Rehabilitation of Children with Ectodermal Dysplasia. Part 2: An International Consensus Meeting

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A consensus meeting was arranged to provide an opportunity to discuss the residual nonconsensus questions following three rounds of a Delphi study. It was hoped that the nonagreements could be resolved to define a comprehensive protocol for the management of ectodermal dysplasia, particularly with respect to the use of dental implants in growing patients. An international panel of expert clinicians in pediatric dentistry, prosthodontics, and orthodontics was invited to be part of the Delphi study to develop agreement on clinical questions through a consensus of ideas. Each expert had been invited to form a study group or team within his or her home institution. As required by the Delphi protocol, a 90-part questionnaire was considered by the collaborating teams and progressed through three iterations with increasing agreement. This process is discussed in part 1 of the study. The residual nonconsensus questions, which represented 10% of the questionnaire, required collaborative interaction for resolution. The consensus meeting was held in London, England, over a 2-day period with support from Nobel Biocare and the British Dental Association. Int J Oral Maxillofac Implants 2013;28:1101–1109. doi: 10.11607/jomi.2981

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The consensus meeting to conclude a Delphi study of the treatment of ectodermal dysplasia (ED) required the participation of team leaders (and where possible an additional team member), and teams from Sweden, France, the United Kingdom, the United States, Hong Kong, and Australia were present. Dr Sophie Watkins represented Professor Richard Palmer (Guy's, Kings, and St Thomas's Medical and Dental Institute, London), and Associate Professor Albert Guckes (University of North Carolina) was not available to attend. Table 1 lists the participants.

It was recognized in the initial proposal that a consensus meeting may be required as the final step in the Delphi study, should there be residual nonconsensus items. As a result, a 2-day consensus meeting was held in London at the British Dental Association (BDA) headquarters and was sponsored by the BDA and Nobel Biocare.

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Table 1 Participants in Consensus Conference on Rehabilitation of Children with Ectodermal Dysplasia

Team	Name
Sweden	Dr Birgitta Birgendal Dr Johanna Norderyd
France	Prof Marie-Cecile Meniere Dr Francois Claus
United Kingdom	**Eastman Dental Institute** Emeritus Prof John Hobkirk
	University of Newcastle Dr Ross Hobson Dr Nick Jepson
	Guy's, King's and St Thomas's Medical and Dental Institute Dr Sophie Watkins (for Prof Richard Palmer)
USA	Prof Clark Stanford Assoc Prof Kenneth Kurtz Prof Arun Sharma
Hong Kong	Prof Nigel King
Australia	Prof Iven Klineberg (co-ordinator) Assoc Prof Angus Cameron
Apologies	Prof Richard Palmer Assoc Prof Albert Guckes

Of the 90 questions and partial questions contained in the questionnaire, consensus was reached for 90% of them, and for 10% there was no consensus.

The study resulted in considerable agreement between the international teams in their approach to management of the dental aspects of ED and the consensus meeting was to attempt to reach a consensus on the remaining items. This would then provide an agreed-upon treatment framework for a multicenter study by providing an in-depth understanding of the questions and dental difficulties in managing this complex clinical problem.

MEETING AGENDA

The discussion was coordinated by a chair (from the Sydney team), and in a collegial atmosphere, constructive discussion addressed the following (Fig 1):

- Nonconsensus items after round three of the Delphi study. Nonconsensus items represented 10% of the items in the questionnaire and are summarized in Figs 1 and 2.
- Consideration of a multicenter clinical trial to apply the specific protocol recommendations agreed by consensus for a specific manifestation of ED. An appropriate descriptive title for the study and the details of the study protocol were to be determined. This information is not included in this paper and will be presented separately.

METHODS

A decision was made by the Sydney team that after three rounds of questionnaire completion/revisions, there was a sufficiently high degree of consensus to justify progression to the next phase. Delphi surveys have reported that three iterations are the usual requirement and that further iterations were unlikely to achieve significant further agreement.[1]

Of the 19 areas of clinical consideration, for which there were 90 questions, the following outcomes were achieved:

- At the end of round 1, there was consensus on 61% of the questions;
- With round 2, there was consensus on a further 21% of the questions;
- In round 3, there was consensus on a further 8% of the questions.

