Quality of life in adolescents who underwent congenital diaphragmatic hernia surgical repair

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ABSTRACT

Objective. The advances made in the surgical and postnatal treatment of congenital diaphragmatic hernia (CDH) have considerably improved patient survival, but morbidity remains significant. The objective of this study was to analyze the effect these sequels have on the health-related quality of life (HRQL) of adolescents and young adults who have survived CDH, and to compare it with that of the general population.

Materials and methods. A transversal descriptive study of patients diagnosed with CDH in our institution from 1997 to 2004 was carried out. Survival, location, hernia size, herniated organs, need for extracorporeal membrane oxygenation, and mechanical ventilation time were analyzed. In addition, a comparative study of the current HRQL of survivors was conducted using the SF-36 survey (36-Item Health Survey Short Form), which assessed physical function, physical role, body pain, general health, vitality, social function, emotional role, and mental health. Data of 24 healthy adolescents was used as a control group.

Results. Of the 29 survivors (70.7%), 21 were successfully contacted, and 16 responded to the survey. They all claimed their overall quality of life was good or very good. The group of adolescents who underwent CDH surgical repair had better results in the vitality ($p=0.001$) and mental health ($p<0.05$) areas, but the overall HRQL score and the remaining health areas were similar. No significant differences were found regarding diaphragmatic size or need for ECMO.

Conclusions. According to adolescent survivors who underwent CDH surgical repair, their quality of life is similar to that of other individuals of their age. Our results are encouraging and may prove useful for future parents of CDH patients.

KEY WORDS: Congenital diaphragmatic hernia; Health-related quality of life; Long-term survivors; Extracorporeal membrane oxygenation.

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CALIDAD DE VIDA EN ADOLESCENTES INTERVENIDOS DE HERNIA DIAFRAGMÁTICA CONGÉNITA

RESUMEN

Objetivo. Los avances en el tratamiento quirúrgico y posnatal han mejorado significativamente la supervivencia de pacientes con hernia diafragmática congénita (HDC). La morbilidad asociada sigue siendo significativa. El objetivo del estudio es evaluar efecto de estas secuelas sobre la calidad de vida relacionada con la salud (CVRS) de adolescentes y adultos jóvenes supervivientes de HDC y compararla con la población general.

Material y métodos. Estudio descriptivo transversal de pacientes diagnosticados de HDC entre 1997 y 2004. Supervivencia, localización, tamaño de la hernia, órganos herniados, necesidad de oxigenación por membrana extracorpórea, tiempo de ventilación mecánica. Estudio comparativo de CVRS actual de pacientes supervivientes mediante la encuesta SF-36 (*36-Item Health Survey Short Form*): función física, rol físico, dolor corporal, salud general, vitalidad, función social, rol emocional, salud mental. Utilizamos datos de 24 adolescentes sanos como grupo control.

Resultados. De los 29 supervivientes (70,7%), fueron localizados 21 y contestaron la encuesta 16, que consideraron tener una calidad de vida global buena o muy buena. El grupo de adolescentes intervenidos tuvieron mejores resultados en las esferas de vitalidad ($p = 0,001$) y salud mental ($p < 0,05$), pero la puntuación de la CVRS global y el resto de dimensiones de salud fueron similares. No se objetivaron diferencias significativas en relación con el tamaño diafragmático ni la necesidad de ECMO.

Conclusiones. Los adolescentes supervivientes intervenidos de HDC consideran tener una calidad de vida similar a jóvenes de su misma edad. Los resultados de nuestro estudio son alentadores y permiten un mejor asesoramiento para futuros pacientes con HDC.

PALABRAS CLAVE: Hernia diafragmática congénita; Calidad de vida relacionada con la salud; Supervivientes a largo plazo; Oxigenación por membrana extracorpórea.

