



## Mental health and psychosocial functioning in adolescents with esophageal atresia

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Received 21 May 2008; revised 6 September 2008; accepted 29 September 2008

### Key words:

Esophageal atresia;  
Mental health;  
Psychosocial functioning;  
Feeding difficulties;  
Dissociative symptoms;  
Chronic illness

### Abstract

**Purpose:** We examined mental health and psychosocial functioning in adolescents with esophageal atresia (EA) and searched for predictors of impaired outcome.

**Methods:** The study group comprised 21 adolescents with EA and 1 or both parents. A comparison group comprised 36 adolescents from the general population. Mental health, self-esteem, psychosocial functioning, and parental/family functioning were assessed by standardized questionnaires and semistructured interviews. Physical health was assessed by growth and clinical symptoms.

**Results:** Mental health and psychosocial functioning did not differ from the comparison group. Seven of 21 EA adolescents had special education. Dilatations of esophagus, birth weight, well-being, and maternal psychological distress were prognostic factors predicting mental health. Height, birth weight, well-being, dissociative symptoms, and family strain were prognostic factors predicting psychosocial functioning.

**Conclusion:** Adolescents with EA adjusted well, and mental health and psychosocial functioning did not differ from controls. Dilatations of esophagus and birth weight were significant predictors of mental health and psychosocial functioning.

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Chronically ill children are at risk for mental health and psychosocial problems. Repeated hospital admissions and surgery in early childhood may have long-term effects on emotional and behavioral adjustment in adolescence [1-7]. Risk factors for psychosocial problems in chronically ill

adolescents include severity of the medical condition, sex, self-esteem, school functioning, and family strain [8-10].

Feeding difficulties and respiratory problems are frequent during infancy and early childhood of esophageal atresia (EA) patients, whereas dysphagia, food impaction, and gastroesophageal reflux (GER) are common later [11-13]. In a survey assessing mental health in infants with EA, psychiatric disturbances; mostly trauma related, were identified by 1 year of age [14]. More than 1 operation, mechanical ventilation beyond 1 day, mother-reported posttraumatic

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stress symptoms, and moderate/severe family strain were prognostic factors predicting the infants' mental health. Despite the challenges child and parents experience in the early years, most adult patients with EA seem to have a good quality of life [15,16]. There is a paucity of studies that have examined long-term psychological outcome of children with EA; and the results reporting psychosocial functioning are contradictory, ranging from comparable with those of healthy controls [11,15,17,18] to more emotional and learning problems [19,20]. Because these studies have few patients in each age group and different assessment procedures have been used, it is difficult to make any firm conclusions. No assessment of mental health and psychosocial functioning has been conducted in adolescents with EA.

Our aim was to assess mental health and psychosocial functioning in adolescents with EA compared with adolescents from the general population. An additional aim was to search for prognostic factors predicting mental health and psychosocial functioning in adolescents with EA.

## 1. Methods

### 1.1. Patients and controls

The sample consisted of all (34) eligible adolescents born in the period 1986 through 1990 with EA treated at Rikshospitalet University Hospital, Norway. The eligibility criteria excluded children with incomplete medical records ( $n = 1$ ) and children who had died ( $n = 9$ ; ie, of serious associated anomalies with respiratory failure, complications of extreme prematurity and major congenital heart disease, or sudden infant death syndrome). Two patients were not invited because of present serious medical condition unrelated to EA (cancer, anorectal anomaly). Of 22 eligible patients, 21 adolescents (96%) and 1 or both parents participated in the study. From the medical records, the patients' physical condition was classified according to the Montreal prognostic classification [21].

Comparison group for the psychiatric and psychosocial assessments was a subsample drawn from a Norwegian epidemiologic study of mental health. The study used a stratified random sampling procedure and a 2-stage design [22]. In the first stage, screening was carried out by means of a standardized questionnaire, the Child Behavior Checklist (CBCL) [23]. In the second stage, screen-positive (CBCL T score  $\geq 60$  according to the American norm) adolescents 13 to 15 years of age and a random sample of screen-negative (CBCL T score  $< 60$ ) adolescents in this age group were invited to participate in a more detailed assessment. A T score of 60 corresponds to the 95th percentile of the frequency distribution for this age group in the Norwegian population. Thirty-six adolescents (19 boys and 17 girls; mean age, 14 years; range, 13-15 years) and their parents participated and are referred to as the *control group*. Six adolescents had

been hospitalized for various medical conditions (eg, congenital dislocation of the hip, asthma, otitis media, or accidents) during their childhood. Two adolescents had chronic illness/eczema and epilepsy. None had neonatal hospital admissions or operations.

