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Lethal fetal abnormalities: how to approach perinatal palliative care?

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Abstract

Objective: Some of the antenatally diagnosed fetal pathologies are unlikely to get compatible with life. Still some women choose to continue with pregnancy. Subsequently, perinatal palliative care (PPC) has become a constructive demarche in such situations. Our study, based on a multicentric survey, reports some cases of fetal pathologies considered as lethal according to perinatal professionals and reveals the decisional process in each case.

Methods: We sent by emails a questionnaire to 434 maternal–fetal medicine specialists and fetal care pediatric specialists at 48 multidisciplinary centers for prenatal diagnosis.

Results: The participation rate was 49.3%. In total, 61 obstetric–gynecologists and 68 neonatologists completed the survey. The results showed that 35.4% of the pregnant women asked for the continuation of pregnancy and 24.7% asked for the termination of pregnancy. More than half of professionals (52.9%) took the initiative of informing women about the options for birth support (including PPC), while 32.7% of obstetric gynecologists did not take this initiative versus 10.2% of neonatologists (p < 0.01).

Conclusion: This study demonstrates the absolute need to provide PPC training for professionals and to standardize its practices.

Keywords

Ethical issue, fetal abnormality, information, palliative care, perinatal medicine

History

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Introduction

Severe congenital morphologic anomalies are one of the main causes of infantile morbidity and mortality. Some of these antenatally diagnosed pathologies are unlikely to get compatible with postnatal survival. Some women choose to continue with the pregnancy. In this context, perinatal palliative care (PPC) seems to be a constructive demarche to offer in such situations [1]. It constitutes, at international level, a new clinical practice where decision dilemmas take part: prognostic uncertainty, possible prolonged survival and attachment to the infant. In a previous study, we revealed the real place given to the concept of neonatal palliative care in the routine antenatal care [2]. That study showed the complexity of this issue and the difficulty in defining it. The practices discussed were multi-challenged (on professional, informational and ethical levels) and, therefore, questioned the necessity of having a multidisciplinary approach. In fact, neonatal palliative care imposes on the medical team and parents to accept the eventuality of the child’s death, even if this is neither imminent nor certain. The professional, being unable to cure, has now to master ‘‘caring’’; it is now a question of taking good care of the child (caring as opposed to curing) [3]. From a purely medical point of view, the conditions leading to a rapid postnatal death are relatively rare. As described by Leuthner [4], the majority of prenatal diagnoses need to be confirmed at birth. For many of these diagnoses, only the clinical progress of the patient will determine whether palliative care is the best or indeed the only option. Not all fetal abnormalities are life-threatening at birth.

As a continuation of our previous work, we aim in this study to report the fetal pathologies considered as lethal by perinatal professionals working in French multidisciplinary centers for prenatal diagnosis (MCPD) and to explore the decisional process in practice with the accordingly implemented information.

Materials and methods

Our research team designed an opinion survey and distributed the questionnaire electronically for a period of nine months. The French Advisory Committee for Information Processing in Health Research expressed their admiration for this research (reference number: 12.154).

We used a database provided by MCPDs (a master file available in the website of each center) in order to select a national sample of perinatal professionals working in MCPD in France. We obtained 434 email addresses from 48 MCPDs. The perinatal professionals were divided into two groups: obstetrician/gynecologist (OBS/GYN) group and neonatologists group. We could develop a questionnaire based on a qualitative study that used focus groups from two French
MCPDs [2]. Anonymity was strictly respected. Parts of this questionnaire focused on how lethal fetal pathology is perceived by the perinatal professionals and explored the associated practices. Each of the investigated professionals reported two cases of lethal fetal pathology and mentioned the information they provided to the concerned woman. This was a simulation exercise; it was approached with this question: “for your last two a diagnosed lethal fetal pathology cases, what was the discussion you made with the pregnant woman?” Then, for each pathology proposed by the professional, seven questions (Yes or No) revealed the information delivered to the pregnant woman: Question 1: Did you personally announce the diagnosis to the patient?; Question 2: According to you, was the information given in a multidisciplinary way?; Question 3: Did you take the initiative of informing the pregnant woman about the options for birth support, without waiting for her own request?; Question 4: Did she spontaneously make a request for termination of pregnancy?; Question 5: Did you spontaneously make a request for continuation of pregnancy?; Question 6: Did you spontaneously clarify the legal framework in this context?; Question 7: Did you spontaneously mention the possible scenarios in case of continuation of pregnancy and birth?