When round 3 had been completed, consensus had not been reached for 10% of the items (9 questions or partial questions).

Some questions raised complex diagnostic and treatment planning issues, and not all groups found it possible to identify one correct answer, as this may be too simplistic. This raised issues that were described in the comments section of the questionnaire, and it was agreed that for such questions the response without discussion was that "agreement cannot be reached." Given the level of scrutiny in applying the algorithm to the answers and the careful consideration of the written responses by the Sydney team, it became clear that a collaborative discussion was required.

RESULTS

The nonconsensus items listed in Fig 2 were considered in order. This information is summarized here with the data presented for each nonconsensus question by indicating: *(1)* the specific question; *(2)* details of the discussion, listed under "Discussion was informed by"; and *(3)* where needed, an agreed-upon rewording of the question for clarity and to facilitate consensus.

Day 1

- Some questions raised complex issues and some teams were unable to identify one answer.
- Feedback comments indicated this.
- Rewording was needed for:
  - Q3: Age for panoramic radiographs?
  - Q7: Orofacial function pretreatment and assessment?
  - Q11: Age range 3 years for RPD?
  - Q13: Ages for implant treatment maxilla/mandible?
    Implants spaced for three-unit or two-unit cantilever fixed prosthesis?
    Implant overdenture more appropriate?
  - Q17: Case study 2: How to treat loss of occlusal vertical dimension with primary molar ankylosis?

Day 2

- Discussion and agreement to initiate a multicenter clinical trial to apply the specific protocol recommendations reached by consensus.
- Agreement that the study be limited to ED with mandibular anodontia (ages 8 to 15 years) as a crossover clinical trial to contrast a two-implant overdenture with a four-implant fixed prosthesis.
- Assessment of oral health–related quality of life with regard to function, nutrition, and psychologic well-being

Fig 1 Consensus meeting agenda: summary of nonconsensus questions.

Q3. Radiographs: At what age—(a) 3 to 5 years (b) 6 to 8 years (c) 9+ years—should each type of radiograph be taken?

(a) Panoramic radiographs — NO CONSENSUS

Q5. Do you agree with the following statements?

(b) Parents often have legitimate but unrealistic demands in relation to the provision of treatment for their child. Is it acceptable to sedate/manage under general anaesthesia a pre-cooperative child to provide treatment for hypodontia to:

1. Is it acceptable to sedate a precooperative child to provide treatment for hypodontia to:
   (i) Take an impression — NO CONSENSUS

Q7. Orofacial function needs to be considered as a patient-specific problem and the assistance of parents is required.

(a) A pretreatment assessment questionnaire is required to be completed by: parent(s) and child. — NO CONSENSUS

Q11. Hypodontia

(a) The following suggests an age range for specific treatment possibilities.

2. 2 to 3 years:
   RPD — NO CONSENSUS
3. 6 to 10 years:
   orthodontics/RPD with implants — NO CONSENSUS

Q13. Implants in growing bone (in the absence of alveolar bone)

In considering the questions below, please indicate how they may apply to each age group, based on clinical experience and outcome data published in the scientific literature. Please consider the placement of implants in children with severe hypodontia as opposed to those children who may have suffered tooth loss and normal alveolar bone is present.

(a) Are there particular ages at which time implants may be successfully placed in children? — NO CONSENSUS

(i) Should implants be spaced as for a three-unit RDP or a two-unit cantilever RDP or placed adjacent to each other, as for separate crowns?

1. Maxilla:
   anterior/posterior — NO CONSENSUS

(k) Is an implant overdenture more appropriate in growing bone?

1. Maxilla: anterior/posterior — NO CONSENSUS

CASE STUDY 2

Q17. Consider treatment for the same patient at 15 years and 19 years. What might be:

(d) the treatment for loss of vertical dimension associated with primary molar ankylosis
At 15 years — NO CONSENSUS

Fig 2 Nonconsensus questions.

