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Our Children Are Not a Diagnosis: The Experience of Parents Who Continue Their Pregnancy After a Prenatal Diagnosis of Trisomy 13 or 18

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Trisomy 13 and trisomy 18 (T13-18) are associated with high rates of perinatal death and with severe disability among survivors. Prenatal diagnosis (PND) may lead many women to terminate their pregnancy but some women choose to continue their pregnancy. We sent 503 invitations to answer a questionnaire to parents who belong to T13 and 18 internet support groups. Using mixed methods, we asked parents about their prenatal experience, their hopes, the life of their affected child, and their family experience. 332 parents answered questions about 272 children; 128 experienced PND. These parents, despite feeling pressure to terminate (61%) and being told that their baby would likely die before birth (94%), chose to continue the pregnancy. Their reasons included: moral beliefs (68%), child-centered reasons (64%), religious beliefs (48%), parent-centered reasons (28%), and practical reasons (6%). At the time of the diagnosis, most of these parents (80%) hoped to meet their child alive. By the time of birth, 25% chose a plan of full interventions. A choice of interventions at birth was associated with fewer major anomalies ($P < 0.05$). Parents describe "Special" healthcare providers as those who gave balanced and personalized information, respected their choice, and provided support. Parents make decisions to continue a pregnancy and choose a plan of care for their child according to their beliefs and their child's specific medical condition, respectively. Insights from parents' perspective can better enable healthcare providers to counsel and support families. © 2013 Wiley Periodicals, Inc.

Key words: trisomy 13; trisomy 18; life sustaining interventions; quality of life; parental opinions; ethics; end of life decision-making

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INTRODUCTION

Trisomy 13 and 18 (T13-18) are chromosomal disorders associated with high rates of neonatal and infant death and profound disability [Baty et al., 1994; Koogler et al., 2003; Parker et al., 2003; Rasmussen et al., 2003; Morris and Savva, 2008; Vendola et al., 2010]. For this reason, these conditions are frequently referred to as being "lethal" or "incompatible with life" in the medical literature. Some children do not have "full" T13-18 but instead have one of many variants of these chromosomal conditions, with outcomes than can vary in severity [Tucker et al., 2007; Griffith et al., 2009]. Prenatal diagnosis (PND) of T13-18 has increased due to improvements in prenatal imaging, higher use of prenatal serum screening, increase in maternal age and lower thresholds in examining the causes of intrauterine growth restriction. The technology behind expanded prenatal testing has developed rather rapidly which has led to changes in how prospective parents and physicians interact during the perinatal period [Hickerton et al., 2012]. Today, the majority of the diagnoses for these conditions occur in the prenatal period [Crider et al., 2008; Parker et al., 2010; Irving et al., 2011]. In the last

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Abbreviations: HCP, Healthcare providers; PND, renatal diagnosis; T13-18, Trisomy 13 and trisomy 18.

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decade, non-invasive prenatal genetic testing has also developed rapidly which suggests that diagnosis of T13-18 in the prenatal period will continue to increase. Non-directive prenatal counseling is recommended by genetics and obstetrics professional associations [ACOG Committee, 2009]. Many women choose termination of pregnancy for these life-limiting conditions. When the pregnancy is continued, the risk of miscarriage or stillbirth is high [Irving et al., 2011; Lakovschek et al., 2011; Loane et al., 2013].

The American Academy of Pediatrics Neonatal Resuscitation Program textbook recommends against newborn resuscitation for "lethal" chromosomal anomalies [AAP, 2010]. The 2010 American Heart Association guidelines make similar recommendations for T13 and 18 on the basis of "unacceptably high morbidity" [Morrison et al., 2010]. A recent study examined the experiences of parents who lived with children with T13-18 [Janvier et al., 2012]. The majority of parents reported that their family was strengthened and enriched since the birth—and often the death—of a child with T13 or 18 regardless of longevity. This difference in perception might cause health care providers (HCPs) to experience tension or confusion if they don't understand a woman's choice to continue her pregnancy. In a small qualitative study consisting of 19 families who received a PND of trisomy 18, parents describe dissatisfaction with health care experiences, mainly regarding communication, expressions of empathy from providers and their lack of participation in medical decision-making [Walker et al., 2008]. A systematic review of the literature related to parental outcomes after diagnosis of fetal anomaly revealed that many parents experience intense grief reactions regardless of the choice they make [Wool, 2011]. However, parents who were offered support and who chose to continue pregnancy described a positive experience in all explorative descriptive studies.