INTRODUCTION

Congenital diaphragmatic hernia (CDH) is a diaphragmatic defect occurring during embryonic development. As a result of it, abdominal organs are introduced within the

thorax, thus impairing adequate lung development (pulmonary hypoplasia). Prevalence is 1 in 2,000-2,500 births, and estimated survival ranges from 50% to 80% according to each healthcare institution[1].

In the last years, the survival of these patients has significantly improved primarily thanks to the improvement of prenatal diagnostic techniques and the advances in neonatal intensive care, such as extracorporeal membrane oxygenation (ECMO), high frequency oscillatory ventilation, permissive hypercapnia, nitric oxide, and delayed surgery until better hemodynamic stabilization has been achieved[2]. Accurately determining complications and sequels is not an easy task as a result of its low incidence and the differences among healthcare institutions and countries regarding perinatal management, surgical techniques, and perinatal intensive care. However, there are multiple well-known studies on this. Since 1995, the CDH Study Group[3] has collected data and answered medical questions from healthcare institutions treating children with congenital diaphragmatic hernia all over the world. Among many other variables analyzed, diaphragmatic defect size, hepatic herniation, and presence of severe cardiac abnormality are currently regarded as the most significant risk factors with an impact on mortality[4,5].

Short- and mid-term complications, which mostly determine overall morbidity, are primarily related to pulmonary involvement (pulmonary hypertension, pulmonary hypoplasia, and chronic pulmonary pathology) and neurodevelopment disorders. In the mid-term, stunted growth and gastrointestinal (mainly gastroesophageal reflux) and musculoskeletal (chest wall deformities and scoliosis) pathologies, which are more frequent in these patients, gain particular importance[6,7]. Associated morbidities may cause significant life deterioration, which becomes particularly visible during adolescence. Studying health-related quality of life (HRQL) in children and adolescents as a way of measuring the long-term results of CDH treatment is key. Understanding physical, psychical, and social consequences should facilitate optimal care of older CDH survivors.

Even though the short-term results of diaphragmatic hernia treatment are well-known, few healthcare institutions have published HRQL studies in children born with CDH, and pre-adulthood literature references are scarce. The objective of this work was to understand quality of life as perceived by a group of adolescents who underwent CDH surgical repair al birth, and to compare the results with a control sample of healthy adolescents.

MATERIALS AND METHODS

An observational study and a comparative study were carried out. First, an observational, descriptive, transversal study of health-related quality of life was conducted. The study population included adolescents and young adults – as of today – who underwent congenital diaphragmatic hernia surgical repair neonatally in our institution from 1997 to 2004. The survey was carried out from January to March 2018.

Parents of CDH survivors and CDH survivors of legal age were contacted by phone, and the objective of the study was individually explained to each of them. Those who accepted to participate were requested to provide a contact email address to fill out the survey and submit written authorization.

Second, a comparative study of quality of life was carried out between the aforementioned sample and a control group of healthy adolescents from the Community of Madrid (Spain). Age and sex characteristics of these individuals, who were not related to our hospital, were similar to those of the sample. A voluntary, anonymous health-related quality of life survey was proposed to reflect the quality of life of a group similar to the sample, but without history of CDH.

Date of birth, sex, and diaphragmatic-hernia-related survival or demise of all patients who underwent CDH surgical repair in that period were collected. This was carried out according to our hospital's protocols on medical history data access for scientific purposes, and with the authorization of the Medical Research Ethics Committee. Regarding the hernia itself, location (right, left, or bilateral), herniated organs within the thoracic cavity (liver, stomach, small bowel, large bowel, spleen, pancreas, and kidney), need for ECMO before surgery, and postoperative mechanical ventilation time were assessed. Operative reports were analyzed in all cases, and each hernia was allocated to one of the 4 categories established by the CDH Study Group classification[3] according to defect size – types A, B, C, and D, with type A being the smallest, and type D the largest.

