Decision-making after ultrasound diagnosis of fetal abnormality

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van een foetale afwijking

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Chapter 1

General introduction

1.1 Congenital abnormalities

Congenital abnormalities are the main cause of infant death in industrialised countries.[1,2] Furthermore, these form the main diagnosis in end-of-life decisions in infants.[3] Congenital abnormalities are frequently diagnosed before birth, as most major congenital abnormalities can be detected by routine prenatal examination, including ultrasound.[4-6] Table 1 gives a general picture of the nature and severity of congenital abnormalities as well as the possibilities and limits of prenatal diagnosis. This table is based on data derived from an unselected British population, which is comparable to the Dutch population.[7]

Ultrasound scanning is considered the most important tool for prenatal diagnosis of structural congenital abnormalities. It detects the majority but certainly not all of the congenital abnormalities. [7] In centres for prenatal diagnosis for example, detection rates are 80-95%. [5,8] However, these vary with the nature of congenital abnormalities. For example, the detection rate of neural tube defects is 98% while congenital heart defects are prenatally identified by ultrasound in 38%.[7] Furthermore, maternal obesity results in considerably lower detection rates.[9]

When severe congenital abnormalities are detected prenatally, couples may request for termination of pregnancy.[7] In the majority of end-of-life decisions, suspicion of fetal abnormality was first aroused after ultrasound scan.[7] Hence, the practice of ultrasound scanning is closely related to that of end-of-life-decisions.

1.2 Ultrasound

Developments in fetal ultrasound

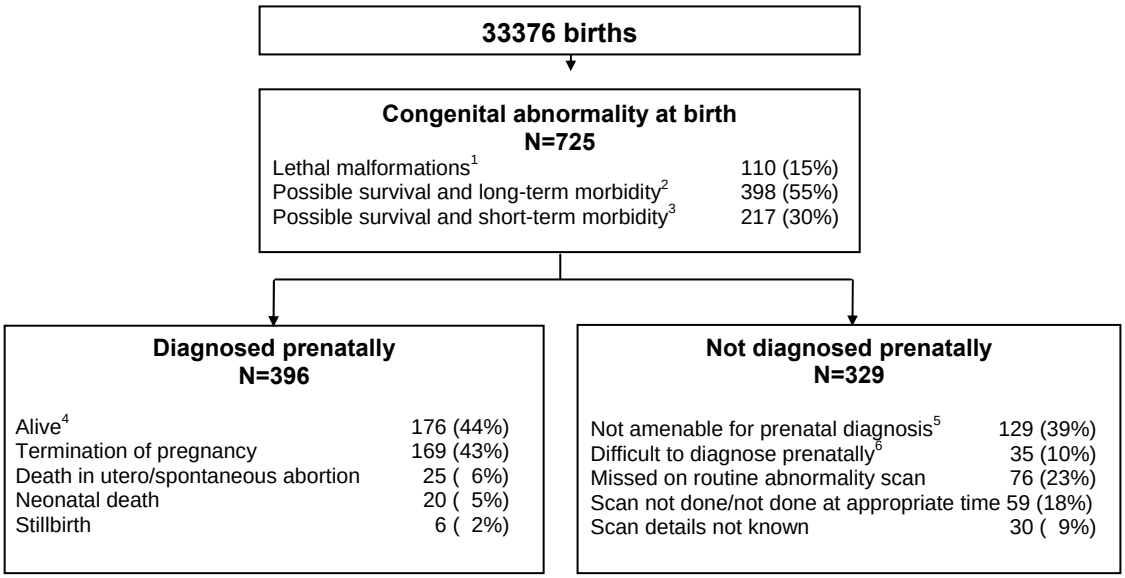
Since the first ultrasound image of a fetus in 1963, ultrasound has evolved into the most important diagnostic tool in fetal medicine.[10] First, only static images of the fetus were available. However, around 1977, the introduction of real-time scanning, allowing for moving images of the fetus, resulted in a further expansion of ultrasound diagnosis. Ultrasound became widely available. In a short period of time a myriad of reports on both normal and abnormal anatomy of the fetus appeared. Nowadays, ultrasound examination during pregnancy is routine practice in most industrialised countries.[10,13,15] This is usually done by a two-scan regime: a dating scan in first trimester of pregnancy and a fetal abnormality scan at approximately 18 to 20 weeks' gestation.[10]

In countries with routine ultrasound screening, more than half of all congenital abnormalities are diagnosed prenatally, including 74% of the major abnormalities (i.e. abnormalities that have implications for the infant's health) and 46% of the minor abnormalities (i.e. abnormalities that have no implications for the infant's health).[7,11]

Ultrasound screening

The potential benefits and limitations of ultrasound population screening for fetal abnormalities has been debated extensively.[5,10] To date, no conclusive data on the benefits of ultrasound screening have appeared.[12] Randomized controlled trials have been done, but have used perinatal mortality and morbidity as outcome variables. Moreover, in the setting of these studies the expertise levels of sonographers varied much. Finally, these studies took place at a time that ultrasound was less advanced than it is now.[13] These methodological problems are reflected in an enormous variation of reported over-all sensitivity for detection of congenital abnormalities, which ranges between 14% and 96%. [4-6] Apart from these methodological flaws, the use of perinatal mortality and morbidity as the only important outcome variable is questionable.[13] Other outcome variables, such as pregnant women being informed, enhancing fetal-maternal attachment and preventing a 'wrongful life' are of relevance as well.[12] These issues reflect the question what should be the aim of screening, the answer of which depends not only on medical data, but on moral choices as well. [13-15]

Table 1. Overview of prenatal detection of congenital abnormality when prenatal screening is offered routinely in an unselected population in Oxford, 1991-1995; births included all births over 20 weeks and all pregnancies terminated because of fetal abnormality detected at any age of gestation.



[1] e.g., anencephaly, trisomy 13, trisomy 18, hypoplastic left heart, renal agenesis, Meckel Gruber syndrome
[2] e.g.,spina bifida, hydrocephalus, Down's syndrome, complex cardiac malformations, diaphragmatic hernia, abdominal wall defects
[3] e.g., non-complex cardia malformations, facial clefts, club foot, hypospadia
[4] two infants died after the neonatal period
[5] e.g.,skin abnormalities, hypospadia, congenital dislocation of the hip, cleft palate, atrial-septal defect
[6] e.g., tracheo-oesofageal fistula or fistula, coarctation of aorta, polydactyly, ambiguous genitalia

In the Netherlands, ultrasound screening has been debated.[15] The government has decided that ultrasound is only to be offered on medical indication, i.e. targeted at women at increased risk of congenital abnormalities in offspring.[15] This attitude should be seen within the Dutch sociocultural context. Dutch law typically forbids screening, unless certain conditions are fulfilled, including the availability of effective treatment for the condition. [15] Furthermore, there are concerns about the implications of prenatal screening in the context of medicalisation of the pregnancy and regarding the societal position of handicapped people.[15] Very recently, however, a trend towards offering all women ultrasound examination at 20 weeks gestational age is seen.

The government's decision not to offer routine ultrasound screening results in large practice variations in antenatal care, e.g., some women have no ultrasound examination at all, other women have a two-scan regime and some women opt for having a 'pleasure scan' in a commercial setting , which may however create false reassurance as many of these ultrasound examinations are not apt for detecting fetal abnormalities[15] [16] Unfortunately, there are no guidelines for timing of the ultrasound, counselling or qualifications of the sonographers. So even if women have had one or multiple ultrasound examinations, potentially detectable major fetal abnormalities can still be missed.

Psychological effects of normal ultrasound

Normal findings at ultrasound examination have strong beneficial psychological effects on the pregnant woman and her partner. For couples, ultrasound is a way of 'meeting' the unborn child (see text box 1). [17-20] The personalisation of the fetus enhances both maternal-fetal bonding and bonding of the pregnant woman and her partner.[19,21,22]. A normal ultrasound reassures parents about the pregnancy.[21,23-26]

The positive effects of ultrasound are stronger when more feedback is provided, such as showing images on a second monitor, and explaining what can be seen.[21,25]

So, fetal ultrasound is highly appreciated by pregnant women and their partners.[20,26-28] For most women it forms an integral part of obstetric care.[27] However, frequently women lack information about the purposes for which an ultrasound is done and what are its technical limitations. Therefore, women are frequently unprepared for adverse findings. [28,29]

Text box 1. An illustrative reaction of a couple to a normal ultrasound

A couples reaction to a normal ultrasound: *'The baby becomes more real…once you see the scan, that all changes. It's no longer your imagination at work, but you have this real image of a little baby. You can see so much detail it is amazing, his little fingers and toes, his eyes, oh, everything. It is magical, so awe inspiring to see'* Puddifoot JE, Johnson MP. The legitimacy of grieving: the partner's experience at miscarriage. Soc Sci Med 1997;45(6):837-45.

Psychological effects of abnormal ultrasound

The results of an abnormal ultrasound frequently come unexpectedly and are intensely shocking for the expecting parents, in particular when major congenital abnormalities are encountered.[28,30] Pregnant women and their partners may have several reactions on the announcement of fetal abnormality. Firstly they may have negative feelings as associated with psychological traumas in general, such as anxiety, grief, anger, loneliness, hopelessness, prostration and guilt. [29-31,33,34] These feelings may be aggravated by the loss of the imagined future, as the pregnancy may end in the daily reality of having no child or a severely handicapped child, requiring readjustments of the entire family. These feelings can be enhanced by the confrontation with reality, when having to decide about very pragmatic issues ' should I decorate the nursery?', 'should I make arrangements for the funeral?', and 'what should I tell my other child?'. Finally, some parents experience a loss of reference. The news of a fetal abnormality in an apparently uneventful pregnancy, usually comes so unexpectedly and is in such contrast with the pleasant experiences that often come with pregnancy, that parents often find it very difficult to grasp the facts. It seems so unreal that the child who is kicking inside the womb is severely disabled or will even die, that all meaning seems lost. When parents consider terminating the pregnancy, the ambivalent feelings they experience may enhance this loss of reference. On the one hand, they are committed to the wanted and intended pregnancy. On the other hand, they want to protect the unborn child, themselves and the family from the burden of severe disability.[35]

1.3 End-of-life-decisions after ultrasound diagnosis of fetal abnormality

End-of-life decisions after ultrasound diagnosis of fetal abnormality

With the burgeoning of ultrasound, questions around the appropriate obstetric management in case of sonographically established fetal abnormalities have arisen. Should we apply all means to keep alive a fetus with a very poor prognosis? Do medical professionals in the field of perinatal medicine agree on fetal prognosis after ultrasound diagnosis of fetal abnormality? How should obstetric and neonatal management be attuned? How do parents view upon end-of-life decisions regarding their unborn infant? These and other issues have opened a new field of research: end-of-life decision-making after ultrasound diagnosis of fetal abnormality. End-of-life decisions are decisions about medical interventions at the end of life, which certainly or probably hasten death.[36] Two kinds of end-of-life decisions can be distinguished after ultrasound diagnosis of fetal abnormality: 1) non-aggressive obstetric management and 2) termination of pregnancy.

Table 2. Studies evaluating determinants of parental decision-making after ultrasound diagnosis of fetal abnormality

Author	Population	N	Method	Outcome variable	Results	
Determinants						
Grevengood et al.[53]	isolated neural tube defect identified < 24 weeks GA normal karyotype	50	analysis decisions in case of anencephaly analysis decisions in spina bifida laesions >T9 analysis decisions in spina bifida laesions <T9	TOP	anencephaly :23/23 TOP spina bifida > T9 : 5/ 5 TOP spina bifida < T9 :16/ 22 TOP	
Pryde et al.[54]	abnormalities identified <24 weeks GA no abnormal karyotype	159	GA stratified into early (<14 weeks GA), mid (15-19 weeks GA), late) (20-24 weeks GA diagnosis Prognostic severity of abnormality stratified into mild, uncertain, severe	TOP	severity of abnormality GA maternal age gravidity parity	p<0.001 n.s. n.s. n.s. n.s.
Sheiner et a.[55]	abnormalities incompatible with life or severe enough to Significantly interfere with normal living in Arab Bedouin population	188	63 cases GA>24 weeks 125 cases GA < 24weeks	TOP	GA previous uncompleted pregnancies central nervous sytem abnormality maternal age gravidity parity previous perinatal death congenital abnormality in family	p<.01 p<.01 p<.01 n.s. n.s. n.s. n.s. n.s.
Schechtman et al.[56]	abnormalities identified <24 24 weeks GA	53630	severity of abnormality stratified on scale 1-5, as evaluated by sonographer	TOP	educational level parents severity of abnormality chromosomal abnormalities central nervous system abnormality maternal age	p < .001 p < .001 p < .001 p < .001 n.s.

