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Impaired motor performance in adolescents with esophageal atresia

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ABSTRACT

Aims: The study prospectively assessed motor development from infancy to adolescence in patients with esophageal atresia (EA).
Methods: At one year of age motor performance was evaluated with the Psychomotor Developmental Index (PDI) of the Bayley Scales of Infant Development, Second Edition (BSID-II), and as adolescents reevaluated with Motor Assessment Battery for Children, Second Edition (MABC-2). Associations to clinical factors were assessed.
Results: 23 EA patients were followed from infancy to adolescence. The median total PDI score in infancy was 102 (56–118) and the corresponding mean z-score was −0.006 (SD 0.995) and not significantly different from the reference values ($p = 0.48$). The median total MABC-2 score in adolescence was 75 (32–93) and the corresponding mean z-score −0.43 (SD 0.998) which is significantly below normal ($p = 0.03$). Children with impaired motor function in adolescence underwent significantly more rethoracotomies than those with normal motor performance ($p = 0.037$); whereas the two groups did not differ with respect to other clinical characteristics.
Conclusion: From infancy to adolescence the motor performance in the group of EA patients deteriorated from within normal range to significantly impaired compared to reference values. Interdisciplinary follow-up programs from infancy to adolescence with close monitoring for motor function is necessary to detect motor impairments.

Level of evidence

Prognostic study, Level II

1. Introduction

Esophageal atresia (EA) is a serious congenital malformation characterized by interruption of the esophagus needing surgical intervention in the neonatal period. About half of the EA neonates have associated malformations like congenital heart disease (CHD), musculoskeletal malformations, anorectal- and intestinal malformations, and genital- urinary malformations. Twenty to forty percent of EA patients are born prematurely and may suffer from morbidities associated to prematurity [1].

The risk for reduced motor performance in toddlers after EA repair has been reported, however, the results are ambiguous. A few studies have reported neurodevelopmental outcome within normal range in one year old EA infants [2-4]. Faugli et al. completed developmental assessment in 36 EA infants and identified developmental delay in 11% [5]. Harmsen et al. evaluated motor development in five years old EA children compared to the normative group. They found reduced gross motor development with persisting problems with gross motor performance when reevaluating at the age of eight years [6]. However, longitudinal follow-up studies on motor development from infancy to adolescence in children with EA after the age of eight years are still missing.

We speculated that patients with EA may develop motor problems. We assessed motor performance longitudinally in children with EA from infancy to adolescence and searched for risk factors which may be associated with deterioration.

Abbreviations: BSID-II, Bayley Scales of Infant Development, Second Edition; CDH, Congenital Diaphragmatic Hernia; CHD, Congenital Heart Disease; ECMO, Extra Corporal Membrane Oxygenation; EA, Esophageal Atresia; GA, Gestational Age; MABC-2, Motor Assessment Battery for Children, Second Edition; PDI, Psychomotor Developmental Index; UK, United Kingdom; US, United States; VACTERL, Vertebral defect, Anal atresia, Cardiac defect, Trachea-esophageal fistula, Renal abnormalities, and Limb abnormalities.

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2. Material and methods

2.1. Population

Between 1999 and 2002, 44 neonates with EA were consecutively treated at Oslo University Hospital. From this group 36 participated at the age of 13 months in a study on mental health and motor performance [5]. All 36 parents accepted at that moment to be invited for further studies, and all were assessed for eligibility to be included in the present follow-up study.

Exclusion criteria were serious medical conditions and age older than 16 years at follow-up ((evaluation instrument Motor Assessment Battery for Children, Second Edition (MABC-2) not suitable [7])).

2.2. Demographics and clinical characteristics

Demographics and medical characteristics as gender, gestational age (GA), birthweight, prematurity (GA < 37 weeks), EA Gross classification, major CHD (including those needing surgery), VACTERL associations, number of days of initial hospital stay, duration of mechanical ventilation, number and age at rethoracotomy /sternotomy during childhood, number of anesthetic procedures and esophageal dilations at both 12 months of age and at adolescence were registered from medical records [8,9].

2.3. Design

A prospective cohort study of motor development in EA children from infancy to adolescence after EA repair.

