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Nutritional Status in Adolescents with Esophageal Atresia

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Objectives To examine factors that may affect nutritional status in adolescents with esophageal atresia.

Study design Anthropometric measurements, blood samples, pH measuring, mapping of dysphagia with a modified Easting Assessment Test questionnaire, 4-day dietary record, and a semistructured interview about eating habits and nutrition counseling were performed in a cross-sectional cohort of adolescents with esophageal atresia.

Results Out of 102 eligible patients, 68 (67%) participated. The median height-for-age Z score was −0.6 (−4.6 to 1.8). Ten (15%) were classified as stunted (height-for-age Z score <−2). Fourteen (21%) were overweight. More than two-thirds reported symptoms of dysphagia (Easting Assessment Test score ≥3) and avoided specific foods. Forty-eight (71%) completed the 4-day dietary record, which showed daily intake of energy below age-appropriate recommendations. One-third reported an energy intake below their estimated basal metabolic rate. Only 24% had received counselling from a dietitian.

Conclusions Adolescents with a history of esophageal atresia have growth below reference values and energy intake below recommendations. Energy intake and its relation to stunting needs to be further studied in patients with esophageal atresia. *(J Pediatr 2020;218:130-7)*.

Esophageal atresia is a rare congenital malformation with major therapeutic challenges and consequences for feeding both in infancy and childhood.[1-5] Known underlying mechanisms for the feeding difficulties include dysphagia expressed through swallowing disorders and esophageal dysmotility, and gastroesophageal reflux disease (GERD).[6-9]

Feeding difficulties in childhood, long-term dysphagia, and gastrointestinal problems may have consequences for the nutritional status and growth potential later in life.[3,7,8,10] The growth of children with esophageal atresia is reported to be below the reference norm during the first years of life.[11,12] Menzies et al have presented prevalence of growth deficiencies in children with esophageal atresia (median age, 3.7 years) with wasting in 18% and stunting in 9%, whereas Vergouwe et al showed that growth was below the reference norm during the first years of life, but had normalized at age 12 years.[11,12]

There are few studies on growth into adolescence and adulthood. Dietary assessments in terms of intake of energy and micronutrients in patients with esophageal atresia are also lacking. The primary aim of the present study was to describe nutrition status including anthropometry, dietary intake, and micronutrient status in adolescents with esophageal atresia.

Methods

Between January 1996 and December 2002, 125 neonates with esophageal atresia were admitted to a tertiary hospital in Norway for surgical repair (**Figure**). Of these, 109 were long-term survivors and eligible for this cross-sectional study performed in 2015-2017. Seven patients were excluded. Exclusion criteria were additional conditions associated with growth disorders (cerebral palsy, muscular dystrophy, Down syndrome) or intellectual disability leaving the esophageal atresia adolescent unable to answer questionnaires. Non-Norwegian speakers were also excluded. All participants signed an informed consent. The study was approved by the Regional Ethical Committee in Norway (reference number: 2014/1224).

Medical records were reviewed for the type of esophageal atresia according to the Gross classification, neonatal data, associated anomalies, length of postnatal hospital stay, and esophageal dilatations for stenosis.[13] The vertebral, anal atresia, cardiac, trachea, esophageal, renal, and limb defects syndrome was defined according to Solomon.[14]

At the follow-up visit, nutritional status was evaluated by anthropometric measurements, biochemical measures, and nutritional intake. The examination also included a semistructured interview regarding eating habits and nutrition counseling, pH-measuring, and assessments of dysphagia and symptoms of GERD.

BMI	Body mass index
BMR	Basal metabolic rate
EAT-10	Easting Assessment Test
GERD	Gastroesophageal reflux disease
HAZ	Height-for-age Z score

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Anthropometry

Participant's height and weight were measured by trained personnel using standardized measuring equipment. Sex- and age-adjusted Z scores were calculated for height (height-for-age Z scores [HAZ]) and body mass index (BMI) by using the Norwegian national reference standards and a growth analyzer calculation tool (*Vekstjournalen*).[15,16] According to the World Health Organization, stunting was defined as a HAZ of <−2. A BMI SD score of <−2.0 and >1.0 were used as cut-off points to classify wasting and overweight (including obesity), respectively.[17] Scoliosis was diagnosed by radiological assessment of the Cobb angle, and a Cobb angle of >40° was defined as severe scoliosis.[18]

Biochemical Measures

Nonfasting blood samples were drawn for evaluation of nutritional status and factors associated with growth. Reference values were based on reference ranges from Oslo University Hospital, except for iron status, where we used World Health Organization's cut-off values. Iron deficiency was defined by serum ferritin of <15 μg/L.[19] Vitamin D deficiency was considered to be present at 25-OH-vitamin D total of <50 nmol/L, whereas severe vitamin D deficiency was defined by a threshold of <25 nmol/L according to European guidelines.[20]

