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A patient led, international study of long term outcomes of esophageal atresia: EAT 1

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ABSTRACT

Introduction: Long term outcomes of esophageal atresia (OA) are poorly understood. The Federation of Esophageal Atresia and Tracheo-Esophageal Fistula support groups (EAT), a collaboration of patient support groups aimed to define patient reported long term outcomes and quality of life (QoL) in a large international cohort of OA patients.

Methods: Questionnaires were designed focusing on patient/parent reported outcomes including surgical history, current symptomatology and quality of life. Members of support groups within EAT were invited to complete questionnaires electronically via SurveyMonkey®.

Results: 1100 patients from 25 countries responded to the questionnaire and 928 were analyzed. 80% had type C anatomy, 19% type A and 1% type E. Patient ages were <5 years (42%), 5–10 years (26%), 11–17 years (16%) and 18 years and older (16%). 49% of all patients reported previous dilatations which was similar across age groups. Reflux symptoms affected 58% of patients and persisted into adulthood. Dysphagia also persisted in the adult population with 50% reporting sometimes or often getting food stuck. Reflux was significantly more frequent in 'long gap' versus 'standard gap' patients ($p < 0.005$). Respiratory symptoms and chest infections decreased in frequency with age. In children median SDS for height was -0.41 (IQR -1.4 to 0.67) and that for weight was -0.63 (-1.6 to 0.67). BMI in adults was 21.5. Quality of life was described as significantly affected by OA in 18% of patients while 25% reported no effect on QoL.

Conclusions: These results highlight the significant long term morbidity suffered by OA patients as children and into adulthood and suggest the need for quality transitional care. The patient designed and reported nature of the study gives a unique perspective to the results and emphasizes the benefits of collaboration.

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1. Background

Outcomes in esophageal atresia (OA) have improved in the last 50 years with survival now of more than 90% [1–3] leading to an increasing population of long term survivors. While our understanding of short term outcomes in infants with OA is good [4–7], longer term functional outcomes are more poorly understood with a limited research base [8–10]. There is increasing interest therefore in longer term outcome studies, and results of such studies potentially impact current practice in management of OA. This may be especially important for transitional care as patients leave the care of the pediatric surgeon for the adult world. The current literature tends to be limited to small national [4,9] and institutional datasets or meta-analysis of such studies [8]. Patient reported outcomes (PROMS), as an adjunct to

reporting of outcomes by physicians and surgeons are becoming increasingly recognized as very important for improving care in a variety of conditions [11,12].

The Federation of Esophageal Atresia and Tracheo-Esophageal Fistula support groups (EAT) is an international federation of patient support groups (Table 1), led by patients and parents aiming to share the international knowledge and experience of OA, promote awareness of the condition across the world and support worldwide research and collation of information concerning the treatment and care of people born with esophageal atresia. EAT therefore has a keen interest in the short and long term outcomes of OA and has first-hand experience of the difficulties faced by many patients and carers at all times in their journey from diagnosis through transition and on into adult care. In order to gain an idea of the patient-reported outcomes in OA, EAT has conducted a patient and carer designed and led study. This paper aims to report the data from this unique study in order to provide insight into patient-reported short and long term outcomes of OA in a large international cohort of patients with OA.

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Table 1
Member organizations of EAT.

Organization		Country
AFAO	Association Francaise de l'Atresie de l'Oesophage	France
ABeFAO	Association Belge Francophone de l'Atrésie de l'Oesophage	Belgium
FATE	Famiglie con Atresia dell'Esofago	Italy
KEKS	Kinder und Erwachsene mit kranker Speiseröhre	Germany
KEKS	Kinder und Erwachsene mit kranker Speiseröhre	Austria
OARA	Oesophageal Atresia Research Association	Australia
OA-Switzerland	OA-Switzerland	Switzerland
TOFS	Tracheo-Oesophageal Fistula Support	United Kingdom
TROAD	Türkiye Özofagus Atrezisi Çocuk ve Aile Destek Derneği	Turkey
VOKS	Vereniging voor Ouderen en Kinderen met een Slokdarmafsluiting	The Netherlands
AQAO	Association Quebecoise de l'Atresie de l'Oesophage	Canada