Part	Topic	Question no.	Round 1
1	Dental disability	1	100%
2	Initial diagnosis	2.3.4	62.5%
3	Global disability	5,6,7,8	50%
4	Oral health	9	100%
5	Dental treatment options		
	Orthodontics	10	100%
	Hypodontia	11	70%
	Anodontia	12	100%
	Implants	13	31.6%
		14,15	40%
6	Case studies		
	3-year-old	16	40%
	15- to 19-year-old	17	83.3%
	3-year-old	18	25%
	5-year-old	19	75%

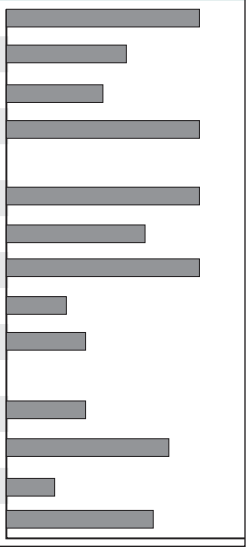

Fig 3 Percentages of questions reaching consensus in round 1.

Part	Topic	Question no.	Round 2
1	Dental disability	1	–
2	Initial diagnosis	2.3.4	12.5%
3	Global disability	5,6,7,8	10%
4	Oral health	9	–
5	Dental treatment options		
	Orthodontics	10	–
	Hypodontia	11	10%
	Anodontia	12	–
	Implants	13	31.6%
		14,15	60%
6	Case studies		
	3-year-old	16	60%
	15- to 19-year-old	17	0%
	3-year-old	18	75%
	5-year-old	19	25%

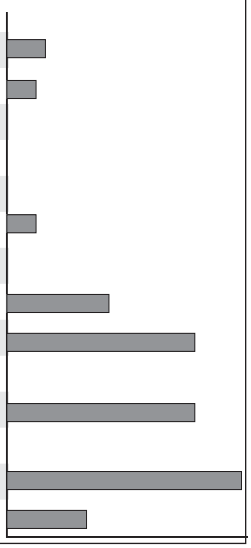

Fig 4 Percentages of questions reaching consensus in round 2.

Question 13 required considerable discussion to agree upon an approach to the optimal use of dental implants in growing bone.

Figures 3 to 5 summarize the progress in agreement among the participants, and Fig 5 highlights the residual questions. The agreed-upon rewording of questions to reach consensus is tabulated and may be found online at http://sydney.edu.au/dentistry/research/article.php.

Item 1, Part 2: Initial Diagnosis

Question 3a. Radiographs: *At what age should panoramic radiographs be taken?*

Discussion was informed by the following:

- It was generally agreed that a panoramic radiograph should be taken at a time deemed appropriate by the clinician.
- Should an age be specified?
- Radiation guidelines and regulations in Sweden and the United Kingdom state that radiographs must be taken for a specific indication.
- A panoramic radiograph should not be taken before age 6 to clarify which teeth are missing.
- Radiographs should not be taken too early in development.
- Radiographs are part of diagnostic assessment—a diagnostic tool—and not part of routine treatment prior to therapy.
- An alternative of 10 intraoral radiographs instead of a panoramic radiograph could be used.
- In Sweden, the radiologist decides what radiographs are needed.
- Radiographs should only be taken when there is clinical justification to support diagnostic assessment. There is no justification for routinely taking radiographs.
- It was agreed that the question was poorly worded.

Agreed rewording of Q3: *When should radiographs be taken for diagnostic or therapeutic purposes?* ***Agreed answer for Q3:*** Radiographs should be only taken when clinically justified to support diagnostic assessment, and there is no justification for the routine use of radiographs in young patients.

Item 2, Part 3: Global Disability

Question 5a_1(ii). *Is it acceptable to sedate a precooperative child to provide treatment for hypodontia to take an impression?*

Discussion was informed by the following:

- It was noted that *precooperative child* is a pediatric term for a child who is at an age or stage of development where reasoning is not possible.
- Sedating a child who is uncooperative is not considered ethical in Sweden and the United States.
- Different countries have different ethical requirements.

- In the United States and Australia, clinicians must have additional university training (a graduate diploma) to provide sedation.
- In France, only conscious sedation is allowed, whereas in the United States, deep sedation is allowed.
- In Australia, pediatric anesthesia requirements have changed, and general anesthesia is now used routinely.
- A child who is not cooperative can be trained to understand the process of impression taking, eg, by introducing the mixing of alginate and placing the child's fingers in the material or by giving the child an impression tray to take home to practice before returning for impressions.
- Study models may be needed as part of the treatment process, and a child may be sedated for ease of impression taking.
- Prosthodontists who were accustomed to using sedation were not always comfortable with it, whereas all pediatric dentists used sedation regularly.