Nutritional Intake

For dietary assessment, participants were instructed by a dietitian to register the intake of all food and beverages over a 4-day period, preferably 3 weekdays and 1 weekend day as recommended.[21] Detailed information about dietary supplements was not mapped through the registration form. The dietary records were completed primary by the adolescents themselves, with help from parents when needed. They were asked to give detailed descriptions of the type of food and beverages, as well as the amount consumed. A validated booklet containing photographs of foods was used to estimate portion sizes.[22,23] Energy and micronutrient intakes were calculated using the software Kostholdsplanleggeren (Norwegian Directorate of Health and the Norwegian Food Safety Authority, Oslo, Norway), which was based on Norwegian food composition tables.[24,25] Energy and micronutrient intake (vitamins A, D, C, thiamin, riboflavin, folate, calcium, and iron) was compared with national recommendations.[26] Micronutrient intake was also expressed as nutrient density of the diet (intake per unit of energy), and by comparison with average requirements.[27]

Participants who completely failed to complete dietary records diaries were excluded from the analysis of nutrient intake. To evaluate participants registered energy intake, we compared estimated intake with calculated individual basal metabolic rates (BMR). The BMR is the amount of energy needed for maintaining vital processes of the body, not including activity and food processing. We used the Scofield equation, which allows us to estimate the BMR based on sex, age, and weight.[28]

Semistructured Interview regarding Eating Habits and Nutrition Counselling

Information about eating habits, use of dietary supplements, and previous counseling by a dietitian was obtained by a questionnaire designed by the dietitian for this study.

Dysphagia

To identify dysphagia, we used a questionnaire slightly modified from the validated tool Easting Assessment Test (EAT-10). The EAT-10 is a self-report dysphagia assessment tool where patients answer if, and to what extent, they experience swallowing difficulties.[29,30] The original EAT-10 consists of 10 questions, where each question is scored from 0 (no problem) to 4 (severe problem). A total score of ≥3 is deemed to be abnormal.[31] In the modified EAT-10 used in this study, 7 of the questions were translated to Norwegian without any adjustments. The last 3 questions were slightly adapted for this specific patient group to obtain information regarding fluid intake during meals, time spent on meals, and whether the adolescent tended to avoid eating together with friends because of his or her swallowing difficulties. The questionnaire was self-administrated and completed in <2 minutes.

pH Testing

The methods for esophageal pH testing followed standard procedures where all antacid medication was stopped 7 days before the planned examination. After probe insertion to the ventricle, the probe was withdrawn with the help of fluorescence until the tip was located above 2 vertebral bodies above the diaphragm. This placement was to make sure the probe was above the diaphragm and above the potentially withdrawn z-line, which may be the case in some patients with esophageal atresia. The following day, all patients were examined with upper endoscopy. Acid exposure to esophagus was considered normal with a reflux index of <3%, intermediate between 3% and 7%, and abnormal at >7%.[32]

GERD

GERD was registered if the participants had troublesome reflux symptoms (regurgitation, vomiting, throat/retrosternal pain), if they used antacid medication on a regular basis, or if they previously had had antireflux surgery performed.

Statistical Analyses

Data were extracted into an electronic spreadsheet and statistical analyses were performed using SPSS version 25.0 (SPSS Ince, Chicago, Illinois). Results are presented as median (range, percentiles) or means with SD. Differences between 2 groups were analyzed using non parametric tests (Mann-Whitney *U* test) and parametric tests (Student *t* test). The Spearman rank correlation coefficient was used to discover the strength of a link between 2 sets of data. Statistical significance was accepted if the *P* value was <.05.