2. Methods

This study was conceptualized as a questionnaire which was then designed by the board of EAT (see author list) with support from a Professor of Quality of Care (MvD). It was designed to be a patient/carer focused study looking at outcomes of specific interest to those groups. The questionnaire was therefore designed to assess the following key areas of interest:

1. Current symptomatology
2. Patient/carer satisfaction
3. Quality of life (QoL)

Demographics and operative details were also obtained as part of the questionnaire in order to describe the OA population surveyed. A questionnaire was then piloted within the EAT board, prepared in multiple languages (English, Dutch, German, French and Italian) and put in an electronic format using SurveyMonkey®. Invitations to participate were sent electronically to all members of 11 different European patient support groups (Table 1). This included patients and carers in 24 different countries, as members of the support groups sometimes resided outside of Europe.

Where possible, patients completed the questionnaire; otherwise this was performed by carers. Completed questionnaires were collected centrally, collated and analyzed anonymously. Initial data were presented by the Chairman of EAT at British Association of Paediatric Surgeons Congress (Cardiff, 2015). Following this the organization approached UCL Institute of Child Health for assistance in data analysis and interpretation. Formal ethics approval was not initially sought as this was a patient group led study. Approval was obtained for anonymized data analysis from ICH/GOSH Research & Development office at (project number 16DS21). Initial data were cleaned and duplicate responses excluded, together with responses where date of birth was not completed. Responses with missing or duplicate data for individual questions were included for those questions answered and percentages calculated from only completed answers.

Patient height and weight were analyzed using standard deviation scores (SDS) using LMS Growth Excel Plug In version 2.69 and UK-WHO Growth Charts in respondents younger than 18 years. Body mass index was calculated for those 18 years and older and compared to the WHO International Classification. Data were compared by Fisher's exact test or Mann-Whitney, regarding $p = 0.05$ as the cut-off for significance.

3. Results

3.1. Demographics

1100 responses were received from an estimated 2500 members of EAT organizations invited to participate. Following exclusions, 928 responses (56% male and 44% female) were analyzed with a patient age

range of 1 month to 60 years. For analysis, patients were divided into the following age groups, <5 years (42% of responses), 5–10 years (26%), 11–17 years (16%) and 18 years and older (16%).

Responses were received from 25 different countries (Fig. 1). Smaller numbers of responses were also received from Canada, USA, Ireland, South Africa, New Zealand, Spain, Guadeloupe, Tunisia, Sweden, Malta, Lithuania, Israel, Hungary, Hong Kong, Greece and Denmark in decreasing order of number of responses. 121 (18%) of responses were completed by patients and 761 (82%) by a parent or carer.

OA with tracheoesophageal fistula (OA-TOF) was the most common defect, reported in 80% of cases, 19% were pure OA and 1% an H type fistula. Participants reported their defect as 'long gap' either as supplementary to reporting the anatomy of their defect or as an isolated diagnosis in 18% of cases. Similar distribution of type of defect was seen in all age groups although there were a significantly higher proportion of pure OA and long gap participants in the adult age group with 13% of cases being pure OA in participants younger than 18 years versus 23% in the adult cohort ($p = 0.004$).

3.2. Surgical care

Overall, the median length of hospital stay following initial surgery was 6 weeks (range 1 to 174 weeks). However, considering only the respondents older than 18 years, the median length of stay was 8 weeks (IQR = 16). When compared by diagnosis, those with OA-TOF had a median LoS of 6 weeks (IQR) versus 8 weeks for those with pure OA (IQR). Long gap cases reported that median LoS was 18 weeks (IQR) versus 5 weeks (IQR) for standard gap OA. Fifty-three percent (0–5 years), 46% (5–10 years) and 52% (11–17 years) of participants in the younger age groups respectively only required 1 operation to repair their defect, whereas in the 18 year and above group only 36% were repaired with a single operation and 45% needed 2–5 procedures.

