Journal of Pediatric Surgery xxx (2017) xxx–xxx

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

Expert surgical consensus for prenatal counseling using the Delphi method☆,☆☆

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ARTICLE INFO

Article history:
Received 12 May 2017
Received in revised form 27 September 2017
Accepted 21 November 2017
Available online xxxx

Key words:
Congenital diaphragmatic hernia
Congenital pulmonary airway malformation
Prenatal counseling
Delphi method
Expert opinion

ABSTRACT

Background: Pediatric surgeons frequently offer prenatal consultation for congenital pulmonary airway malformation (CPAM) and congenital diaphragmatic hernia (CDH); however, there is no evidence-based consensus to guide prenatal decision making and counseling for these conditions. Eliciting feedback from experts is integral to defining best practice regarding prenatal counseling and intervention.
Methods: A Delphi consensus process was undertaken using a panel of pediatric surgeons identified as experts in fetal therapy to address current limitations. Areas of discrepancy in the literature on CPAM and CDH were identified and used to generate a list of content and intervention questions. Experts were invited to participate in an online Delphi survey. Items that did not reach first-round consensus were broken down into additional questions, and consensus was achieved in the second round.
Results: Fifty-four surgeons (69%) responded to at least one of the two survey rounds. During round one, consensus was reached on 54 of 89 survey questions (61%), and 45 new questions were developed. During round two, consensus was reached on 53 of 60 survey questions (88%).
Conclusions: We determined expert consensus to establish guidelines regarding perinatal management of CPAM and CDH. Our results can help educate pediatric surgeons participating in perinatal care of these patients.
Level of Evidence: V.

Birth defects are one of the leading causes of infant mortality, accounting for more than 20% of all infant deaths, and creating an ongoing source of morbidity for many afflicted children who survive infancy [1]. Prenatal counseling plays an increasingly important role in influencing decision making during pregnancy. For surgical anomalies, referral to appropriate surgical specialists in addition to maternal–fetal medicine consultants is critical. There are several reports in the literature of prenatal consultation for certain congenital anomalies leading to increased anxiety or unrealistic expectations. In some cases, decisions regarding termination of pregnancy may be based on erroneous information [2,3].

Congenital diaphragmatic hernia (CDH) and congenital pulmonary airway formation (CPAM) are two diagnoses for which prenatal consultation can have an important impact on decision making, ranging from prenatal intervention to location and mode of delivery [4,5]. Technical advances in prenatal diagnosis have led to increased accuracy and the ability to detect anomalies earlier in pregnancy, and many of these conditions are surgically correctable [6]. Pediatric surgeons, along with maternal fetal medicine specialists and neonatologists, play an essential role in interpreting these findings, conducting prenatal consultation, and participating in perinatal decision making [7]. This can lead to important decisions, ranging from where and how the baby will be delivered to considering an in utero intervention or terminating the pregnancy [1,8]. Although there are some data to drive this decision-making process, there is lack of clear consensus in the literature on

☆ Conflicts of interest: None.
☆☆ This work was supported by an Institutional Development Award (IDeA) from the National Institute of General Medical Sciences of the National Institutes of Health under grant number U54-GM104941 (PI: Binder-Macleod).
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https://doi.org/10.1016/j.jpedsurg.2017.11.056
0022-3468/

many aspects of these decisions. Care of these patients is therefore often guided by expert opinion and experience. It is critical that pediatric surgeons, upon completion of their training, be able to provide evidence-based information in a way that will allow future parents to make decisions consistent with their values, and set up realistic expectations as to how the anomaly will impact the life of their child.

There are several obstacles to accomplishing this important goal. First, there are limited long-term outcome data on children with congenital anomalies; therefore, much of what may be shared is the anecdotal experience of the surgeon. Second, a recent survey of pediatric surgery fellowship graduates showed that 47% of pediatric surgeons felt underprepared to perform prenatal consults, citing that there was inadequate exposure to prenatal consultation during fellowship (with 54% of them participating in five or fewer prenatal consultations during their training), and a lack of clear resources to guide prenatal decision making and counseling [9].

Eliciting feedback from pediatric surgical experts is integral to defining best practice and standardizing fetal counseling content and intervention. The Delphi method is a validated scientific approach to solving a complex problem through expert consensus [10,11]. It builds consensus by submitting a series of questionnaires to a panel of identified experts, permitting involvement of geographically distant and informed individuals [12,13]. Anonymity is inherent to the method, designed to offset conventional means of pooling opinions and avoid the introduction of bias through a particular vocal or dominant individual or group of individuals. In contrast to other data-gathering and analysis techniques, the Delphi method employs multiple iterations in the feedback process, allowing and encouraging the selected experts to reassess initial judgments about the information provided in previous iterations. Thus, in a Delphi study, the results of previous iterations regarding specific statements and/or items can change or be modified by individual panel members in later versions based on their ability to review and assess the comments and feedback provided by the other Delphi panelists. A Delphi method expert guides the scientific process of defining expert consensus.