#### 1.1.1. Procedures

The EA adolescents and 1 or both parents visited the hospital for follow-up assessment. A pediatric surgeon different from the surgeon operating the patients in the neonatal period and a specialized nurse performed the standardized physical examination and assessments. A child and adolescent psychiatrist performed the mental health and psychosocial assessments, all standardized and international well-known instruments with well-established reliability and validity.

### 1.2. Study instruments

#### 1.2.1. Mental health

We used standardized questionnaires from the Achenbach System of Empirically Based Assessment, including competence scales, problems scales, and a trauma-related dissociative symptoms scale [24-29]. The adolescents completed the Youth Self-Report (YSR/11-18 version 1991) [30]; and the mothers, the CBCL for ages 4 to 18 years (CBCL/4-18 version 1991) [23]. The CBCL and YSR are internationally the most frequently used standardized questionnaires for assessing competencies and behavioral and emotional problems in children and adolescents. Higher scores imply more emotional or behavioral symptoms/problems and better academic or social competencies. Raw scores of subscales were used in the analysis. Multi-culture studies document support for the taxonomic generalizability of the CBCL/YSR syndromes across diverse societies, including Norway [25,26].

Psychopathology was assessed by a semistructured diagnostic interview, Schedule of Affective Disorders and Schizophrenia for School-Age Children, providing *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition* (DSM-IV), Axis I psychiatric diagnosis [31]. The interview was videotaped. The reliability and validity of the instrument are well established [31,32]. A semistructured interview based on the Parental Account of Children's Symptoms [33] with 1 parent was performed to obtain information on the adolescents' behavior and emotional problems as seen by their parents. To test interrater reliability of the DSM-IV, classification in the present study, 10 videotaped interviews with the adolescents were chosen at random and scored independently by another experienced child psychiatrist. All 10 patients were evaluated similarly by the 2 raters. This correspond to a  $\kappa$  of 1.0.

#### 1.2.2. Psychosocial functioning

The overall psychosocial function assessment of the adolescents was based on information from both adolescent and parent interviews and rated in accordance with the

Children's Global Assessment Scale (CGAS) [34], with 100 representing excellent functioning in all areas and 0 implying severe malfunctioning. Interrater reliability, discriminant validity, and clinical usefulness of the CGAS are well documented [35,36]. Scores greater than 70 are designated as indicating normal function [34]. A second child psychiatrist scored blind CGAS on 10 randomly selected cases based on videotaped adolescent interviews. Intraclass correlation for CGAS in the present study was .77.

### 1.2.3. Self-esteem

Self-esteem was assessed by the Self-Perception Profile for Adolescents (SPPA) [37-39]. The SPPA was chosen because it has been used in other studies with similar populations of chronically ill adolescents [39]. The SPPA contains 7 subscales: scholastic competence, social acceptance, athletic competence, physical appearance, romantic appeal, close friends, and global self-worth. Each subscale contains 4 items, providing a mean score of perceived competence ranging from 1 (lowest level of self-esteem) to 4 (highest level of self-esteem).

### 1.2.4. Well-being

The adolescents' experience of subjective well-being was assessed by asking patients to indicate how well they felt on a 100-mm visual analogue scale (VAS) anchored by 2 extremes: "my life is extremely unpleasant" (0) and "my life feels very good" (100) [40].

### 1.2.5. Parental and family strain

The parental interview in the EA group was supplemented with questions covering past and present views of the esophageal malformation. Maternal psychological distress was assessed by the General Health Questionnaire, 30-item version (GHQ-30) [41,42]. The GHQ scores based on Likert score (item score 0-1-2-3) were calculated, with higher scores indicating increased distress level. Overall psychological distress was measured by total GHQ-30 sum Likert score (range, 0-90) [43,44].