2.4. Motor evaluation in infancy

At one year of age the patients were evaluated with the standardized Bayley Scales of Infant Development, Second Edition (BSID-II) using the United State (US) reference norms obtained in 1700 children aged 1–42 months [10]. A calculated raw score is converted into a Psychomotor Developmental Index (PDI score) with a mean score of 100 and a standard deviation (SD) of 15. In a normally distributed population, 95% will obtain a PDI score in the range of 70–130. A score between 85 and 114 is classified as within the normal range. An achieved PDI score between 70 and 84 is classified as at risk for delayed motor performance, and a score of 69 or below is classified as definitely delayed motor performance [10]. Motor evaluation was carried out by a teacher certified to perform these tests.

2.5. Motor evaluation in adolescence

As adolescents the patients were reevaluated with the Motor Assessment Battery for Children, Second Edition (MABC-2) [7]. MABC-2 is designed to describe and identify motor impairments in children and adolescents from 3 to 16 years of age. Motor performance is divided into fine and gross motor skills. MABC-2 consists of three different categories: manual dexterity (fine motor skills), ball skills, and static and dynamic balance (gross motor skills). MABC-2 is a normed referenced evaluating tool validated in 1170 children and adolescents from the United Kingdom (UK), 566 boys (48.3%), representing different ethnic groups and parental educational level. We used the 11–16 years age band. According to the UK norms, a raw score for each of the three motor categories are summarized into a total test score, the higher number of total test score, the better motor performance. The total test score is converted into a standard score and a percentile rank. According to the manual, we classified motor performance as normal (percentile score >15), at risk for motor delay (percentile score 6–15), and motor delay (percentile score < 6) [7]. The evaluations with MABC-2 were completed by an experienced pediatric physical therapist.

2.6. Physical therapy

In a semi structured interview, the adolescents and their parents were asked if they had been offered physical therapy; continuously from infancy to adolescence, as toddlers only, or as adolescence only (yes/no).

2.7. Organized recreational activity

During the semi structured interview, the adolescents were asked if they at present took part in organized recreational activities (yes/no). All aspects of organized activities were registered from sports to music lessons. A random sample of 50 Norwegian healthy adolescents recruited from different schools in Oslo area, mean 15.6 years (range 12–20), participated as reference group.

2.8. Statistical analysis

Normative data values for specific percentiles (1, 2.5, 5, 10, 25, 50, 75, 90, 95, 97.5, 99) were Box-Cox transformed to remove skewness and meet the requirements for a Gaussian distribution. Subsequently estimates for the expectation value μ and standard deviations were calculated. Patient data values (X) were treated likewise. From a transformed value Y the z-score = (Y- μ)/s was found and incorporated in normality plots. Comparing with the reference values, a total test scores for participants in infancy (PDI) and adolescence (MABC-2) were converted to z-scores.

Descriptive analyses are presented with median values and total range. The Mann-Whitney test was used for comparing baseline data of participants and non-participants, and for comparing participants with normal MABC-2 total score and impaired MABC-2 total score. Statistically significant value was accepted at the level $p = 0.05$. Data are analyzed with the use of SPSS Statistics version 25 (IBM, Armonk, NY) in collaboration with the statistician.

2.9. Ethics

Informed written parental consent was obtained at both ages and also from adolescents of 13 years and older. The project has obtained approval from the National Regional Ethical Committee for Medical Research (2014/1224 REK) and from the Data protection officer reg. no: 2014/9344.

3. Results

3.1. Patients

Thirty-six participants were studied in infancy at a median age of 13 months (range 12–17). One child had died, and two children could, according to the manual of MABC-2, not fulfill the test reliability because of serious medical morbidities [7]. Of the 33 patients eligible for the study, three were too old for MABC-2. Thus, 30 adolescents were invited, and seven adolescents refused to participate (Fig. 1). Consequently, 23 EA patients were evaluated at the median age of 16 years (range 13–16); in 21 of them (91%) longitudinal data were obtained (Fig. 1).

3.2. Clinical characteristics

Among 36 infants, seven (19%) had PDI scores below normal (two infants significantly delayed and five at risk). Of these seven infants, only two were reevaluated in adolescence (Fig. 1).