Agreed rewording of Q5: *Is there an indication to sedate a precooperative child to provide treatment for hypodontia?* ***Agreed answer for Q5:*** There are jurisdictional differences that define the ethical requirements in different countries that needed to be acknowledged, and that, where appropriate, a conservative approach that includes child training is preferred to sedation.

Item 3, Part 3: Global Disability

Question 7a. Orofacial function needs to be considered as a patient-specific problem and the assistance of parents is required; a pretreatment assessment questionnaire is required to be completed by the parent(s) and child.

Discussion was informed by the following:

- All agreed that orofacial function should be considered; however, its assessment is problematic.
- Questionnaires are not essential for clinical assessment and diagnosis; however, without them, some information may be overlooked.
- Questionnaires are accepted as research instruments but not for routine patient care.
- All agreed to keep the question but reword it.
- A Scandinavian network of dentists and speech/language pathologists have developed a screening method for orofacial function (Nordic Orofacial Test–Screening[2]; see also www.mun-h-centre.se). This has been used with ED children and adults in 46 centers in Sweden. It was found that there were problems in many of the 12 domains of orofacial function (speech, dryness of the mouth, chewing, etc), that may not be necessary for clinical purposes. As a result, it was concluded that a screening method was a quick and easy way to determine the patient's problem, but it was not a requirement for clinical judgment.
- Questionnaires indicate a formal approach for data collection and may also be used as a research tool.
- All agreed that orofacial function needed to be considered as a patient-specific problem and that assistance from the parent or caregiver was required; therefore, a rewording of the statement was required.

Part	Topic	Question no.	Round 3
1	Dental disability	1	–
2	Initial diagnosis	2.3.4	12.5%
3	Global disability	5,6,7,8	20%
4	Oral health	9	–
5	Dental treatment options		
	Orthodontics	10	–
	Hypodontia	11	0%
	Anodontia	12	–
	Implants	13	21%
		14,15	–
6	Case studies		
	3-year-old	16	–
	15- to 19-year-old	17	16.7%
	3-year-old	18	–
	5-year-old	19	–

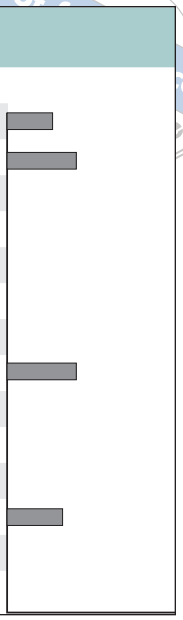

Fig 5 Percentages of questions reaching consensus in round 3.

Agreed rewording of Q7: *A pretreatment assessment is required.* ***Agreed answer for Q7:*** Orofacial function needed to be considered as a patient-specific problem, and that assistance from the parent or caregiver was required to obtain this information.

Item 4, Part 5: Dental Treatment Options

Question 11a_2. Hypodontia: The following suggests an age range for specific treatment possibilities: 2 to 3 years: removable partial dentures (RPDs).

Discussion was informed by the following:

- Making an RPD for children age 2 to 3 years was rarely indicated.
- The objective of providing dentures confers a psychologic advantage when children begin school.
- At preschool or early school age, children normally lose their primary teeth, so it is not uncommon for children in general to have missing teeth at that age.

- Psychologists advise that children at 7 to 8 years of age are more self-aware.
- The problem is not only psychologic but may also be functional.
- It was agreed that the sentence should be reworded.

Agreed rewording of Q11a_2: *The age range for specific treatment.* ***Agreed answer for Q 11a_2:*** At age 2 to 3 years, a possible treatment option is an RPD.

Item 5, Part 5: Dental Treatment Options

Question 11a_3. Hypodontia: The following suggests an age range for specific treatment possibilities: 6 to 10 years: orthodontics/RPD with implants.