TOP: termination of pregnancy; GA: gestational age; n.s.: not significant

A non-aggressive obstetric management refers to an obstetric management, in which interventions needed to sustain fetal life are forgone, because of poor fetal prognosis. A non-aggresive obstetric management was first reported in 1989, when Chervenak and McCullough described such management in 13 cases. They regard a non-aggressive obstetric management as permissible, and even preferable, when there is certainty of death or absence of cognitive developmental capacity as outcome of the congenital abnormality.[37,38] Chervenak and McCullough argue that in such cases, the fetus does not benefit from obstetric intervention, whereas such intervention may harm the pregnant woman and interfere with her autonomy.[37,38] However, empirical data that are needed for a balanced professional and societal debate about forgoing fetal life-sustaining treatment, are scarce.[37,39,40]

Termination of pregnancy is a management, in which the pregnancy is terminated with the explicit intention of hastening fetal death. Termination of pregnancy is done by induction of labour, which may be preceded by fetal intracardial potassium injection. The first termination of pregnancy after ultrasound diagnosis of fetal abnormality was reported by Campbell et al. in 1972. This concerned a fetus with anencephaly.[41] Termination of pregnancy has far-reaching implications. It bears life-long lasting consequences and evokes very strong emotions of the couples involved. [42,43]

End-of-life decision-making after ultrasound diagnosis of fetal abnormality has to

take into account both the interest of the fetus and the pregnant woman. Therefore, it is usually done by multidisciplinary teams.[44-48] These typically consist of obstetricians, neonatologists, paediatric surgeons, and other paediatric specialists, such as paediatric urologists, paediatric neurologists, and paediatric neurosurgeons.[44-48] However, little is known about how decisions are being taken in these teams.

Parental decision-making after ultrasound diagnosis of fetal abnormality remains largely unknown. Table 2 shows the studies to date, which evaluate determinants of parental decision to choose for termination of pregnancy. Lower gestational age, a more severe abnormality, involvement of the central nervous system, previous uncompleted pregnancies, lower maternal educational level, and the presence of chromosomal abnormalities have been related with a higher rate of women deciding upon termination of pregnancy. However, these data are not conclusive and do not reflect why and in what way some factors are important for the parents. Sandelowski has studied parental decision-making concerning prenatal diagnosis of fetal abnormality in more depth. [49] She interviewed 15 women and 12 of their partners. This study shows the concept of choice is contested. Women in comparable circumstances feel differently about whether or not they have a choice about the future of their pregnancy. [49]

Legal context and current guidelines

In the Netherlands, law prohibits termination of pregnancy at a gestational age of 24 weeks and beyond. However, in case of good clinical practice, physicians typically are not prosecuted by the public prosecutor. [50-52] Good clinical practice is described in guidelines that were made by a collaborative group of the Ministry of Health, Ministry of Justice and the Dutch Society of Obstetricians and Gynaecologists. According to these guidelines, when parents insistently request for termination of pregnancy, this will be granted in case of extremely poor fetal prognosis. Fetal prognosis should be: 1) the infant has no chance of survival and the abnormalities not be treated or the infant has a chance of extra-uterine survival but post-natal use of life-prolonging medical treatment is considered futile.[51]

1.4 Research objectives

This thesis aims at exploring decision-making after ultrasound diagnosis of fetal abnormality. The following issues and research questions are addressed:

1) The frequency of end-of-life decisions after ultrasound diagnosis of fetal abnormality

1. What is the frequency of end-of-life-decisions in a tertiary referral centre?
2. What is the estimated frequency of fetal end-of-life decisions in the Netherlands?

2) The process of decision-making after ultrasound diagnosis fetal abnormality

Physicians

1. To what extent can fetal prognosis after ultrasound diagnosis of fetal abnormality be classified, in the context of end-of-life decision-making?
2. What is inter- and intra-observer agreement about fetal prognosis after ultrasound diagnosis of fetal abnormality?
3. To what extent does decision-making in a multidisciplinary perinatal team enhance consensus about obstetric management decisions, neonatal management decisions, and hospital of delivery?
4. To what extent are decisions regarding obstetric and neonatal management attuned prenatally?
5. What is neonatal management in case of infants that are born alive after a non-aggressive obstetric management?

Pregnant women

6. How do pregnant women make decisions about the fetus?

3) Consequences of end-of-life decisions after ultrasound diagnosis of fetal abnormality

1. What is survival in utero, during delivery, and during the neonatal and post-neonatal period in case of a non-aggressive obstetric management after ultrasound diagnosis of fetal abnormality?
2. What is the health status of children who survived after a non-aggressive obstetric management?

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Decision-making by physicians

Chapter 2

Ultrasound diagnosis of fetal abnormalities: an analysis of perinatal management of 318 consecutive pregnancies in a multidisciplinary setting

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Abstract

Objectives

The objectives of this study are to analyse the perinatal management decisions made in a multidisciplinary setting following the prenatal diagnosis of fetal abnormalities and to evaluate to what extent, in clinical practice, decisions about obstetric management are attuned to those about neonatal management.

Methods

Data on perinatal management of 318 consecutive singleton pregnancies presented to a multidisciplinary perinatal team in a tertiary centre were collected retrospectively.

Results

The multidisciplinary perinatal team decided upon non-aggressive obstetric management in 20% of the cases and consented to termination of pregnancy in 10% of the cases. The multidisciplinary perinatal team decided upon neonatal management in 112(36%) of all fetuses. In 100(89%) of these fetuses standard neonatal management and in 12(11%) no neonatal life-sustaining treatment was decided upon. Implementation of the clinical-ethical decisions of the multidisciplinary perinatal team on the various management modalities ranged from 88% to 100%.

Conclusion

The multidisciplinary perinatal team worked well in making decisions about obstetric management. In 30% this concerned end-of-life-decisions. However, for the majority of cases, the perinatal team did not plan neonatal management before birth and thereby did not attune obstetric and neonatal management to each other. This probably reflects different attitudes towards end-of-life-decisions between obstetricians and neonatologists. However, to ensure a consistent perinatal management, a multidisciplinary perinatal team has to make prenatal decisions about both obstetric and neonatal management

Introduction

The increasing possibilities of prenatal diagnosis confront us with raise important questions concerning medical management during pregnancy, delivery and the neonatal period. In case of a poor fetal prognosis, the appropriateness of interventions aimed at sustaining life can be questioned. [1-4] Ethical aspects, diagnostic uncertainty, limited time and the involvement of parents whose future will be deeply affected by the decisions taken, complicate planning of perinatal management. [5-7] As both obstetric and neonatal interventions affect outcome,often a multidisciplinary approach is widely adopted. [7-12] However, different perspectives of the specialties involved may impede this process.[13]

To date, there are few studies evaluating to what extent, in clinical practice, obstetric and neonatal management are attuned to each other. Especially when end-of-life-decisions have to be taken, different attitudes of neonatologists and obstetricians may lead to inconsistencies in perinatal management. The objectives of this study are to analyse the perinatal management decisions made inclinical-ethical a multidisciplinary setting following the prenatal diagnosis of fetal abnormalities and to evaluate to what extent, in clinical practice, decisions about obstetric management are attuned to those about neonatal management.

Patients and methods

All singleton pregnancies scanned at the Fetal Medicine Unit of the Erasmus Medical Centre, University Hospital Rotterdam and presented to a multidisciplinary perinatal team between January 1996 and February 2001 were included. Obstetricians in the southwest region of the Netherlands refer women with a suspected fetal abnormality to the Fetal Medicine Unit for fetal abnormality scanning. Following the sonographic diagnosis of a fetal abnormality, the case is discussed by a team of physician-sonographers and obstetricians of the department of obstetrics and gynaecology to determine obstetric management. Subsequent referral to the multidisciplinary setting and inclusion in this study occurred for three reasons: (i) uncertainty about the diagnosis and/or prognosis; (ii) to discuss parental requests for pregnancy termination beyond 24 weeks of gestation which is the upper legal limit for pregnancy termination in the Netherlands; iii) when the infant is expected to be referred to the paediatric department after birth. The multidisciplinary perinatal team consists of medical specialists involved in perinatal care, such as obstetricians, neonatologists, paediatric surgeons, paediatric intensive care specialists, paediatric cardiologists, paediatric neurologists and clinical geneticists. Typically, decisions were made as follows: following the presentation of the case by a physician-sonographer, the multidisciplinary perinatal team establishes the diagnosis as well as the fetal prognosis. After discussing both the medical and ethical aspects, the perinatal team makes a decision about obstetric management, neonatal management and whether or not the infant should be delivered in a tertiary centre. Decision-making is based on both scientific evidence and clinical experience. End-of-life-decisions are made in accordance with national guidelines. [14-16]

A non-aggressive obstetric management or a termination of pregnancy after 24 weeks is only considered when the abnormalities are incompatible with life or for which the use of post-natal life-prolonging treatment is futile. In these cases, termination of pregnancy is only considered if requested for by the parents. Generally, decisions are as much as possible made in accordance with parental preferences. For the obstetric part of perinatal management three options are considered: standard management, non-aggressive management or termination of pregnancy. Standard management is defined as management aimed at the delivery of an infant in good physical condition and non-aggressive management as management in which not all interventions needed to sustain fetal life are made, because of poor fetal prognosis. For the neonatal part of perinatal management two options are considered: standard management or no life-sustaining treatment.

Retrospective collection of data was carried out by a physician (HB) experienced in prenatal care, who was not clinically involved in any of the cases. Ultrasound reports, minutes of multidisciplinary perinatal meetings as well as obstetric and paediatric records were reviewed. The nature of the decisions on perinatal management and the subsequent adherence to these decisions were studied. Fetal prognosis was assessed by expert opinions.

Two physicians (HB and HW) experienced in prenatal diagnosis classified the prognosis for all cases according to an adapted version of the classification system of the Working Party on Late Pregnancy Termination in the Netherlands.[14]

Fetal prognosis was classified as poor, good or uncertain. A fetus with a lethal abnormality or with a non-lethal but severely disabling abnormality was classified as having a poor prognosis. A prognosis was classified as uncertain when at least one of the experts was not able to categorise the fetal abnormality as poor or good or when there was a disagreement between both experts on fetal prognosis. A close inter-observer-agreement was found (percentage of agreement 90%, kappa=0.78).[17]

Results

A total of 318 consecutive singleton pregnancies was included. Five cases were excluded from the analysis, because relevant data were not available.

Mean maternal age was 30.2 years (sd 5.4 years) and mean gestational age at the time of the first fetal abnormality scan was 28.4 weeks (range 12.6-40.0 weeks). In 225(72%) women, gestational age was more than 24 weeks. Fetal karyotyping was performed in 281(90%) women, the results of which were known at the time of the multidisciplinary perinatal meeting in 166(56%) women. An abnormal karyotype was found in 17 fetuses. All karyotypes that were not yet known at the time of the perinatal meeting turned out to be normal.

Table 1. Management decision (in number and percentage) by prognosis of fetal abnormality

	Prognosis			
	Poor *n=81*	*Good* *n=168*	*Uncertain* *n=64*	*Total* *n=313*
Standard management	2 (26%	15 (93%	3 (50%	21 (67%
Non-aggressive	3 (46%	8 (5%)	1 (30%	64 (20%
Termination of pregnancy	2 (25%	2 (1%)	1 (16%	32 (10%
Other	3 (4%)	1 (1%)	3 (5%)	7 (2%)

Table 1 shows the relationship between fetal prognosis and obstetric management as decided upon by the multidisciplinary perinatal team. Obstetric management was aimed at sustaining life in 210(67%) cases. In 96(30%) of the cases the multidisciplinary perinatal team decided upon pregnancy termination or non-aggressive obstetric management. In two cases, pregnancy classified as having a good fetal prognosis was terminated before 24 weeks, for reasons of respect for parental autonomy.

