Developing a core outcome set for the health outcomes for children and adults with congenital oesophageal atresia and/or tracheo-oesophageal fistula: OCELOT task group study protocol

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ABSTRACT

Introduction

Heterogeneity in reported outcomes of infants with oesophageal atresia (OA) with or without tracheo-oesophageal fistula (TOF) prevents effective data pooling. Core outcome sets (COS) have been developed for many conditions to standardise outcome reporting, facilitate meta-analysis and improve the relevance of research for patients and families. Our aim is to develop an internationally-agreed, comprehensive COS for OA-TOF, relevant from birth through to transition and adulthood.

Methods and analysis

A long-list of outcomes will be generated using(i) a systematic review of existing studies on OA-TOF, and (ii) qualitative research with children (patients), adults (patients) and families involving focus groups, semi-structured interviews, and self-reported outcome activity packs. A two-phase Delphi survey will then be completed by four key stakeholder groups:(i) patients (paediatric and adult); (ii) families;(iii) healthcare professionals; and (iv) researchers. Phase 1 will include stakeholders individually rating the importance and relevance of each long-listed outcome using a nine-point Likert scale, with the option to suggest additional outcomes not already included. During phase 2, stakeholders will review summarised results from phase 1 relative to their own initial score and then will be asked to re-score the outcome based on this information.

Responses from phase 2 will be summarised using descriptive statistics and a predefined definition of consensus for inclusion or exclusion of outcomes. Following the Delphi process, stakeholder experts will be invited to review data at a consensus meeting and agree on a COS for OA-TOF.

Ethics and dissemination

Ethical approval was sought through the Health Research Authority via the Integrated Research Application System, registration no. 297026. However, approval was deemed not to be required and so study sponsorship and oversight were provided by Alder Hey Children's NHS Foundation Trust. The study has been prospectively registered with the COMET Initiative. The study will be published in an open access forum.

WHAT IS ALREADY KNOWN ON THIS TOPIC

- Typical for rare conditions, evidence from research on OA-TOF is limited and data are often of low methodological quality
- Heterogeneity of outcome measures between existing data sets, combined with the effect of biases and a lack of consensus on which outcomes are important for patients and families, makes for poor clinical validity and data synthesis is challenging

WHAT THIS STUDY ADDS

- Short and long-term outcomes will be highlighted through participation of a broad representation of key stakeholders, including patients (children and adults), families, healthcare professionals and researchers
- International collaboration aims to improve intercontinental and transcultural validity of the final COS across healthcare systems

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE, OR POLICY

- Benefits of a COS for OA-TOF include: (i) improved relevance of research for patients, families, healthcare professionals and researchers; (ii) standardisation of outcome reporting; (iii) reduction in outcome reporting bias; and (iv) facilitation of meta-analysis
- Ultimately this will improve the quality of OA-TOF research and permit development of guidelines that are truly evidence-based and patient-centred
- We envisage that an OA-TOF COS will inform database and registry studies, as well as guiding best practice for clinical governance and multidisciplinary team initiatives

INTRODUCTION

Oesophageal atresia (OA) is a congenital malformation where there is interruption in the continuity of the oesophagus. It occurs in approximately 1 in 3500 to 1 in 4200 live births [1, 2]. The most common OA variant (85%) consists of a blind-ending upper oesophagus and a lower segment connected to the trachea (tracheo-oesophageal fistula [TOF]); there are less common subtypes relative to the location and presence of OA or TOF.

OA-TOF may be suspected antenatally, typically with polyhydramnios and less commonly through detection of a small or absent fetal stomach [3]. More often it is diagnosed shortly after delivery. Most babies present with inability to swallow saliva and milk feeds. They may aspirate, causing choking and respiratory distress. Diagnosis is typically confirmed by inability to pass a feeding catheter from the mouth or nose into the stomach. Rather, the catheter will coil in the upper oesophageal atretic pouch, demonstrable on a plain chest radiograph. Presence of abdominal bowel gas suggests the presence of a distal TOF.

