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PRENATAL **DIAGNOSIS**

ORIGINAL ARTICLE

Does website-based information add any value in counseling mothers expecting a baby with severe congenital diaphragmatic hernia?

Alexander C. Engels[1], Philip DeKoninck[1,2,3], Johannes L. van der Merwe[4], Tim Van Mieghem[1,2,3], Petra Stevens[3], Beverley Power[5], Kypros H. Nicolaides[6,7], Eduard Gratacos[8,9] and Jan A. Deprest[1,2,3]*

[1]Centre for Surgical Technologies, Faculty of Medicine, Katholieke Universiteit Leuven, B-3000 Leuven, Belgium
[2]Department of Obstetrics and Gynaecology, Division Woman and Child, Fetal Medicine Unit, University Hospitals, B-3000 Leuven, Belgium
[3]Department of Development and Regeneration, Research Unit Fetus Placenta and Neonate, Faculty of Medicine, Katholieke Universiteit Leuven, B-3000 Leuven, Belgium
[4]Department of Obstetrics and Gynaecology, Stellenbosch University and Tygerberg Hospital, Cape Town, South Africa
[5]CDH-UK, The Denes, Norfolk, UK
[6]Harris Birthright Research Center, King's College Hospital, London, UK
[7]Department of Fetal Medicine, University College Hospital London, London, UK
[8]Department of Maternal–Fetal Medicine, ICGON, Hospital Clinic-IDIBAPS, University of Barcelona, Barcelona, Spain
[9]Centre for Biomedical Research on Rare Diseases (CIBER-ER), Barcelona, Spain
* Correspondence to: Jan Deprest. E-mail: jan.deprest@uzleuven.be

ABSTRACT

Objective We aimed to measure whether website-provided information about congenital diaphragmatic hernia (CDH) and fetal therapy for severe cases provides added value compared with clinical counseling of parents.

Methods This is a single center study in 102 couples who earlier opted for fetoscopic endoluminal tracheal occlusion (FETO) because of isolated severe CDH. They were asked to fill out an anonymized web-based survey of 12 questions. Then, they were offered access to information on the web pages of the randomized Tracheal Occlusion to Accelerate Lung Growth (TOTAL) trial. One week later, their appreciation was measured again by a second questionnaire.

Results Eighty-two (80%) parents completed the first questionnaire, and 48 (47%) completed the entire survey. Several items became more clear to the parents after reading the website, such as the length of hospital stay (23.2% prior to web information, 60.4% after; $P = 0.004$), maternal risk, or the requirement of fetal anesthesia for FETO (43.9% resp. 79.2%; $P = <0.001$).

Conclusion Complementing prenatal counseling on CDH and FETO by standardized information via website is perceived by parents as of added value. Maternal risks and the need for fetal medication need more clarification during the verbal counseling prior to prenatal interventions. © 2013 John Wiley & Sons, Ltd.

Supporting information may be found in the online version of this article.

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INTRODUCTION

Congenital diaphragmatic hernia (CDH) is a sporadic defect with a prevalence at birth of approximately 2.7 in 10,000. According to the international CDH registry, mortality in fetuses diagnosed with isolated CDH is around 30% mainly because of pulmonary hypoplasia and pulmonary hypertension.[1,2] To improve the prognosis in those, attempts were made to accurately prenatally predict lethal pulmonary hypoplasia, as well as doing a prenatal intervention to reverse pulmonary hypoplasia. Current clinical fetal surgery programs essentially use ultrasound and/or MRI for patient selection by determining the position of the liver and measuring the lung size.[3,4] The latter is typically performed using 2-D ultrasound to measure the lung-to-head ratio (LHR) and expressed as a function of what would be expected in a gestational aged matched normal control (observed/expressed LHR; in %).[5,6] There is already ample experience with percutaneous fetoscopic endoluminal tracheal occlusion (FETO) with a detachable balloon.[7–9] In the European programs, FETO actually involves two steps, that is, balloon placement at around 26–29 weeks and its removal at 34 weeks (‘plug–unplug’ procedure);

the latter is essentially performed to improve lung maturation after stimulated growth.[10–12] In our experience, prenatal balloon removal more than 24 h before birth significantly increased survival, although the group of Ruano obtains similar survival rates of around 50% by removing the balloon at the time of birth.[9,13,12]