Table 2 and table 3 show obstetric and neonatal management as well as the place of delivery as advised by the multidisciplinary perinatal team. A non-aggressive approach was adopted in 26(32%) cases with multiple abnormalities and in 38(17%) cases with a single abnormality. Termination of pregnancy was decided upon in 18(22%) of the cases with multiple abnormalities and in 14(6%) of the cases with single abnormalities. Within the group of fetuses with multiple abnormalities, when an abnormal karyotype was found, the multidisciplinary perinatal team never advised a standard obstetric management. When multiple abnormalities and a normal karyotype were found, standard obstetric management was advised in 22(54%) cases. In case of a single abnormality, the decision of the multidisciplinary perinatal team was related to the nature of the fetal abnormality. In case of intra-abdominal abnormality, abdominal wall defect, diaphragmatic hernia or lung abnormality, the multidisciplinary perinatal team usually advised a standard obstetric management. Finally, in instances of renal abnormality, intracranial abnormality, spina bifida, hydrocephalus or cardiac abnormality, standard obstetric management was advised in 22(73%), 13(62%), 8(47%), 6(35%) and 11(73%) cases respectively.

Table 2. Management decisions (in number and percentage) are given according to karyotype result

	Multiple structural abnormalities			
	Normal karyotype n=41	*Abnormal karyotype[1] n=13*	*Karyotype unknown n=28*	*Total n=82*
Obstetric management				
Standard management	22 (54%	- -	1 (46%)	3 (43%
Non-aggressive	13 (32%	2 (15%)	1 (39%)	2 (32%
Termination of pregnancy	5 (12%	11 (85%)	2 (7%)	1 (22%
Other	1 (2%)	- -	2 (7%)	3 (4%)
Neonatal management				
Standard management	12 (29%	- -	7 (25%)	1 (23%
No life-sustaining	3 (7%)	- -	2 (7%)	5 (6%)
No decision	26 (63%	13 (100%	1 (68%)	5 (71%
Hospital of delivery				
Tertiary center	27 (66%	5 (39%)	1 (64%)	5 (61%
No tertiary center	10 (24%	5 (39%)	8 (29%)	2 (28%
No decision	4 (10%	3 (23%)	2 (7%)	9 (11%

[1]Trisomy 18 (4) , trisomy 13 (3), triploidy (3), trisomy 21 (1), monosomy 7 trisomy 10 (1), 46xy,ad(q)(p22) (1)

Table 3: see next page

Table 4. Actual obstetric management (in number and percentage) by planned obstetric management, actual neonatal management (in number and percentage) by planned neonatal management and actual hospital of delivery (in number and percentage) by planned hospital of delivery.

	Planned obstetric management				
	Standard management n=210	*Non-aggressive management n=64*	*Pregnancy termination n=32*	*Other n=7*	*Total n=313*
Actual obstetric management					
Standard management	207 (99%)	6 (9%)	-	1 (14%)	214 (68%)
Non-aggressive management	2 (1%)	57 (89%)	1 (3%)	1 (14%)	61 (19%)
Pregnancy termination	1 (1%)	1 (2%)	31 (97%)	2 (29%)	35 (11%)
Other				3 (43%)	3 (1%)
	-	-	-		

	Planned neonatal management[1]			
	Standard management n=100	*No life-sustaining treatment n=12*	*No decision n=201*	Total *n=313*
Actual neonatal management				
Standard management	96 (100%)	1 (9%)	130 (87%)	227 (87%)
No life-sustaining treatment	-	9 (91%)	20 (13%)	29 (11%)
Stillbirth/Death during delivery	4	2	51	57

	Planned hospital of delivery			
	Tertiary centre n=188	*No tertiary centre n=90*	*No decision n=35*	Total *n=313*
Actual hospital of delivery				
Tertiary centre	176 (94%)	7 (8%)	24 (31%)	207 (66%)
No tertiary centre	12 (6%)	83 (92%)	11 (69%)	106 (34%)

[1] Percentages of actual neonatal management were calculated after subtracting the infants that were not born alive.

Table 3. Management decisions (in number and percentage) in case of single structural abnormalities are given according to nature of the abnormality

	Single structural abnormalities																					
	Intra-abdominal abnormality n=50		*Renal abnormality n=30*		*Abdominal wall defects n=21*		*Intracranial abnormality n=21*		*Diaphragmatic hernia n=18*		*Spina bifida n=17*		*Hydrocephaly n=17*		*Cardiac abnormality n=15*		*Lung abnormality n=11*		*Other abnormality n=31*		*Total n=231*	
Obstetric management																						
Standard	50[1]	(100%)	22	(73%)	20	(95%)	13	(62%)	16	(89%)	8	(47%)	6	(35%)	11	(73%)	9	(82%)	20	(65%)	175	(76%)
Non-aggressive	-	-	4	(13%)	-	-	4	(19%)	1	(6%)	6	(35%)	11[4]	(65%)	4	(27%)	2	(18%)	6[3]	(19%)	38	(17%)
TOP	-	-	4	(13%)	-	-	2	(10%)	-	-	3	(18%)	-	-	-		-	-	5	(16%)	14	(6%)
Other	-	-	-		1[2]	(5%)	2	(10%)	1	(6%)	-	-	-	-	-		-	-	-		4	(2%)
Neonatal management																						
Standard	27	(54%)	13	(43%)	3	(14%)	7	(33%)	12	(67%)	4	(24%)	3	(18%)	2	(13%)	3	(27%)	7	(23%)	81	(35%)
No LST	-	-	-	-	-	-	4	(19%)	-	-	1	(6%)	1	(6%)	-	-	-	-	1	(3%)	7	(3%)
No decision	23	(46%)	17	(57%)	18	(86%)	10	(48%)	6	(33%)	12	(70%)	13	(76%)	13	(87%)	8	(73%)	23	(74%)	143	(62%)
Hospital of delivery																						
Tertiary center	27	(54%)	15	(50%)	19	(90%)	7	(33%)	16	(89%)	7	(41%)	12	(71%)	12	(80%)	11	(100%)	7	(23%)	138	(60%)
No tertiary center	19	(38%)	14	(47%)	1	(5%)	11	(52%)	-	-	7	(41%)	3	(18%)	-	-	-	-	17	(55%)	67	(29%)
No decision	4	(8%)	1	(3%)	1	(5%)	3	(14%)	2	(11%)	3	(18%)	2	(12%)	3	(20%	-	-	7	(23%)	26	(11%)

[1] Trisomy 21 in one case ; [2] Trisomy 18; [3] trisomy 21 in one case;[4] balanced translocation (13;14) in one case; TOP: termination of pregnancy, LST: life-sustaining treatment

Table 4 shows agreement between planned and actual perinatal management. Neonatal management was planned prenatally in 112(36%) cases. If planned prenatally, neonatal management was aimed at sustaining life in 100(89%) cases. If the 32 cases in which pregnancy termination was decided upon are disregarded, neonatal management was planned prenatally in 110/281(61%) of the fetuses and was aimed at sustaining fetal life in 100/110(91%). If one disregards the cases in which pregnancy termination was decided upon, obstetric management was aimed at sustaining life in 210/281(75%) of the fetuses. Implementation of the decision of the multidisciplinary perinatal team regarding the various management modalities ranged between 88% and 100%.

Outcome one week after delivery was known for 312 infants. Twelve(4%) pregnancies ended in stillbirth, 45(14%) infants died during delivery (28 as result of termination of pregnancy), 59(19%) died in the first week of life (8 after termination of pregnancy). A total of 196(63%) infants was alive one week after delivery.

Discussion

Limited time and lack of diagnostic information, such as fetal karyotype and therefore prognosis, often complicated the process of perinatal management in the presence of single or multiple fetal abnormalities. In 10% of all cases the multidisciplinary perinatal team consented to pregnancy termination. In 20% of all cases the multidisciplinary perinatal team advised a non-aggressive obstetric management. In case of poor fetal prognosis the multidisciplinary perinatal team decided upon non-aggressive obstetric management in 46% of the cases and consented to parental request for termination of pregnancy in 25% of the cases. The multidisciplinary perinatal team more often decided to limit obstetric intervention aimed at sustaining fetal life in case of multiple abnormalities and in case of specific 1single abnormalities, such as central nervous, neural tube and renal abnormalities, as described by others. [1,18-20]

Neonatal management was planned prenatally only in 36% of the cases. If planned prenatally, neonatal management was aimed at sustaining the infant's life in 89%, whereas prenatally planned obstetric management was aimed at sustaining fetal life in 67%. If one disregards the cases in which termination of pregnancy was consented to, neonatal management was aimed at sustaining life in 91% and obstetric management in 75% of the cases. So, with or without taking into account termination of pregnancy, in case of prenatally detected fetal abnormalities, neonatal management was less often planned in advance than obstetric management and, if planned, was more often directed towards sustaining life. Different perspectives of obstetricians andchildcare specialists neonatologists might account for these differences. Firstly, the prognosis of a specific abnormality might be perceived differently. Neonatologists meet encounterthe infants whichinfants who at least survived until after birth, whereas obstetricians generally seesee a population with a worse prognosis. [21-22] Secondly, uncertainty of ultrasound diagnosis is probably perceived differently. Whereas for an the obstetrician ultrasound usually provides a considerable amount of information otherwise not available, for a childcare specialist the neonatologist, diagnostic possibilities of prenatal ultrasound are rather limited compared to diagnostic possibilities after delivery. Thus, it might be that a specific diagnosis is perceived as more certain by the obstetrician than by the childcare specialistthe obstetrician perceives a specific diagnosis and its prognosis with more certainty than the neonatologist. Furthermore, there may be a discussion over whose best interests should be primarily served, the mother's' or the infant's.. For the obstetrician, the pregnant womans woman's' beneficience well-being might be paramount, whereas the childcare specialistneonatologist will tend to focus on the neonate.[13]

Further, consequences of refraining from medical interventions are more tangible for the childcare specialistneonatologist than for the obstetrician. In case of (complete) non-aggressive obstetric management the infant is not monitored and the obstetrician will not be confronted with signs of fetal distress. However, the childcare specialistneonatologist may be confronted with a live child in distress with a need for either therapeutic intervention or comfort care. Our data probably reflect differences between obstetricians and child care specialistneonatologists, resulting in a tendency of childcare specialistneonatologists to give a fetus the benefit of the doubt and to postpone clinical decisions until after birth. However, both obstetric and neonatal

management affect the infant's well-being and they should be considered together in order not to worsen outcome, such as may occur when obstetrician labour-induced hypoxic damage is superimposed upon the initial problem, leading to severe disability.

The actual perinatal management usually wawas in accordance with the decision of the multidisciplinary perinatal team, indicating its importance for clinical practice. Our study has several limitations. Firstly, due to its retrospective nature, only the outcome of the multidisciplinary meeting could be studied, ignoring the process and the arguments that played a role. For future research we recommend that the decision making process in the multidisciplinary perinatal team is evaluated prospectively. Secondly, follow-up extended to one week after delivery and was assessed only as being alive or not. A more prolonged follow-up, including data on quality of life of the infant as well as experiences of the parents, is needed in order to further evaluate medical management. Thirdly, this study was done in one centre in the Netherlands, which may limit generalisation to other centres. However, in many perinatal centres end-of-life-decisions are taken in a multidisciplinary context.[2,23,24]

In the Netherlands there is a rather open societal and professional debate about end-of-life decision making, which is reflected in the accessibility of data on end-of-life-decisions in obstetric and neonatal records. Therefore, the Dutch setting provides the opportunity to evaluate differences between specialists which may remain undiscovered in settings where end-of-life-decisions are less widely accepted.

Conclusion

We have described the the clinical-ethical decisions on perinatal management of fetuses with one or more sonographically established fetal abnormalities as provided by a multidisciplinary perinatal team in a tertiary hospital setting. Planning perinatal management in case of ultrasound diagnosis of fetal abnormalities is often complicated by limited time and scarce diagnostic information Limited time and scarce diagnostic information often complicated planning perinatal management in case of ultrasound diagnosis of a fetal abnormality. The multidisciplinary perinatal team worked well in making decisions about obstetric management. In 30% this concerned end-of-life-decisions. However, for the majority of cases, the perinatal team did not plan neonatal management before birth and thereby did not attune obstetric and neonatal management to each other. This probably reflects different attitudes towards end-of-life-decisions between obstetricians and neonatologists. However, to ensure a consistent perinatal management, a multidisciplinary perinatal team has to make prenatal decisions about both obstetric and neonatal management. For clinical practice, this indicates the importance of a mutual understanding of the several specialities as well as the importance of joined decision-making.