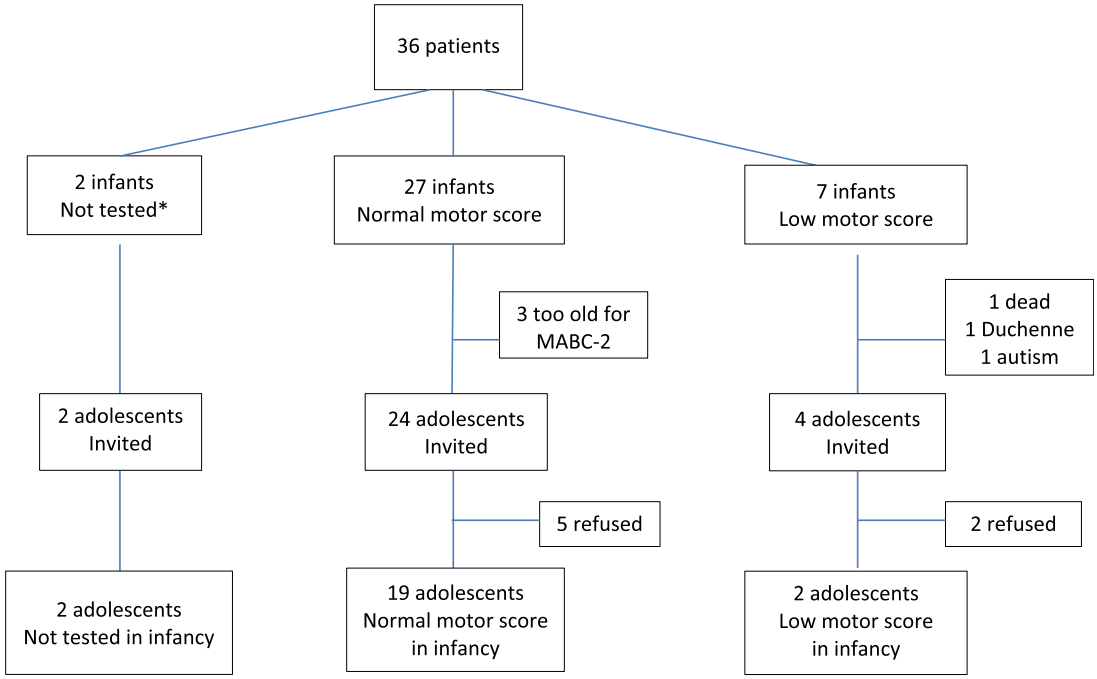

Fig. 1. Flow chart of the included and not included patients. A total sample of the 23 esophageal atresia (EA) adolescents participated.
MABC-2= Motor Assessment Battery for Children, Second Edition.
*MissingPDI score in infancy.

Table 1
Demographic and clinical characteristics of the 33 esophageal atresia (EA) patients eligible for the follow-up study.

Variables	Included $n = 23$	Not included $n = 10$	p-value
Male, n (%)	16 (70)	6 (60)	.598
Birthweight, grams, median (range)	2820 (1690–4570)	3485 (585–4020)	.457
Prematurity, n (%)	6 (26)	4 (40)	.431
Gestational age, weeks, median (range)	38 (30–40)	41 (27–42)	.551
EA Gross A, n (%)	1 (4)	1 (10)	.538
EA Gross C, n (%)	19 (83)	8 (80)	.860
EA Gross D, n (%)	3 (13)	1 (10)	.808
CHD, n (%)	1 (4)	1 (10)	.538
VACTERL, n (%)	5 (22)	0 (0)	.115
Initial hospital stay, days, median (range)	21 (10–177)	21 (14–116)	.505
Mechanical ventilation, days, median (range)	0 (0–41)	0 (0–18)	.089
Rethoracotomy, n (%)	4 (17)[a]	2 (20)	.860
PDI score, median (range)[b]	102 (56–118)	90 (70–121)	.228
No. anesthesias < 12 months, median (range)	1 (1–30)		
No. anesthesias total, median (range)	1 (1–36)		
No. esophageal dilations < 12 months, median (range)	0 (0–25)		
No. esophageal dilations total, median (range)	0 (0–26)		

CHD, congenital heart disease. VACTERL, vertebral defect, anal atresia, cardiac defects, trachea-esophageal fistula, renal abnormalities, and limb abnormalities.
[a] Rethoracotomy: complex cardiac anomaly ($n = 1$), anastomotic revision ($n = 3$).
[b] Two missing PDI (Psychomotor Developmental Index) data in the included group and three missing in the not included group.

Clinical characteristics of the 23 EA included adolescents are shown in Table 1. Registered variables and PDI score did not differ between participants and non-participants (Table 1). Three participants had a rethoracotomy during the first year of life ((refistula at three months of age, cardiac surgery (complex cardiac anomaly) at five months of age, surgery because of esophageal stenosis at nine months of age)), and one patient (recurrent fistula) at four years of age. All neonatal reconstructive surgeries were performed by posterolateral right sided thoracotomy.