OA-TOF requires surgical intervention shortly after birth. In most cases (when present), this begins with TOF ligation to prevent gastric ventilation and airway contamination from gastric secretions. Where possible, this is followed by oesophageal anastomosis. In cases of OA without TOF (~10%) or more rarely OA with a proximal TOF, there is typically a long gap between the atretic ends of the oesophagus, making primary anastomosis more challenging or impossible [4]. In such cases, a gastrostomy is typically formed in the early neonatal period and/or delayed oesophageal anastomosis or replacement is performed. When primary oesophageal anastomosis is not feasible, surgical options for oesophageal continuity are numerous, but broadly can be divided into: (i) delayed primary anastomosis following a period of growth (with or without techniques to lengthen the native oesophagus); or (ii) replacement of the oesophagus with the stomach, colon, or a small bowel graft segment [4]. Irrespective of surgical technique(s) and the success of primary oesophageal anastomosis, children with OA-TOF often have mechanical and functional abnormalities with significant morbidity and potentially life-long impact for the patient and family [5, 6].

Problems after OA surgery are multifactorial. For optimal healthcare, children should ideally be managed in specialist clinics staffed by multidisciplinary teams, yet access to such specialists appears variable amongst centres [7, 8]. Recurrent respiratory tract infections and tracheomalacia are frequently seen and consequential long-term poor growth is recognised [9, 10]. Gastrointestinal complications are common, including gastro-oesophageal reflux, oesophageal stricture, oesophagitis and Barrett's oesophagus [11-13]. Psychosocial impact on the patient and family, including in later life, can be profound [14, 15].

Typical for rare conditions such as OA-TOF, evidence from clinical trials and other research is limited and data are often of low methodological quality, particularly relative to outcomes. Comparable to other neonatal surgical pathology, heterogeneity of outcome measures exists between data sets; this, combined with the effect of biases and a lack of consensus on which outcomes are important for patients and families, makes for poor clinical validity and data synthesis is challenging [16, 17].

It is particularly important that reported outcomes should be relevant, accurately represent the studies' findings and be synthesisable through meta-analysis. A core outcome set (COS) is defined as an agreed minimum set of outcomes that should be measured and reported in all studies in a specific condition [18]. Benefits of a COS for OA-TOF include: (i) improved relevance of research for patients, families, healthcare professionals and researchers; (ii) standardisation of outcome reporting; (iii) reduction in outcome reporting bias; and (iv) facilitation of meta-analysis.

METHODS AND ANALYSIS

Scope

Our aim is to develop an internationally agreed and comprehensive COS for OA-TOF relevant to all ages, from birth through to health service transition and adulthood. Our objectives are as follows:

- Determine and consolidate outcomes currently reported in OA-TOF studies
- Identify outcomes that patients (children and adults), families, healthcare professionals (HCP) and researchers regard important following surgical repair for OA-TOF
- Prioritise outcomes that patients, families, HCP, and researchers think should be included in a COS for OA-TOF
- Reach final consensus between key stakeholders on a COS for OA-TOF applicable to both research and routine clinical practice

Study oversight

A steering committee has been established to oversee this work and will meet regularly during this process, ensuring the study runs in accordance with good research practice and guidelines are upheld. The committee includes ten HCP, one researcher from the COMET Initiative and two patient representatives (one of whom is also an HCP).

Stakeholders

Four main stakeholder groups will be involved in the COS development. All stakeholder groups will have both United Kingdom (UK) and international involvement:

1. Patients (children, young people and adult representatives)

Children, young people and adults with OA-TOF will be invited to participate through social media platforms and charitable groups (including *TOFS* and *EAT: Federation of Esophageal Atresia and Tracheo-esophageal Fistula Support Groups*), with invitations to join focus groups, interviews, or the Delphi process [19-20].

2. Families (parents, carers, children, siblings, spouse/partner)

Whilst the focus will be on parents of children with OA-TOF, we will also seek families of adults with OA-TOF and welcome their thoughts on key outcomes. This group may have a unique insight into the later impacts of OA-TOF, including psychosocial impact. Families will again be recruited via social media platforms and charitable organisations.

3. Healthcare professionals

HCP may have different perspectives to patients and families and include paediatric and neonatal surgeons (some with specific interests in thoracic and upper gastrointestinal surgery, as well as antenatal counselling), respiratory physicians, otolaryngologists, gastroenterologists, neonatologists, general paediatricians, specialist nurses, speech and language specialists, physiotherapists, dieticians, general practitioners and psychologists. HCP will be invited to participate through professional channels (including principal existing OA-TOF organisations such as European Reference Network for Rare Inherited Congenital Anomalies [ERNICA], International Network of Esophageal Atresia [INoEA] and TOFS), society or college emails and personal communication. Good representation across these groups will be ensured by targeting invitations to under-represented disciplines as required. To improve international relevance, at least one HCP from each continental region is included to provide population representation.