Discussion was informed by the following:

- Implants are an option but not a requirement, and the question should be reworded.
- As implants in the maxilla pose many problems, discussion was focused on implant treatment in the mandible.
- The suggestion that orthodontics and treatment with an RPD should be combined was confusing but may be considered as a case-specific requirement.
- It was suggested to remove "with" and add "or" provision of orthodontics "or" a partial denture are options and implants may be included.

Agreed rewording of Q11a_3: *The age range for specific treatment.* ***Agreed answer for Q11a_3:*** At 6 to 11 years, a possible treatment is an RPD and/or mandibular implants.

Item 6, Part 5: Dental Treatment Options

Question 13a. Implants in growing bone (in the absence of alveolar bone): Are there particular ages at which implants may be successfully placed in children?

Discussion was informed by the following:

- The problems were that *(1)* neither the mandible nor the maxilla was necessarily indicated for implants; and *(2)* the number of teeth present or absent did not necessarily prescribe the use of implants; the specific requirements need to be defined.[3,4]
- Different responses were received for different ages; some participants recommended implant treatment as early as 5 to 6 years.
- Clarification was required regarding the earliest age at which implant treatment may be successfully used in *(1)* the maxilla and *(2)* the mandible.
- The earliest age for implant treatment was agreed to be 7 or 8 years in the anterior mandible and older in the maxilla.
- In the edentulous maxilla, implants may be placed, but grafting is usually required, irrespective of age.
- When there is little alveolar bone and no adjacent teeth, implants may be placed earlier. However, if there are adjacent teeth, implants cannot be placed until growth is completed.
- If there is no alveolar bone but there is still growth potential, then there is no urgent need to place implants.
- The jaw relationship with implants placed early is likely to require maxillary advancement when growth is completed. The position of the maxilla would need to be changed regardless of whether implants were placed.
- Parents could be required to make a decision on a major undertaking such as implant placement in the maxilla, where the outcomes are questionable, as opposed to the anterior mandible, where implants are more predictable.
- It was questioned whether, in the case of the maxilla, treatment with an implant-retained RPD was preferable up to a particular age or until adequate growth had occurred. There was considerable discussion about the optimum age at which implants may be used in the maxilla, since a recommendation would have significant implications that some clinicians would use to justify implant placement irrespective of factors other than the patient's age. A definitive statement against using implants in an edentulous maxilla in this cohort would be a statement against recommending implants, but is restrictive. The group did not wish to recommend absolutely that it opposed the idea of providing an 8-year-old with an edentulous maxilla with implants. However, the group was cautious about giving clinicians the authority in general to use implants on the basis that they may be indicated in some circumstances.
- It was acknowledged that, in the maxilla, until adequate growth had occurred, RPDs have proven to be successful and implants need not be considered.
- There is not yet sufficient evidence to either recommend or not recommend the use of implants in the maxilla at age 8.
- It was noted that, in one center, treatment of six edentulous maxillae with overdentures at 6 to 9 years of age had been successful, but long-term follow-up data were not available. In that patent cohort, there were no negative outcomes; however, this raised the question of whether treatment would be perceived as reflecting the patient's or the caregiver's needs. The parents clearly wanted their child treated, but this is a separate issue. This potential negative impact on the success of implants in those children needed to be considered.
- A recent Scandinavian consensus conference on implants[4] that used the Scottish Network for Evidence-Based Dentistry and chose recommendation levels

suggested that the recommendations were based on little evidence and few publications.
- It is necessary to differentiate between the placement of implants in the mandible from 6 years of age if there are no teeth, and the maxilla, for which there are few data. There are no major studies on implants in the mandible either, but more case studies have been reported.
- A 20-year follow-up of data from Sweden was published in 2008 based on 40 implants in six patients. Complications occurred with 52% of the implants, with more complications in the maxilla than the mandible: 11% fractured and 15% were lost. Another study had a rate of 36% for prosthetic complications, which was expected, but the fracture rate was also high.
- With respect to bone grafting, a study is in progress in France to collect all available data about ED patients who have received bone grafts in the maxilla. Although data are limited, the failure rate of the grafted cases was 50%. The ED patients were treated by different teams, and each was highly specialized in bone grafting. The reasons for this very high failure rate are unresolved. If implants in the maxilla are being considered, it is necessary to also consider bone grafting and the preferred technique. The donor site of the graft may also influence the implant outcome. It was suggested that the current consensus for the maxilla in this cohort is to wait until growth is complete, as there were insufficient data to recommend the use of implants.