The proportion of patients receiving dilatation following repair is summarized in Fig. 2. Forty-nine percent of all respondents had esophageal dilatation and the number of dilatations received was broadly similar across each age group. If dilatation was performed the median number was 4 dilatations and was the same for all age groups.

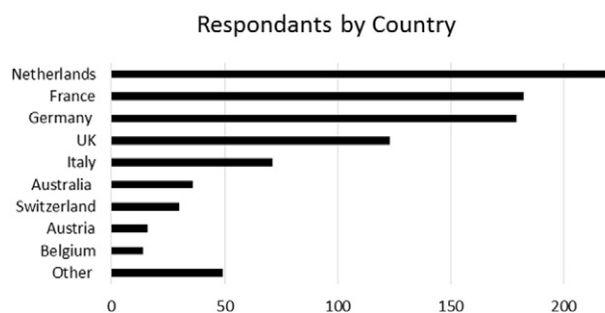


Fig. 1. Respondents by country.

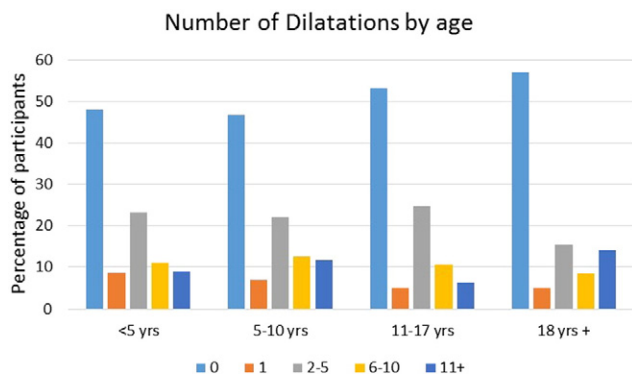


Fig. 2. Numbers of dilatations by age.

The median number of reported dilatations in participants with long gap OA was 2 (IQR 0–10) and was significantly higher ($p < 0.001$) than in those with standard gap who reported a median of 0 dilatations (IQR 0–5). Those respondents reporting reflux symptoms had a significantly higher median number of dilatations at 1 (IQR 0–5) than those without symptoms where the median number of dilatations was 0 (IQR 0–3; $p = 0.01$).

3.3. Symptomatology

Across all participants, 65% reported that they could 'eat anything' with no restrictions on type of food. In those younger than 5 years, this figure was only 50% which was significantly less than those older than 5 years in which 75% reported they could 'eat anything' ($p < 0.0005$). In those with long gap defects, only 35% of those younger than 10 years reported that they could eat without restriction.

Fig. 3 shows how frequently participants in different age groups reported food getting stuck in the esophagus. The patterns across age groups are broadly similar with 16% of participants often getting food stuck and only 22% reporting never having food stick. More than 50% of adult patients (18 years and older) report sometimes or often getting food stuck in the esophagus.

58% percent of respondents reported gastroesophageal reflux symptoms and a similar incidence was seen in all age groups; 65% in <5 years, 53% in 5–10 years, 54% in 11–17 years and 60% in adults. When analyzed by gap length, 78% of those with long gap defects reported suffering from reflux symptoms versus 55% of respondents with standard gap length ($p < 0.005$). The use of antireflux medication showed a decreasing trend with age. Reflux medication was reported being taken by 60% of those younger than 5 years but only 30% of adult participants, despite a similar proportion in each age group reporting symptoms.