3.3. Motor development from infancy to adolescence

The median total MABC-2 test score for the 23 EA adolescents was 75 (32–93), and the corresponding mean z-score −0.43 (SD 0.998), which is significantly below normal ($p = 0.03$). The distribution of motor function level (normal, at risk, and delayed) in total MABC-2 test scores, with subcategories of fine and gross motor skills, are shown in Fig. 2.

Children with impaired motor function in adolescence underwent significantly more rethoracotomies than the group with normal motor performance ($p = 0.037$) (Table 2). However, the two groups did not differ with respect to other clinical characteristics.

The median total PDI score for the 21 infants with longitudinal data was 102 (56–118), and the corresponding mean z-score for total PDI score in infancy for the 21 patients was −0.006 (SD 0.995) and not significantly different from the reference values ($p = 0.48$) [10].

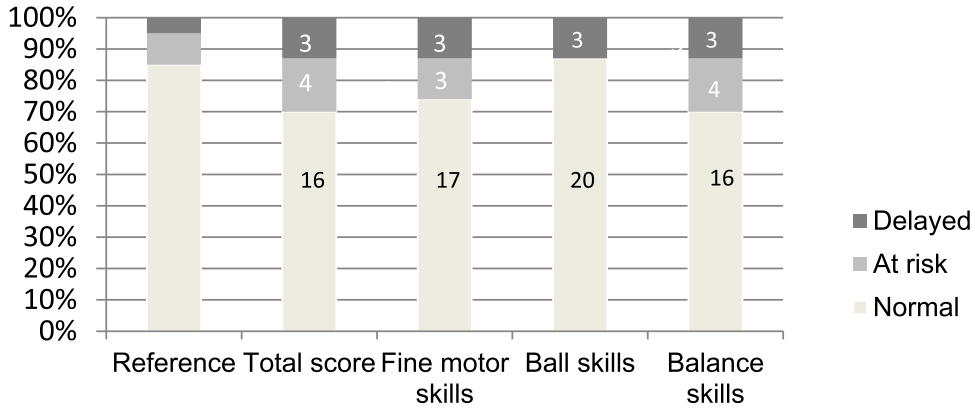

Fig. 2. Motor performance (normal, at risk, and delayed) evaluated with Motor Assessment Battery for Children, Second Edition (MABC-2) in esophageal atresia (EA) adolescents. Reference data (percentile rank), total motor score, fine-, and gross (ball and balance) motor skills.

Table 2
Clinical characteristics in esophageal atresia (EA) patients registered with Motor Assessment Battery for Children, Second Edition (MABC-2) total score within normal range and within impaired range.

Variables	Normal MABC-2 total score ($n = 16$)	Impaired MABC-2 total score ($n = 7$)	p-value
Birthweight, grams, median (range)	2830 (1690–3664)	2740 (1900–4570)	.593
Gestational age, weeks, median (range)	38,5 (30–40)	37 (34–40)	.173
Prematurity, n (%)	3 (19)	3 (43)	.236
VACTERL, n (%)	3 (19)	2 (29)	.607
Initial hospital stay, days, median (range)	20 (14–50)	22 (14–178)	.867
Mechanical ventilation, days, median (range)	0 (0–3)	0 (0–41)	.431
Rethoracotomy, n (%)	1 (6)	3 (43)[a]	.037[b]
No. anesthesias < 12 months, median (range)	1 (1–7)	2 (1–30)	.407
No. anesthesias total, median (range)	3.5 (1–12)	6 (1–36)	.363
No. esophageal dilations < 12 months, median (range)	0 (0–4)	0 (0–25)	.517
No. esophageal dilations total, median (range)	0 (0–9)	1 (0–26)	.224

VACTERL, vertebral defect, anal atresia, cardiac defects, trachea-esophageal fistula, renal abnormalities, and limb abnormalities.
[a] Complex cardiac anomaly $n = 1$, esophageal anastomotic complication $n = 2$.
[b] Statistically significant at $p = 0.05$.

Thus, from infancy to adolescence the motor performance in the total group of EA patients deteriorated from within normal range to significantly impaired compared to reference values.

As infants, 19 of the 21 children (90%) had total motor performance within normal range, and 15 of them (79%) still scored in the normal range as adolescents. Deterioration of total motor performance from infancy to adolescence occurred in 4/21 (19%) children. Persistent impairment of total motor performance as adolescents was shown in both children with motor function delay in infancy: One child with hydrocephalus had delayed motor performance at both evaluations. Another child was born prematurely, and classified at risk for motor delay in infancy and as delayed in adolescence.