A global assessment of chronic family difficulties [45,46] was based on information from parental interview (Parental Account of Children's Symptoms) including present family situation. Chronic family difficulties were scored on an interval scale from 0 to 6, where 0 implies none and 6 implies severe family strain or difficulties.

### 1.2.6. Physical health

Somatic data were recorded retrospectively from the medical report. Medical history and health problems in the previous 12 months with emphasis on gastrointestinal and respiratory symptoms were obtained from the patient and the parent(s). The frequency of dysphagia, esophageal food impaction, and GER symptoms was recorded. Dysphagia was judged based on careful history of frequency of swallowing problems and sensation of a food bolus becoming lodged in the esophagus and categorized as

daily, occasional, or no. *Symptoms of GER* were defined as retrosternal discomfort (heart burn) and categorized as more than once a week, occasional, or no. *Bronchitis* was defined as an episode of worsening cough, rattling, and constitutional upset lasting for 5 days or more. *Loose stools* were defined as stool frequency exceeding 3 daily.

Height (in centimeters) and weight (in kilograms) were recorded. Growth values were expressed to age/sex-adjusted growth charts [47-49] and body mass index. In addition, standard deviation scores (SDS) were calculated for height. *Stunted or wasted* (growth failure) was defined as less than the 2.5th percentile.

### 1.2.7. Ethics

Informed written consent was obtained from the adolescents and the parents. The study was approved by the Regional Ethics Committee for Medical Research.

## 1.3. Statistical analysis

When comparing 2 groups, response variables with a normal distribution were analyzed with a 2-sample *t* test (EA group/control group) or a 1-sample test (EA group/population group), whereas nonnormally distributed continuous variables were analyzed with a Mann-Whitney test. Simple linear regression was conducted to assess the effect of individual, clinical (medical), and environmental covariates on the response variables "mental health" (CBCL/YSR total problem score) and "psychosocial functioning" (CGAS). A final multiple linear regression was fitted to find prognostic variables for adolescents' mental health and detect possible confounders. Covariates were checked for collinearity before

**Table 1** Sample characteristics of 21 patients with EA and 36 adolescents from the general population (no significant differences)

Characteristics	EA	Control
Age (y), median (range)	13 (12-17)	14 (13-15)
Boys, n (%)	12 (57)	19 (53)
Montreal high-risk class II, n (%)	4 (19)	–
Serious associated anomalies, n (%)	6 (29)	–
Proportion living with mother, n (%)	21 (100)	36 (100)
Proportion living with single biological parent, n (%)	7 (33)	10 (29)
School scale score (academic competence) (YSR)	2.1 (.3)	2.2 (.4)
School scale score (academic competence) (CBCL)	4.2 (1.2)	4.8 (1.1)
Maternal education (y), mean (SD)	13.2 (2.8)	13.6 (3.6)

being entered into the final models, and the assumptions underlying regression analyses were checked. Goodness of fit of the final model was assessed by adjusted  $R^2$ .

Differences in outcome between the EA and control groups were controlled for age differences by multiple linear regression, with response variable as dependent and group and age as independents. Fisher's Exact test was used to check results for small data sets. SPSS 16 (SPSS, Chicago, IL) was used for statistical analyses.

## 2. Results

### 2.1. Patient characteristics

Sample characteristics are shown in Table 1. There were no significant differences between the EA group and the control group concerning age, sex, and sociodemographic characteristics. Mean birth weight of the EA patients was 2513 g (range, 1000-3340). The median duration of the first hospital stay was 28 days (range, 12-154). All patients had tracheoesophageal fistula, and esophageal anastomosis was performed within 2 to 3 days after birth. One had an additional fistula to the upper pouch. Three patients had gap greater than 3 cm. Two patients had scoliosis requiring treatment by physiotherapist, whereas 3 had "light" scoliosis.