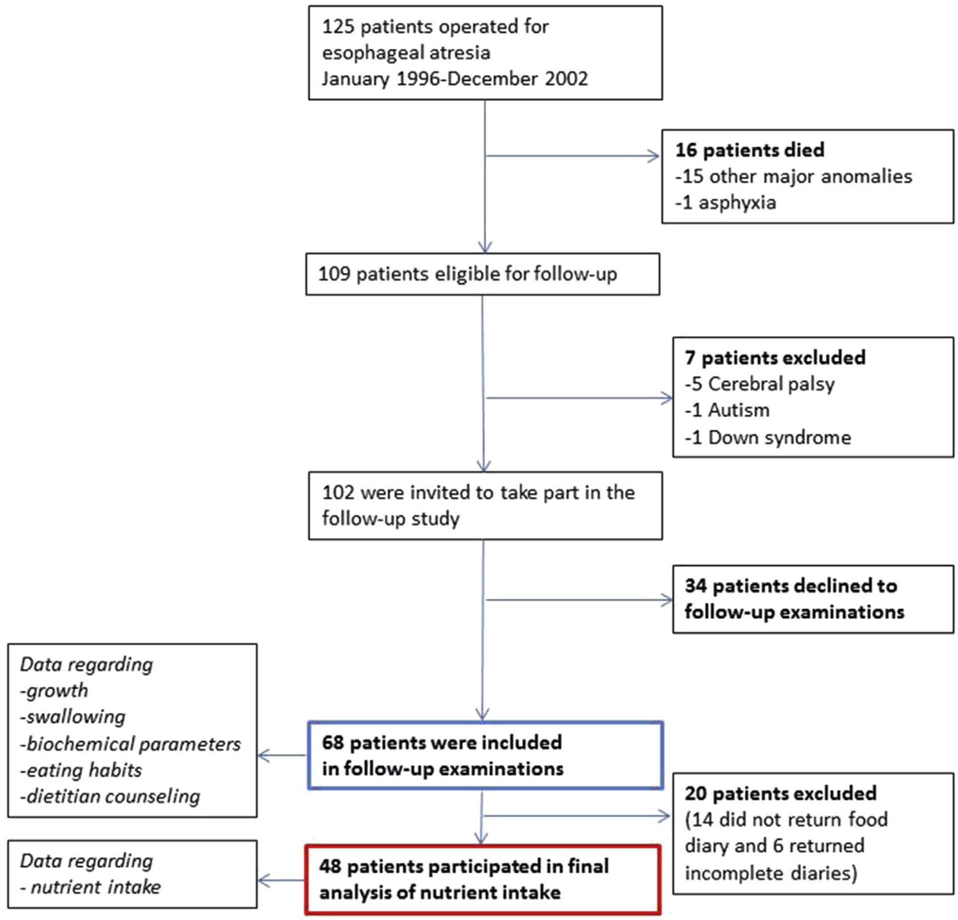

Figure. Study population.

Results

Of the 102 adolescents invited to participate in the study (**Figure**), 34 did not respond and 68 participated yielding a total response of 67%. There were 28 females and 40 males, and the median age at follow-up was 16 years (range, 13-20 years); 50 patients were 14-17 years of age and 18 were >18 years of age. Of the total of 68 participants, 20 were excluded from analysis of nutrient intake because 14 did not return food diaries and 6 returned incomplete diaries (**Figure**).

There were no significant differences between patients with esophageal atresia who participated and this who did not participate in the study with respect to sex, gestational age, type of esophageal atresia, or associated anomalies (data not shown). Characteristics of the participants are presented in **Table I**. According to the Gross classification, esophageal atresia type C occurred in 58 patients (85%), and among the remaining patients 3 (4.5%) were type A, 4 (6.0%) type D, and 3 (4.5%) type E. Data about growth and dietary intake were analyzed in age groups according to growth references and recommended dietary intakes (14-17 years and >18 years).

Table I. Characteristics of 68 adolescents with esophageal atresia (n = 68)

Characteristics	Values
Birth weight, grams	2810 (1380-4570)
Esophageal atresia with distal TEF	58 (85)
No. of esophageal dilatations at <12 months of age	0 (0-25)
Total number of dilatations during lifetime	0 (0-108)
Congenital heart disease	17 (25)
VACTERL association	13 (19)
Anorectal malformation	8 (12)
Diaphragmatic hernia	2 (3)
Length of postnatal hospital stay in days	22 (8 to 264)
Nissen fundoplication	9 (13)
Gastrostomy (previous)	12 (18)
Characteristics at follow-up	
HAZ	−0.6 (−4.6 to 1.8)
BMI Z score	−0.03 (−3.9 to 3.1)
EAT-10 score ≥3	47 (69)
RI by 2- hour pH-metry*	
Normal (RI <3%)	39 (66)
Intermediate (RI 3%-7%)	13 (22)
Abnormal (RI >7%)	7 (12)

RI, reflux index; *SDS*, SD score; *TEF*, tracheoesophageal fistula (classified as Gross C); *VACTERL*, vertebral, anal atresia, cardiac, trachea, esophageal, renal, and limb defects.
Values are median (range) or number (%).
*Of 68 participants, 59 performed 24 hours pH-metry at follow-up.

Anthropometry

Height and BMI Z scores at follow-up are shown in Table I. Ten adolescents (15%) were defined as stunted, including 3 with severe scoliosis. Exclusion of the participants with scoliosis did not change the median HAZ −0.6 (range, −3.3 to 1.8) without scoliosis and −0.6 (range, −4.6 to 1.8) in the total group. Adolescents with stunting had significantly lower birth weight (P = .015), longer postnatal hospital stay (P < .001), a greater number of esophageal dilatations before 12 months of age (P < .001), a greater total number of esophageal dilatations (P = .002), a history of gastrostomy feeding (P < .001), and more dysphagia based on results from the EAT-10 (P = .026). Height Z score in males was significantly higher in the youngest (14-17 years) compared with the oldest (>18 years; P = .004). In females, there were no differences in HAZ between the age groups (P = .941). Neither height nor weight were correlated with the presence of associated anomalies.

Only 2 adolescents (3%) were categorized as wasted (BMI Z score of <−2), whereas 14 (21%; 7 females and 7 males) were overweight (BMI Z score of >1). The HAZ among the overweight adolescents was not different from the rest of the group.

Biochemical Measures

Median values for biochemical measures are given in Table II. Most of the micronutrient measures were within normal range, except for vitamin D, folate, and iron status. Thirty patients (44%) were diagnosed with vitamin D deficiency and 3 (4%) had severe vitamin D deficiency. We did not find any association between vitamin D deficiency and HAZ (P = .072). According to World Health Organization cut-off values, 7 participants (10%) had iron deficiency (ferritin of <15 μg/L), of which 18% were females and 5% were males. When evaluating all biochemical indicators of iron status (hemoglobin, ferritin, serum iron, transferrin, total iron-binding capacity, transferrin saturation, and transferrin receptor), 15% of the participants were considered to have insufficient iron status.