Recently, the American College of Medical Genetics has suggested broader applications to non-invasive genetic testing [Levinson, 2013]. As healthcare providers diagnose prenatal conditions more frequently, it is crucial to ensure ethical counseling and optimal communication. The overall goal of this article is to gain a better understanding of parents who decided to continue their pregnancy after a PND of T13-18. Prenatal counseling and perinatal support may be improved by acquiring knowledge about parental experiences.

METHODS

This study is part of a larger study; the general results of this study were reported, and a detailed description of the methodology can be found in this previous publication [Janvier et al., 2012]. For the purpose of this study, the analysis is focused on the experiences of the subgroup of parents who received a PND of T13-18.

Participants and Questionnaire

A computer assisted self-completion questionnaire was designed using expert opinion, including three focus groups and two pretests involving 10 parents. One of the collaborators in this study is a parent (BF). The 18 English websites and Facebook groups dedicated to T13-18 [Janvier et al., 2012; Trisomy support groups, online access 2011] were contacted and 570 email addresses of individuals whom had made their e-mail addresses accessible were obtained. Our inclusion criteria were: parents of children who live(d) with full T13-18, mosaicism, and other structural variations involving chromosomes 13 and 18 (called variants in this article). Our exclusion criteria were: respondents other than the parent, diagnoses other than T13-18, families who experienced in utero deaths and incomplete questionnaires.

The 503 possible participants received an invitation to participate in the study with the Internet link to the study site. They were then sent three reminders, with 3 weeks in between each reminder. The last reminder was sent in January 2011. The first page of the questionnaire informed respondents about the nature of the study and asked for their consent to participate. An informed consent box had to be checked in order to access the survey. A one-use link to the survey was generated for each participant and only responses obtained with this link were accepted to ensure that each individual parent could only participate once. All respondents were asked 10 open-ended and 12 demographic questions. Questionnaires were counted as complete if six specific questions had been answered: respondent identification (mother or father); diagnosis of "full" T13 or 18 or other variant; birth date of child, whether the diagnosis was made prenatally or postnatally, level of medical intervention provided; whether the child died before initial discharge home, and whether child was living at the time of the survey. The answers to these questions determined which additional questions were presented: from a minimum of 31 to a maximum of 106 questions. At the end of the questionnaire, parents were informed they could communicate with the principal investigator (AJ) or the parent representative (BF) if they had any questions or comments about the study, or if they wanted to communicate any further information they considered important.

Some multiple choice questions were asked to all parents, regardless of PND. Questions related to the child fell in the following categories: genetic diagnosis, congenital anomalies, medical problems, medical interventions, hospitalizations, medical needs at home, and neurodevelopment. Questions related to the family fell in the following themes: demographic information, effects of the diagnosis for the family, interactions with healthcare providers, and decision-making regarding interventions and level of care. Some parents in the two pilot studies were unfamiliar current medical terms used to describe levels of care. The term "interventions as for any other child" was the one best understood by all parents to define life sustaining interventions or "full interventions." The term comfort care was understood by parents and was used to describe withholding or withdrawing life sustaining interventions while focusing on the child's comfort. We included a third category (in between comfort care and "full interventions") because some interventions, such as tube feeds or oxygen, were considered to be in between "full" interventions and comfort care by parents in our pilot group. Other questions were only asked to parents who had a PND, for example, related to their decision to continue the pregnancy and their hopes.

In this article, we analyzed the answers to the following open-ended questions:

– "Please tell us up to three reasons why you chose to continue the pregnancy."

–"When you learned about the diagnosis of trisomy, what was your hope for [name of child]?"
–"Would you like to share more about the special healthcare provider you met at the time of the diagnosis (prenatal)?"
–"Tell us about up to three most helpful comments or actions made by healthcare providers (in the prenatal setting)."
–"Tell us about up to three of the most unhelpful/insensitive comments or actions made by healthcare providers (in the prenatal setting)."

Confidentiality

This questionnaire was designed with significant parental input. The questionnaire was anonymous in terms of respondent (parent) identification, but parents in the two pilot studies unanimously recommended we use the name of their child in open-ended questions. The questionnaire started by asking the name of their child, which was then mentioned throughout the questionnaire. Parents consented to the use of their child's first name in survey results. Parents who sent us pictures of their child did not want the pictures to be anonymous.

Analysis of Data

In this article, the analysis is focused on the experiences of the subgroup of parents who received a PND of T13-18. For relevant data, we compared the answers of two groups of parents: those who experienced PND versus those who learned about their child's condition after birth.