**Table 2** Physical outcome at follow-up based on symptoms preceding 12 months of 21 adolescents with EA

	n (%)
<i>Respiratory symptoms</i>	
Daily cough	6 (29)
Cough during sleep	3 (14)
Cough related to meals	5 (24)
Dyspnea and wheezing	13 (62)
Bronchitis once	2 (10)
Bronchitis >1	3 (14)
Total with respiratory symptoms	16 (76)
<i>Gastrointestinal symptoms</i>	
Upper	
Dysphagia/food impaction	15 (71)
GER symptoms	6 (29)
Total with upper gastrointestinal symptoms	15 (71)
Lower	
Loose stools	13 (62)
Total with gastrointestinal symptoms	19 (91)
<i>Growth<sup>a</sup></i>	
Weight by height	
<10th percentile	4 (20)
<2.5th percentile	2 (10)
Height by age	
<10th percentile	6 (30)
<2.5th percentile	1 (5)

<sup>a</sup> n = 20.

**Table 3** Mental health outcome in adolescents with EA

	EA group	Control group
YSR <sup>a</sup> , mean (SD)		
Total competence score	18.9 (4.3)	13.4 (2.8) *
Total problems score	24.3 (15.1)	27.5 (25.1)
Internalizing score	6.7 (4.6)	8.9 (9.5)
Externalizing score	7.7 (5.9)	9.2 (8.9)
Dissociative symptoms score	2.2 (1.7)	2.8 (2.8)
CBCL <sup>b</sup> , mean (SD)		
Total competence score	22.7 (4.2)	17.1 (3.5) *
Total problems score	14.6 (11.9)	14.9 (18.1)
Internalizing score	5.6 (4.2)	3.9 (4.6)
Externalizing score	3.3 (4.8)	5.8 (8.7)
Dissociative symptoms score	1.7 (1.7)	1.5 (2.2)
Psychiatric diagnosis <sup>c</sup> , n (%)	3 (15)	5 (14)
Psychosocial functioning <sup>c</sup> (CGAS), mean (SD)	79.2 (11.7)	82.6 (11.1)

<sup>a</sup> n = 17.

<sup>b</sup> n = 19.

<sup>c</sup> n = 21.

\*  $P < .001$ .

Eleven patients (52%) needed a median number of 3 (range, 1-24) esophageal dilatations. None of the adolescents had experienced separation from their mother for 1 week or more during infancy or preschool years.

There were no significant differences on academic competence scores (CBCL and YSR) between the groups. Seven patients (33%) had special education: 5 of 17 from the prognostic low-risk group and 2 of 4 from the prognostic high-risk group (Montreal).

### 2.2. Physical health at follow-up

Physical outcomes based on symptoms in the preceding 12 months are shown in Table 2. Thirteen of 15 adolescents with dysphagia reported occasional dysphagia/food impaction, for example, when eating too rapidly. Certain types of food, particularly fresh bread, raw vegetables, and meat, occasionally inconvenienced these patients. None had experienced episodes of food impaction requiring medical help in the previous year, and the adolescents described strategies they had developed to improve swallowing. Five adolescents (24%) were completely asymptomatic concerning respiratory and upper gastrointestinal symptoms. Three (14%) had Nissen fundoplication. Loose stools were reported by 13 (62%) of the adolescents; 2 of them had Nissen fundoplication.

Mean height (SD) was 159 cm (12), and mean weight (SD) was 46.7 kg (14.6). Mean body mass index (SD) was 17.9 (3.6). Six adolescents (30%) had height by age below the 10th percentile, and 4 (20%) had weight by height below the 10th percentile. One was stunted, and 2 were wasted.

**Table 4** Comparison of self-esteem between adolescents with EA and population norm assessed by the Harter SPPA (higher scores represent better self-esteem)

SPPA subscales, mean (SD)	EA <sup>a</sup>	Population norm <sup>b</sup>
Self-worth	3.30 (.50)	2.87 (.54) **
Scholastic competence	2.89 (.42)	2.84 (.54)
Athletic competence	2.69 (.50)	2.44 (.63)
Social acceptance	3.35 (.45)	3.09 (.49) *
Close friends	3.44 (.54)	3.18 (.59)
Physical appearance	3.13 (.63)	2.60 (.68) **
Romantic appeal <sup>c</sup>	2.83 (.37)	2.62 (.58)

<sup>a</sup> n = 18.<sup>b</sup> n = 11,315.<sup>c</sup> n = 5 of 6 (only adolescents ≥14 years were included).\*  $P < .05$ .\*\*  $P < .01$ .