Respiratory problems were commonly reported by participants and show a decreasing trend with age (Fig. 4). Patients with long gap OA more often reported respiratory symptoms with 42% of younger than

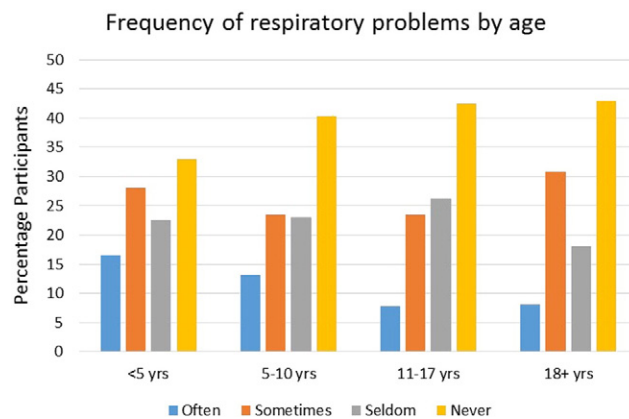


Fig. 4. Frequency of respiratory problems.

5 years and 24% of 5–10 years old with long-gap reporting 'often' having symptoms compared to 16% and 8% respectively in standard gap patients. There is a similar decreasing trend with age in the incidence of chest infections with 30% of children younger than 5 years reporting having more than 3 chest infection per year but only 14% of adults reporting the same frequency of infections (Fig. 5). Long gap patients did not report more frequently occurring chest infections than standard gap patients except in those younger than 5 years.

Regular antibiotic usage has shown similar decreasing trend in the use of with 17% of 0–10 year olds reporting regularly needing antibiotics, falling to 9% (11–17) and 5% (>18) in the older age groups respectively.

3.4. Anthropometry

On average, children <18 years with OA were slightly less than average weight and height, with median SDS scores of -0.41 (IQR -1.4 to $+0.67$) for height and -0.63 (IQR -1.6 to 0.17) for weight, where population averages are zero (Fig. 6A). BMI in respondents 18 years and older is shown in Fig. 6B. Two percent of adult respondents reported weight and height that put them in the obese BMI category, 15% were overweight, 62% were normal weight and 21% were underweight. The mean BMI was 21.5.

3.5. Care provision

Current care provision is shown in Fig. 7. Twenty-two percent of children younger than 5 years are reported not to be under the care of a pediatric surgeon and there is a trend of fewer participants being looked after by pediatric surgeons with increasing age. There is also an increasing trend with age in those with no current care provider with

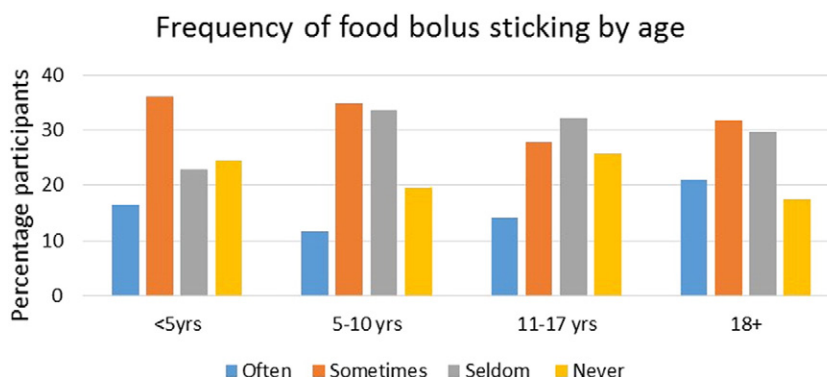


Fig. 3. Frequency of food bolus sticking by age.

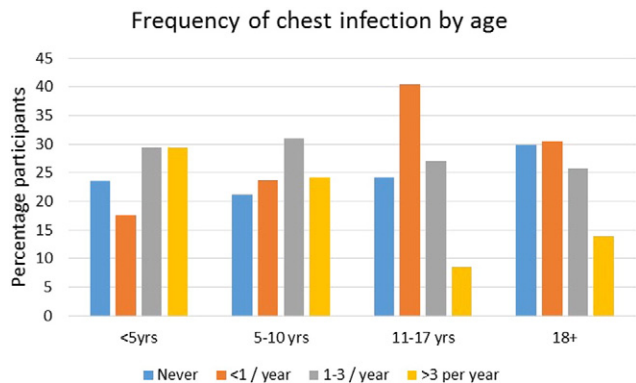


Fig. 5. Frequency of chest infections by age.