This is a study using mixed-methodology. Quantitative data were analyzed using Excel statistical software package. For questions related to parental perspectives we analyzed responses from all respondents. For questions related to clinical outcomes, we analyzed data on children with full T13-18 and who were still alive. When two parents answered questions for the same child, we used only maternal answers. We used descriptive statistics for quantitative data. Chi-square was used to compare proportions between groups.

All answers to the five open-ended questions were analyzed using NVivo 9 qualitative software package (QSR international). Open-ended questions were analyzed using thematic analysis [Denzin and Lincoln, 2000; Creswell, 2003; Hsieh and Shannon, 2005]. The themes were developed simultaneously by two research teams ("Eastern" team: AJ and BF; "Western team": BW, JG, and TK). These two teams developed themes independently and finalized the main themes, nodes and sub-nodes that would subsequently be used for coding. Specifically, using a thematic qualitative content analysis approach, themes and coding definitions were developed on the basis of the analysis of 30 respondent answers for each question by each team [Hsieh and Shannon, 2005]. This open coding was discussed between the investigators and each code was strictly defined to ensure thoroughness. After rigorous definitions of themes, node and sub-nodes, the coding of each question was simultaneously done by two primary coders (either two out of AJ, BF, JG, and BW) depending on the question analyzed. Coding of each question was then compared between the two coding researchers. Discrepancy in coding was either done by consensus between the two coders or by involving a third researcher. We used the basic matrix and modeling functions of NVivo 9 to generate comparisons between quantitative data and answers to open-ended questions. An expert in mixed methods was consulted to assist with this analysis.

This study obtained ethics approval from Sainte-Justine Hospital.

RESULTS

Demographic Information

Between October 2010 and January 2011, 503 parents received an invitation to participate in the study. Three hundred fifty-four surveys were returned and 332 were considered to be complete. Sixteen percent of diagnoses of T13-18 were made prenatally before 2000, compared to 49% after 2006, and 100% after 2008. This analysis is mainly focused on the experiences of the 128 (39%) parents who received a PND of T13-18, which consisted of 30 fathers and 98 mothers. Of those, 21 were couples and had an affected child. The majority of these parents (83%) were from the US, 5% from Canada, 5% from the UK, and 7% from 12 other countries. When their child was born, parents' median age was 38 years old: 4% were less than 20 years old and 25% older than 40. Seventy-eight percent already had children and 30% had three children or more. All parents completed high school, and the majority (73%) completed at least one university degree, with 21% also completing postgraduate studies. Parents generally described themselves as religious (85%), with 57% of the parents attending religious services.

They answered questions for 107 children: 75% of children were born after 1999 and 52% were born after 2006. Of the 107 children, 97 (91%) had full T13 or 18. Of the 97 children with full T13-18 who were diagnosed prenatally, 47% lived to go home, 28% lived more than 3 months, and 19% more than 1 year. At the time of the survey, the median age of survivors was of 3 years.

The remainder of the results falls into four areas: (1) reasons for continuing the pregnancy; (2) parents interaction with HCPs; (3) parental hopes and plans; (4) outcomes and family experiences; (5) comparison of parental experiences between PND and postnatal diagnosis.

Reasons for Continuing the Pregnancy

Reasons parents gave were analyzed and four main themes were identified: reasons related to moral beliefs, child-centered reasons, parent-centered reasons, and practical reasons (see Fig. 1).

Reasons related to moral beliefs. The most common reason parents cited for continuing their pregnancy related to their moral beliefs (77%) These beliefs were related to either personal values (68%) and/or explicit religious beliefs (48%). Sixteen percent of parents gave reasons that fell into both subthemes. Personal values included moral beliefs about abortion (42%) and fatalism (36%). The description of moral beliefs about abortion related to comments in which abortion was stated as not being acceptable, without religious reference. These comments were often accompanied with recognition of the fetus as a person:

> "*She was alive and kicking. Termination was not an option to me. It was the right thing to do, morally.*"

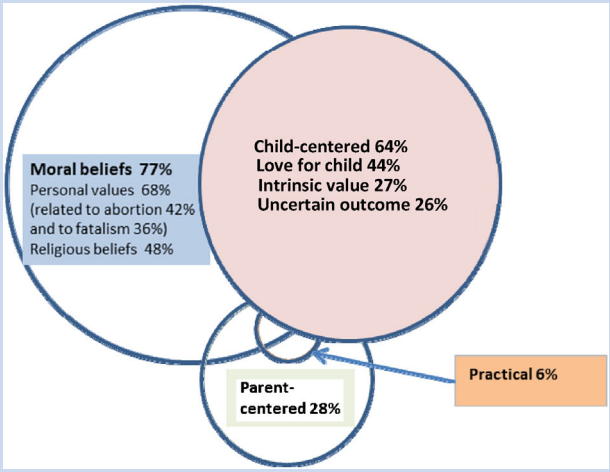

FIG. 1. Thematic reasons parents gave for continuing pregnancy.