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Chapter 3

Obstetrician's agreement on fetal prognosis after ultrasound diagnosis of fetal abnormalities

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Abstract

Introduction

After ultrasound diagnosis of a severe fetal abnormality is made, difficult decisions may arise regarding obstetric management. Guidelines have been developed to support obstetricians in decision-making. However, it is unknown to what extent in the clinical situation guidelines are actually supportive.

Objectives

We aimed at: i) determining whether obstetricians in the presence of a fetal abnormality are able to classify fetal prognosis according to guidelines; ii) establishing inter- and intra-observer agreement regarding fetal prognosis.

Methods

We used three categories of fetal prognosis: category 1: the infant has no chance of survival and the abnormalities cannot be treated (1.1); or the infant has a chance of extra-uterine survival but post-natal use of life-prolonging medical treatment is considered futile (1.2); category 2: the infant has a chance of extra-uterine survival and post-natal use of life-prolonging medical treatment, if necessary, is considered beneficiary. Five senior obstetricians categorized 100 case descriptions of severe fetal abnormalities, which were classified again after five months.

Results

Four obstetricians were able to classify 98% or more of cases. In 67% of cases, four or all obstetricians agreed on fetal prognosis. Overall kappa-coefficient was 0.48 (moderate agreement). The differences between obstetricians represented systematic differences in opinions on how to classify cases. Intra-observer agreement was 82-97%.

Conclusion

Obstetricians were usually able to classify fetal prognosis according to guidelines, but in a substantial number of cases there was disagreement, reflecting systematic differences between obstetricians.

Introduction

Whenever an ultrasound diagnosis of a severe fetal abnormality is made, difficult decisions may arise regarding obstetric management. In general, obstetric management involves applying all means to deliver the infant in the best possible condition. However, a 'non-aggressive' policy may be adopted when the prognosis is extremely poor. In that case all interventions that involve a risk for the mother and serve no benefit to the infant will be avoided. In exceptional cases termination of pregnancy may be considered because of parental wish in the case that prolongation of the pregnancy is considered not to be in the interest of the child. In many countries, the conditions under which termination of pregnancy is allowed are legally regulated. In the Netherlands, termination of pregnancy is permitted until 24 weeks of gestation. Termination beyond this gestational age has nevertheless been shown to occur, in the Netherlands as well as in other countries.[1-4]

Decisions about unborn infants with severe abnormalities have far-reaching consequences. They require a careful decision-making process, the quality of which is enhanced by the use of guidelines.[5] For this purpose, several guidelines have been developed.[6,7]. However, it is unknown to what extent in the clinical situation guidelines are actually supportive. Furthermore, in making an informed decision, parents rely heavily on their obstetricians' views on fetal prognosis. Therefore, accurate and objective information about fetal prognosis and hence on the best obstetric management is of great importance. However, it is unknown whether obstetricians actually share their views on fetal prognosis. Therefore we aimed at: i) determining whether obstetricians in the presence of a fetal abnormality are able to classify fetal prognosis according to guidelines; ii) establishing inter- and intra-observer agreement regarding fetal prognosis.

Methods

In our Rotterdam tertiary referral centre for fetal abnormality scanning, women with an ultrasound diagnosis of one or several fetal abnormalities are discussed within a multidisciplinary team for several reasons: (i) there is uncertainty about the diagnosis and/or fetal prognosis; (ii) when the infant is expected to be referred to our hospital after birth; (iii) in case the woman requests termination of her pregnancy beyond 24 weeks of gestation. The multidisciplinary team consists of specialists in obstetric, neonatal and paediatric care. A random sample of 100 singleton pregnancies was drawn from all 313 patients with an ultrasound diagnosis of fetal abnormality who were presented to the multidisciplinary team between January 1996 and January 2001. The fetal karyotype was available in 50/100 fetuses, of which 6 were abnormal. 27/100 fetuses displayed multiple structural abnormalities. Gestational age varied between 19.6 and 39.7 weeks (median: 32.5 weeks). Five senior obstetricians from five tertiary prenatal referral centres in the Netherlands were asked to classify these 100 cases. All obstetricians had more than five years of clinical experience in the field of fetal abnormality scanning. Obstetrician 4 classified the cases together with an equally experienced colleague from the same department.

For this study, we used the national guidelines of the Netherlands. In these guidelines a classification of fetal abnormalities is proposed which takes into account both the chance of survival and the chance that an acceptable quality of life will be achieved after birth. This non-standard approach of obstetric management is considered acceptable if there is either no chance of survival and no possibility to treat the abnormality (e.g., in case of anencephaly), or if the quality of life after birth will be unacceptably poor and treatment of the abnormality is either impossible or generally considered to be futile (e.g., in case of severe hydrocephaly). According to the Dutch national guidelines the following two categories could be defined (1.1 and 1.2) (see box 1) [8]: category 1: the infant has no chance of survival and the abnormalities cannot be treated (1.1); or the infant has a chance of extra-uterine survival but post-natal use of life-prolonging medical treatment is considered futile

(1.2); category 2: the infant has a chance of extra-uterine survival and post-natal use of life-prolonging medical treatment, if necessary, is considered beneficiary. Classification by the five obstetricians was based on the description of the ultrasound findings, the fetal karyotype (if available) and gestational age (see box 2) No information about parental preferences was given. All obstetricians were asked to classify each case again five months later to determine the level of intra-observer agreement after all cases had been arranged in a new order.

Inter- and intra-observer agreement on whether cases were classified in category 1 (1.1 or 1.2) or category 2 was established by calculating the number of agreeing obstetricians per case and percentages of agreement between pairs of obstetricians. Kappa coefficients for pairs of obstetricians and an overall kappa coefficient were determined according to Landis and Koch[9]: slight agreement: 0-0.20; fair agreement: 0.21-0.40, moderate agreement: 0.41-0.60, substantial agreement: 0.61-0.80 and almost perfect agreement: 0.81-1.0. The Wilcoxon ranking test was used to establish whether variability in categorization resulted from random or systematic differences between obstetricians. Both kappa coefficients between pairs of obstetricians and the Wilcoxon ranking test were calculated using the Statistical Package for the Social Sciences, version 10.0. The overall kappa coefficient was calculated using Stata, version 7.0.

Box 1. Examples of the case descriptions as provided to the obstetricians. In the instruction it was explained that all structures that were not mentioned in the case description had a normal appearance on ultrasound.

Case 33.
Lumbosacral spina bifida of 7-8 vertebrae. Club feet. Ventriculomegaly. Arnold-Chiari-malformation. Gestational age 21 1/7 weeks. Karyotype: unknown.

Case 56
Intra-uterine growth retardation, holoprosencephaly, micrognathy, hypotelorism, cleft lip. Heart: ventricular septal defect, overriding aorta. Gestational age 28 1/7 weeks. Karyotype: trisomy 13.

Case 77
Double bubble. Polyhydramnion. Gestational age 32 1/7 weeks. Karyotype: normal.

Box 2. Definitions of the categories of classification of fetal prognosis.

Category 1.1
No chance of survival; the abnormalities can not be treated.

This category includes fetal abnormalities, which are expected to inevitably lead to death during or immediately after delivery. In most cases, death will occur during or immediately after delivery, but exceptionally survival is somewhat longer.
Examples: severe lung hypoplasia, some severe and non-operable cardiac abnormalities, some skeletal dysplasias, bilateral renal agenesis, trisomy 13, trisomy 18, anencephaly.

Category 1.2
Chance of survival after delivery, but post-natal life-prolonging treatment is considered futile.

This category includes fetal abnormalities in the fetus, which lead to severe and non-treatable functional impairments. There is a (often limited) chance of survival. According to current medical opinion life-sustaining treatment after delivery will only prolong an existing situation, which is considered hopeless for the infant. Regarding the dismal prognosis, life-sustaining treatment can even be considered harmful.
Examples: very severe spina bifida, very severe hydrocephalus.

Category 2.
Chance of survival after delivery and post-natal life-prolonging treatment after birth, if necessary, is considered beneficiary.

This category includes abnormalities, which have a good chance of survival after birth. Neonatal life-sustaining treatment is either not necessary or, if necessary, is regarding the chance of survival and prognosis, considered beneficiary according to current medical opinion
Examples: isolated intra-abdominal cyst, unilateral hydronephrosis.

prognosis was impossible (n=4), or a combination of these reasons (n=2). The number of cases that was classified as category 1 (no chance of survival or postnatal treatment is considered futile) ranged from 17 to 50. Obstetricians 1, 2 and 3 systematically classified cases more often as category 1 than obstetricians 4 and 5 (table 1).

Table 1. Categorization by the obstetricians

	Obstetrician						
	1	*2*	*3*	*4*	*5*	*Total*	
	n	n	n	n	n		
Category 1	50	43	33	17	28	171	34%
Category 2	49	54	42	83	70	298	60%
Could not be classified	1	1	24		2	2	6%
Missing values		2	1			3	

Wilcoxon ranking test comparing the classifications of the obstetricians: obstetricians 1and 2: p= 0.09, 1 and 3: p=0.5, 1 and 4: p=0.00, 1 and 5: p=0.00, 2 and 3: 0.2, 2 and 4: p=0.00, 2 and 5: p=0.01, 3 and 4: p=0.00, 3 and 5: p=0.02, 4 and 5: p=0.01

Table 2 shows that the median percentage of cases for which pairs of obstetricians were in agreement was 76% (range 67%-93%). The overall kappa coefficient was 0.48 (moderate agreement), while the kappa coefficients for pairs of obstetricians varied between 0.31 and 0.86.

In 67/100 (67%) cases there was agreement on the classification between four or all obstetricians (table 3). If a single structural abnormality was present (n=73), in 51/73 (69%) cases four or all obstetricians agreed on the fetal prognosis. In this subset, fetal prognosis was classified as category 1 in 13/51(25%) cases. This included five out of six cases of spina bifida and three out of six cases of hydrocephaly. In the cases representing an intra-abdominal cyst (n=7), double bubble (n=5), echodense kidneys (n=2), gastroschizis (n=2) or intestinal dilatation (n=2), four or all obstetricians agreed that the fetal prognosis should be classified as category 2 (table 3). In the presence of multiple structural abnormalities (n=27), in 16(59%) cases four or all obstetricians agreed on the fetal prognosis. In this subset, in 9/16 (56%) cases fetal prognosis was classified as category 1. In the four cases with multiple structural abnormalities associated with an abnormal karyotype (i.e. trisomy 13 or trisomy 18) four or all obstetricians agreed that fetal prognosis should be classified as category 1.

In 82-97% of cases, the repeat classification was similar to the first classification (table 4). The median kappa coefficient between the first and repeat classification was 0.65 (range 0.64-0.93).

Discussion

The sample of 100 cases, which was used to evaluate a classification schedule for fetal prognosis, was drawn from a population of complicated cases for which discussion within a multidisciplinary team of a tertiary referral centre was considered necessary. Nevertheless, information about ultrasound findings, gestational age and karyotype (if available) was in the majority of cases sufficient for four out of five senior obstetricians to classify them into one of two main prognostic categories. One obstetrician was not sure about the fetal prognosis in a significant number of cases. On average, the obstetricians classified 34% of the cases in category 1, i.e. cases with abnormalities that are incompatible with life or for which the use of post-natal life-prolonging treatment is considered futile. This percentage is rather high due to the selection of cases. Since in the Netherlands no routine ultrasound is offered, ultrasound abnormalities are often not discovered until late in pregnancy or even after birth. [10-12]

In two thirds of the cases, most or all obstetricians were in agreement on the classification of fetal prognosis. On an individual level, the obstetricians were

Table 2. Obstetricians' inter-observer agreement.

Obstetricians	*n*	p_o	*Kappa*
1-2	96	82%	0.65
1-3	75	86%	0.73
1-4	99	67%	0.34
1-5	97	74%	0.48
2-3	73	93%	0.86
2-4	97	71%	0.38
2-5	95	72%	0.40
3-4	75	72%	0.39
3-5	73	75%	0.47
4-5	98	76%	0.31
Median		76%	
Overall kappa			0.48

p_o :percentage of observed agreement

Table 3. Degree of agreement for the different abnormalities.

