Welbury RR. Paediatric Dentistry, 2nd edn. Oxford: Oxford University Press, 2001: 277–278.
- 2 Crawford PJ, Aldred M, Bloch-Zupan A. Amelogenesis imperfecta. *Orphanet J Rare Dis* 2007; **2**: 17.
- 3 Aldred MJ, Crawford PJM, Savarirayan R. Amelogenesis imperfecta – a classification and catalogue for the 21st century. *Oral Dis* 2003; **9**: 19–23.
- 4 Sapp JP, Eversole LR, Wysocki GP. Contemporary Oral and Maxillofacial Pathology, 2nd edn: Mosby, 2004: 1–44.
- 5 Coffield KD, Phillips C, Brady M, Roberts MW, Strauss RP, Wright JT. Psychosocial impact of developmental dental defects in people with hereditary amelogenesis imperfecta. *J Am Dent Assoc* 2005; **136**: 620–630.
- 6 Witkop CJ, Sauk JJ. Heritable defects of enamel. In: Stewart R, Prescott G (eds). Oral Facial Genetics. St. Louis: Mosby, 1976: 151–226.
- 7 Wright RJ. Redefining the principles of restorative dentistry: lessons from microscope-assisted dentistry. *Dentistry Today* 2000; **19**: 66–68, 70–5.
- 8 Moretti AB, Sakai VT, Oliveira TM *et al.* Oral management of a child with mixed dentition affected by amelogenesis imperfecta. *J Dent Child* 2007; **74**: 157–160.
- 9 Poulsen S, Gjørup H, Haubek D, Haukali G, Hintze H, Lovschall H. Amelogenesis imperfecta – a systematic literature review of associated dental and oro-facial abnormalities, and impact on patients. *Acta Odontol Scand* 2008; **66**: 193–199.
- 10 Marshman Z, Gibson B, Robinson PG. The impact of developmental defects of enamel on young people in the UK. *Commun Dent Oral Epidemiol* 2008; **37**: 45–57.
- 11 Ritchie J, Lewis J. Qualitative Research Practice. A Guide for Social Science Students and Researchers. London: SAGE, 2006.
- 12 Glaser BG, Strauss AL. The Discovery of Grounded Theory: Strategies for Qualitative Research. Chicago: Aldine de Gruyter, 1967.
- 13 Black N, Brazier J, Fitzpatrick R, Reeves B. Designing and using patient and staff questionnaires (Chapter 5). Health Services Research Methods – A Guide to Best Practice, 1st edn. London: BMJ Books, 1998: 46–51.
- 14 Jokovic A, Locker D, Stephens M, Kenny D, Tompson B, Guyatt G. Validity and reliability of a questionnaire for measuring child oral-health related quality of life. *J Dent Res* 2002; **81**: 459–463.
- 15 Jokovic A, Locker D, Guyatt G. Short forms of the Child Perceptions Questionnaire for 11–14-year-old children (CPQ11–14): development and initial evaluation. *Health Qual Life Outcomes* 2006; **4**: 4.
- 16 Marshman Z, Hall M. Oral health research with children. *Int J Pediatr Dent* 2008; **18**: 235–242.
- 17 Beaune L, Forrest CR, Keith T. Adolescents perspectives on living and growing up with Treacher Collins syndrome: a qualitative study. *Cleft Palate Craniofac J* 2004; **41**: 343–350.
- 18 Giles H, Hewstone M. Cognitive structures, speech and social situations: two integrative models. *Lang Sci* 1982; **4**: 187–219.
- 19 Quteish Taani DSM. Dental anxiety and regularity of dental attendance in younger adults. *J Oral Rehabil* 2002; **29**: 604–608.
- 20 Abdel-Kader HM. Psychosomatic norm in orthodontics: problems and approach. *World J Orthod* 2006; **7**: 394–398.
- 21 Cunningham SJ, Hunt NP, Feinmann C. Perceptions of outcome following orthognathic surgery. *Br J Oral Maxillofac Surg* 1996; **34**: 210–213.
- 22 Rodd HD, Barker C, Baker SR, Marshman Z, Robinson PG. Social judgements made by children in relation to visible incisor trauma. *Dent Traumatol* 2009; **26**: 2–8.
- 23 Hassebrauck M. The visual process method: a new method to study physical attractiveness. *Evol Human Behav* 1998; **19**: 111–123.
- 24 Prokhorov AV, Perry CL, Kelder SH, Klepp KI. Lifestyle values of adolescents: results from Minnesota Heart Health Youth Program. *Adolescence* 1993; **28**: 637–647.
- 25 McKiernan EX, McKiernan F, Jones ML. Psychological profiles and motives of adults seeking orthodontic treatment. *Int J Adult Orthodon Orthognath Surg* 1992; **7**: 187–198.