Nutritional Intake

Data on nutritional intake from the 48 adolescents who completed the dietary records are shown in Table III. None of the participants had a gastrostomy. Energy intake was 6.4 ± 1.5 megajoule (MJ)/day in females 14-17 years, 8.3 ± 3.0 MJ/day in females >18 years, 9.7 ± 3.0 MJ/day in males 14-17 years, and 10.1 ± 3.0 MJ/day in males >18 years. Energy intake was lower than the age-appropriate recommendations for females and males in both age groups, and especially low for the youngest females (58% of recommended intake) (Table III). The energy intake in adolescents with cardiac malformations was not different from the rest of the group. There were no correlations between energy intake and WAZ (r = 0.01), HAZ (r = 0.20), or BMI Z score (r = 0.20). The median ratio energy intake/estimated BMR for all participants was 1.3 (range, 0.8-2.4), and 15 (31%) had an energy intake below their estimated BMR.

On average, macronutrients (proteins, fats, and carbohydrates) contributed to total energy intake in the following way: 18% from protein, 37% from fat, and 46% from carbohydrates. Diet composition of macronutrients was in accordance with recommendations, except for saturated fat and sugar.[26] One adolescent (2%) met the recommended intake of saturated fatty acids (<10% of total energy intake) and 29 (60%) for sugar (<10% of total energy intake).

Nearly one-half of the adolescents (47%) used dietary supplements, but no one reported to consume any kind of energy-containing supplements. Because of insufficient information about brand names and dosages, dietary

Table II. Levels of serum micronutrients in relation to sex and age-group and proportions deficient adolescents in total group

	Male		Female		
Biochemical parameters	14-17 years (n = 31)	>18 years (n = 9)	14-17 years (n = 19)	>18 years (n = 9)	Reference values*
Hemoglobin (g/dL)	14.5 (13.2-16.7)	15.3 (13.2-16.4)	15 (5-30)	13 (8-28)	12-15†/13-17‡
Ferritin (μg/L)	47 (14-121)	94 (8-129)	22 (5-156)	28 (17-75)	15-150†/200‡
Iron (μmol/L)	18 (6-28)	13 (4-27)	15 (5-30)	13 (8-28)	9-22
Transferrin (g/L)	2.9 (2.1-3.7)	2.9 (1.3-3.3)	3.0 (2.3-4.0)	2.8 (2.2-4.1)	2.0-3.3
Transferrin saturation (%)	0.25 (0.10-0.47)	0.21 (0.05-0.37)	0.22 (0.01-0.39)	0.16 (0.12-0.38)	0.15-0.57
Transferrin receptor (mg/L)	3.3 (2.0-5.2)	3.7 (1.9-5.9)	3.0 (1.6-9.8)	2.4 (1.7-3.4)	1.9-4.4†/2.2-5.0‡
Cholesterol (mmol/L)	3.7 (2.3-4.6)	3.8 (2.9-4.6)	4.2 (3.3-5.6)	4.3 (3.5-6.1)	3.0-5.5
TSH (mIE/L)	1.9 (0.7-4.5)	1.4 (1.1-3.3)	1.7 (0.7-3.5)	1.9 (0.5-2.6)	0.5-4.4
FT4 (pmol/L)	16 (11-21)	16 (14-25)	16 (13-111)	16 (13-20)	12-22
Vitamin B_{12} (pmol/L)	357 (187-635)	360 (281-535)	393 (169.719)	325 (186-639)	150-650
Folate, B_9 (nmol/L)	12 (6-36)	18 (5-31)	19 (6-30)	15 (8-38)	>10
Zinc (μmol/L)	14 (11-18)	15 (12-16)	13 (11-22)	13 (13-15)	10-17
Vitamin A (μmol/L)	1.6 (1.0-2.6)	1.7 (0.8-2.9)	1.6 (0.9-2.5)	1.7 (1.6-2.7)	1.2-3.6§
P-25-OH-vitamin D total (nmol/L)	54 (21-96)	49 (24-68)	45 (21-84)	50 (27-133)	>50
Vitamin E (μmol/L)	21 (13-27)	25 (17-26)	24 (19-33)	24 (15-39)	17-45§
Growth hormone, IGF-1 (nmol/L)	40 (26-85)	47 (20-70)	45 (27-76)	37 (19-75)	13-61

FT4, fraction of thyroxine; *IGF1*, insulin-like growth factor 1; *TSH*, thyroid-stimulating hormone.
Values are median (minimum-maximum), or range.
*Oslo University Hospital reference values. World Health Organization reference values were used for hemoglobin and ferritin.
†Females.
‡Males.
§Lower values are expected in children.