	Agreement			
	3 or less obstetricians	*4 or all Obstetricians*		Total
		category 1	category 2	
Multiple structural abnormalities				
normal karyotype	6	2	6	14
abnormal karyotype	-	4	-	4
karyotype unknown	5	3	1	9
Single structural abnormalities				
intra-abdominal cyst	-	-	7	7
diafraphmatic hernia	2	-	5	7
renal abnormality	3		8	11
spina bifida	1	5		6
hydrocephaly	1	3	2	6
double bubble	-	-	5	5
intrathoracic cyst	3	-	1	4
no stomach filling	1	-	3	4
intestinal abnormality	1		3	4
gastroschizis	-	-	2	2
cardiac abnormality	2	-	-	2
skeletal dysplasia	1	1	-	2
encephalokele	1		1	2
other	6	4	1	11
Total	33	22	45	100

Table 4. Obstetricians' intra-observer agreement.

Obstetrician	*n*	p_o	*kappa*
1	99	82%	0.64
2	97	91%	0.81
3	67	97%	0.93
4	99	89%	0.65
5	97	87%	0.65
median		89%	0.65

p_o :percentage of observed agreement

consistent in assessing fetal prognosis based on the information provided. The substantial intra-observer agreement that was found in this study indicates that the case descriptions were adequate and that the differences between obstetricians represented systematic differences in opinions on how to classify cases. Some obstetricians appeared to be more inclined to consider an extremely poor prognosis than others.

Several factors may account for these systematically disparate opinions on fetal prognosis. First of all, obstetricians may have interpreted some case descriptions, in particular the sonographic information, differently and may therefore have come to different conclusions about the diagnosis. Secondly, the distinction in two prognostic categories, as proposed in the Dutch national guidelines, is based upon the diagnosis and the possibility to treat abnormalities after birth. It may not be obvious in all cases whether or not post-natal treatment for a specific diagnosis is possible.[13] For example, opinions may differ as to whether a hypoplastic left heart or a major spina bifida can be treated. Thirdly, the classification system used in this study relates to national guidelines for end-of-life-decisions in neonates, for which treatment is considered futile in case of no chance of survival or when the expected quality of life is extremely poor. [14-16] Examples of poor quality of life are life-long dependency on intensive medical treatment, physical suffering (pain and dyspnoea) and lack of ability to communication with the environment. However, judgments of what quality of life is unacceptably poor may vary between obstetricians. Moreover, whereas any prognosis involves a degree of uncertainty, the level of uncertainty that is accepted with regard to classifying abnormalities as incompatible with life or for which the use of postnatal life-prolonging treatment is considered futile may differ amongst obstetricians. The finding that obstetricians especially disagreed on the fetal prognosis in the presence of multiple congenital abnormalities with a normal or unknown karyotype supports this. Obstetrician four remarked that in case of prognostic uncertainty, even in case of severe abnormalities, he/she would give the fetus the benefit of the doubt by classifying the prognosis as category 2. The classification of obstetrician four may have been influenced by the fact that classification of all cases took place with another colleague.

It is not unlikely, that the obstetricians based their classification not only on the prognosis they thought to be most likely, but also on the type of obstetric management they thought was indicated.[17] Our comments concerning the factors underlying differences in opinion concerning the appropriateness of neonatal treatment also hold for obstetric management. Rational decision-making and prevention of undesirable variability could be improved by aiming at an evidence-based evaluation of fetal diagnosis and prognosis, using data, which are as objective as possible. Furthermore, medical and moral factors should be clearly distinguished in the assessment of prognosis and in the choice of obstetric management.[13,18-20]

A number of other issues have to be taken into account when evaluating the results of this study. Firstly, this study was carried out in the Netherlands, where there is a rather open societal and professional debate about end-of-life decision making.[21] As a result, Dutch obstetricians may be more familiar with the concept of futility of medical treatment and quality of life than most of their colleagues in other countries. Secondly, in clinical practice, the opinion of paediatric specialists is important in evaluating fetal prognosis as well. [22-24] Finally, the views of parents are obviously very important in end-of-life decision-making.

In conclusion, our study shows that obstetricians are usually able to classify fetal abnormalities into prognostic categories. However, we found systematic differences between the obstetricians' classifications, which may be explained by different interpretations of the ultrasound findings, different attitudes towards the acceptability of poor levels of quality of life, and different attitudes towards diagnostic uncertainty. Differences in the classification of fetal prognosis between obstetricians may result in differences in obstetric management between obstetric centres.

Rational decision-making and prevention of undesirable variability could be improved by aiming at an evidence-based evaluation of fetal diagnosis and prognosis, using data, which are as objective as possible. Furthermore, medical and moral factors should be clearly distinguished in the assessment of prognosis and in the choice of obstetric management. Finally, possibilities of neonatal treatment and its consequences for survival and quality of life should be taken into account before deciding on obstetric management.

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Chapter 4

Survival after non-aggressive obstetric management in cases of severe fetal abnormalities

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Abstract

Objective

Obstetricians may choose to refrain from interventions aimed at sustaining fetal life, (i.e., non-aggressive obstetric management) in some cases of extremely poor fetal prognosis. But as this approach does not always result in the death of the infant during delivery, neonatal management must now be considered.

We sought to provide empiric data concerning such perinatal end-of-life decisions. Firstly, to describe survival during delivery and after birth following non-aggressive obstetric management in pregnancies complicated by severe fetal anomalies. Secondly, to describe neonatal management in the live-born infants with severe anomalies.

Design

Retrospective descriptive study.

Setting

Tertiary centre.

Population

Eighty consecutive cases that were managed non-aggressively after the diagnosis of a severe fetal anomaly.

Methods

Data were collected from obstetric and neonatal records, as well as ultrasound reports.

Main outcome measures

Survival, neonatal management and health status after birth.

Results

Relevant data were available for 78/80(98%) infants. Six (8%) infants died in utero, 16 (21%) died during delivery (11 from cephalocentesis), and 56 (72%) were born alive. In 29 (52%) of the live-born infants, life-sustaining neonatal treatment was initiated. Twenty three of 29 (79%) infants died within 6 months of birth in case neonatal life-sustaining treatment was initiated, compared with 25/27 (93%) who did not receive neonatal life-sustaining treatment. Eight infants survived; each was profoundly handicapped.

Conclusion

Life sustaining neonatal support after non-aggressive obstetric management in case of severe fetal malformations has little to no beneficial impact on survival.

Introduction

The ultrasound diagnosis of a fetal anomaly with a poor prognosis provokes questions about future obstetric management. For example, caesarean delivery seems inappropriate for a lethal anomaly since it does not convey any benefit to the fetus whilst it is associated with iatrogenic maternal morbidity[1]. Here, the obstetrician may refrain from obstetric interventions. Such management, referred to as non-aggressive obstetric management, may result in the birth of a live infant. In this situation, prenatal management may have impaired the child's condition at birth. Furthermore, the birth of a live infant mandates a decision as to whether or not initiate life-sustaining neonatal treatment.

The debate on non-aggressive obstetric management so far has has centred around the ethical aspects. [1-3] Chervenak argued that non-aggressive obstetric management is acceptable when there is (1) a very high probability of the diagnosis and either (2a) a very high probability of death or (2b) survival with a very high probability of severe and irreversible deficit of cognitive developmental
Capacity. [4] However, clinical information about the sequelae of non-aggressive obstetric management is scarce. It is reported that some infants are born alive after non-aggressive obstetric management, but information on outcome is lacking[2,3]. The objective of the present study is to provide such information as a guide to the validity of this practice.

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Methods

The Erasmus MC is the largest tertiary referral centre for fetal anomaly scanning in the Netherlands, serving 3.5 million inhabitants and 35000 newborns/year. Between December 1995 and January 2003 consecutive women receiving non-aggressive obstetric management after the diagnosis of a severe fetal malformation were included. Cases are typically discussed after the ultrasound diagnosis by a multidisciplinary perinatal team consisting of obstetricians, neonatologists, paediatric surgeons and organ specialists, such as paediatric neurosurgeons, paediatric neurologists, paediatrics cardiologists and paediatric urologists. The multidisciplinary perinatal team, meeting weekly, decides as a team on obstetric and neonatal management. Non-aggressive obstetric management is defined as management in which obstetric interventions aimed at sustaining fetal life are partially or completely refrained from. Generally, such management is adopted when: i) the fetus has no chance of survival (i.e. certain death, mostly perinatal but in exceptional cases the child may reach the age of one year) or ii) the anomaly has some chance of survival, but health status is expected to be so severely affected that neonatal treatment is considered futile and in some cases even harmful. Families were counselled, and standard obstetric management adopted if the parents objected to a non-aggressive obstetric management. We obtained permission from the Ethics Committee of our hospital for this study.

Infants were included if: i) a fetal anomaly was diagnosed by ultrasound and ii) non-aggressive obstetric management was adopted. The minutes of the multidisciplinary perinatal team were used to establish which fetuses were selected for non-aggressive obstetric management. A physician (HB) experienced in prenatal diagnosis, but not involved in the clinical management collected the data retrospectively from obstetric and neonatal records, as well as ultrasound reports. In case the woman delivered in another hospital, relevant data were requested from the obstetric, neonatal and paediatric departments of the delivering hospital. Data points included survival in utero, during delivery, and during the neonatal and post-neonatal period. Furthermore, we evaluated whether or not neonatal life-sustaining treatment was initiated for live-born infants, as well as the perinatal characteristics. Finally, we

ascertained the health status of the children who had survived for a period of at least six months.

Table 1. Diagnostic characteristics in fetuses for whom a non-aggresive obstetric management was adopted.

Fetal abnormality	No chance of survival	Some chance of survival, but with severely affected health status	Total
	n=14	n=64	
Intracranial abnormalities	1	24	25 (32%)
Hydrocephaly	-	*15*	
Encephalocele	-	*2*	
Vein of Galen malformation	-	*1*	
Hydranencephaly	*1*	-	
Arterio-venous malformation	-	*1*	
Holoprosencephaly	-	*2*	
Multiple intracranial abnormalities	-	*3*	
Multiple structural abnormalities(1)	-	15	15 (19%)
2 abnormalities	-	*4*	
3 abnormalities	-	*8*	
4 abnormalities	-	*2*	
5 abnormalities	-	*1*	
Chromosomal abnormalities	8	-	8 (10%)
Trisomy 18	*4*	-	
Trisomy 13	*1*	-	
Trisomy 21 +hydrops fetalis	*1*	-	
Trisomy 22	*1*	-	
Monosomy 7, trisomy 10	*1*	-	
Spina bifida	-	6	6 (8%)
Bilateral kidney abnormality	-	6	6 (8%)
Cardiac abnormality	-	6	6 (8%)
Skeletal dysplasia	2	2	4 (5%)
Osteogenesis imperfecta type II	*2*	-	
Jeune syndrome	-	*2*	
Fetal akinesia syndrome	3	-	3 (4%)
Other	-	5	5 (6%)
CCAML	-	*2*	
Fetal hypokinesia+hydrops fetalis	-	*1*	
Congenital rubella infection	-	*1*	
Diaphragmatic hernia	-	*1*	