"*Ethically it just felt it was the right thing to do for him.*"

"*I was already 5 months pregnant with JayLynn so she was already a person to me.*"

The description of fatalism included parents who believed it was better to let nature takes its course or a sense of destiny/natural order (this was "meant to be"):

"*We were going to let Alaina determine the outcome of her life, no intervention in one way or the other*"

"*If my son was going to pass away, I wanted it to be on his time, and not at my choosing.*"

"*We could not, we felt that this pregnancy was meant to be. We had very strong feelings prior to conception that we were supposed to open our home to another child.*"

Forty-eight percent of parents reported continuing their pregnancy for religious reasons, which included religious beliefs towards abortion:

"*We believe that God had entrusted us with his life and we weren't the ones who had the right to take that away—only God can.*"

"*Only God can take a life.*"

"*All children are a gift from God. It is our role to accept them as they are.*"

Child-centered reasons. The majority of parents (64%) expressed child-centered reasons for continuing the pregnancy. This theme includes love for their child (44%), intrinsic value of their child (27%), uncertain outcomes (26%), and giving the child a chance to live (36%). These sub-themes were often invoked in the same sentence. The most common sub-theme was the love that parents felt for their child:

"*Lucas is my son. The diagnosis made no difference to the fact I loved him. I wanted him to have his chances.*"

"*She was our baby, we loved her. She was already a member of our family. The kids read her stories at night and she was a part of our life.*"

Child-centered reasons also included the intrinsic value parents saw in their child, regardless of disability:

"*We loved her as we love our other children. Our children have value. We do not love our kids because of their accomplishments. We love our kids because they are our kids.*"

"*Luke's life was still valuable to me, regardless of his trisomy13.*"

Some parents discovered there are uncertain outcomes with consideration of the child's anomalies and wanted to give their child a chance:

"*After much research online, I found that there were children that were living with Trisomy 18. I also had many prenatal ultrasounds that found that she didn't have any incredibly horrible malformations that would keep her from possibly living.*"

Parent-centered reasons. Twenty-eight percent reflected a parental desire or benefit to the parent in continuing their pregnancy. The subthemes include the desire to meet their child, and spend time with their child, regardless of how long they believed their child would live:

"*I wanted a chance to meet my son alive and tell him I loved him and give him a cuddle and a kiss.*"

"*I wanted to build a relationship with my child no matter how short the time was with her.*"

Others did not want to experience future regrets if they terminated:

"*I was going to grieve anyway-(if terminating) so I might as well have a chance to have some good times as well as the grief.*"

"*We thought about it and discussed the option of termination but we both agreed that the regret we would have felt down the road could have possibly torn us apart, where raising a child with Trisomy 18 may have been challenging, it would have definitely brought us closer together.*"

"*I believe that continuing my pregnancy was beneficial to my long term emotional health because it allowed a more natural grieving process (vs. termination).*"

Practical reasons. A few parents (6%) had practical considerations for choosing not to terminate. These related to the inability to obtain a termination of pregnancy:

"*I was 28 weeks and too far along to terminate.*"

Parents' Interaction With Healthcare Providers

The majority of parents (63%) felt some HCPs did not view children with T13-18 as unique children and that they did not look beyond the grim statistics of these conditions (84%). Following the diagnosis, parents report being told by a HCP that their baby would likely die before or at the time of birth (94%), that their baby would not live for more than a few months (88%), that the condition of their baby was lethal or incompatible with life (93%), that their child would be a vegetable (55%), that their baby would destroy their family or their marriage (28%), and that if their baby survived, he would live a meaningless life (55%) or a life of suffering (68%). However, parents were also told that some children live for many years (47%), that their child might enrich their lives (12%) and that some children live a short, meaningful life (51%). The information and predictions provided to parents were similar regardless of whether the fetus had full or variant T13-18, holoprosencephaly, a heart anomaly, intrauterine growth retardation, or any combination of the last three findings mentioned. Providers of perinatal hospice/palliative care were reported to have provided similar information about T13-18 as other providers.