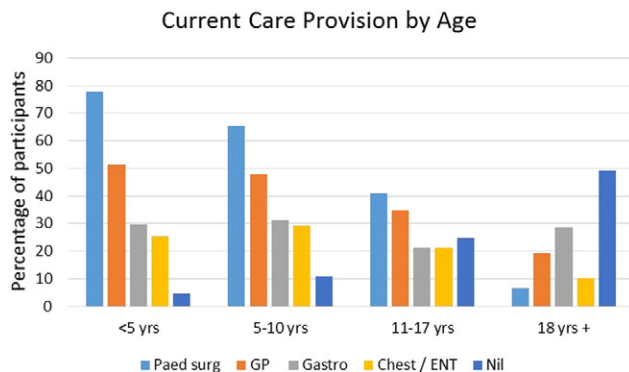


Fig. 7. Current care provision by age group.

50% of adults not reported being looked after by a general practitioner or equivalent primary care physician.

3.6. Quality of life

The graphs in Fig. 8A–D show responses to the questions related to quality of life in the study. They demonstrate that while 20–25% of respondents report that OA has had no effect on their own or their families QoL a similar proportion of patients suggested that their QoL was significantly affected. More than 55% of respondents report ‘some’ or a ‘significant’ effect on their family’s QoL.

4. Discussion

The EAT 1 survey offers unique, international patient directed short, medium and long term follow-up data on one the largest cohorts of OA patients in the literature to date. It highlights the clinical long term morbidity that OA patients suffer from and how these may change with age. It also importantly examines the often ignored quality of life issues faced by this patient group. The large size of this study group makes it more representative of the wider international population of OA patients than most of the existing literature which is limited to smaller national [4,10] and institutional data series [3,7,13] or meta-analyses [8]. The study participants are broadly representative of the wider population based on anatomy of original defect in comparison to EUROCAT figures [14]. The significantly higher incidence of pure and long gap OA in the adult group probably represents the demographics of adult patients accessing support groups, i.e., those with more complex conditions and a similar pattern of recruitment is seen in the German support group (KEKS) quality of life (QoL) study

[15]. While ongoing care is expected for most complex patients during their adult life, this is also one of the limitations of studies like ours in which responders are recruited via a patient support association and should be taken into account during counseling. The age range of participants studied again makes this study unique including infants up to the oldest subject who was 60 years of age at completion of the survey. The fewer patients in the older age range may be the result of several factors including the previous limited survival of babies in the early years of OA surgery, the relatively new development of patient support groups – through which recruitment took place – and the perceived need of such support groups in older patients. The geographic spread of participants represents the principal patient support groups involved in the study and membership in each of those countries.

The anthropometric data collected in this study are consistent with existing literature [16–18] suggesting that as children, patients with OA tend toward being underweight and to a less extent shorter than the normal population. Interestingly, we showed a significantly lower mean score for height in children younger than 5 years than older children. It may represent a sampling weakness as these were self-recorded and reported outcomes in an age group in which it may be difficult to get accurate length/height measurements. Chetcuti and Phelan [17] reported that there is ‘catch up’ growth as children become adolescents and then adults. The median BMI of the adult group is within the normal range at 21.5 suggesting catch-up growth, although there are still 21% of adults classed as underweight by BMI and a much lower level of obesity (2%) in this patient population than in the normal European population (15–30%).