Table III. Median daily energy and nutrient intakes (without supplements), by sex and age groups (n = 48)

	14-17 Years (n = 38)*		>18 Years (n = 10)†		Recommendations,‡ 14-17/>18 years
Nutrients	Median	25-75 Percentiles	Median	25-75 Percentiles	
Energy (MJ)					
Male	9.4	7.5-10.8	8.8	8.4-13.2	11.8/11.7§
Female	5.8	5.5-7.3	6.8	6.1-11.3	9.8/9.4§
Energy (kcal)					
Male	2250	1790-2580	2100	2000-3160	2820/2800
Female	1390	1320-1750	1630	1460-2700	2340/2250
Total fat (% TE)					25-40
Male	38	33-41	40	39-50	
Female	33	31-36	33	32-41	
SFA (% TE)					<10
Male	15	13-18	18	16-19	
Female	13	11-15	14	13-17	
Total carbohydrates (% TE)					45-60
Male	45	40-50	43	37-45	
Female	48	46-50	51	43-53	
Total sugars (% TE)					<10
Male	9	5-12	8	7-10	
Female	9	6-12	11	7-17	
Total proteins (% TE)					10-20
Male	17	15-19	17	14-18	
Female	18	17-21	16	14-17	
Fiber (g/MJ)					3 g/MJ
Male	1.7	1.5-2.1	1.9	1.4-2.1	
Female	2.2	1.8-2.5	2.1	1.5-3.5	
Vitamin A (μg Retinol equivalent)					
Male	590	426-1142	858	634-1049	900
Female	489	287-621	615	499-2268	700
Vitamin D (μg)					10
Male	6.7	2.9-8.2	7.9	7.2-12.8	
Female	4.2	1.7-5.3	4.1	2.8-5.9	
Vitamin C (mg)					75
Male	52	33-101	41	23-115	
Female	44	16-85	57	49-102	
Thiamin (mg)					
Male	1.4	1.2-1.9	1.6	1.3-1.9	1.4
Female	0.9	0.8-1.6	1.1	1.0-2.3	1.2/1.1
Riboflavin (mg)					
Male	1.8	1.1-2.3	1.7	1.2-2.0	1.7/1.6
Female	1.0	1.0-1.2	1.2	1.0-2.3	1.4/1.3
Folate (μg)					
Male	229	194-271	223	155-305	300
Female	141	120-221	219	173-297	400
Calcium (mg)					900
Male	1011	655-1391	1396	899-1754	
Female	780	499-1015	675	564-1244	
Iron (mg)					
Male	9.1	6.8-11.7	7.9	7.0-11.8	11/9
Female	5.7	4.8-8.1	8.9	6.7-12.0	15

SFA, saturated fatty acids; *TE*, total energy.
*Includes 25 males and 13 females.
†Includes 4 males and 6 females.
‡The Norwegian Directorate of Health, Anbefalinger om kosthold, ernæring og fysisk aktivitet (2014).
§Medium physical activity.

supplements were not included when analyzing the participants' micronutrient intake.

Based on the 4-day dietary records median intake of all micronutrients, except thiamin, riboflavin, and calcium intake in males, was found to be below recommendations (Table III). Intakes of micronutrients below average recommendations were most commonly seen for vitamin C, vitamin D, and iron. Nutrient density (intake per unit of energy) was, compared with the recommendations, sufficient for most nutrients, but inadequate for iron, folate, and vitamin D.[27]

Nutrition Counseling

Sixteen patients (24%) had received nutrition counseling from a dietitian in early childhood. The proportion of adolescents with stunting was higher among those who had visited a dietitian ($P = .003$), but otherwise there were no differences in any aspect of nutrition and health between those who had visited a dietitian and those who had not.

Eating Habits

According to the semistructured interview 48 participants (71%) avoided specific foods. Twenty-four (50%) reported food texture as the reason for avoidance, and especially meat and meat-related products were reported to be challenging. Fourteen (21%) and 19 (28%) reported food dislikes as the reason for avoidance of fruits and vegetables, respectively. Ten (15%) reported prolonged mealtime duration (>30 minutes) for dinner, but to a lesser extent for other meals.

Dysphagia and Gastroesophageal Reflux

The prevalence of self-reported swallowing difficulties and GERD are shown in **Table I**. Twenty-four females (86%) and 23 males (59%) had current swallowing difficulties according to the modified EAT-10 questionnaire. The most frequently reported challenges were food getting stuck during swallowing (65%), need for extra liquids to manage swallowing (59%), difficulties with swallowing solid foods (56%), and pills (47%). None of the participants reported pain to be associated to swallowing. Swallowing difficulties expressed as total EAT-10 score was not significantly correlated with total energy intake (P = .81), energy intake related to body weight (MJ/kg, P = .98), or ratio of energy intake to estimated BMR (P = .69). Nine adolescents (13%) did not want to undergo pH-metry. Thirty-nine adolescents (66%) had normal reflux index, 13 (22%) had intermediate, and 7 (12%) had a reflux index of >7%. Fourteen (14/44 [32%]) with symptoms corresponding with GERD (regurgitation, vomiting, throat and retrosternal pain) used medical antireflux therapy on a regular basis. We did not find any statistical correlation between pH-metry and nutrient intake (MJ/kg; P = .65) nor between pH-metry and HAZ (P = .06).

Discussion

We assessed the nutritional intake of adolescents with a history of esophageal atresia. We found that calculated energy intake was below age-appropriate recommendations, and below estimated BMR in one-third of the participants. The estimated intake of micronutrients based on dietary records, not including dietary supplements, also revealed low intake, especially for vitamin D and iron.