### 2.3. Mental health and psychosocial functioning

The EA adolescents adjusted well behaviorally and emotionally, and they did not differ from the adolescents in the control group on several parameters (Table 3). The mean CBCL scores of both groups were close to the mean of the Norwegian population [26]. The total competence score of the EA group was significantly better compared with controls.

There was no significant difference between EA adolescents and controls on the psychosocial functioning scale score (CGAS) (Table 3). One adolescent requested psychological treatment, and one had recently started such treatment.

### 2.4. Self-esteem and well-being

The EA adolescents reported significantly better self-worth, social acceptance, and physical appearance than the

general population [38] (Table 4). Among EA adolescents, the median “self-experienced well-being” score (VAS) was 95 (range, 35-100).

### 2.5. Parental and family strain

The most challenging period for the EA parents was during infancy (10/21) and toddlerhood/preschool years (7/21). Feeding difficulties and lung infections caused worries. Furthermore, parents felt it was difficult to let others take care of their child during feeding. If they currently should advise other parents with an EA infant, the adolescents’ parents used terms like “it is allowed to be anxious, but do not exaggerate”, “do not stress during feeding time”, “the child will not die if choking”, “have confidence in yourself”, and “it will be easier as the child grows older, have faith!”. A contact person at the hospital to call in case of insecurity was emphasized by 8 parents. Parents of 17 adolescents had no concerns about the future of their EA adolescent and expected them to be comparable with healthy adults. Parents of 2 adolescents had mild concerns, and only parents of 1 adolescent had serious concerns.

Mean (SD) maternal GHQ-30 sum Likert score was 22.6 (6.2). A “chronic family difficulties” score of at least 4 was observed in 4 families (19%).

### 2.6. Predictors of mental health and psychosocial functioning

More than 1 esophageal dilatation had significant negative effect on mental health as assessed with self-rated “total problems score” (YSR), in addition to “maternal psychological distress” (GHQ-30) (Table 5). Subjective well-being (VAS) was positively associated with mental health

**Table 5** Unadjusted and adjusted *B* for mental health assessed by YSR “total problems” among adolescents with EA

	Univariate linear regression		Multiple linear regression	
	<i>B</i> (unadjusted) (95% confidence interval)	<i>P</i> value	<i>B</i> (adjusted) (95% confidence interval)	<i>P</i> value
Individual characteristics				
Sex (male = 1, female = 2)	-14.1 (-29.1, .87)	.063		
Birth weight (g)	.003 (-.015, .021)	.744		
Somatic data and well-being at follow-up				
Montreal class (low = 1, high = 2)	-1.0 (-20.0, 18.0)	.909		
Operations >1 (no = 1, yes = 2)	13.1 (-1.6, 27.8)	.077		
Dilatation of esophagus >1 (no = 1, yes = 2)	16.0 (2.4, 29.5)	.024	17.2 (5.4, 29.1)	.008
Weight (kg)	.01 (-.57, .58)	.985		
Height (SDS) (continuous)	1.6 (-8.1, 11.3)	.753		
Well-being (VAS) (continuous)	-1.1 (-2.1, -.01)	.047	-1.1 (-2.0, -.3)	.013
Environment				
Chronic family difficulties	2.0 (-4.3, 8.4)	.506		
Maternal psychological distress (n = 16)	1.9 (.73, 3.0)	.003		