Qualitative variables are presented as numbers and percentages, quantitative variables as means and standard deviations. Comparisons were made using the chi-square test or analysis of variance (ANOVA), depending on the type of variable. Nonparametric tests were applied for variables with non-normal distribution. The threshold of significance of the tests was fixed at 5%. Statistical analyses were carried out using SPSS version 18 software (Marseille, France).

Results

Forty-two MCPDs took part (87.5%) in the study and the participation rate was 49.3%. In total, 61 OB/GYN and 68 neonatologists completed the survey, yielding 73.5% of professionals who responded (total responses: 212 professionals from different medical specialties). The mean age was 43.9 years (SD: 11.4) and 62.8% were women. About 72.5% of the professionals had more than 10 years of experience in their field (80.3% for OB/GYN vs. 64.7% for neonatologists). There was no significant difference in sociodemographic characteristics between the two professional groups.

The fetal pathologies considered as lethal by the professionals were classified according to the organ damage and listed according to their prevalence: 1. Central nervous system abnormalities (i.e. acalvaria, anencephaly, exencephaly, lissencephaly, meningomyelocele); 2. Cardiac abnormalities (i.e. total anomalous pulmonary venous return, complex heart disease, single ventricle); 3. Other structural birth defects (i.e. giant omphalocele, severe intrauterine growth restriction, syndromic congenital diaphragmatic hernia); 4. Kidney diseases (with oligo/anhidramnios and pulmonary hypoplasia, that is, bilateral renal agenesis, posterior urethral valve); 5. Trisomy 18; 6. Chromosomal abnormalities and genetic disorders (i.e. pentalogy of cantrell, trisomy 13, thanatophoric dysplasia, triploidy); 7. Prematurity (i.e. 22 weeks of gestation) and 8. Hypoplastic left heart syndrome (Figure 1).

Figure 1. Types and prevalence of lethal fetal abnormality. 1. Central nervous system abnormalities; 2. Cardiac abnormalities; 3. Other structural birth defects; 4. Kidney diseases (with oligo/anhidramnios and pulmonary hypoplasia); 5. Trisomy 18; 6. Chromosomal abnormalities and genetic disorders; 7. Prematurity; 8. Hypoplastic left heart syndrome.

Table 1. Obstetric and pediatric talks conduct.

<table>
<thead>
<tr>
<th>Professionals</th>
<th>Obstetric gynecologist, n (%)</th>
<th>Neonatologist, n (%)</th>
<th>p Values</th>
</tr>
</thead>
<tbody>
<tr>
<td>Question 1</td>
<td>80 (65.5)</td>
<td>60 (44.1)</td>
<td>0.003</td>
</tr>
<tr>
<td>Question 2</td>
<td>96 (78.6)</td>
<td>104 (76.4)</td>
<td>NS</td>
</tr>
<tr>
<td>Question 3</td>
<td>70 (57.3)</td>
<td>66 (48.5)</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Question 4</td>
<td>48 (39.3)</td>
<td>14 (10.2)</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Question 5</td>
<td>39 (31.9)</td>
<td>53 (38.9)</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Question 6</td>
<td>60 (49.1)</td>
<td>41 (30.1)</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Question 7</td>
<td>82 (67.2)</td>
<td>87 (63.9)</td>
<td>0.013</td>
</tr>
</tbody>
</table>

’n’ signifies the number of professionals who answered ‘‘yes’’ to the statement.