Agreed answers for Q13a: *(1)* There is currently insufficient clinical or experimental data to support a recommendation for the placement of implants in the maxilla in this cohort; *(2)* it was agreed that, beginning at age 7 to 8, implants may be placed in the anterior mandible when clinically justified, recognizing currently available data.

Item 7, Part 5: Dental Treatment Options

Question 13i. Implants in growing bone (in the absence of alveolar bone): Should implants be spaced, as for a three-unit or a two-unit cantilever RDP, or placed adjacent to each other as for separate crowns: maxilla: anterior/posterior?

Discussion was informed by the following:

- There is no need to place an implant for every two teeth.
- The real issue is that, because the bone in growing patients may have different characteristics from mature bone, as the biomechanical response may be different. However, as these views indicate, a number of variables need to be considered when deciding the optimum implant configuration, including whether there is the potential for bone to have different properties (mechanical or biologic).
- Are there differences between an individual of 50 years or more who has lost all teeth and all alveolar bone and children with ED? Participants questioned whether the basal bone in these edentulous patients with alveolar resorption behaves in exactly the same way in patients who lack alveolar bone because of hypodontia. Clinical experience suggests that the behavior of bone in patients with ED may be different biologically. There is no research evidence on bone characteristics, despite the fact that such patients are sometimes potentially better treated with multiple implants for function and esthetics. Such an option is currently considered unacceptable because of a lack of evidence regarding bone behavior. It can be stated that: *(1)* the decision regarding the implant configuration used is based on multiple factors; *(2)* there is not enough evidence to support the decision, as the bone may be different in patients with ED from bone versus those who have lost teeth and alveolar bone but do not have ED or hypodontia.
- There is some evidence to indicate that basal bone has similar mineralization levels in dentate and edentulous mandibles. However, the data point to genetic and bone mineral differences in dentate and edentulous jaws.
- Could the differences be a result of functional rather than biologic factors?
- The anterior and posterior regions are different because the former involves esthetic issues that might require implants that are spaced rather than adjacent to each other. In the posterior maxilla, implants placed in a straight line may lead to problems during occlusal loading/function, so a higher number of implants in the posterior maxilla should be considered, as well as their configuration.
- In these patients there is limited bone height, indicating a need for shorter implants. However, there is no evidence for the use of mini-implants.
- A general statement in relation to differential management of the anterior and posterior maxilla is required because of: *(1)* loading requirements and *(2)* prosthodontic considerations. However, in the absence of specific data it is prudent to consider the possibility of a cantilever fixed dental prosthesis in the anterior mandible for its esthetic advantages and individual implants in the posterior regions.
- Some clinicians prefer to splint implant restorations. Given functional and parafunctional loading expectations, one implant per tooth in the posterior mandible may be advantageous. It was *not agreed* that an implant to replace each tooth in posterior

regions was required; it *was agreed* that cantilevers should be of minimal length.
- The designing of cantilevers requires an understanding of how to manage the occlusion and consideration of the opposing arch.

Agreed answer for Q13i: There is insufficient evidence to support the use of implant configurations that are different from those used in patients who do not have this condition.

Item 8, Part 5: Dental Treatment Options

Question 13k. Implants in growing bone (in the absence of alveolar bone): Is an implant overdenture more appropriate in growing bone?

Discussion was informed by the following:

- Regarding an implant overdenture versus a fixed implant dental prosthesis: Are implants to be placed in the maxilla for an overdenture or a fixed dental prosthesis?
- Regarding linking five spaced implants with a bar: Is this an appropriate option for growing bone? ([i] Maxilla: anterior/posterior)
- The bar could be divided into separate sections and the prosthesis designed as an overdenture that attaches to the bar.
- The question for the maxilla is for an overdenture linked to implants as a RPD.
- Consensus was reached on k1 and k2 concerning rehabilitation in the mandible, and it was therefore not necessary to consider this question.
- Question (k) to be deleted.