**Table 6** Unadjusted and adjusted *B* for psychosocial functioning (CGAS) among adolescents with EA

	Univariate linear regression		Multiple linear regression	
	<i>B</i> (unadjusted) (95% confidence interval)	<i>P</i> value	<i>B</i> (adjusted) (95% confidence interval)	<i>P</i> value
<b>Individual characteristics</b>				
Sex (male = 1, female = 2)	-7.7 (-18.1, 2.7)	.137		
Birth weight (g)	.01 (.00, .02)	.006	.01 (.001, .016)	.029
Dissociative symptoms score (CBCL) (continuous)	-4.6 (-7.5, -1.8)	.003	-3.6 (-6.3, .89)	.012
Academic competence score (CBCL) (continuous)	4.2 (-9, 9.1)	.098		
Academic competence score (YSR) (continuous)	3.3 (-11.3, 18.0)	.635		
<b>Somatic data and well-being at follow-up</b>				
Montreal class (low = 1, high = 2)	-3.0 (-16.9, 10.8)	.654		
Operations >1 (no = 1, yes = 2)	2.3 (-8.9, 13.5)	.670		
Dilatation of esophagus >1 (no = 1, yes = 2)	-4.4 (-15.1, 6.4)	.405		
Weight (kg)	.39 (.05, .72)	.026		
Height (SDS) (continuous)	9.2 (15.5, 13.0)	<.001	6.6 (3.1, 10.0)	.001
Well-being (VAS) (continuous)	.53 (.28, .78)	<.001	.33 (.12, .55)	.004
<b>Environment</b>				
Chronic family difficulties (ordinal)	-5.6 (-8.8, -2.4)	.001	-4.7 (-8.4, -.99)	.016
Maternal psychological distress (GHQ) (ordinal)	-1.1 (-2.0, -.30)	.011	-.61 (-1.4, .21)	.134

(YSR) (Table 5). Based on the regression analysis, adolescents with birth weight not exceeding 2500 g on average had 12.4 higher “total problems score” (CBCL).

Lower birth weight (in grams) and higher trauma-related dissociative symptom score (CBCL) had significant negative effect on the EA adolescents’ psychosocial functioning (CGAS) (Table 6). Moreover, declined height (SDS) had significant negative effect on psychosocial functioning (CGAS); and subjective well-being (VAS) was positively associated with CGAS (Table 6). Increased chronic family difficulties had significant negative effect on psychosocial functioning (Table 6).

Among potential prognostic covariates for mental health as assessed with “total problems score” (YSR), a final multiple linear regression model was fitted. Dilatations of esophagus and subjective well-being (VAS) were significant prognostic factors predicting mental health (adjusted  $R^2 = .508$ ) (Table 5).

To find the best prognostic factors predicting psychosocial functioning (CGAS), a final multiple linear regression was fitted with “birth weight” and “trauma-related dissociative symptom score” (Table 6). Both were significant prognostic factors predicting psychosocial functioning (CGAS) (adjusted  $R^2 = .508$ ). In addition, a final multiple linear regression was fitted with “height (SDS)” and “well-being.” Both were significant prognostic factors predicting psychosocial functioning (adjusted  $R^2 = .726$ ) (Table 6). Lastly, a final multiple linear regression was fitted with the environmental covariates “chronic family difficulties” and “maternal psychological distress.” Chronic family difficulties was the single significant prognostic factor predicting psychosocial functioning (adjusted  $R^2 = .479$ ), and so this documents confounding (Table 6).

### 3. Discussion

We found that adolescents with EA adjusted well. Parent- and adolescent-reported mental health problems as well as a clinician-based assessment of psychosocial functioning were similar in the study group and general population-based control group. Global self-worth, social acceptance, and physical appearance were even better among adolescents with EA than population norms.

The findings of our study contradict those of Bouman et al [19] who reported that EA children aged 8 to 12 years had significantly more parent-reported emotional and behavioral problems compared with population norms. In accordance with our findings, children in the Bouman et al [19] study did not report more emotional and behavioral problems than the population. Our results are in line with studies of mental health and psychosocial functioning in adolescents with Hirschsprung disease [50] and juvenile chronic arthritis [51], but contradictory to studies on adolescents with anorectal anomalies [52] and bladder exstrophy and epispadia [53]. We hypothesize that specific challenges related to the different diseases contribute to the different outcomes. A meta-analysis of psychological adjustment in pediatric physical disorders showed that the effect of having chronic disease on psychological adjustment decreased when matched for age, sex, and socioeconomic status [6]. Comparisons with normative data on self-concept measures showed a tendency toward “normalizing” self-concept problems or even more positive self-esteem among children with physical disorders [6].