Parents expecting a baby with a severe congenital anomaly, such as CDH, want and should obtain as much information as possible about the condition, its natural history, and its management options.[14,15] An active, informed role of the parents in decision making can improve the compliance and therefore have a positive effect on the outcome of prenatal management of a fetus with a congenital anomaly.[16] Typically, a multidisciplinary team will provide as comprehensive as possible information to the family.[14] Next to that, families may seek further information. A recent study reported that 64% of parents are regular users of the Internet for medical information.[17] Having that in mind, we aimed to complement the verbal information given to the parents at the time of evaluation and counseling, with information via the Internet. For that purpose, we developed a website on isolated CDH and its prenatal management, as an additional tool for counseling women. This website was developed by physicians and reviewed initially by the midwife-case manager of our fetal therapy program, a number of midwives and representative lay people, familiar with CDH such as parents working at the British patient group 'CDH-UK'. To evaluate the added value by this website, we set up a web-based survey amongst parents we earlier saw at our unit prior to fetal surgery for CDH. The design of this website was sponsored by a European Commission 6th framework project on perinatal tissue engineering (www.eurostec.eu).

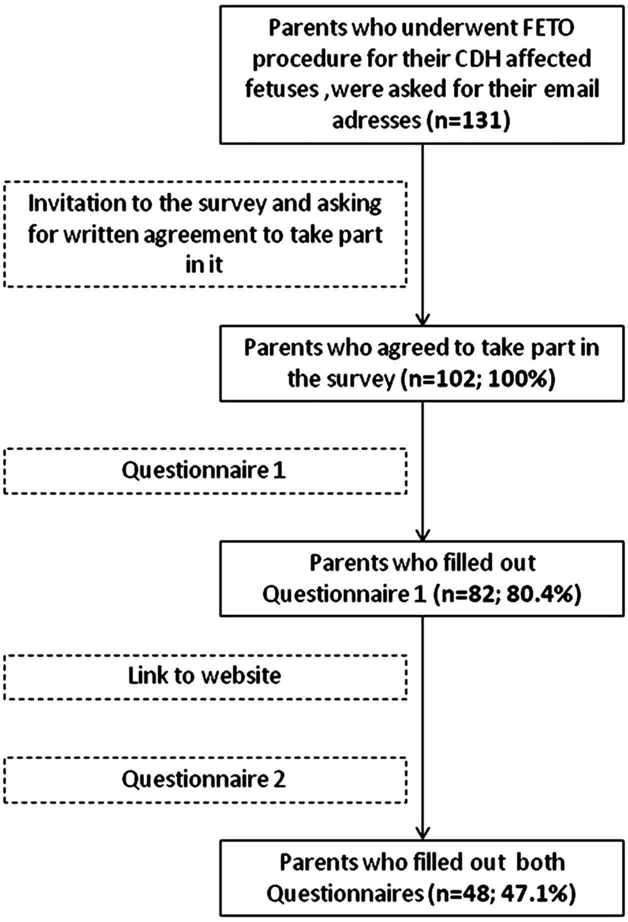

Figure 1 Diagram of the survey process. Boxes with discontinuous lines show what has been given by the survey authors to the participants