4. Researchers

Academics with a specialist interest in this field will be included and provide insight on how a COS can frame future studies.

Identifying outcomes

There will be four stages to the COS development study (see figure 1):

1. Systematic review

Methodology will be guided by the COMET Initiative handbook principles throughout and recommends a systematic review to inform phase 1 of the Delphi process [21]. Following PRISMA guidelines, a systematic review will be performed of all literature on the OA care process published between 1st January 2015 and 1st October 2023 to highlight already reported outcomes in existing research on OA-TOF. Medline, Embase and Cochrane databases will be searched using the term '(o)esophageal atresia' combined with the following search terms: morbidity; mortality; survival; outcome; complication. All papers concerning any aspect of the main OA care process will be included. Editorials, reviews, guidelines and case reports or case series with <10 patients will all be excluded. Two researchers will independently extract all outcomes. Upon agreement, similar studied parameters will be categorised and merged into overarching terms. Definitions and standardised instruments used to measure these outcomes will also be extracted. The final report will include a complete list of outcomes described in recent OA research.

2. Qualitative research

Opinions on important outcomes for OA-TOF will be sought from patient (child and adult) and family stakeholder groups in the form of online focus groups, semi-structured interviews, and child patient self-reported outcome packs. A core focus of this study is the inclusion of patient and family perspectives to ensure the holistic issues faced by this population are represented in the final COS. The range of methods aims to offer choice to children and adults on how they want to share their views. The involvement of patients and families in this stage is highlighted according to age of patient in table 1.

Table 1. Qualitative research: patient and family involvement

Data collection	**Target population**
Focus groups	Children/young people born with OA-TOF (aged 7-15 years) Adults born with OA-TOF (aged 16+ years) Parents/carers/families of people with OA-TOF (any age)
Semi-structured interviews	All patient and family groups of all ages
Child patient self-reported outcome packs (digital or paper)	Children/young people born with OA-TOF (aged 7-11 years)

We aim to gain maximum variation sampling from all stakeholder groups, with diversity in geographical location, age, and HCP occupation. Participants can choose to attend a focus group (different focus groups were held for each participant group to facilitate flexibility and focussed discussion), interview, or complete a self-reported outcome pack. Focus groups and interviews will be held using digital platforms (e.g. Zoom©) and facilitated by multiple team members.

The steering group will consolidate all responses and categorise as per Dodd's classification [22]. Any similar items will be discussed by the steering group and a decision made whether to amalgamate items or remove one. If a unanimous decision is not reached, both items will remain. Outcomes will then be submitted to the Delphi process.

Whilst focus groups and interviews described in stage 2 of the study will be carried out in the UK with UK participants, the importance of international involvement is recognised. At least one HCP for each continental region will be invited to join a working group.

International collaborators will be invited to contribute to this long-list to ensure any specific geographical variance in outcomes are included. Multi-language Delphi survey will not be feasible, but 2 surveys, an English language and a Spanish language survey are proposed. One of the international collaborators will be responsible for translation of documents to ensure consistency amongst surveys.

3. Delphi survey

A long-list of outcomes will be formulated from the systematic review and qualitative research from stages 1 and 2, which will be submitted to a two-phase online Delphi survey. In each phase, participants from all stakeholder groups will be asked to score each item on a nine-point Likert scale ranging from 1-9 (1-3 labelled *'not that important'*, 4-6 labelled *'important but not critical'* and 7-9 labelled *'critical')*. For each outcome, participants will also have the option of *'unable to score'*. There will also be the option to comment on the reason for their score. A plain language description will be provided for each outcome. At the end of phase 1, participants will have the option to suggest additional outcomes they think are important that were not included in the survey. The steering committee will review all the additional outcomes and decide if they should be added to phase 2 of the Delphi survey.

In phase 2 of the Delphi survey, responses for each stakeholder group will be summarised for each outcome and displayed graphically as the percentage of each group who have given each score. All outcomes scored in phase 1 will be retained for phase 2. Participants will be able to view the grouped responses together with their own score in phase 1 and will be asked to re-score the outcome based on this information using the same Likert scale. Participants may choose to change their score or keep it the same. Participants will also be asked to score any additional outcomes that have been added from phase 1.