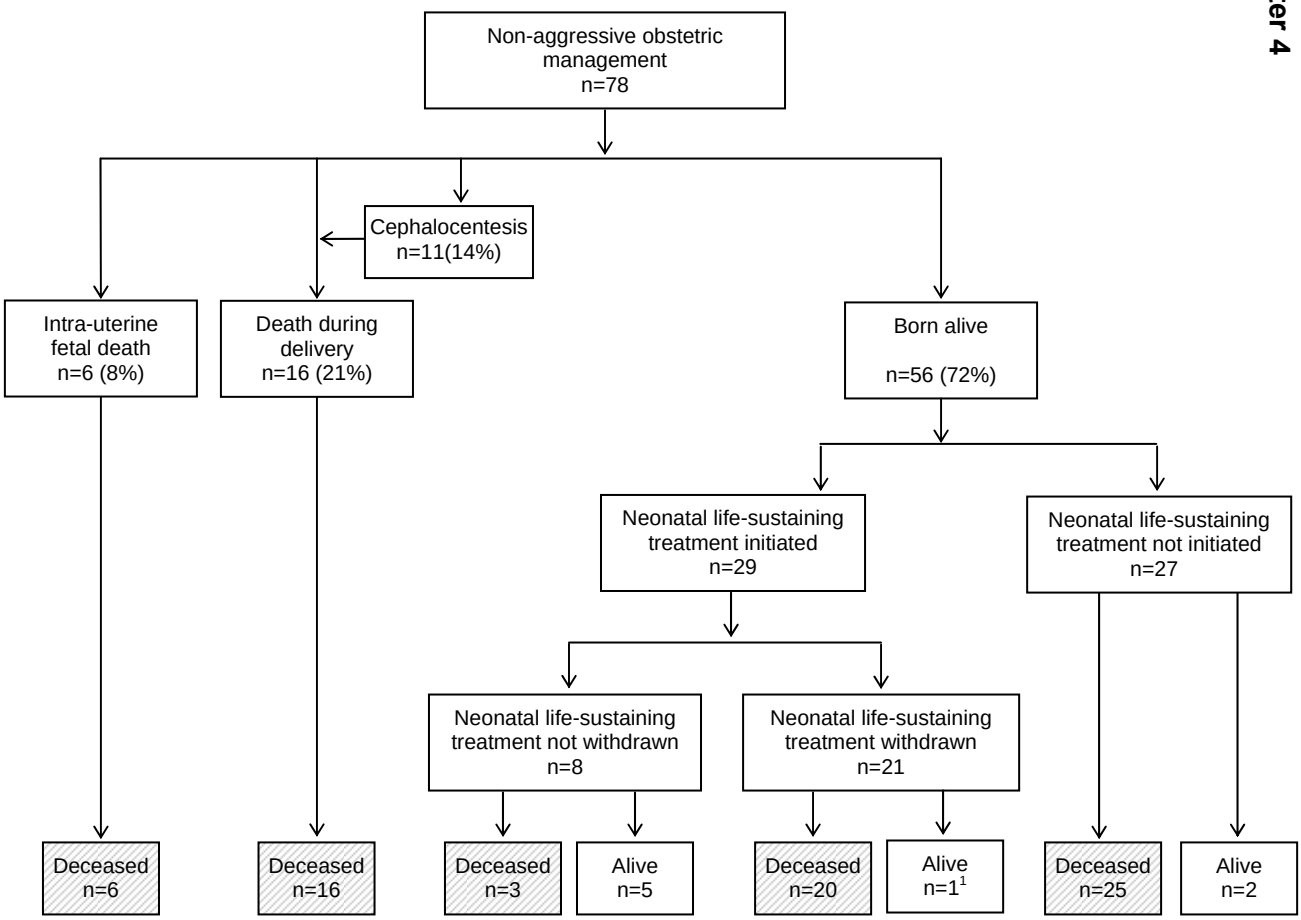
(1) normal or unknown karyotype

Table 2. Survival in utero, during delivery and after birth of infants with sonographically diagnosed severe fetal abnormalities in case of a non-aggressive obstetric management with and without neonatal life-sustaining treatment

Survival		Neonatal treatment initiated		Neonatal not initiated	Total
		withdrawn	continued		
n=78	n=22	n=21	n=8	n=27	n=78
Stillbirth	6	NA	NA	NA	6 (8%)
Death during delivery	16	NA	NA	NA	16 (21%)
Death within 24 hours after birth		4	1	19	24 (31%)
Death within first week after birth		11	2	4	17 (22%)
Death within six months after birth		5	0	2	7 (9%)
Alive after six months after birth		1	5	2	8 (12%)

NA: not applicable

Figure 1. Survival of infants with sonographically diagnosed severe fetal abnormalities in case of a non-aggressive obstetric management.



[1] withdrawal of neonatal life-sustaining treatment in this case consisted of an advance directive for DNR-management

Results

Non-aggressive obstetric management was adopted in 80 pregnancies with a severe fetal anomaly (table 1). Two pregnancies were excluded because follow-up data were not retrievable.

The median maternal age was 29.4 years (range 17.6-43.0 years) and the median gestational age at diagnosis was 30.6 weeks (range 12.6-38.0 weeks). Diagnostic characteristics are provided in table 1. In 14/78 (18%), the fetus was considered to have no chance of survival, and in 64/78 (82%), the fetus was thought to have some chance of survival but with an extremely poor prognosis (table 1). In three instances, non-aggressive obstetric management consisted of refraining from a caesarean section (on fetal indication). In the remaining 75 pregnancies, the recommendation was to refrain from all obstetric interventions to sustain fetal life. In total, 16/78(21%) infants were born before 34 weeks of gestation; 49/78 (63%) infants were delivered in the tertiary centre; in 6/78 (7%) cases, the delivery was assisted for maternal reasons.

Figure 1 and table 2 illustrate the survival of the fetuses for whom non-aggressive obstetric management was adopted. Six of 78 (8%) infants died in utero and 16/78 (21%) died during delivery, 11 of which as result of cephalocentesis.

Fiftysix of 78 (72%) infants were born alive. In 29 (52%) of the live-born infants, one or more life sustaining neonatal treatments were initiated including artificial ventilation (n=21), invasive procedures (n=11) and resuscitation (n=3) (table2). Twelve of 14 (86%) infants diagnosed prenatally as having no chance of survival were born alive; in one, life-sustaining treatment was initiated. Fortyfour of 64 (69%) who were diagnosed as having some chance of survival were born alive. In 28/44 (64%), life-sustaining neonatal treatment was initiated. Infants subjected to life-sustaining treatment had a higher median gestational age, birth weight and umbilical cord arterial pH compared with infants for whom no such treatment was initiated (table 3).

Table 3. Perinatal characteristics of fetuses in case of a non-aggressive obstetric management with or without initation of neonatal life-sustaining treatment

Perinatal characteristics	Neonatal treatment initiated	Neonatal treatment not initiated
	n=29	n=27
Gestational age (weeks;median range)	38.4 (28.9-43.6)	35.4 (29.3-42.9)
Birth weight (grams; median, range)	2975 (1415-4620)	1990 (1010-3380)*
pH (median, range)	7.27 (7.08-7.37)†	7.11 (6.90-7.30)‡
Apgar-score 1 minute (median, range)	5 (1-10) §	3 (1-9)¶
Delivery in tertairy centre (number, %)	20/29 (69%)	18/27(67%)

*2 missing values †11 missing values ‡ 19 missing values § 4 missing values ¶ 3 missing values

Of 29 infants receiving neonatal life-sustaining treatment, 22 (76%) died within six months (table 2). Of the 27 infants for whom no neonatal life-sustaining treatment was initiated, 25 (93%) died within six months. Life-sustaining treatment was withdrawn in 21/29 (72%) infants at a later stage. The stated reasons for withdrawal included: a poor prognosis of the congenital anomaly as already recorded before birth (n=8), maximum life-sustaining treatment had failed to be beneficial (n=7), postnatal diagnostic results confirmed a poor prognosis (n=3), neonatal complications (n=2) or unknown (n=1).

Table 4 describes the infants who survived. Long-term follow-up was missing for one child. The median follow-up time was 5.0 years (range 2.3-7.6 years). Five children were diagnosed with hydrocephaly, with or without spina bifida or multiple intracranial anomalies. Five children had a limited life expectancy and were dependent on medical care for daily activities. Six children had a developmental delay, five children will never live independently and two children had severely impaired ability to communicate. None of the children suffered from major hypoxic consequences, and none were considered to have cerebral palsy.

Discussion

We describe 78 fetuses with severe structural anomalies for whom non-aggressive obstetric management was adopted. About one quarter of these fetuses died in utero or during delivery, thus the majority was born alive. Life-sustaining neonatal treatment was initiated in about half. But whether or not neonatal life-sustaining treatment was initiated, most infants died soon after birth. Whilst 8/78 infants survived for at least six months, all had severe health problems. An earlier study noted that neonatal management is often not planned prenatally. [5] And when it was planned prenatally, in only 11% of the cases it was decided to refrain from life-sustaining neonatal treatment. [5]. We describe a substantial number of cases of non-aggressive obstetric management, and in doing so confirm the reasonableness of the approach. [3,6]

Table 4. Diagnosis, condition and follow-up period of children with sonographically diagnosed severe abnormalities born after a non-aggressive obstetric management

			(years)
1.	Spina bifida (Th8-L4), severe hydrocephaly	Paralysis from Th7, wheelchair-dependent, difficulties with sitting, urinary and fecal incontinence, special education	7.6
2.	Spina bifida (thoracolumbar), syringomyely, hydrocephaly	Paralysis from Th8, wheelchair-dependent, urinary incontinence, special care, normal psychomotoric development	6.3
3.	Severe hydrocephaly	Special education, developmental delay	6.8
4.[1]	Severe hydrocephaly, corpus callosum agenesia, atrophy occipital brain	Hemiparesis, impaired sight, impaired hearing, developmental delay of one year	4.3
5.	Multiple intracranial abnormalities, bilateral cataract	Tetraplegy, epilepsy, impaired swallowing, severe developmental delay, special nursing care	4.1
6.	Spina bifida (Th12-L5), hydrocephaly	Walks with support, urinary and fecal incontinence, some developmental delay	5.7
7.	Multiple structural abnormalities	Long-term follow-up missing.	
8.[1]	Hypoplastic left heart syndrome	Norwood-I surgery, further surgery not possible. Severely impaired physical endurance	2.3

The management of pregnancies complicated by severe fetal anomalies can either be standard, that is, all interventions necessary to keep the fetus in an optimal condition until birth are applied, or non-aggressive, that is, interventions that may harm the pregnant woman are avoided. The upper legal limit for termination of pregnancy in the Netherlands is 24 weeks of gestation. [7,8] After that, termination of pregnancy is only allowed when the fetus has no chance of survival. [7,8] Therefore, the options for obstetric management in case of a diagnosis of severe fetal anomaly after 24 weeks of gestation are often limited to either standard or non-aggressive management.

Only 12 live-born infants in our sample were considered to have no chance of survival. Non-aggressive obstetric management was associated with death in all cases, usually soon after birth. Life-sustaining neonatal treatment was initiated for only one of these infants.

Unfortunately, the majority of cases involved infants with anomalies that did not preclude survival, but were expected to convey severe health problems. Such problems may include the absence of cognitive development, daily dependence on medical care, lack of communicative possibilities, and physical suffering, such as pain or dyspnoea. [8-11] The prognosis of these infants was so poor, that obstetric interventions aimed at sustaining the life of the fetus were considered not to be in its best interest. Most of these infants were born alive, and birth leads to the initiation of life-sustaining neonatal treatment in a substantial number of cases. In accordance with previous findings, neonatologists tended to initiate life-sustaining treatment especially when the infants seemed vital, as reflected by good Apgar scores, normal birth weight and maturity. [12]

Thus, postnatal management seeking to sustain life may follow prenatal management that avoids life-sustaining interventions. There are several explanations for this. Firstly, interventions aimed at sustaining life after birth may be applied while the diagnosis made before birth is confirmed. Sustaining the life of the newborn infant makes diagnostic testing possible and allows follow-up of the newborn's vital condition and disease course. Whereas, any intervention before birth carries risks for the pregnant woman, the desire to confirm the diagnosis postnatally does not negate the decision to avoid life-sustaining interventions before birth. Secondly, the newborn infant may be (or seem to be) more vital than expected before birth. Thirdly, neonatologists rather than the multidisciplinary perinatal team decide the medical treatment after birth, and their view concerning the appropriate management may be

different from what they agreed upon before birth. To what extent neonatal interventions have prolonged futile suffering in these cases cannot be concluded from our study. Neither can we be sure either that non-aggressive obstetric management resulted in a less than optimal condition of the infant at birth and to a less than optimal health status after birth. Whether or not standard obstetric management would have improved the health status of the eight long-term surviving children is unclear, but seems unlikely considering the individual circumstances.

This study was carried out in the Netherlands where the societal and professional debate about end-of-life decision making is rather open, thus facilitating research and debate about decisions to limit life-sustaining obstetric interventions. [13] However, we used a retrospective approach to collect cases, because non-aggressive obstetric management is quite rare. This resulted in limited data about the dynamics of the decision-making process or the role of the parents.

Conclusion

Most anomalous infants are born alive after non-aggressive obstetric management, stressing the need to consider the neonatal approach in advance. In half the live-borns, life-sustaining treatment is initiated, reflecting differences between the prenatal and postnatal perspective. Life sustaining neonatal support after non-aggressive obstetric management for severe fetal malformations has little to no beneficial impact on survival. Only 10% of the infants for whom a non-aggressive obstetric management was adopted survived, and all have severe health problems.

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Chapter 5

Impact of decision-making in a multidisciplinary perinatal team

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Abstract

Objectives

1. To describe the characteristics of decision-making about management of unborn infants with serious abnormalities by a multidisciplinary perinatal team
2. To evaluate the impact of multidisciplinary team discussions on the degree to which decisions about the management of unborn infants with serious abnormalities are supported.
3. To evaluate the impact of the team discussions on the arguments used by physicians for their preferences concerning management.