It was agreed that question 13k could be deleted.

Item 9: Case Study 2

Question 17d. Consider treatment for the same patient at 15 years and 19 years. What might be the treatment for loss of occlusal vertical dimension associated with primary molar ankylosis?

Discussion was informed by the following:

- Because primary teeth are present and the plan is to place implants later, what strategy should be used? There is very little support in the literature for an implant approach, and there is an increased risk of losing bone if the teeth are extracted.
- Implants should be placed as close to the extraction sites as possible; however, the literature is based only on clinical opinion.
- It is acceptable to place implants immediately following tooth extraction.
- With a patient with missing teeth and a mandibular third molar, the orthodontist would be asked to attempt to reduce the space mesiodistally.
- Is transplanting a tooth possible in this case, and is it common in some parts of the world?
- Tooth transplantation is very successful if planned and executed carefully but is particularly operator-sensitive.
- The general statement should be refined as a response to question 17.

Agreed answer for Q17: A number of treatment options are available for this patient, which are dependent on: clinical evidence (not yet available), the views of the patient and their caregiver, available resources, and the experience and skill of the team. A number of treatment options are appropriate depending on circumstances and may not be mutually exclusive.

The agreed wording for all questions to reach consensus has been tabulated and can be seen on the dedicated website at http://sydney.edu.au/dentistry/research/article.php.

DISCUSSION

The meeting was informed by a welcomed breadth of discussion. It was apparent that expert groups varied in their clinical experiences with dental implants; however, there was common agreement regarding interdisciplinary management of ED patients. There was also a willingness to discuss and recognize possible clinical applications of implants based on the successes reported in adult oral rehabilitation. It was also accepted that there was emerging data on the use of implants in growing bone.

The question regarding implants for a particular age range and with specific anatomical locations in the jaws was of special interest. It was agreed that the decision was necessarily based on patient-specific requirements and clinical indications in growing bone, and that dental and oral rehabilitation should desirably be conservative and minimally invasive for this patient cohort.

Nonconsensus items are summarized in Fig 2. The difficulty in reaching consensus during the Delphi study iterations was partly a result of the wording of some questions and of differences in clinical protocols for children across the groups, which required discussion. Questions requiring rewording were questions 5 and 11, and the specific rewording is described in the results; it was also agreed that question 13k be deleted, as there was no evidence on which to provide informed comment.

Discussion on the other residual questions allowed careful consideration of the implications of particular wording. The group wished to ensure that there

was an absence of ambiguity and felt a responsibility to make recommendations based on evidence from published data, rather than expert opinion. It was acknowledged that available data from clinical studies were not sufficient to provide all required answers, and as a result the agreed wording was conservative. In particular, this applied to the question of maxillary implants in growing bone, where it was acknowledged that data were lacking to support general recommendations. As a result, this particular aspect of questions 13a was emphasized and 13k was deleted, as it could not be considered in an evidence-based context in the absence of appropriate clinical data.

It was recognized that there may be biologic and structural differences in growing bone in ED patients compared with regular patients, and there are emerging data to describe the ultrastructural differences.[5,6] Further data are required to clarify these differences and their implications for treatment planning.

CONCLUSIONS

A total of 11 international teams, identified as leading groups in the field by the coordinating team at the University of Sydney, Faculty of Dentistry, contributed to a Delphi study. The protocol was applied constructively to address, by questionnaire, questions relating to the assessment and management of patients presenting with varying expressions of ED and edentulism.

Following each round of questionnaire completion, there was progressive agreement in the questions that followed clarification of the initial nonconsensus questions that emerged by way of the Delphi study protocol. Clarification was possible as a result of the specific responses to questions from each team, which allowed the nonconsensus questions to be reworded and returned as the following round in the protocol. After three iterations of the questionnaire, there were residual items that could not be adequately addressed without a face-to-face discussion at a consensus meeting.

The consensus meeting brought together team leaders and key members of the teams. Eleven teams were represented and contributed to a 2-day discussion of the residual questions. This proved to be a successful interaction where, although the discussion identified team differences based on their specific experiences and expectations, agreement was reached by consensus regarding the rewording of some questions for clarification. Following consensus on all questions, plans were considered for a multicenter study.

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