3.4. Physical therapy

Overall, 16 (70%) of EA adolescents had been offered physical therapy after neonatal period; eight as toddlers only, four as adolescence only, and four continuously from infancy to adolescence. No patients had been offered systematic gross motor therapy.

3.5. Organized recreational activities

Twenty (87%) EA adolescents participated in organized recreational activities versus 39/50 (78%) in the reference group. The three EA adolescents not participating in organized recreational activities had total motor performance within normal range, whereas those with impaired motor performance participated in organized recreational activities.

4. Discussion

This is the first longitudinal assessment of motor performance from infancy to adolescence in children with EA. We speculated that patients with EA may worsen in motor performance as they get older. Our results show that motor performance changed from within normal range in infancy to significantly impaired compared to reference values in adolescence. One third of the adolescents scored below normal range (i.e. < 16 percentile level). Especially gross motor skills were impaired. The only factor related to reduced motor performance in adolescence was rethoracotomy.

Our result in EA infants seem to be in accordance with earlier studies. Aite et al. using BSID-III and Gischler et al. using the Dutch version of BSID, reported motor skills within normal range in one year old EA infants [2,4]. Walker et al. applying BSID-III, found lower motor score in one year old EA patients compared to the control group, but the difference was not significant [3]. We used the BSID-II and reference data from healthy US children. Although different instruments and different reference values have been used in the other studies, we still think that our results are in line with the previous studies in EA infants [2-4].

One third of the EA adolescents in our study showed impaired motor function with lowest scores for the gross motor skills. There are few studies on motor performance in EA adolescents. Harmsen et al. evaluated EA patients longitudinally at five and eight years using MABC [6]. They found in accordance with our results impaired motor skills, particularly in gross motor tasks concerning ball and balance skills at both evaluations. However, it is important to emphasize that the results so far are based on a small sam-

ple size, 45 and 23 participants respectively in Harmsen's and our study. Therefore, we need to be careful in concluding.

Assuming that overall motor development is normal or slightly impaired in infancy, and that approximately 30% of children with EA had gross motor scores below normal at preschool age, school age, or adolescence, we wanted to know who is at risk and needs follow-up of motor performance at older age [2-4, 6].

Looking at our own data, rethoracotomy seems to be a risk factor for impaired motor development. One of our four patients with rethoracotomy was a five months old child with a complex cardiac anomaly operated by sternotomy. Complex cardiac anomalies may also be a risk factor for motor function problems in childhood as reported by Holm et al. [11]. Furthermore, Harmsen et al. reported that total duration of anesthetic exposure in the first 24 months of life is negatively associated with motor performance as assessed by MABC at five and eight years [6]. However, in the present study, no association between anesthetic exposure and motor performance could be identified. All our patients had neonatal surgery by right sided posterolateral thoracotomy with affection of the serratus anterior and latissimus dorsi muscles. This surgical procedure may later cause musculoskeletal asymmetry which may increase during growth [12,13]. We wonder if this anatomical deformity may cause less trunk stability and disturb motor function, particularly gross motor skills as balance skills. Balance skills were the most affected skills in our study (Fig. 2). Number of surgical interventions may increase the prevalence of musculoskeletal morbidity as chest wall deformity and scoliosis [14]. Therefore, we speculate if rethoracotomy will increase the risk for asymmetric muscular morbidity and cause impaired gross motor function. More studies are needed to conclude in this speculation.

Interestingly, a study conducted by Mazer et al., assessing 105 patients with non-cardiac congenital anatomical anomalies including 15 EA patients, reported early motor development as predictor of development at five years, with number of congenital anomalies as predictive of outcome over time [15]. Among the 21 patients examined at both occasions in our study, four patients deteriorated in motor performance from infancy to adolescence, and two patients were impaired at both occasions. It seems obvious that motor performance in EA patients may be impaired at both an early and late age. Unfortunately, there were too few patients in our study to conclude on predictive value of number of congenital anomalies or early motor development. Furthermore, our results do not seem to support Mazer, reporting early developmental delay predicting development at age five [15]. Thus, we will still recommend that all EA children should be followed up with standardized instruments during childhood, even if they seem to have normal motor performance in infancy.

It can be assumed that parental overprotection contributes to inexperience with physical activity and reduced motor skills in children with serious congenital malformations. However, in our study 87% of patients participated in organized recreational activities. It is also confirmed by Toussaint-Duyster et al. that children with EA are physically active [16]. They reported in their study of 63 eight years old EA patients that 79.4% took part in sport activities. Harmsen's group reported that sport participation is positively associated to motor performance at five and eight years [6]. In our study the sample size is too small to conclude on the effect of sports activity on motor performance. Moreover, none of the studies information was obtained on the nature and intensity of the physical activities.