Question 1: Did you announce the diagnosis to the patient personally?; Question 2: According to you, was the information given in a multidisciplinary way?; Question 3: Did you take the initiative of informing the pregnant woman about the options for birth support, without waiting for their own request?; Question 4: Did she spontaneously make a request for termination of pregnancy?; Question 5: Did you spontaneously make a request for continuation of pregnancy?; Question 6: Did you spontaneously clarify the legal framework in this context?; Question 7: Did you spontaneously mention the possible scenarios in case of continuation of pregnancy and birth?

The second part of the study consisted of exploring and analyzing the information given to the pregnant women when confronting a situation of lethal fetal pathology. In this context, 35.4% of these women asked for the continuation of pregnancy and 24.7% asked for the termination of pregnancy. More than half of the professionals (52.9%) took the initiative of informing these women about the options for birth support (including PPC option), while 32.7% of OB/GYN versus 10.2% of neonatologists did not, p <0.01. Almost half of OB/GYN (49.1%) explained the legal framework of TOP and/or palliative care, while only 30.1% of neonatologists did, p <0.01 (Table 1). We made the analysis by groups of fetal pathologies; there was no significant difference between the two groups of professionals in regard to Trisomy 18, all seven...
questions included. In particular, for question 3 (Did you take the initiative of informing the pregnant woman about the options for birth support, without waiting for her own request?), 50% versus 40% of OB/GYN and neonatologists, respectively, did not inform women about the options for birth support in case of Trisomy 18. For the other groups of pathologies, the difference was significant ($p < 0.01$) between OB/GYN and neonatologist for six questions out of seven; the exception was question 2 (According to you, was the information given in a multidisciplinary way?) where 78.6% of OB/GYN versus 76.4% of neonatologists answered ‘‘YES.’’ At that point, for all described pathologies, the professionals agreed on the fact that, whatever the decision of the pregnant woman was, the information was given in a multidisciplinary way.

**Discussion**

Our French national survey evaluates the perception of lethal fetal pathology by perinatal professionals and reveals its impact on the information given to parents (specifically concerning PPC). It shows the variation in the perception of this theme and the effect of this variation on clinical practice. So, our study does not aim to establish a list of eligible diseases in PPC but to explore the ways to apprehend them. Inevitably, this study has some limitations. The participation rate could have been higher if the questionnaire were more concise but, in spite of that, this rate was equivalent to the rate in many other qualitative pediatric studies [5]. We hypothesize that PPC, a relatively new field in neonatology, might conflict with personal convictions and with the complexity in managing such complicated pregnancies [2]. Some doctors also stated that their nonparticipation was due to their lack of knowledge in this field.

Voluntary interruption of pregnancy for medical reason in the second or third trimester has some particularities. Decision is not to be taken but according to medical reasoning and the concerned physicians are just a third party. This issue in France relies on clear indications that will determine the appropriate approach [6]. These indications are based on health-related criteria that will justify each procedure and show the decisive function of physicians in the final process. Subsequently, based on such indications, the diagnosis of trisomy 18 is a flagrant example illustrating the divergence of perceptions between caregivers and parents. In our study, the group ‘‘Trisomy 18’’ is the only among lethal fetal pathologies not to encounter inconsistency between the professionals in the decisional process and the information giving. In fact, although described as lethal or incompatible with life in literature [7], trisomy 18 is variable in its genetic presentation and then in its outcome at the short, medium and long term [8]. Already well experienced in the ante and postnatal, some parents of children diagnosed with trisomy 18 shared their own experience. Pauline Thiele, a mother who continued with her pregnancy despite the fetal diagnosis of trisomy 18, told her experience in an article entitled: “He was my son, not a dying baby” [9]. Furthermore, the recent article of Annie Janvier was about many families who experienced a prenatal diagnosis of trisomy 13 or 18 and decided to continue with the pregnancy in spite of that. The parents reported facing a permanent judgment because of the decision they made, blaming their comprehension and, sometimes, leading to medical abandonment, “the best thing would be if their baby dies” [10]. In this context, Dominic Wilkinson talked about “Fatal Fetal Paternalism’’ [11] after the study that Cara Heuser published on 2011 in which the decisions taken by obstetricians concerning lethal fetal pathologies were reported: In case of anencephaly or trisomy 18, when the pregnant woman asks for continuing with the pregnancy and for a successful delivery, 7% of the interrogated professionals encourage or support her, one-third refuse and more than the half try to dissuade her from this decision [12]. In a previous national survey for our team, half of the interrogated professionals considered medical termination of pregnancy as the appropriate step taken for trisomy 18 (50.1%, $n = 112$) and anencephaly (55.6%, $n = 119$) [13]. Besides, a recent study in Quebec showed that 75% of perinatal professionals would go themselves to medical termination of pregnancy if ever confronted with an antenatal diagnosis of trisomy 18 [14].