The postoperative management of OA has improved over the last 2 decades and this may explain the shorter length of stay seen in the younger participants. However, the groups are not evenly matched for type of defect and gap length and the higher incidence of long gap and

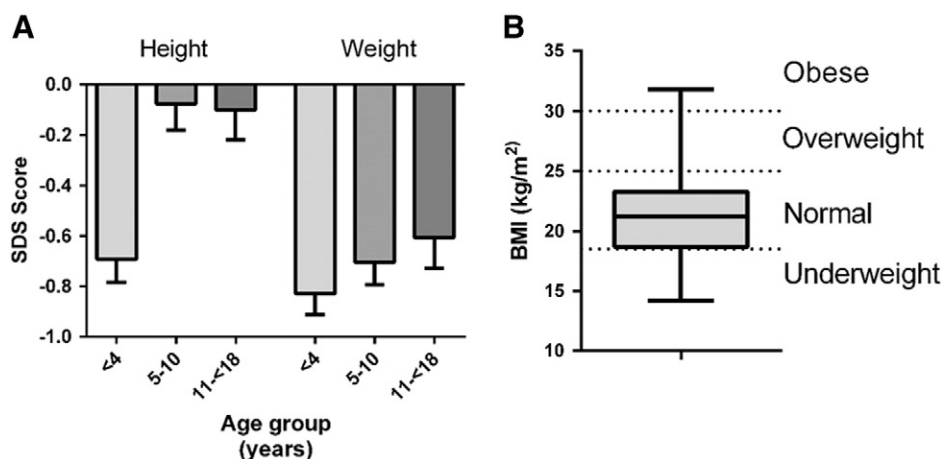


Fig. 6. Height and weight by age. (A) SDS score in patients younger than 18 years. (B) BMI in adult patients.