The EA adolescents reported that they perform at least as good as or even better than controls. This may reflect the possibility that the EA adolescents and their families, after many years of struggling, have developed a more optimistic look of life in general and their health in

particular than the controls. Similar observations have been found in studies of children with cancer [54]. Others have reported that effective management of the illness may enhance self-esteem [55].

The EA adolescents did not differ from controls concerning academic competence. However, 33% had needed special education, reflecting vulnerability to learning problems. This finding is in accordance with others [19]. Proper assessment of cognitive development and search for predictors in controlled studies of children and adolescents with EA are required to explore this finding.

A survey assessing mental health among 1-year-old infants with EA identified psychiatric disturbances, mostly traumatic related [14]. In the present study, the EA adolescents' trauma-related dissociative symptoms were a prognostic factor for their psychosocial functioning. There were no records of traumatic experiences in early childhood from the medical reports in this study, and very few parents did recall any traumatic experiences for their child during infancy or early childhood. However, dilatations of esophagus were a prognostic factor predicting the EA adolescents' mental health. Emerging data suggest that children often experience medical treatment (eg, invasive, frightening, and/or painful treatment procedures) as traumatic [56,57]. Our findings may reflect that early traumatic experiences may create a vulnerability to mental health and psychosocial functioning problems later in life [58]. Prospective studies are needed to explore the impact of early trauma related to physical illness later.

Less chronic family strain had positive effect on EA adolescents' psychosocial functioning. This is in accordance with previous studies reporting that chronic family strain predicts mental health in EA infants [14]. This may reflect the nurturing effect of a supportive environment and parental care for children with EA. Positive parental attention may have a greater impact on the outcome of emotional and psychological adjustment than the actual illness itself.

The EA adolescents with shorter stature showed less optimal psychosocial functioning. The association between height and psychosocial functioning has been assessed in mixed diagnostic populations, and the results are divergent [59,60]. In the Wessex Growth Study [61,62], a prospective population-based cohort study, stature did not affect psychosocial adaptation in children or young adults. However, children with organic disease were excluded from the study. Others have highlighted the etiology of short stature as an important factor for the psychological adjustment of children [63].

The fact that the EA patients seemed to be sensitive about their smaller stature may indicate that the SPPA is not sensitive on the aspect of physical appearance. Others have discussed what the concept global self-worth according to SPPA really measures, and a better understanding about the determinants of the adolescents' perception of their physical appearance is requested [39]. In accordance with this, our

findings support that perceived physical appearance is determined by other factors than merely looks or stature.

Strengths of the present study were the use of standardized diagnostic interviews in addition to well-validated and reliable questionnaires, and the comparison with a control group. The data set is small, and statistical analyses may therefore be assessed as unstable. However, we used different methods and confirmed the significance of *t* tests using Mann-Whitney; and  $\chi^2$  tests were checked using Fisher's Exact test. We therefore maintain that the results in this study are consistent and stable.

Frequent esophageal dilatations and maternal and family distress have negative impact on EA adolescents' mental health and psychosocial functioning. Psychosocial experts at hand as part of the health care team are recommended. Pediatric surgeons should be informed about the characteristics of patients and families "at risk" for psychosocial dysfunctioning to refer them early for psychosocial support. Based on our findings, we would emphasize early intervention and support to EA families with increased chronic strain and infants requiring dilatations. Increased knowledge about effects of early traumatic experiences is needed among pediatricians and pediatric surgeons.

The results require replication, and there is need for multicenter studies. To gain more insight into the predictors of adjustment, large longitudinal studies are recommended. Self-esteem as a mediator of psychosocial adjustment in adolescents with EA would be an interesting variable to investigate more thoroughly.

## Acknowledgments

The study was supported financially by the National Research Council, Region Centre for Child and Adolescents Mental Health, Eastern and Southern Norway, and Southern Norway Regional Health Authority. Particular thanks to Thore Egeland, PhD, for statistical help and Astrid Aasland, PhD, for scoring the videos used in assessing interrater reliability.

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