Meeting a special HCP. Parents were asked if they met at least one special healthcare provider at the time of the PND and in the perinatal period. Fifty-six percent reported meeting at least one special HCP, with all specialties and professionals involved in PND including nurses, ultrasound technicians, genetic counselors, and physicians in genetics, obstetrics, neonatal medicine, cardiology, palliative care were represented. Thirty-six percent of parents met a palliative care provider following PND and 68% of these found it helpful. When asked to describe their special HCP, several themes were identified. Several themes were often present in parental responses. These themes were: providing information in a balanced manner, respecting decisions, treating their unborn child as an individual, and allowing the parents to have reasonable hope.

"Our first OB refused to continue seeing us if we would not abort, so we found a new OB. He was wonderful, always called Jordan by name. He allowed us to have long ultrasounds so we could have time seeing Jordan alive."

"*We were fortunate to be able to work with a specialist who was very concerned about making sure that we had accurate information of both the positive and negative potential outcomes. Her main concern was to help us understand what was happening in Aaron's own unique case, not just what happens to the average T18 baby.*"

"*We found a pediatrician that had never had a trisomy 13 baby, but was very willing to work with us. He was at our bedside within 45 minutes of being admitted to L&D, and stayed until Nathan passed. He never left, and was very comforting.*"

"*The cardiologist was the only doctor who treated Carly with respect and called her by name before she was born. He said, "I can help you.""*

"*We had a consultation with a neonatologist to discuss the philosophy, viewpoints, and options we had once Madison was born. He discussed limitations that we would reach and the philosophy of the hospital but also gave us a chance to share our perspective and our wishes for Madison's care.*"

Supportive actions made by healthcare providers. Parents noted many actions as being very beneficial to them throughout the perinatal period. Parents cherished sonogram pictures and keepsakes of their fetus knowing that they could miscarry or only have a short time with their child. They appreciated that their provider called their child by name. Parents liked their pregnancy to be managed in the same way as other pregnant women, including monitoring, blood tests and ultrasound exams. Parents appreciated being prepared for the life or the death of their child in a practical sense. Practical information was thought to be invaluable: the importance of pictures, breastfeeding, spiritual or religious concerns the family may have, the role/presence of the other children or family members. After birth, some parents noted that they were touched when a HCP held or spoke to their child.

Pressure to terminate pregnancy. The majority (61%) of parents reported feeling pressure to terminate the pregnancy. In the open-ended questions, 50% described their experience and gave specific examples:

"I was told by the geneticist that the only way I could get an appointment with the main Obstetrician was if I was booking in for a termination."

"The obstetrician encouraged abortion, saying that we would never find any doctors to treat her. We would be doing her a favor by saving her from suffering…"

"I was told many times that abortion was definitely the best option for us and I had full support to have an abortion right up until my 26th week of pregnancy but hardly any support for wanting to carry on the pregnancy."

"After we confirmed again we would not terminate, we got told that the best thing that can happen now is if your baby dies then you can get over this and try again."

Parental Hopes and Plans

Hopes for child at time of PND. Parents described their hopes when they first heard about the diagnosis. The most common hope was that the child would be born alive and that parents would have a modest amount of time to spend with their child (80%):

"Based on our research, all we really hoped for was that he was with us long enough to hold him."

"My hope was that Lilly would live. That my family and I would be able to spend some time with her. That she would be born to term, instead of miscarry[ing] or be stillborn like all the Specialists said would happen."

Some parents (20%) hoped that their child would exceed expectations or be one of the survivors:

"We hoped she would be one of the 10% who lived for a year";

"I expected the worst but hoped for the best. I was going to get every service available to help Candace progress as far as she could. I was hoping when she grew up she would be able to live with assistance."

Some parents reported that they hoped that their child would remain comfortable throughout their life, without experiencing any pain or suffering (10%). Others simply hoped that their child would feel or know that were loved (12%). Only a few expressed the hope that their child was either misdiagnosed or that a miracle would occur (4%).

Choosing the plan of care. After the diagnosis, 53% of parents chose comfort care for their child, 25% chose interventions "as for any other child" and 22% chose interventions "between comfort care and full intervention" (see Table I); 64% of parents who chose some degree of intervention felt judged for the decisions they were making.

The plan of care was not associated with the parents' education, religiosity or any other demographic factor. On the other hand, an association between plan of care and reasons provided by parents for continuing the pregnancy was observed. All parents who had practical reasons for continuing pregnancy chose comfort care. Parents who provided only child-centered reasons to continue pregnancy (n = 47) chose "full interventions" 40% of the time whereas parents who provided only parent-centered reasons (n = 12) never chose full interventions.