In the current study, the adolescents' energy intake was lower than national recommendations. However, national recommendations are not the same as reference values on dietary intake. Comparing our results with data from dietary surveys, the intake in patients with esophageal atresia does not seem so discordant. In a Norwegian dietary survey 13-year-olds reported a mean energy intake of 7.4 ± 2.6 MJ/day for females and 8.6 ± 2.7 MJ/day for males, compared with mean values of 6.4 ± 1.5 MJ/day and 9.7 ± 3.0 MJ/day in our study.[33] Another national study summarized mean energy intake in adults 18-29 years to be 8.1 ± 2.5 MJ/day for females and 12.8 ± 4.0 MJ/day for males, compared with a mean intake of 8.3 ± 3.0 MJ and 10.1 ± 3.0 MJ for the oldest females and males in our study.[34] This means that findings in our study are consistent with research showing adolescents' energy intake to be lower than national recommendations.

The low ratio energy intake/estimated BMR (median, 1.3) found in our study indicates an energy intake not compatible with normal physical activity. Unfortunately, physical activity was not registered in this study. According to Nordic nutrient recommendations 2012 a ratio of total energy expenditure/estimated BMR of <1.66 refers to a low physical activity in adolescents 10-18 years.[27] Thus, a possible explanation for the low ratio may be that adolescents with esophageal atresia are less active than healthy adolescents. Another possible explanation may be underreporting of energy intake. Challenges in evaluation and validation of self-reported energy intake are well-known, and there is no reason to assume that adolescents with esophageal atresia are different from others adolescents in respect of this.[35,36]

The estimated intake of vitamin D and iron was below recommendations. Low vitamin D intake corresponds with results from national dietary surveys, and vitamin D deficiency has also been reported to be evident throughout the European population.[35-37] Norwegian surveys are scarce, but a recent study from the northern part of Norway reported 60.2% of healthy adolescents (15-18 years old) to be vitamin D deficient.[35] Iron deficiency in adolescents with esophageal atresia also seems to be comparable with healthy, Norwegian adolescents. Handeland et al presented in a recent study that 9.7% of participants (n = 478; mean age, 14.6 years) had iron deficiency (a ferritin of <15 μg/L).[37] Thus, based on the fact that frequencies of deficiencies in vitamin D and iron were similar to those observed in the general population, it does not seem appropriate to attribute the observed deficiencies only to the disease under study.

Longitudinal data on growth in patients with esophageal atresia are few.[10] A recent study demonstrated restricted growth in childhood, with height improvement in early adolescence, and Chetcuti and Phelan have reported normal height in adults with esophageal atresia.[11,38] In contrast, the rate of reduced growth in our study is in accordance with earlier reports with Okuyama et al also reporting adults with esophageal atresia to be short.[38-40] The HAZ was lower in the oldest males compared with the younger group in our study. We do not find any obvious reasons to assume that the differences between age groups are caused by changes in surgical or rehabilitations factors in recent years, but advances in neonatal medical care may have had an impact.

A majority of the adolescents in our study had swallowing difficulties and reported food textures as a factor influencing food choice. Previous studies have identified dysphagia and feeding behaviors as main causes of nutritional problems in patients with esophageal atresia.[1,6,12] We found a relation between stunting and dysphagia, but surprisingly there were no associations between self-reported dysphagia and nutrient intake. The reason for this discrepancy may be that the dysphagia have had influence on growth in childhood. During life, the adolescent may have developed compensatory eating behaviors like limiting bolus size, slowing the rate of eating, and diet modification enabling the patient to normalize nutrient intake.

In this study, we focused on nutritional intake, but there may of course be additional reasons for both dysphagia and impaired growth in patients with esophageal atresia. Among these comorbidities, esophageal dysmotility, GERD, and esophagitis are the most relevant in patients with esophageal atresia. Even though it is possible that stunting is caused by nutritional conditions in early childhood, other unidentified factors related to the primary diagnosis may also contribute to deviant development in children with esophageal atresia. In our rather small series of patients, we found no significant relations to any of these conditions, but these factors may be relevant in larger groups of patients.

Despite challenges with growth, feeding, and nutrient intake, only 24% of the participants had ever consulted a dietitian, which is in accordance with other studies in patients with esophageal atresia. Menzies et al reported that 75% of patients with esophageal atresia were reviewed by a dietitian and/or speech therapist at their initial appointment after discharge, but long-term evaluation is lacking.[12] The pubertal growth phase is considered an additional window of opportunity for nutritional intervention, which support a need for multidisciplinary follow-up, including with a dietitian, to prevent and treat feeding difficulties and nutritional challenges.[7,41-43] The strengths of the present study include the long-term perspective in adolescents with esophageal atresia and the relatively high number of patients. However, information regarding growth development and nutrient intake from early childhood is lacking. Another limitation of this study is the lack of knowledge about parental height, and the participants' physical activity level to evaluate and validate their reported energy intake. The population in our study was heterogeneous from 13 to 20 years of age, including different types of malformations and different lasting comorbidities, but the prevalence of different types of malformations and associated anomalies was comparable with previous studies on patients with esophageal atresia.[40,44] Underreporting of energy intake was potentially a great challenge of our study.