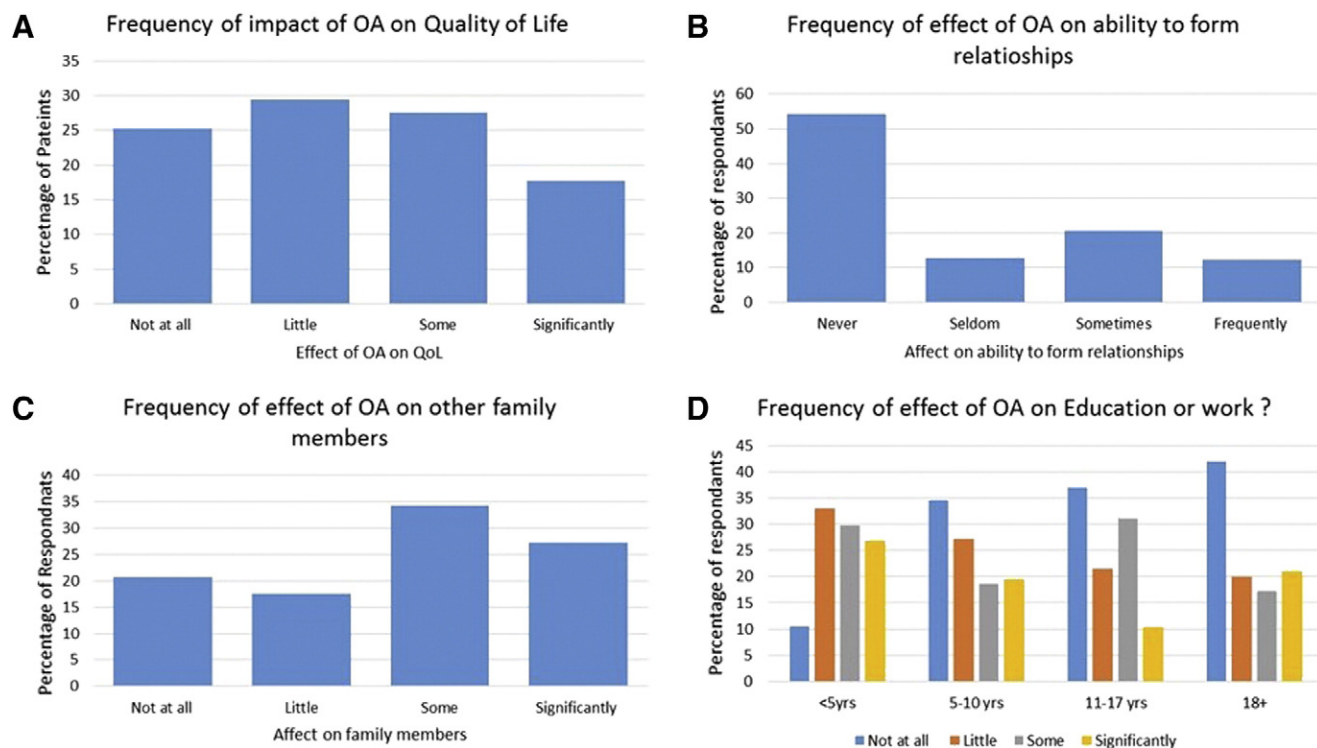


Fig. 8. Quality of life. Frequency of impact of OA on (A) quality of life, (B) other family members, (C) ability to form relationships and (D) education or work.

pure OA in the adult group may account for the increased median length of stay and number of operation required.

With increasing clinical and research interest in the long term follow-up of OA patients, reported outcomes relating to symptomatology and quality of life in such a large patient cohort are important additions to the literature. Dysphagia and reflux are well reported in the short and long term after OA repair and we see a high reported incidence of food 'getting stuck', a surrogate patient reported outcome for dysphagia, in agreement with published literature [8] [16]. Previous studies have reported reflux symptoms in nearly 50% of children younger than 5 years with a decreasing frequency with age. We however found a higher reported incidence of reflux in children younger than 5 years (65%) and this did not decrease significantly with age with 60% of adults reporting symptoms. Meta-analysis reports a lower rate (40%, range 18–64%) of reflux symptoms but recognizes the variation in the definition of GOR across studies analyzed. We are also limited in our analysis by the lack of definition of our patient reported outcomes but our findings add to the increasing body of literature highlighting long term morbidity in adult OA patients. Use of reflux medication was limited in our adult cohort (30%), despite the high reported incidence of symptoms (60%) and is similar to that reported [19]. Schneider et al. [19] went on to endoscope a cohort of adult OA patients and concerningly found that 67% of patients had histological evidence of esophagitis and 43% Barrett's esophagus. Several other studies report an increased risk of Barrett's [9,20] in OA and while a definitive increased risk of esophageal cancer in OA patients has yet to be proven [8,21] the frequency of reflux symptoms in this patient group and histological metaplastic changes mean that more attention needs to be paid to the consideration of surveillance endoscopy and use of antireflux medication in the adult aged OA population.

Respiratory symptoms are another important morbidity following OA repair, with 38% of all patients and 39% of adult patients reporting sometimes or often suffering from respiratory symptoms. Respiratory infections however do appear to be less frequent in the older patients with nearly 30% of those younger than 5 years having >3 chest infections per year, falling to 8% and 13% in the older age groups. Connor

et al. [8] suggested an overall prevalence of 24% for respiratory tract infections but with a range of 9.5–51%. It is difficult to compare accurately the data from our study and to calculate the prevalence but there is a similar message that while respiratory tract infections may decrease in frequency with age a third of patients with OA report ongoing respiratory symptoms through school years and adulthood.

The ongoing gastroenterological and respiratory morbidity in OA patients leads to the question of who provides care for this patient group. While there is international variation in the way care is provided to these patients what is clear is that while only 5–10% of children younger than 10 years have no documented care provider (which remains concerning), this increases to 49% in the adults. Although it is possible that some adult patients have no medical input because they are asymptomatic and feel that they do not 'need' regular medical input, the high incidence of reflux and respiratory symptoms seen in this and other studies may suggest that more patients should have a regular care provider with experience in OA/TOF, and that the higher number of adults not under regular care may reflect the relatively recent emphasis that is being put on transitional care and long term outcomes. This is an area of great concern to many patients.