Methods

Prospective analysis of 78 cases discussed within the multidisciplinary perinatal team of a tertiary centre by means of an anonymous one-page questionnaire with structured questions pertaining to the opinion of the responder on medical management of each case.

Results

We did not find systematic differences in between specialties prior to the discussion of cases. However, discussion with the multidisciplinary perinatal team improved decision-making about management of unborn infants with serious abnormalities by enhancing the degree of support for the decisions taken. The team discussions did not change the arguments physicians mentioned for their preferences.

Conclusion

Multidisciplinary team discussions improve decision-making about management of unborn infants with serious congenital abnormalities

Introduction

The diagnosis of serious fetal abnormality can raise difficult questions about perinatal management. In case of a poor fetal prognosis, the appropriateness of interventions aimed at sustaining life can be questioned. [1-4] Ethical aspects, diagnostic uncertainty, limited time and the involvement of parents whose future will be deeply affected by the decisions taken, complicate planning of perinatal management.[5-7] As both obstetric and neonatal interventions affect outcome, a multidisciplinary approach is widely adopted.[6,8-13] Different team members can analyse the case from different perspectives and add their own expertise. So far, little is known about the role of multidisciplinary team discussions in this field. Crombleholme, evaluating 221 cases of fetal abnormality in which prenatal surgical consultation was sought, found that the decision to terminate pregnancy was changed in 3.4%.[9] Regarding intellective decisions in health care, multidisciplinary team discussions generally are thought to lead to better decisions than those made by a single individual. [14] However, group discussions tend to focus on shared rather than unshared information.[15,16] Furthermore, group interaction is shown to suppress rather than enhance the expression of disparate opinions, thus impeding the possible beneficial influence of new views. [17] Finally, groups have been shown to make riskier decisions than do their individual members alone. [18] The aims of this study were 1) To describe the characteristics of decision-making about management of unborn infants with serious abnormalities by a multidisciplinary perinatal team 2) To evaluate the impact of multidisciplinary team discussions on the degree to which decisions about the management of unborn infants with serious abnormalities are supported, and 3) To evaluate the impact of the multidisciplinary team discussions on the arguments used by physicians for their preferences concerning management.

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Materials and methods

The Erasmus MC University Hospital Rotterdam is a regional tertiary referral centre for the southwest of the Netherlands, encompassing 3.5 million inhabitants and 35,000 newborns per year. The perinatal team of our hospital consists of a variable group of physicians, including physician-sonographers, obstetricians, neonatologists, paediatric surgeons, and other paediatric specialists, in the fields of neurosurgery, neurology, cardiology and urology. Midwives, general practitioners and nurses do not participate in the multidisciplinary perinatal team. Pregnancies complicated by fetal abnormalities are discussed by the perinatal team in case there is uncertainty about the diagnosis and/or foetal prognosis, in case the infant is expected to be referred for paediatric care after birth, or to discuss a request of parents to terminate a pregnancy that is already beyond 24 weeks of gestation, which is the upper legal limit for pregnancy termination in the Netherlands.

The multidisciplinary perinatal team (or 'the team') meets weekly and makes decisions on obstetric and neonatal management as well as on the intended place of delivery (i.e., whether or not the delivery should take place in a tertiary centre). Options for obstetric management include standard management, non-aggressive management and termination of pregnancy. Options for neonatal management include standard management or refraining from life-sustaining treatment. During the team's meeting, each case is presented by a physician-sonographer. Subsequently, the team discusses the case and makes decisions.

In our study, we prospectively included 78 consecutive cases, which were discussed by the team in a period of 17 months. For each case, we used a systematic checklist to register characteristics of the foetus, mother and pregnancy as presented, and characteristics of the discussion and decision-making. Further, the physicians participating in the meeting were requested to fill in anonymously a one-page questionnaire with multiple choice questions pertaining to their opinion about the most appropriate type of obstetric management (standard, non-aggressive, termination of pregnancy, no opinion (as yet), other), neonatal management (standard, no neonatal life-sustaining treatment, no opinion (as yet), other), their arguments for their preferences for both obstetric and neonatal management, and their opinion on where delivery

should take place (home/hospital with midwife/general practitioner, non-tertiary hospital with obstetrician, tertiary hospital with obstetrician, no opinion (as yet)). The questionnaire was filled in twice for each case: firstly, immediately after the presentation of the case (i.e. before the team discussion) and, subsequently, after the team discussion. The physicians also filled in their specialty, their position, and whether or not they had participated actively in the discussion, i.e. raised a question or expressed a view.

As no golden standard for a good standard decision exists for for decision-making in this field, we used the degree of support (consensus) for the decision as outcome measure. This is based both on the assumption that high degree of physician support reflects good quality of the decision and the fact that in clinical practice maximum support for the final decision is necessary for pragmatic reasons. The degree to which different specialists tend to have disparate opinions was measured by comparing percentages of physicians within each group that supported either of the management options, both before and after the team discussion. We also evaluated the percentage of physicians of the different specialties supporting the various options by means of correlation coefficients. We evaluated the impact of the team discussions in several ways. Firstly, we calculated the differences in mean percentages of physicians supporting the final decisions in the three areas of decision-making before and after the team discussions. Secondly, we assessed the number of cases in which consensus increased, decreased of stayed the same. Thirdly, we calculated the mean percentage of phycisians changing their opinion. Finally, we looked if characteristics of the discussions during meetings with a large increase in the degree of support, arbitrarily defined as an increase of 25% or more, differed from those during meetings with a lesser increase in the degree of support. Finally, we compared the arguments used within the specialties to support specific types of management, both before and after the discussions.

All analyses were done using the Statistical Package for the Social Sciences, version 11.0.

Results

Characteristics of cases and of the decision-making were registered during 36 meetings, during which the team discussed 78 cases. On average, 17 physicians attended these meetings. They received in total 1432 questionnaires, of which 1328 (93%) were completed and returned.

Table 1 shows the characteristics of the team meetings. Most discussions concerned third trimester pregnancies (mean gestational age 32.3 weeks (sd 4 weeks)). On average, 74% of the participants were senior medical specialists; a mean of 33% of all participants actively took part in the discussion. The presentation, discussion, and the final decision-making took, on average, 10 minutes per case (sd 6 minutes). The physician-sonographer usually presented information about the ultrasound findings, gestational age, and obstetric history, and showed the ultrasound findings on videotape. The cases discussed concerned multiple congenital abnormalities in 22/78 (28%) cases and single abnormalities in 56/78 (72%) cases. Three fetuses with multiple congenital abnormalities had chromosomal abnormalities (trisomy 13 (2) and marker chromosome (1)). In the remaining 19 fetuses chromosomes were normal (6) or unknown (13), because the results weren't known at the time of the meeting or because the parents did not want chromosome analysis to be done. A wide variety of single abnormalities was discussed: cardiac abnormality (9), double bubble (7), diaphragmatic hernia (6), gastroschizis (4), hydrocephaly/ventriculomegaly (4), intra-abdominal cyst (4), CCAML/lung sequester (3), bilateral renal abnormality (3), spina bifida (2), skeletal dysplasia (2), encephalocele/skull abnormality (2), sacrococcygeal teratoma (2), and other (8). Foetal prognosis was mentioned explicitly in the presentation of 51 cases (65%); uncertainty about the diagnosis or prognosis was mentioned in about half of the cases. Parental preference was mentioned explicitly in 30 (39%) cases. The team discussed specific alternatives for management in 33 cases (42%). Non-aggressive obstetric management or termination of pregnancy after 24 weeks were discussed in 34 cases (44%). The team most often decided upon standard obstetric management (60 cases, 77%) and standard neonatal management (52 cases, 67%). In 10 cases (13%) the team chose for non-aggressive obstetric management and in 8 cases

Table 1. Characteristics of participants, information provided at presentation, discussion and decisions of a multidisciplinary perinatal team of a tertiary centre.

Participants	mean	(sd)
mean number of participants per perinatal meeting	17	(6)
Specialty		
mean % participants of neonatalogy/paediatrics	30%	(16%)
mean % participants of obstetrics	32%	(15%)
mean % participants of paediatric surgery	35%	(16%)
mean % of particpants of other specialties		
Position		
mean % medical specialists	74%	(16%)
mean % residents	26%	(16%)
Active participation		
mean % participants taking an active part	33%	(18%)
mean % participants not taking an active part	67%	(18%)
Information provided at presentation		
cases in which was/were mentioned:	n	%
ultrasound findings	78	(100%)
gestational age	78	(100%
obstetric history	72	(92%)
whether or not obstetric complications other than fetal abnormality were present	68	(87%)
videotape images of ultrasound findings	68	(87%)
fetal growth	55	(71%)
indication for fetal abnormality scannning	53	(68%)
(explicite) fetal prognosis	51	(65%)
uncertainty of fetal prognosis	43	(55%)
differential diagnosis of the fetal abnormality (e.g.genetic syndrome)	36	(46%)
uncertainty of diagnosis	35	(45%)
parental preferences for management	30	(39%)
prior discussion of management between parents and gynaecologist	23	(30%)
prior consultation of paediatric specialist	25	(32%)
Discussion		
cases in which:	n	%
consensus about obstetric management was reached	78	(100%
several management alternatives were considered explicitly	33	(42%)
the consequences of the alternative options were discussed	25	(32%)
a non-aggressive obstetric management was considered	20	(26%)
termination of pregnancy > 24 wks gestational age was considered	14	(18%)
ethical issues were mentioned explicitly(1)	2	(3%)
Decision		
Obstetric management		
standard obstetric management	60	(77%)
non-aggressive obstetric management	10	(13%)
termination of pregnancy	8	(10%)
no decision		
Neonatal management		
standard neonatal management	52	(67%)
no initiation of life-sustaining treatment	4	(5%)
decision on management after evaluation of postnatal situation	4	(5%)
no explicit decision (2)	18	(23%)
Place of delivery		
tertiary centre	49	(63%)
non-tertiary centre	26	(33%)
no explicit decision	3	(4%)

Management in 6 cases, non-aggressive obstetric management in 4 cases

Table 2. Percentage of participants of each specialty that supported the different management modalities

	Obstetricians				Paediatricians/neonatologists				Paediatric surgeons			
	before		after		before		after		before		after	
	n=398		n=395		n=370		n=360		n=437		n=450	
	mean	*sd*	mean	*sd*	mean	*sd*	mean	*sd*	mean	*sd*	mean	*sd*
Obstetric management												
Standard	67	*41*	72	*42*	74	*38*	75	*40*	73	*38*	73	*40*
Non-aggressive	20	*35*	16	*32*	14	*29*	15	*32*	10	*22*	12	*29*
Termination of pregnancy	6	*22*	6	*22*	5	*19*	6	*22*	7	*22*	7	*25*
No opinion (yet)/other	8	*18*	6	*13*	12	*18*	4	*13*	10	*17*	6	*16*
Neonatal management												
Standard	64	*41*	72	*40*	71	*38*	72	*40*	71	*38*	72	*38*
No life-sustaining treatment	23	*36*	20	*36*	18	*32*	22	*37*	15	*28*	18	*33*
No opinion (yet)/other	13	*32*	9	*19*	12	*18*	6	*13*	13	*19*	11	*18*
Place of delivery												
No tertiary center	37	*38*	35	*41*	39	*39*	36	*42*	32	*33*	32	*38*
Tertiary center	59	*39*	63	*41*	56	*38*	62	*43*	63	*33*	66	*39*
No opinion (yet)/ other	5	*11*	3	*8*	5	*12*	1	*6*	6	*13*	2	*8*

Table 3. Mean percentage of consensus before and after discussion for each management modality

Final management decision	Maximum support			
	Before discussion		After discussion	
	mean	sd	mean	sd
Obstetric management (1)				
standard (n=60)	87%	20%	94%	14%
non-aggressive (n=11)	71%	26%	85%	20%
TOP (n=5)	80%	17%	90%	10%
Neonatal management (2)				
standard(n=61)	84%	21%	90%	17%
no LST (n=16)	72%	19%	83%	18%
Place of delivery 93)				
no tertiary center(n=25)	68%	20%	83%	15%
tertiary center(n=50)	78%	19%	89%	13%

(1) additionally, in one case the consensus concerned 'other management', and in one case the consensus for 'standard obstetric management' was equal to the consensus for 'non-aggressive obstetric management'.
(2) additionally, in one case the majority stated they had not yet an opinion on neonatal management
(3) additionally, in 3 cases, the consensus for 'tertiary center' was equal to the consensus for 'no tertiary center'

(10%) for the termination of pregnancy beyond 24 weeks gestational age. In four cases (5%), the team decided prenatally to refrain from life-sustaining treatment after birth.