We cannot conclude on the effect of physical therapy because our patients did not have systematic and focused follow-up. However, just to create awareness of problems by measuring may motivate children with EA to be physically active.

Toussaint-Duyster et al. evaluated the parent reported motor performance MABC-2-Checklist [17]. They concluded that just a parent reported questionnaire is not a sufficient tool to identify motor impairment in children. Proper measurement by physical therapist followed by counselling and tailor-made advice is still the gold standard of follow-up to prevent deterioration [18].

Other physical problems such as pulmonary morbidity may have implications for motor performance, and we know that children with EA may have persistent airflow obstruction and abnormal lung function [16,19]. Pulmonary problems may also withhold children from being active at older age. Toussaint-Duyster et al. found in their study reduced exercise capacity in 55 eight years old EA patients [16]. We have in the current study not assessed lung function, but we think that pulmonary problems may contribute to avoidance of physical activities at older age in children with EA. Some support may also be sought from the study of van Cammen et al. in 254 extra corporal membrane oxygenation (ECMO) survivors showing deterioration in motor performance [20]. Among these patients, a substantial number were patients diagnosed with congenital diaphragmatic hernia (CDH) who like EA, is a foregut anomaly. It is possible that the foregut anomalies have a common tendency to develop similar comorbidities. The CDH patients were assessed with MABC-1 at five, eight, and 12 years of age, and showed gradually deterioration in motor performance at all the test points [20]. We speculate that persistent respiratory morbidity and parents who are reluctant to stimulate physical activities in early childhood, may lead to physical inactivity and a negative spiral with increasing motor problems and physical deconditioning.

We considered prematurity as a risk factor for impaired motor function. However, prematurity as the explanation for our results is unlikely, as most children were born after 32 weeks, and they were not overrepresented in the subgroup of children with impaired motor function.

Furthermore, one may speculate if the patients are just growing into deficits in motor performance because of critical neonatal illness. Recently Rudisill et al. suggested that delayed brain growth occurred during the perioperative period of repair of long gap EA [21]. Schiller et al. proposed a common neurodevelopmental pathway to neuropsychological impairment following neonatal critical illness [22]. Such vulnerabilities may also be risk factors for impaired motor development in EA patients [22].

4.1. Strengths

The strengths of the study are the longitudinal design, following the same patients from infancy to adolescence. Additionally, all participants were treated in one center.

The instruments are both validated on different ethnic groups and social classes as the Norwegian population is more and more becoming alike. The motor instruments used in this study are well known and much used in clinical practice and research within and outside Norway.

4.2. Limitations

The small sample size is a limitation; only 23 EA patients participated and only 21 were evaluated twice. This means that the influence of potentially important variables such as prematurity, CHD, or other associated anomalies and medical factors is difficult to evaluate.

It is also important to point out that the observed decline in motor skills from infancy to adolescence may be hidden in the design of the two different motor instruments being used at the two different test times. BSID at one year is a test focusing on observed milestones, and fine details in motor skills may be difficult to register. In contrast, MABC-2 consists of more complex tasks, and subtle motor problems may be easier to detect as the children get

older and better to take instructions and cooperate with the examiner.

Different versions of Bayley Scales of Infant and Toddlers Development used in the studies may also introduce a bias in comparing results. Walker and Aite have used the 3rd Edition of Bayley Scales of Infant and Toddlers Development, and Gischler applied the Dutch version of the Bayley Scales of Infant Development.

Another weakness in the present study is that we have not used control group based on Norwegian national reference values. Our results are based on reference data from healthy US and UK children which may differ from the level of motor performance in Norwegian children. Other studies also used different comparison groups [2-4]. Therefore, the outcome of the comparison of results between these different studies must be interpreted with caution.

However, even though the study samples are small and different versions of BSID and different types of reference data have been applied, we think the results are in line with previous studies and therefor bring up important new data on natural course of motor function in EA patients over time.

5. Conclusion

In early years it is hard to distinguish who will develop motor impairments during childhood. Consequently, interdisciplinary follow-up programs from infancy to adolescence with close monitoring for motor function with standardized motor evaluation instruments, is necessary to detect motor impairments and offer tailor-made interventions.

Declarations of Competing Interest

None

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