For the quality of life study of these patients and those in the control group, the Short Form-36 (SF-36) quality of life survey was used. This survey has been validated for the Spanish population and may be completed by patients aged ≥ 14 years old. It consists of 36 items exploring 8 health areas[8,9]:

- **Physical function:** extent to which health limits physical activities such as self-care, walking, climbing stairs, leaning down, grabbing or carrying weight, and moderate and intense efforts.
- **Physical role:** extent to which physical health interferes at work and in other daily activities, including underperformance, restrictions in the type of activities undertaken, and difficulty in the normal development of such activities.
- **Body pain:** pain intensity and impact on daily work, either outside or at home.
- **General health:** personal health assessment including current health, future health perspectives, and resistance to disease.

- **Vitality:** sensation of energy and vitality vs. fatigue and exhaustion.
- **Social function:** extent to which physical or emotional health issues interfere in normal social life.
- **Emotional role:** extent to which emotional issues interfere at work or in other activities, underperformance, and less effort put at work.
- **Mental health:** general mental health, including depression, anxiety, behavioral control, and general well-being.

In addition, health condition changes observed in each patient over the last year were collected.

Data was collected and analyzed using the SPSS statistical software, version 15.0 for Windows. Contingency tables were used to assess medical history data. Regarding the SF-36 survey, the items generated a scale for each area which was recoded to come up with a 0 (worst quality of life) to 100 (best quality of life) value, using the algorithms recommended by survey score and assessment guidelines[10,11]. The 8 areas were analyzed by means of a descriptive study of frequencies, and mean and standard deviation were calculated. Owing to the number of patients who underwent CDH surgical repair, non-parametric tests were used for comparative study purposes – Kruskal-Wallis test for continuous variables, and Mann-Whitney U test for independent groups with quantitative variables.

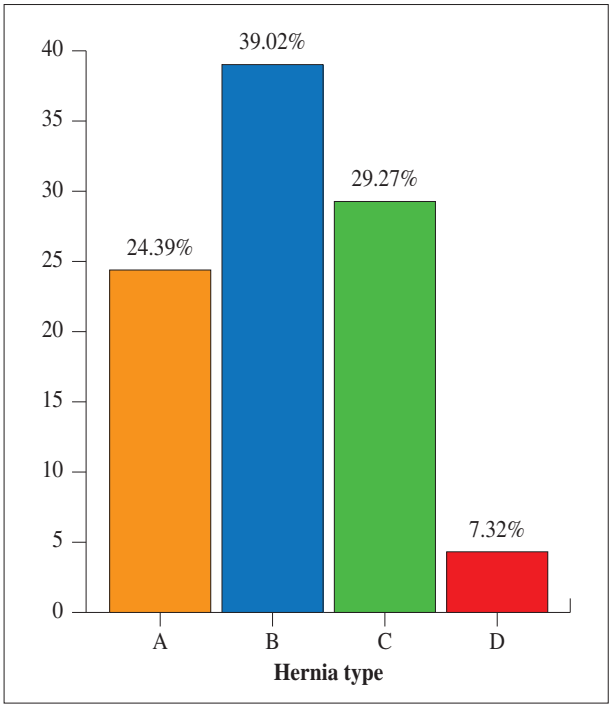

Figure 1. Hernia type distribution in our series according to the CDH Study Group classification..

RESULTS

Demographic data

From June 1997 to April 2004, a total of 41 patients (26 male, and 15 female) underwent congenital diaphragmatic hernia surgical repair. 12 of them died, which means overall survival was 70.73%.

There were 32 left hernias, 8 right hernias, and 1 bilateral hernia (78.9%, 19.5%, and 2.4%, respectively). According to the CDH Study Group hernia size classification, most patients had type B hernia (39%) (Fig. 1).

In 40 patients (97.6%), small and large bowel herniation was observed. Stomach herniation was found in 20 cases (48.8%), and liver herniation was noted in 28 cases (7 right lobe herniations, 11 left lobe herniations, and 2 full liver herniations). The spleen was herniated in 27 patients (65.9%).