Table 2 shows the mean percentage of obstetricians, paediatricians/neonatologists and paediatric surgeons supporting each management option, before and after discussion of the case. The degree of support for the different options was comparable between the different specialties, both before and after discussion. The percentage of physicians supporting a specific management option highly correlated between the different specialties, both before (Pearson correlation-coefficients, range 0.65-0.93) and after (Pearson correlation-coefficients, range 0.87 –0.92) the discussion.

The mean percentage of physicians changing their opinion on obstetric management after discussion of the case was 16% (sd 20%) while the mean percentage of those changing their opinion on neonatal management and on the place of delivery was 15% (sd 17%) and 19% (sd 19%) respectively.

Table 3 shows the mean percentage of physicians supporting a decision, before and after the team discussion. Before the discussion, the degree of support varied between 68% and 87%, and after the discussion it varied between 83% and 94%.

Overall, support for decisions on obstetric management increased with 6% (sd 15%). The degree of support for obstetric decisions increased in 34 (45%) cases (mean increase 18%, sd 14%), decreased in 13 cases (mean decrease 12%, sd 8%) and remained similar in 28 cases (37%): in 26 of the latter, the decision was already supported by the whole team before the discussion (figure 1).

In 10 cases (13%), the degree of support increased more than 25%. This relatively large increase often occurred after team discussions of the appropriateness of non-aggressive obstetric management (discussed in 9/10 (90%) of the cases for which the degree of support increased with more than 25%, versus 11/68 (16%) of remaining cases), of specific alternative management options (10/10 (100%) versus 23/68(34%)) and of the preferences of the parents (8/10 (80%) versus 22/68 (32%)).

Overall, support for decisions on neonatal management increased with 5% (sd 16%). The degree of support for neonatal management increased in 32 cases (43%), decreased in 16 cases (21%) and remained similar in 27 cases (36%): in 23 of the latter, the decision was already supported by the whole team before the discussion. In 6 cases (8%), the degree of support for decisions on neonatal management increased with more than 25%: In these cases, the degree of support for decisions on obstetric management also increased with more than 25%.

Finally, overall, support for decisions on place of delivery increased with 11% (sd15%) . The degree of support for decisions on the place of delivery increased in 56 cases (75%) (Figure 1). In 8 cases (10%), the degree of support for the place of delivery increased with more than 25%.

The arguments for supporting one of the obstetric and neonatal management modalities as reported by each of the physicians before the team discussions did not differ from those given after the team discussions (table 4). However, there were considerable differences between the different management options (Chi-square: $p<0.00$ for all arguments, both before and after the discussions). ‘There is no reason to depart from standard management’ was the most frequently mentioned argument to support standard obstetric management, whereas physicians who preferred non-aggressive obstetric management most often marked ‘poor prognosis in terms of quality of life’. For pregnancy termination, ‘limited life-expectancy’ and ‘parental preference’ prevailed. The most frequently mentioned argument to prefer standard neonatal management was that ‘there is no reason to depart from standard management’. The most frequently mentioned argument to prefer refraining from neonatal life-sustaining treatment was ‘poor prognosis in terms of quality of life’, followed by ‘limited life-expectancy’.

Discussion

This empirical study shows that discussion in a multidisciplinary perinatal team results in an increase of consensus about management of unborn infants with serious abnormalities, ranging from 6% to 15%. The increase in the degree of support was most prominent when regarding of end-of-life decisions, such as non-aggressive management and termination of

pregnancy. Hence, discussion in a multidisciplinary perinatal team results in better decisions about the management of unborn infants with serious abnormalities, thereby assuming that a decision based on a high degree of support is better than one based on less degree. [19,20]

This study was not designed for in-depth analysis of the psychological mechanisms influencing the group process in decision-making about the management of unborn infants with serious abnormalities. Hence, phenomena influencing group decision-making, such as suboptimal sharing of information and the tendency to make riskier decisions as a member of the team than as an individual, were not taken into account. The study rather focused on describing whether team discussion was beneficial, despite the possible occurrence of these phenomena and confirmed the value of team discussion for clinical consensus. For clinical practice, it is advised to collect factual information as well as participants' opinions systematically in order to avoid bias resulting from overrelying on shared information and generally accepted opinions. An impartial chairman may be needed to secure this process.[21,22] Furthermore, confusing moral matters with medical ones should not obscure the discussion. It has been shown that perception of medical facts, such as fetal prognosis, may be influenced by personal and institutional moral values.[23,24] However, moral matters should be discussed as such and not be disguised in discussion about medical facts.[25,26]

Interestingly, we did not find systematic differences of preferences between specialties prior to the team discussions (table 2). It can be questioned, therefore, to what extent the multidisciplinary team serves to attune disparate views between the different specialties.[27-29] In our study, most physicians attending the meetings were senior medical specialists who had already been involved with the perinatal team and its goal of multidisciplinary decision-making for several years. Possibly, these physicians were already used to consider perspectives from the viewpoints of other specialties due to long-term learning experiences. Hence the value of discussion in a multidisciplinary team may be by finding a common language for the team's participants rather than by attuning individual participants' opinions in specific cases. Furthermore, the perinatal team does not only contribute to the aim of making joint decisions, but also to the education of both novel apprentices and colleagues from different specialties, who have to get acquainted with the vocabulary, common knowledge and specific reasoning within different specialties.[14] In most cases, the team decided to apply standard obstetric and neonatal management. These decisions were typically motivated by the 'negative' argument that there was no reason to deviate from standard treatment, that is, treatment aimed at keeping the foetus alive and in the most optimal condition. Decisions to apply non-aggressive obstetric management or even to terminate a pregnancy, and decisions not to apply life-sustaining treatment for the newborn infant were less common. These decisions were usually taken because of an extremely poor prognosis in terms of quality of life, a limited life expectancy even if treatment would be applied, or because of the parents' wish to adopt such management.

Parental preference was mentioned explicitly in 30 (39%) of the cases. In general, parents are asked their preference for obstetric management at the time of diagnosis, especially when end-of-life decisions could be at stake. Termination of pregnancy after 24 weeks gestational age is only considered at explicit parental request. After the multidisciplinary perinatal team has made a decision about perinatal management, this decision will be discussed with the parents. In case parents object to a non-aggressive obstetric management, this will generally be converted to a standard obstetric management.

Few empirical studies have been done on the process and impact of medical decision-making in a multidisciplinary team and the most appropriate methodology for such studies has not clearly been established. Our study has to be considered as an attempt to contribute to the development of such methodology and our findings therefore have to be interpreted with caution.

Further, in a clinical setting, the characteristics of the decision-makers, the cases and the decision-making process are highly variable, and cannot be controlled for. This may explain that

Table 4. Arguments given for each obstetric and neonatal management modality. The participants could choose multiple arguments

	Obstetric management						Neonatal management			
	Standard		Non-aggressive		Termination of pregnancy		Standard		No LST	
	Before	After	Before	After	Before	After	Before	After	Before	After
	n=910	n=922	n=202	n=207	n=85	n=85	n=875	n=883	n=255	n=270
Life expectancy	190 (21%)	200 (21%)	116 (57%)	120 (58%)	58 (68%)	62 (73%)	192 (22%)	194(22%)	153 (60%)	175 (64%)
Quality of life	215 (23%)	199 (21%)	150(74%)	150 (72%)	52 (61%)	44 (52%)	213 (24%)	209 (23%)	184 (72%)	185 (68%)
Parental preference	100 (11%)	129 (14%)	50 (25%)	57 (27%)	42 (49%)	49 (58%)	87 (10%)	114 (13%)	64 (25%)	78 (29%)
No reason to depart from standard management	581 (63%)	587 (63%)	7 (4%)	2 (1%)	-	-	568 (64%)	567(63%)	-	3 (1%)
No opinion (yet)	17 (2%)	5 (1%)	-	-	-	1 (1%)	6 (1%)	5 (1%)	3 (1%)	1 (0%)
Other	46 (5%)	53 (6%)	3 (2%)	3 (1%)	5(5%)	2 (2%)	30 (4%)	36 (4%)	3 (1%)	4 (2%)

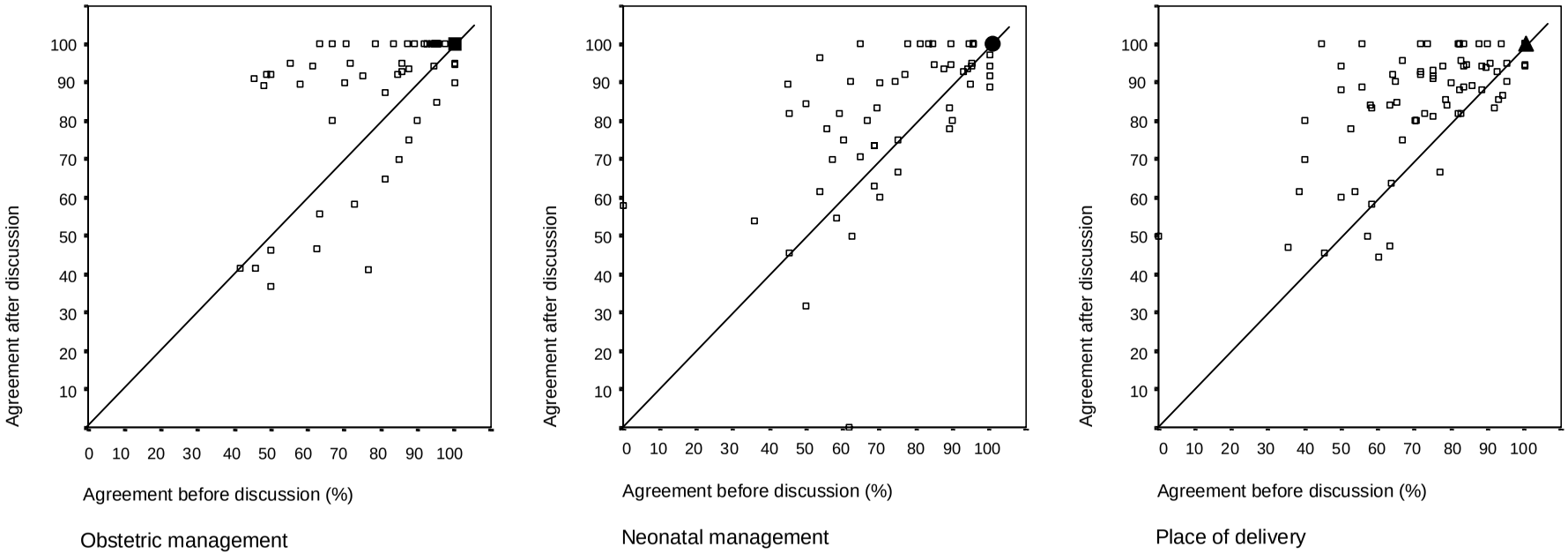

Figure 1. Agreement before and after discussion for obstetric management, neonatal management and hospital of delivery. Each square represents a case discussed by the multidisciplinary team. ■ represents 26 cases (obstetric management), ● represents 24 cases (neonatal management), ▲represents 9 cases (hospital of delivery). The black line indicates equal agreement before and after discussion. In the cases to the left of the black line, agreement has increased after discussing the case. In cases to the right of the black line, agreement has decreased after discussing the case.

we have not identified group characteristics contributing significantly to large increases in the degree of support for decisions within the team.

The team that was studied here shares many characteristics with perinatal teams in other tertiary centres: task-oriented, including obstetric and paediatric specialties and a variable number of physicians with variable levels of expertise and experience. Differences may exist in the participation of other health care professionals, such as social workers, midwives, nurses and general practitioners, the openness of the debate on end-of-life decisions and the legal regulations. These factors may influence the process of decision-making in other centres.

In conclusion, our study has shown that multidisciplinary team discussions about perinatal decisions have a considerable effect on the degree to which decisions are supported. Multidisciplinary decision-making appears to enhance the inclination of physicians to *a priori* consider perspectives from other specialties.

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