Patients with esophageal atresia seem to have persistent problems with dysphagia and growth below reference values. Based on dietary assessments, we suspect an inadequate intake of energy in adolescents with esophageal atresia. Patients with esophageal atresia should be followed into adolescence by a dietitian, and growth and nutritional intake should be recorded in larger studies. ■

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References

1. Gottrand M, Michaud L, Sfeir R, Gottrand F. Motility, digestive and nutritional problems in Esophageal Atresia. Paediatr Respir Rev 2016;19:28-33.
2. Blakstad EW, Moltu SJ, Nakstad B, Veierod MB, Strommen K, Juliusson PB, et al. Enhanced nutrition improves growth and increases blood adiponectin concentrations in very low birth weight infants. Food Nutr Res 2016;60:33171.
3. Ramsay M, Birnbaum R. Feeding difficulties in children with esophageal atresia: treatment by a multidisciplinary team. Dis Esophagus 2013;26:410-2.
4. Conforti A, Valfre L, Falbo M, Bagolan P, Cerchiari A. Feeding and swallowing disorders in esophageal atresia patients: a review of a critical issue. Eur J Pediatr Surg 2015;25:318-25.
5. Mahoney L, Rosen R. Feeding difficulties in children with esophageal atresia. Paediatr Respir Rev 2016;19:21-7.
6. Mahoney L, Rosen R. Feeding problems and their underlying mechanisms in the esophageal atresia-tracheoesophageal fistula patient. Front Pediatr 2017;5:127.
7. Krishnan U, Mousa H, Dall'Oglio L, Homaira N, Rosen R, Faure C, et al. ESPGHAN-NASPGHAN guidelines for the evaluation and treatment of gastrointestinal and nutritional complications in children with esophageal atresia-tracheoesophageal fistula. J Pediatr Gastroenterol Nutr 2016;63:550-70.
8. Ijsselstijn H, van Beelen NW, Wijnen RM. Esophageal atresia: long-term morbidities in adolescence and adulthood. Dis Esophagus 2013;26:417-21.
9. Friedmacher F, Kroneis B, Huber-Zeyringer A, Schober P, Till H, Sauer H, et al. Postoperative complications and functional outcome after esophageal atresia repair: results from longitudinal single-center follow-up. J Gastrointest Surg 2017;21:927-35.
10. Ijsselstijn HI, Gischler SJ, Toussaint L, Spoel M, Zijp MH, Tibboel D. Growth and development after oesophageal atresia surgery: need for long-term multidisciplinary follow-up. Paediatr Respir Rev 2016;19:34-8.
11. Vergouwe FW, Spoel M, van Beelen NW, Gischler SJ, Wijnen RM, van Rosmalen J, et al. Longitudinal evaluation of growth in oesophageal atresia patients up to 12 years. Arch Dis Childhood Fetal Neonatal Ed 2017;0:F1-6.
12. Menzies J, Hughes J, Leach S, Belessis Y, Krishnan U. Prevalence of malnutrition and feeding difficulties in children with esophageal atresia. J Pediatr Gastroenterol Nutr 2017;64:e100-5.
13. Gross R. The surgery of infancy and childhood. Philadelphia: W.B. Saunders; 1953.
14. Solomon BD. VACTERL/VATER association. Orphanet J Rare Dis 2011;6:56.
15. Juliusson PB, Roelants M, Eide GE, Moster D, Juul A, Hauspie R, et al. [Growth references for Norwegian children]. Tidsskr Nor Laegeforen 2009;129:281-6.
16. Turck D, Michaelsen KF, Shamir R, Braegger C, Campoy C, Colomb V, et al. World Health Organization 2006 child growth standards and 2007 growth reference charts: a discussion paper by the committee on Nutrition of the European Society for Pediatric Gastroenterology, Hepatology, and Nutrition. J Pediatr Gastroenterol Nutr 2013;57:258-64.
17. World Health Organization (WHO). Growth reference 5-19 years. 2007. www.who.int/growthref/who2007_bmi_for_age/en/. Accessed March 7, 2018.
18. Reamy BV, Slakey JB. Adolescent idiopathic scoliosis: review and current concepts. Am Fam Physician 2001;64:111-6.
19. UpToDate. Iron requirements and iron deficiency in adolescents. 2018. www.uptodate.com/contents/iron-requirements-and-iron-deficiency-in-adolescents?topicRef=7150&source=see_link. Accessed May 10, 2018.
20. Braegger C, Campoy C, Colomb V, Decsi T, Domellof M, Fewtrell M, et al. Vitamin D in the healthy European paediatric population. J Pediatr Gastroenterol Nutr 2013;56:692-701.
21. Thomas B. Manuals of dietetic practice. 3rd ed. Hoboken (NJ): Blackwell Publishing; 2001.
22. Lillegaard IT, Andersen LF. Validation of a pre-coded food diary with energy expenditure, comparison of under-reporters v. acceptable reporters. Br J Nutr 2005;94:998-1003.
23. Lillegaard IT, Overby NC, Andersen LF. Can children and adolescents use photographs of food to estimate portion sizes? Eur J Clin Nutr 2005;59:611-7.
24. Kostholdsplanleggeren, a diet tool from the Norwegian Directorate of Health and the Norwegian. Food Safety Authority: Norwegian Food