In our series, 10 patients (24.4%) required extracorporeal membrane oxygenation before surgery. All patients needed mechanical ventilation postoperatively, with mean ventilation time being 13 ± 14.3 days (range: 1-90 days).

Surgical closure data was collected in 36 patients – the surgical report was not available in 4 of them as these cases were far back in time. In most cases (66.6%), primary closure was used. Braided suture was employed in 13 patients (12 polyester sutures and 1 silk suture, 2/0-4/0), and monofilament (polypropylene) suture was applied in 11 patients. A total of 12 patients required a polytetrafluoroethylene patch (Gore-Tex® Suture). Of the 29 survivors, recurrence was observed in 3 patients (10.3%), 1 of whom had a second recurrence.

When comparing hernia characteristics between survivors and non-survivors, significant differences were noted regarding need for preoperative ECMO, type of closure – primary closure or patch closure (Table I) –, and hernia size according to the CDH Study Group classification (Fig. 2).

Quality of life surveys

Most contact data of patients who underwent surgery more than 20 years ago was outdated. Of the 29 survivors, 8 could not be contacted – addresses and phone numbers had changed over the years, and they could not be contacted through social media, health insurance cards, or social security numbers. Difficulty was greater in older patients, and especially in those from other regions. Phone/email contact was successful in 21 out of the 29 survivors, 16 of whom accepted to complete the survey. The responses of 24 healthy adolescents within the control group were collected. Table II features the demographic data of both groups.

Regarding respondents who underwent CDH surgical repair, 13 had left diaphragmatic hernia, and 3 had right diaphragmatic hernia. In terms of size, most of them (9

Table I. Hernia characteristics and differences between survivors and non-survivors.

	Survivors	*Non-survivors*	*p value*
Number of cases	29	12	
Hernia side			0.088
• Left	25 (86.2%)	7 (58.3%)	
• Right	4 (13.8%)	4 (33.3%)	
• Bilateral	–	1 (8.3%)	
Preoperative ECMO	3 (10.3%)	7 (58.3%)	**0.03****
Postoperative mechanical ventilation time (days)	11.83 ± 7.27 (2-33)	15.92 ± 24.57 (1-90)	0.414
Hernia size			**0.001****
• A	10 (34%)	–	
• B	14 (48%)	2 (17%)	
• C	5 (17%)	7 (58%)	
• D	–	3 (25%)	
Type of closure			**0.001****
• Primary	24 (82.8%)	–	
• Patch	5 (17.2%)	7/7 (100%)*	

**5 patients died before surgery. **Statistical significance: p< 0.05.*

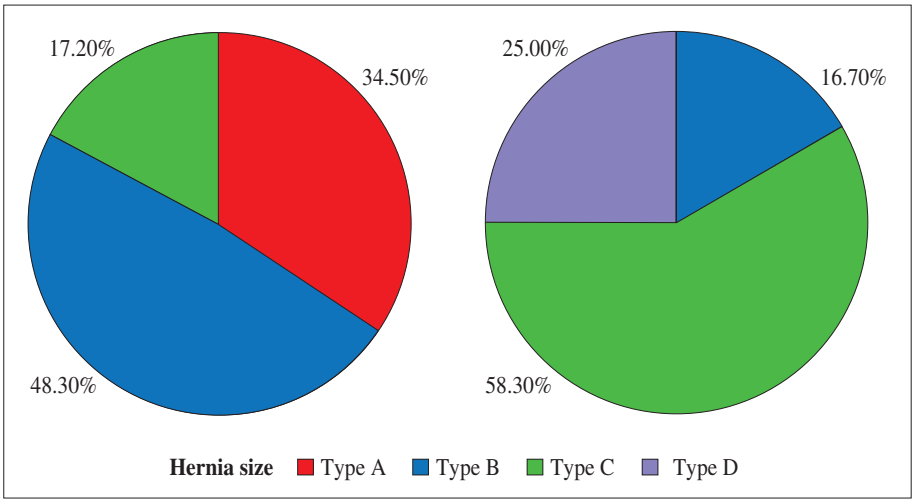

Figure 2. Hernia size in survivors and non-survivors according to the CDH Study Group classification (p=0.001).