MATERIAL AND METHODS

We first contacted by telephone all mothers and, where applicable, their partners who underwent fetoscopic surgery for severe CDH in the fetal medicine unit in Leuven until end of 2010 ($n = 131$; Figure 1). At that time, they were counseled about the procedure, its known maternal side effects and known fetal outcomes. During this consultation, we did not yet have a specific website with information on the pathology or the procedure. They were asked whether they were willing to participate in this study and whether they had access to Internet and had an email account. In case of verbal agreement ($n = 102$; 77.9%), immediately after, a web link with a consent form was sent to them, which they could confirm in writing their participation. This initialized access to a first empirically designed questionnaire. It contained three groups of questions with a total of 14 items (Supporting information 1). The first group of questions was dedicated to polling about how patients felt if they were informed by their physicians and if they understood the given information (example: 'Did you understand what the risks of a balloon insertion procedure were for the fetus?') at the time of preoperative evaluation and counseling at our fetal medicine unit. We used six 5-point Likert scales (Table 1) with optional answers ranging from 'Yes, I understood exactly' to 'No I didn't understand at all'. A second group of questions was testing their knowledge about CDH and the option of prenatal therapy for this condition. There were five items that could be answered either by *yes*, *no*, or *unsure*. One such question was, for example, 'Congenital Diaphragmatic Hernia is a common disease in live born children'. The third group was a single question about the overall satisfaction with the verbal information given by the physician at the time of the preoperative evaluation and counseling. The answer was categorized as either *yes* or *no*, and the parents were invited to add some free text on this item.

One week after completion of the questionnaire ($n = 82$; 80.4%), a new link was sent that brought the parents to the public pages of the Tracheal Occlusion to Accelerate Lung Growth (TOTAL) trial website, which offers information on CDH to expecting parents, as well as on the concept, and further details on the randomized so-called TOTAL trial (www.totaltrial.eu/?id=13). The information was available in English, French, German, and Dutch. This website also contains drawings about the condition and how the fetal intervention is performed. They were asked to read the information on that website carefully. One week after the participants obtained that link, they were asked to fill out

Table 1 Statistical analysis of questions 1, 2, 3, 4, 5, and 6. The Mann–Whitney rank sum test was used for this purpose. A lower rank indicates a higher knowledge, as answers as 'Yes, I understood exactly' was grade 1 and answers as 'No, I didn't understand at all' were grade 5. This table indicates the quality of information either at the time of evaluation (questionnaire 1) or after been additionally been informed by the website (questionnaire 2)

Question	Median rank			
	Questionnaire 1	Questionnaire 2	Z value	P value
1. If your child was operated before birth: Do you know the reasons why your child was operated before birth?	69.6	58.5	−3.014	0.003*
2. Did you know ahead of time how long you would have to stay in hospital for the procedure of balloon insertion?	72.5	53.5	−2.880	0.004*
3. Did a doctor/counselor explain you clearly the condition of your fetus?	62.4	70.8	−1.686	0.092
4. Did you understand what the risks of a balloon insertion procedure were for the mother?	75.5	48.3	−4.206	<0.001*
5. Did you understand what the risks of a balloon insertion procedure were for the fetus?	62.9	69.8	−1.335	0.182
6. Where you informed that there are no known future risks for you as the mother?	70.6	56.7	−2.173	0.030*

again a second questionnaire, containing questions identical to the initial questionnaire; however, the third group of questions, relating to the satisfaction of women with the provided information, was expanded with two additional questions about the parent's satisfaction with the information offered on the TOTAL website. The first additional item asked whether the information provided by the website offered any *additional* information or knowledge. The second additional item was about whether, in retrospect, the website had an influence on their thoughts about fetoscopic balloon insertion for this condition. The latter two items could again be answered by either *yes* or *no*, and the parents were invited to add some additional free text. In the second questionnaire, the mothers were also asked if they read the whole website, to ensure that they had visited the web-based information.

All questionnaires were filled out via the Internet over a period at the choice of the parents. We used a token system so that each invited participant could only answer once. Reminders were sent if answers were lacking, 1 week after the initial invitation. The answers came back to us in an anonymous way. In other words, the investigators were blinded to the individual outcome of the pregnancy, the time point the women were treated, and, at the second questionnaire, to the answers given to the first questionnaire. The web-based survey was created with the software LimeSurvey (v. 1.92, The LimeSurvey Project Team, GPL). Survey invitations were also sent to the participants by this survey application.