The aim of this study was to create and facilitate a model of online Delphi consensus survey on perinatal management, using CDH and CPAM as examples. We identified areas of discrepancy and controversy in the literature and assembled a panel of experts to complete a Delphi consensus survey to develop consensus-based recommendations regarding terminology, diagnostic work-up, delivery plans, postnatal management, and content that should be discussed during prenatal consultation for CDH and CPAM.

1. Methods

1.1. Study population

Members of the American Pediatric Surgical Association (APSA) Fetal Diagnosis and Therapy Committee, the North American Fetal Therapy Network (NAFTNet) Steering Committee and Executive Board, the Society for Maternal and Fetal Medicine, and the American Academy of Pediatrics (AAP) Committee on the Fetus and Newborn were contacted via email. Each of these individuals was asked to name other individuals who they would consider experts in prenatal consultation. Only pediatric surgeons (total of 78 surgeons) were included in the final expert list. We chose to limit participation to pediatric surgeons since our goal in conducting this survey was to define content to be discussed during pediatric surgical consultation.

1.2. Survey development and administration

The current literature on the perinatal surgical care of CPAM and CDH was reviewed. Areas of discussion regarding prenatal imaging practice, interpretation of imaging findings, indication for fetal intervention, and critical points of prenatal counseling, were identified. A questionnaire was created to address these questions with the help of an expert in prenatal consultation and an expert in the Delphi method. Questions included binary (yes/no) responses followed by scaled measures to assess a participant's strength of agreement. Select multiple-choice questions evaluated optimal time frames for diagnosis and treatment; cutoffs for test results; and optimal choice of diagnostic tests, intervention, and treatment (Table 1). The survey was then pilot-tested by two additional pediatric surgeons who were experts in prenatal consultation.

The survey was administered online using Research Electronic Data Capture (REDCap), a secure web application for building and managing online surveys and databases [14]. An invitation email was sent followed by a series of weekly reminder emails. After the initial round of the survey, items that achieved consensus were summarized and included in the second round, allowing respondents to agree or disagree. Items that did not reach consensus in the first round were broken down into additional questions based on comments from first-round respondents. Final consensus was defined as 80% agreement for binary responses and greater than 50% agreement for multiple-choice questions. After round two, it was determined that consensus had been reached, or that two answers to a question had equal weight (divided expert opinion), and no additional rounds were administered. Open-ended questions were included to explore experts' opinions when they did not agree with any of the answer choices offered. All research procedures were approved by the institutional review board.

1.3. Data analysis

The statistics used in Delphi studies are measures of central tendency (means, median, and mode) and level of dispersion (standard deviation and inter-quartile range) to present information concerning the collective judgments of respondents [15]. Consensus on a topic was determined based on the percentage of responses within a prescribed range and by measuring the stability of subjects' responses in successive iterations. There were certain items for which we did not reach the 80% threshold or >50% agreement, but dissenters voiced their reasons in the open-ended response section included for each survey item, and these views are represented in the final summary of findings. For those items that lacked consensus, multivariable logistic regression (consensus—yes/no) was used to assess whether there were demographic characteristics that were associated with lack of consensus. Demographic characteristics of the respondents included age, gender, race,

Table 1
Delphi survey content.

CDH	• What imaging should be obtained prenatally? How often?
	• How should prenatal imaging be used to stratify severity?
	• When should fetuses be referred to fetal treatment center? Where should fetuses be delivered? Scheduled vs spontaneous? How does this vary based on stratification of severity?
	• What should be discussed during prenatal consultation in terms of long-term outcomes?
	• Should minimally invasive surgery (MIS) repair be considered?
CPAM	• What is the proper terminology for discussing congenital lung lesions?
	• What is the natural history in terms of growth, plateau, and regression prenatally?
	• What imaging should be obtained prenatally? How often?
	• How should severity be stratified?
	• What is the threshold for referral to a fetal treatment center? For maternal steroid administration? For fetal intervention for solid and cystic lesions?
	• Where and when should the mother deliver? Scheduled vs spontaneous? How does this vary based on stratification of severity?
	• Should observation without plan for resection be offered in asymptomatic patients?
	• Should segmentectomy rather than lobectomy be performed if technically feasible?

CDH, congenital diaphragmatic hernia; CPAM, congenital pulmonary airway formation.

Please cite this article as: Berman L, et al, Expert surgical consensus for prenatal counseling using the Delphi method, J Pediatr Surg (2017), https://doi.org/10.1016/j.jpedsurg.2017.11.056