Table II. General data of the two respondent groups: patients who underwent congenital diaphragmatic hernia (CDH) surgical repair, and healthy adolescents (control group).

	CDH surgical repair	*No CDH surgical repair*
Number of cases	16	24
Age (years) (mean ± standard deviation)	16.6 ± 2.07	15.8 ± 0.68
Age (years) (range)	14.2-20.8	14.3-17.3
Sex (male/female)	12/4	16/8

patients) had type B defect, 4 had type C defect, and 3 had type A defect. Only 2 respondents required ECMO neonatally.

Table III and Table IV feature survey data in each of the 8 areas. Values are expressed as mean ± standard deviation for the two groups studied, along with reference values in the relevant populations.

In all areas studied, perceived quality of life was lower in healthy adolescents than in patients who underwent CDH surgical repair, with differences being statistically significant in the vitality and mental health areas.

In the group of patients who underwent CDH surgical repair, no significant differences were found regarding quality of life according to sex, right or left hernia, need for ECMO neonatally, or hernia size.

Table III. Data found in the SF-36 survey physical areas.

	CDH surgical repair	*No CDH surgical repair*	*p*
Number of cases	16	24	
Physical function	96.25 ± 5.91	95.88 ± 5.83	0.838
Physical role	90.62 ± 25.62	90.44 ± 18.47	0.980
Body pain	79.56 ± 22.57	70.64 ± 19.53	0.186
General health	81.25 ± 22.21	69.85 ± 18.19	0.080

Values expressed as mean ± standard deviation.

Table IV. Data found in the SF-36 survey mental areas.

	CDH surgical repair	*No CDH surgical repair*	*p*
Number of cases	16	24	
Vitality	76.25 ± 21.87	58.82 ± 12.73	**0.008***
Social function	89.06 ± 20.85	83.08 ± 19.67	0.344
Emotional role	75.00 ± 37.52	63.72 ± 37.93	0.331
Mental health	78.00 ± 20.55	64.70 ± 16.46	**0.032***

*Values expressed as mean ± standard deviation. *Statistical significance: p< 0.05.*

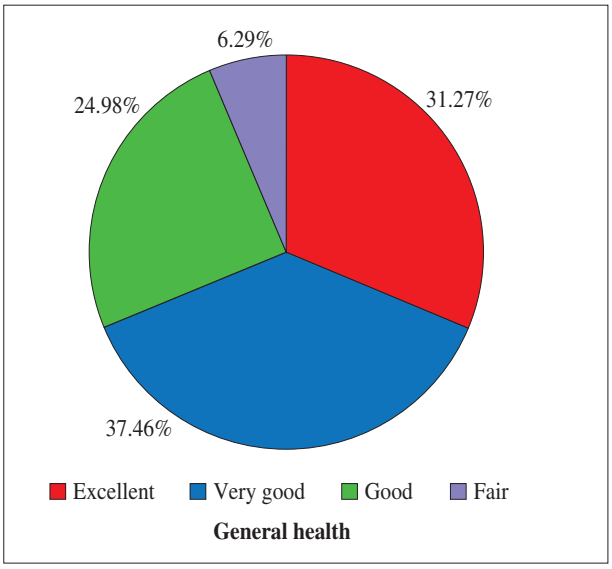

Figure 3. General health question results in patients who underwent CDH surgical repair.