Statistical analyses were performed using IBM SPSS Statistics v.20 (IBM Software Inc., Armonk, NY, US). Mann–Whitney rank sum test was applied to the items from questionnaires 1 and 2, which were ordinal Likert scale scores. A lower rank indicates a higher quality of information (answers = 'Yes, I knew/understood exactly', rank = 1), whereas an answers such as 'No, I didn't understand at all' generates a rank = 5. The data displayed were, in that case, the median of the rank sum. Nominal questions (yes, no, and unsure) were tested using the Pearson's chi-squared test (questions 7, 8, 9, 10, and 11). A P value below 0.05 was considered significant. Ethical approval for this study was obtained from the Institutional Ethics Committee.

RESULTS

Of the 131 initially contacted parents by telephone, 102 (77.9%) were consenting and hence became actual study participants. The median maternal age was 32 years, and the average interval between the preoperative evaluation and the survey was 2.4 years (range: 0.5–5.5 years). The total survival rate of babies from study participants was 41%, but as this was an anonymized study, we could not correlate pregnancy outcome to any of the answers given. The first questionnaire was answered by 82/102 (80.4%), whereas 48/102 (47.1%) answered the second questionnaire. In Figure 2, we display the different answers observed between the two questionnaires for the items 1 to 11. The anonymous nature made it also impossible to make a pairwise analysis of answers to questionnaire 1 and 2.

Answers to four out of six (questions 1, 2, 4, and 6) questions on the provided information were suggesting that additional web-based information was perceived as an added value (Table 1). More in detail, the parents felt that the website added content and/or quality of information about (1) the reasons for the fetal operation, (2) the need to stay in the hospital, (3) the limited maternal risks of the operation and (4) on her future fertility. Whereas 82.9% of respondents said they knew exactly why their child was operated before birth, 100% said so after they had read the website ($P = 0.003$). Before reading the website, only 23.2% knew how long they would have to stay in the hospital, which surged to (yet only) 60.4% after reading the digital information ($P = 0.004$). 41.5% of the parents were unsure if and what the risks of balloon insertion were for the mother. After consulting the website, 10.4% remained unsure about that, whereas 58.3% quoted that they now had totally understood the risks for the mother ($P = <0.001$). In contrast, the parents felt that the clarity of the information on the condition itself (Q3) or the fetal risks of the operation (Q5) was no different from what was offered when being counseled verbally by the physician. As to their *knowledge* about CDH and the fetal intervention, apparently, the issue of the need to anesthetize the fetus was unclear during the initial