The responses from phase 2 of the Delphi survey will be summarised using descriptive statistics and a predefined definition of consensus and outlined in table 2. Responses will be included in the analysis if a participant assesses more than 50% of the outcomes. However, the steering committee will review this approach based on the phase 1 response rate. Reminder emails will be sent to minimise attrition. At least 1 reminder email will be scheduled for each round with additional reminders determined by the response rate and any extensions to the duration of the round.

Table 2. Definitions on consensus

Consensus classification	**Description**	**Definition**
Consensus in	Consensus that outcome should be included in the COS	≥70% in each stakeholder group scoring as 7-9 (*'critical'*) and <15% participants in each stakeholder group scoring as 1-3 (*'not that important'*)

Consensus out	Consensus that outcome should not be included in the COS	≤50% scoring 7-9 (*'critical'*) in each stakeholder group
No consensus	Uncertainty about importance of outcome	Anything else

4. Consensus meeting

Following completion of the Delphi process, a consensus meeting will be held to reach final agreement on the final COS. The results of the Delphi survey will be discussed in an online meeting chaired by an independent facilitator. A sample of participants who completed both phases of the Delphi survey and expressed an interest in attending the consensus meeting will be invited to attend, ensuring similar numbers from each stakeholder group.

Prior to the consensus meeting, participants will receive written information about what to expect from the day, attendance at the meeting will be considered as consent to participate. The consensus meeting will ratify the results of the Delphi survey to confirm outcomes that have met the definition of inclusion or exclusion from the COS after phase 2. All other outcomes that have not reached consensus during the Delphi process will then be discussed and participants of the consensus meeting invited to re-score the outcome, using electronic voting software. Stakeholder groups will score the outcomes separately, using the 1-9 Likert scale, and the same inclusion criteria used for the Delphi survey will be applied here (i.e., 70% or more participants in each stakeholder group scoring the outcome 7-9). If a final core outcome set has not been agreed at the end of the first consensus meeting, subsequent meetings will be arranged.

Patient and public involvement statement

Patient and public involvement is integral to our study, and they are defined key stakeholders in this COS development protocol. Patient and family experiences and opinion directly inform our qualitative research, delphi survey and consensus meeting through their involvement as key stakeholders. They also indirectly inform our systematic review through analysis of prior research on reported outcomes. Study design and oversight is provided by our steering committee, which as outlined includes patient representatives. Study publicity via principal existing OA-TOF organisations and charitable groups is intended to maximise patient and family recruitment to the project. The

study will be published in an open access forum and made available to all key stakeholder groups, including patients and their families.

RESEARCH ETHICS APPROVAL AND DISSEMINATION

Ethical approval has been sought for this work from the Health Research Authority (HRA) through the Integrated Research Application System, registration no. 297026. Following review by the HRA, it was deemed that approval was not necessary as recruitment used methods outside the UK National Health service. Review, study sponsorship and oversight were provided by Alder Hey Children's NHS Foundation Trust. The project has also been prospectively registered with the COMET Initiative.

Written assent/consent (and parental consent where necessary) will be obtained for all focus group and interview participants. Both parental consent and patient assent will be obtained for children under 16 years of age. Electronic consent for the Delphi survey will be obtained at the start of the survey and participants will be unable to move from registration to the survey participation without completing this. Formal consent is not required for the consensus meeting and assumed consent will be used by participants having freely provided their opinions and input.

The study will be published in an open access forum and made available to all key stakeholder groups. We envisage that an OA-TOF COS will inform database and registry studies, as well as guiding best practice for clinical governance and multidisciplinary team initiatives.

AUTHORS' CONTRIBUTIONS

The study was conceptualised and designed by the steering group especially RT, NL, GS, AL, SG, JF and NH. The design of the PPIE was mainly by LBr and JF. The entire steering group fed into the planning of the study. SG led on the protocol design pertaining to the Delphi survey. The first draft and final protocol manuscript was written by JD with input from RT and NL. All co-authors critically reviewed previous versions and approved the final manuscript.

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COMPETING INTERESTS STATEMENT

None declared.

LEGENDS

Figure 1. The four stages of core outcome set development

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