A cross-interdisciplinary approach aims to involve different medical and paramedical specialists in opening up to sharing their thoughts in general and for every situation in particular. Involving participants from different professions allows to reach a more global understanding [2]. In our study, pluridisciplinarity is found to be a base in the information delivery; the OB/GYN–pediatric duo agrees on its importance.

In front of this uncertainty in fetal medicine, the medical team has to inform the pregnant woman about all possible evolutions, step by step. Uncertainty about the future of the baby is a reality that needs to be transmitted to parents in the prenatal period in order to take it into consideration when thinking about medical termination of pregnancy [15]. In fact, a neonatal pathology that leads to therapeutic renunciation is rarely progressive or spontaneously fatal. In this context, using strong dosages exclusively for infant comfort is not frequent. Hence, these pathologies considered incurable might go along with indeterminate prolonged survival, leading, absolutely, to death. The duration of this postnatal period is unpredictable; categorizing conditions as surely and rapidly fatal is prone to discussion [16]. The ideal prognosis might benefit the anticipation, which is important for medical work; however, this prognosis is not guaranteed yet. Additionally, Dominic Wilkinson has recently emphasized that pathologies easily considered as lethal [17] can lead to a prolonged survival [18,19]. The most impressing example is trisomy 18 where survival can go up to 50 years of age [20]. In spite of the instrumentalization in the perinatal medicine, which aims for the well being, there is always a space of uncertainty concerning the future of these babies, leaving it to professionals to open the floor for discussion. So it is up to the perinatal professional to integrate this uncertainty with his act of care, to share it, accept it, realize the risks and develop strategies in collaboration with the ‘‘subject.’’ These strategies, without assuming erasing uncertainty, can help to deal better with the doubts of caregivers and to improve the management of patients (woman and neonate). According to Charlotte Wool, declaring this medical uncertainty is not deleterious for the future parents [1]. In contrary, it is a sign of
goodwill and a source of imagination allowing them to fully live their story.

We highlight the following essential points:

- The antenatal anticipation is described, in our context, as giving detailed information about different possible scenarios in the antenatal period and at birth and as elaborating with the future parents about the possible actions. In practice, introducing all individuals and services involved in the healthcare of the infant to the family seems to be a factor of reinsurance and confidence. The pluridisciplinarity in the management is explained in details and the different intervening individuals are presented. This gives the parents an idea about the quality of surveillance, its periodicity and duration. This also shows them that even if the prognosis is reserved and we cannot predict the degree of sequelae, the latter will be followed and estimated as early as possible for an immediate management.

- Training perinatal professionals on palliative care will lead to making qualified teams for this demarche: caring for any doubt with a high level of confidence and respect, a good level of communication, and with a capability, in front of the unpredictable, to rearrange functions, which means to rearrange the usual hierarchical order for the benefit of the concerned reality and situation.

- We have to avoid the too demanding and rational procedure and/or pre-established protocols that frustrate individual’s thoughts and to propose thoughtful and collaborative demarches that can lead to an adapted decisional process.

Faced with all uncertainties inherent to medicine and the future of the baby, healthcare teams must inform the parents about the different possible outcomes step by step. The consistency in reflection and the intentionality of the care are essential in order to facilitate parental support for any possible fatal outcome. This study demonstrates the clear need to provide PPC training for professionals and to standardize PPC practices.

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Declaration of interest

The authors report no declarations of interest.

References