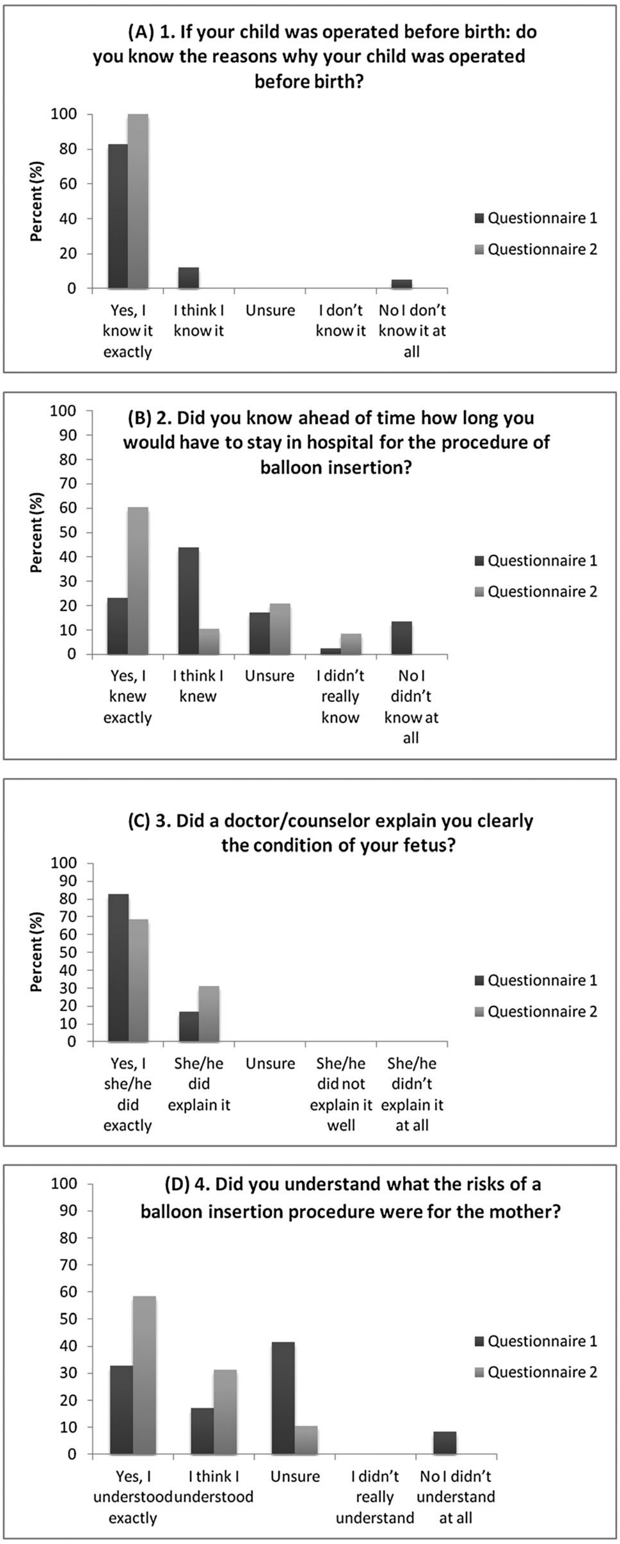

Figure 2 (A–K) Results of the 11 questions asked within the survey. The results are shown in percentages for each answer

counseling, hence became obvious by reading the website. Of the respondents, 43.9% answered that the statement, 'For fetal balloon insertion, it is necessary to anesthetize the mother but not the fetus', is false prior to reading the website, and that went up to 79.2% ($P = <0.001$) (Table 2). Before reading the website, 15.9% did not really know that there are no known future risks for the mother undergoing FETO. After reading the website, still only 45.8% knew that exactly ($P = 0.030$). For the other questions, there were no significant differences.

There were two questions that were only asked in the second questionnaire. Of the respondents, 100% said that the website information was of added value. Conversely, only 4.1% said that the information did change their thoughts about fetal therapy for this condition and therefore might have changed their decision to undergo FETO. In the free-text field, one mother wrote the following: 'We did not learn about fetal anesthesia', 'I didn't know what substances were administrated to the baby before the procedure', and 'On these webpages I found out the baby received drugs in order to sleep and not feel uncomfortable.' These were what one respondent answered explaining what additional value the website meant. Another one experienced the verbal consultation as 'too fast'. Additionally, all patients quoted that they had read the whole website.

DISCUSSION

This study shows that offering additional web-based, written information adds value to the verbal information given by prenatal counselors to parents considering fetal tracheal occlusion. More in particular, women were incompletely aware of fetal anesthesia and maternal risks. In our study population, the additional information would in retrospect not have altered their decision making.

Strikingly, our data demonstrate that the maternal aspects were underestimated when counseling our patient prior to the intervention, despite our perception that we counsel unbiased and fair. Fortunately, it was possible to improve the understanding of maternal risks of the FETO procedure by offering an informative website ($P = <0.001$) as well as improving the understanding of the maternal future risks ($P = 0.030$). Further, though perhaps less clinically relevant, giving more extensive information about the duration of hospitalization was named by study participants as added value ($P = 0.004$). Such information may indeed be important in terms of logistic and financial consequences to parents opting for fetal surgery. Therefore, we emphasize now more on these aspects during the counseling.