Current general health assessment

The results from the SF-36 survey questions assessing current general health in the group of patients who underwent CDH surgical repair were the following:

- *Generally speaking, would you define your health as "Excellent," "Very Good," "Good," "Fair," or "Poor?"* Most patients (68.8%) who underwent CDH surgical repair replied their current health was "Excellent" or "Very Good" (Fig. 3).
- *In the last 4 weeks, to what extent have your physical health or emotional issues interfered in social activity – plans with relatives, friends, neighbors, or other people? "Nothing At All," "A Little," "Fairly," "Quite A Bit," or "A Lot."* 12 out of the 16 patients (75%) who underwent CDH surgical repair responded "Nothing At All," 2 patients (13%) responded "A Little," 1 patient responded "Fairly," and 1 patient responded "Quite A Bit."
- The answers to the other direct health assessment questions are featured in Figure 4.

DISCUSSION

This is the first Spanish study carried out in adolescents and young adults who underwent congenital diaphragmatic hernia surgical repair in the neonatal period. The objective was to assess and analyze quality of life in this particular population group.

Diaphragmatic hernia characteristics and neonatal management in our series were consistent with those found in the literature and in studies with a larger patient cohort. Most patients – 86.2% of survivors and 58.3% of non-survivors –had left diaphragmatic defect[13]. The others had right diaphragmatic defect, except for one patient from the non-survivor group who had bilateral diaphragmatic defect as part of Fryns syndrome[14]. Bilateral cases are known to be frequently associated with other congenital malformations[15]. In our series, most survivors had a small diaphragmatic defect (type A or B), whereas most non-survivors had a large diaphragmatic defect (type C or D). Therefore, hernia size was statistically related to worse prognosis[4].

Quality of life survey results

In our study, patients who underwent CDH surgical repair had a good perception of their own quality of life. Perception was better than that of patients without history of CDH from the control group. Surprisingly, no statistically significant differences were found between responses from patients with a more severe pathology at birth – larger defect size or need for ECMO neonatally – and responses from patients with less severe hernia. However, these results should be addressed with caution since the patient cohort was not large.

In all areas studied, the results were better in patients who underwent CDH surgical repair than in healthy

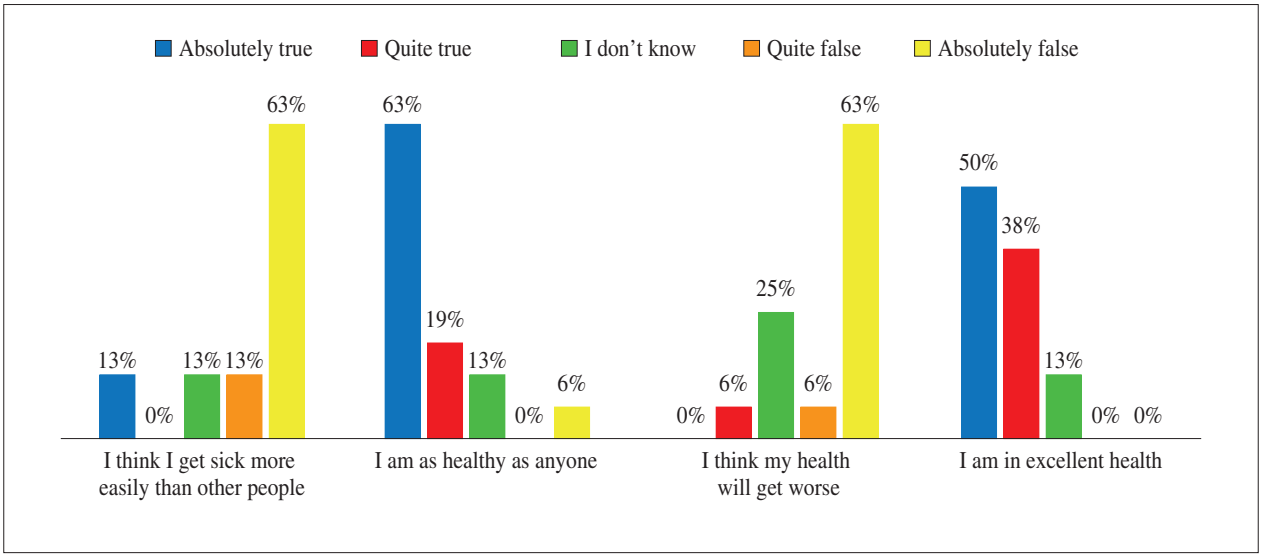

Figure 4. Horizontal axis: health assessment survey claims. Vertical axis: proportion of patients with CDH surgical repair who chose each answer.