Safety Authority. 2018. www.kostholdsplanleggeren.no/. Accessed January 10, 2018.
25. The Norwegian Food Composition Table, Norwegian Food Safety Authority: Norwegian Food Safety Authority. 2018. www.matvaretabellen.no/. Accessed January 10, 2018.
26. Anbefalinger om kosthold, ernæring og fysisk aktivitet, Norwegian Directorate of Health. 2014. https://helsedirektoratet.no/publikasjoner/anbefalinger-om-kosthold-ernering-og-fysisk-aktivitet. Accessed April 7, 2018.
27. Norden. Nordic nutrition recommendations 2012. Copenhagen (Denmark): Nordic Council of Ministers; 2012.
28. Schofield WN. Predicting basal metabolic rate, new standards and review of previous work. Hum Nutr Clin Nutr 1985;39(Suppl 1):5-41.
29. Plowman EK, Tabor LC, Robison R, Gaziano J, Dion C, Watts SA, et al. Discriminant ability of the Eating Assessment Tool-10 to detect aspiration in individuals with amyotrophic lateral sclerosis. Neurogastroenterol Motil 2016;28:85-90.
30. Rofes L, Arreola V, Mukherjee R, Clave P. Sensitivity and specificity of the Eating Assessment Tool and the Volume-Viscosity Swallow Test for clinical evaluation of oropharyngeal dysphagia. Neurogastroenterol Motil 2014;26:1256-65.
31. Wilmskoetter J, Bonilha H, Hong I, Hazelwood RJ, Martin-Harris B, Velozo C. Construct validity of the Eating Assessment Tool (EAT-10). Disabil Rehabil 2019:549-59.
32. Vandenplas Y, Rudolph CD, Di Lorenzo C, Hassall E, Liptak G, Mazur L, et al. Pediatric gastroesophageal reflux clinical practice guidelines: joint recommendations of the North American Society for Pediatric Gastroenterology, Hepatology, and Nutrition (NASPGHAN) and the European Society for Pediatric Gastroenterology, Hepatology, and Nutrition (ESPGHAN). J Pediatr Gastroenterol Nutr 2009;49:498-547.
33. Ungkost 3. Landsomfattende kostholdsundersøkelse blant elever i 4. -og 8. klasse i Norge, 2015. Norwegian Institute of Public Health. University of Oslo. The Norwegian Directorate of Health. Oslo: The Norwegian Food Safety Authority; 2016
34. Norkost 3, En landsomfattende kostholdsundersøkelse blant menn og kvinner i Norge i alderen 18-70 år, 2010-2011. Oslo: The Norwegian Directorate of Health, University of Oslo, The Norwegian Food Safety Authority; 2012. Contract No.: IS-2000.
35. Cashman KD, Dowling KG, Skrabakova Z, Gonzalez-Gross M, Valtuena J, De Henauw S, et al. Vitamin D deficiency in Europe: pandemic? Am J Clin Nutr 2016;103:1033-44.
36. Oberg J, Jorde R, Almas B, Emaus N, Grimnes G. Vitamin D deficiency and lifestyle risk factors in a Norwegian adolescent population. Scand J Public Health 2014;42:593-602.
37. Handeland K, Kjellevold M, Wol Markhus M, et al. A Diet Score Assessing Norwegian Adolescents' Adherence to Dietary Recommendations-Development and Test-Retest Reproducibility of the Score. Nutrients 2016;8.
38. Chetcuti P, Phelan PD. Gastrointestinal morbidity and growth after repair of oesophageal atresia and tracheo-oesophageal fistula. Arch Dis Child 1993;68:163-6.
39. Okuyama H, Tazuke Y, Uenoa T, Yamanaka H, Takama Y, Saka R, et al. Long-term morbidity in adolescents and young adults with surgically treated esophageal atresia. Surg Today 2017;47:872-6.
40. Presse N, Taillefer J, Maynard S, Bouin M. Insufficient body weight of adults born with esophageal atresia. J Pediatr Gastroenterol Nutr 2016;62:469-73.
41. Prentice AM, Ward KA, Goldberg GR, Jarjou LM, Moore SE, Fulford AJ, et al. Critical windows for nutritional interventions against stunting. Am J Clin Nutr 2013;97:911-8.
42. IJsselstijn H, Gischler SJ, Toussaint L, Spoel M, Zijp MH, Tibboel D. Growth and development after oesophageal atresia surgery: need for long-term multidisciplinary follow-up. Paediatr Respir Rev 2016;19:34-8.
43. Beverly EA, Ritholz MD, Brooks KM, Hultgren BA, Lee Y, Abrahamson MJ, et al. A qualitative study of perceived responsibility and self-blame in type 2 diabetes: reflections of physicians and patients. J Gen Intern Med 2012;27:1180-7.
44. Sfeir R, Michaud L, Salleron J, Gottrand F. Epidemiology of esophageal atresia. Dis Esophagus 2013;26:354-5.