Another striking observation is that not all women understood the need for fetal immobilization and anesthesia. Fetal immobilization is a requirement for this procedure, and most fetal medicine specialists would provide the fetus pain relief as well, if the procedure may be a painful stimulus to the fetus.[18] The percentage of participants who knew that the fetus would require anesthesia raised from 43.9% to 79.1% by giving additional information, which confirms that the concept of fetal pain is not well spread in the community.

Though a first step to improved understanding of perspective patient's perception of fetal surgery, this study has a number of shortcomings, and its findings cannot be generalized. First, it is a retrospective and uncontrolled study. Also, it was conducted anonymously, which precluded us from doing a pairwise analysis of results, nor could we link patient's perception

Table 2 **Patient's knowledge about the condition 'Congenital Diaphragmatic Hernia' and fetal therapy for it, either at the time of evaluation (questionnaire 1) or after been additionally been informed by the website (questionnaire 2). For the statistical analysis of questions with 'true', 'false', and 'unsure' as answers, we did the Pearson's chi-squared test**

	Right statements (%)		
Question	Questionnaire 1	Questionnaire 2	*P* value
7. Congenital Diaphragmatic Hernia (CDH) is a common disease in life-born children.	75.6	79.1	0.299
8. CDH is characterized by a defect in the diaphragm of the fetus.	100	100	1.0
9. Although CDH babies have a defect in the diaphragm, their lungs are essentially normal.	92.7	100	0.055
10. For fetal balloon insertion, it is necessary to anesthetize the mother but not the fetus.	43.9	79.2	<0.001*
11. The balloon is preferentially removed before birth because it has a number of advantages.	91.5	100	0.115

to the actual outcome of pregnancy. However, most women pointed to the added value of the written information. Further, only a limited number of women gave very specific misconception of their prenatal understanding of very specific fetal (pain) and maternal (complications) aspects of the procedure. Also, it focuses on only one single anomaly and a very specific minimally invasive fetal surgical procedure, with a less than ideal outcome and unproven benefit. Obviously, other interventions, with different maternal risks, or expected improvements of postnatal outcome, as well as the presence of level I evidence of the efficacy of the offered fetal therapy, do play a role in the nature of information provided to women. The relatively low response rate at the second questionnaire also needs to be mentioned. This might reflect that only positive responders from the first questionnaire filled in the second questionnaire and thus could alter the outcomes of the study. The knowledge that there will be a second questionnaire after reading the website might have led to a more detailed study of the website; hence, the results of our study may not be generalized to a wider population. As the questionnaires were answered online at home, it was not possible for us to completely prevent the usage of foreign information sources, although we strongly asked the mothers in the introduction of the questionnaires not to use any other information other than the ones provided.

CONCLUSION

In summary, we demonstrated that specific aspects of maternal safety as well as fetal pain relief were underemphasized during our counseling and/or were not well understood by the women to undergo FETO. This lack of information can be solved by a website with evidence-based information, although it logically leads to immediate adaptation of the verbal information we provide to expecting parents, prior to FETO.

WHAT'S ALREADY KNOWN ABOUT THIS TOPIC?

- Parents expecting a baby with CDH should receive as much information as possible about this condition and the treatment options to improve their compliance and therefore the children's outcome. As more and more parents are using the Internet as an information source, we should use the Internet as a complementation in prenatal consultation.

WHAT DOES THIS STUDY ADD?

- This study provides evidence that website-based information can complement verbal prenatal counseling on fetoscopic procedures as the FETO treatment for CDH. Especially, the maternal risks and the need for fetal medication need to be explained more clearly to the patients during prenatal consultation.

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