As expected in our study there is a decrease with age in the number of patients looked after by pediatric surgeons. This may be a result of differences in practice across the countries in the study, where the longer term care of OA is led by gastroenterologists as opposed to surgeons or that if children are well they may be referred back to primary care. Adult patients appear to be discharged or transitioned to the gastroenterologist who provide the care to 28% of those older than 18 years in our study. How transition is managed in the future is an important issue for OA care and other chronic pediatric conditions, as the population of long term survivors continues to increase. International studies such as this alongside work from Australia [17], Finland [9,10] Holland [22,23] and more recently the USA [24] are key in highlighting the need for careful planning of patient centered transitional and adult care. This may include the need for some stratification of transition and follow-up. Our results demonstrate a wide spectrum of morbidity experienced by OA patients, many may therefore need minimal long

term follow-up while others with more significant morbidity will require regular tertiary level or hospital follow-up. Early characterization of patients 'at risk' of long term problems from more detailed future studies may help streamline the transition process.

Forward thinking centers are already looking at ways to improve the transitional care process with German centers offering education sessions for OA patients coming up to transition [25] and there is an increasing research effort examining what improves transition across all conditions [26].

Quality of life in OA patients is an outcome that had limited attention from surgeons in the past but is of increasing interest. These data show that 17% of patients/parents report a 'significantly' affected QoL and demonstrate more attention needs to be paid in this area—especially as 21% and 27% also report 'significant' effects on education or work and other family members respectively. QoL in OA has been reported as being 'comparable' with the normal population by the German support group (KEKS) [15]. They did however also find that health related quality of life (HRQoL) in adults is negatively affected by disease specific symptoms such as reflux and respiratory symptoms, a result also seen in other QoL studies [27,28]. It is difficult to make direct comparisons between this and other QoL studies, as this study used patient-led questionnaires whereas other studies have used validated QoL tools. Nevertheless, the parent/patient led description of impaired QoL, and the finding that nearly 90% of parents of children younger than 5 years reporting an effect on their child's education demands attention.

Comparison of our results to that of the KEKS study is useful as both studies examine outcomes of patients identified through patient support groups. However the use of support groups for recruitment may bias results and make comparison to the wider OA population difficult as their membership may be made up of those who require more 'support' and have a higher severity of disease. The variability in recruitment in different counties probably represents the activity and membership of the different national support organizations of EAT as opposed to proportional size of OA populations in each country. Therefore comparison of outcomes between countries is not possible and generalization of results across countries must be done with caution.

The questionnaire used in this study was specifically patient designed and easy to understand and complete and while this maximized recruitment it did lead to data being limited in detail for example of comorbidities and specific surgical detail. As a pilot study however its strengths lie in the numbers of participants recruited across more than 20 countries and firmly establishing the scale and types of morbidity seen across all ages of the OA population.

Another important message from this work is that of collaboration, which has been the key to this project as the patient support groups of Europe worked together toward a common goal. In the literature to date collaboration is limited in OA research as centers have tended to publish their own limited series. More recently larger national and international collaborations have led to more representative studies and with projects such as the European Reference Network of Rare Inherited and Congenital Anomalies (ERNICA) being developed it is hoped that clinicians and research groups can learn from the example set by this patient led project and achieve better quality studies and outcomes through wider collaborative work. The EAT study is testimony to the commitment and drive of the OA patient community and support groups to work together and produce clinically relevant and effective research in attempts to improve clinical care. Now that the EAT 1 study has given a broad baseline of the scale of gastroenterological, respiratory and QoL morbidity seen in OA patients, a more detailed and focused study of a smaller cohort of these patients is being designed.

Acknowledgments

This collaborative work has involved many different people from the design of the study to data collection and subsequent analysis.

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