patients, with differences being significant in the vitality and mental health areas. In our view, this is a result of various factors. First, responses from patients who underwent surgery were more extreme – "never," "always," "absolutely true," "absolutely false" – than those from healthy adolescents, which were mostly intermediate – "quite a bit," "sometimes," "I don't know." Possibly, our patients wished to demonstrate their current situation is normal and their CDH history does not impact quality of life. Second, understanding their personal experience with CDH and to what extent they are impacted by this pathology – which is treated at a very young age – is quite complex. Many are premature, and most of them have required few subsequent hospitalizations. In addition, even though contact with the hospital is maintained throughout childhood and their pathology is frequently and openly discussed with them and their families, the most traumatic moment occurs in a period when children have limited consciousness. Third, we ignore why non-included patients declined to complete the survey. Possibly, some of these adolescents have a lower quality of life and refuse to be exposed to any kind of survey – even an anonymous one. Fourth, especially in the past, an important "selection" was made in the first week of life as a result of high mortality rates in patients with severe pulmonary hypoplasia and treatment-resistant pulmonary hypertension. In other words, patients in a worse condition may have probably not survived[16].

Few studies in Europe and the US assess mid- to long-term quality of life of patients who underwent CDH surgical repair through surveys, and their results are similar to ours. In a study carried out in Finland with adult patients who underwent CDH surgical repair, 75% of respondents had high health-related quality of life (HRQL) scores, with no statistically significant relationship between hernia size and quality of life[17]. A study conducted in the Netherlands analyzed short-, mid-, and long-term quality of life, both in patients with anorectal malformations and with CDH. In this study, the SF-36 survey was used in patients ≥ 16 years old, with results being similar to ours in terms of perceived quality of life as compared to the general population[16]. In Texas, the quality of life of patients prenatally diagnosed with CDH and aged 2-11 years old at the time of the study was analyzed. Regardless of defect size, patients had a good quality of life, similar to that of the healthy population[18]. A study carried out in Sweden with patients aged 9-19 years old concluded that children who underwent CDH surgical repair had a health-related quality of life (HRQL) similar to that of healthy Swedish children. However, the relationship between defect severity and quality of life could not be excluded[19]. Understanding quality of life of adolescents and young adults with history of congenital diaphragmatic hernia can help advise parents regarding CDH fetal diagnosis. It will also allow parents to have a clear idea of what their babies' lives will be like once discharged from hospital.

Given that our study had a small sample size – with only 16 patients who underwent CDH surgical repair completing the survey –, these results should be addressed with caution. Most patients from other Spanish regions could not be contacted more than 15 years later, and 5 out of the 21 who were successfully contacted refused to complete the survey. Probably, data was collected from patients with less severe CDH or fewer sequels, which means it cannot be extrapolated to the whole CDH population and should be carefully dealt with. In addition, the sample was collected in a single institution, which means

perinatal management, surgical techniques, and postoperative management and follow-up may slightly differ from those applied in other healthcare facilities – despite using widely known, tried and tested protocols. The study should be extended to other institutions dealing with large amounts of CDH cases in Spain to increase conclusion validity and extrapolate results to the whole population of patients who underwent CDH surgical repair, including a greater number of more severe cases.

In conclusion, the results from our study are similar to those from the studies conducted in other countries with larger patient cohorts[16-19], which allows a modest positive approach to the quality of life of these patients to be adopted. Even though increased survival of CDH patients has caused long-term morbidity to surge, adolescent survivors claim their health-related quality of life is similar to that of youngsters of their age. Additionally, greater severity of initial CDH is not seemingly associated with lower quality of life in the future.

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