Plan of care was significantly associated with the child's anomalies. Parents whose child had neither cardiac defects nor holoprosencephaly were more likely to choose full interventions than when the child had both anomalies (47% vs. 10%; $P < 0.05$). Variant T13-18 (as opposed to full T13-18) was associated with more parents choosing interventions (64% vs. 22%; $P < 0.05$). Of the 107 children who were born, 38 (36%) received care from a specialized perinatal hospice or palliative care service. Of those, 25 (66%) received comfort care, 6 (16%) full interventions while the remaining 7 (18%) had a birth plan parents identified as "in between" comfort care and full interventions.

Outcomes and Family Experiences

Survival of children. Of the 107 children who had PND, 25% received "full intervention as for any child" and 53% received comfort care (see Table I). Children who received comfort care were more likely to die in their first day of life compared to children who received interventions ($P < 0.001$) (Table I). Children who received interventions were also more likely to live longer than 1 year than children who received comfort care ($P < 0.05$) (Table I).

Family experiences. The majority of parents whose child died describe the overall experience of their child's life as being positive, irrespective of the length of their lives. Parents whose child lived longer than 3 months described happy children who enriched their lives although they did admit to various challenges (Table II).

Parents were informed they could communicate with the principal investigator (AJ) or the parent representative (BF). Parents sent us many pictures of their children and families: some of the babies in the pictures lived minutes; others lived longer than 1 year, with or without medical interventions. All parents gave permission to use their child's picture and furthermore wanted their child's name next to the picture. We observed a stark comparison between the pictures provided by parents and those that are found in medical articles (Figs. 2 and 3).

Parents were asked what they would do if faced with another PND of T13 or 18: 91% answered they would not terminate the pregnancy and of these, 11% would not pursue prenatal testing, 5% were unsure, and 3% report they would terminate the pregnancy (see Table II).

Comparison of PND and Postnatal Diagnosis

Parents who experienced PND and postnatal diagnosis were told the same information about their child, although interaction with HCP was described as being more difficult for those with a PND than those diagnosed postnatally. For example, parents who chose some degree of intervention (as opposed to comfort care) were more likely to feel judged if they experienced PND (64% vs. 24%, $P < 0.05$). There was a higher proportion of children diagnosed in

TABLE I. Outcome of Children Who had a Prenatal Diagnosis of Trisomy 13 or 18 According to Level of Intervention

Level of care	Lived <1 day	Went home	Lived >3 months	Lived >1 year
All levels (n = 107)	34% (36)	50% (54)	32% (34)	23% (25)[a]
Comfort care (n = 57)	53% (30)	40% (23)	25% (14)	18% (10)
In between full interventions and comfort care (n = 23)	9% (2)	70% (16)	30% (7)	17% (4)
Full intervention, as with any child (n = 27)	15% (4)	56% (15)	52% (13)	41% (11)

[a]Median age of surviving children is 3 years.

TABLE II. Perspectives of Parents Who Received a Prenatal Diagnosis of Trisomy 13 or 18

For all respondents n = 128	
I feel that some providers don't see T13-18 as unique children.	63% agree
I feel that some providers don't look beyond the grim T13-18 statistics.	84% agree
The effect on my marriage/ relationship was:	75% positive, 2% separate/divorce
If I was pregnant again with a baby with T13-18 I would continue pregnancy.	91% yes or no test, 7% unsure, 2% no
For respondents whose child died (n = 97)	
How would you describe the overall experience of your child's life?	91% positive
Did you do the right amount of medical interventions?	77% yes, 21% not enough interventions
For respondents whose child lived >3 months (n = 42)	
My child is a happy child.	91% agree
My child enriches our family life.	97% agree
How do you think having a special needs child has affected siblings? (n = 28)	82% positive
My child experience more pain/discomfort than other children.	25% agree
Caring for my child is more difficult than I thought it would be.	34% agree
My family experiences significant financial challenges.	43% agree

utero when holoprosencephaly was present (32% for PND vs. 16%; $P < 0.05$).

DISCUSSION

We describe the perspectives of 128 parents who experienced a PND of T13 or 18 and decided to continue their pregnancy. These parents describe various reasons to continue pregnancies. Most frequently, they felt that it was the best choice for their child and family or because termination conflicted with their personal values. They loved their child, already felt a connection and many already considered him a part of their family. Some parents did not want to take an active role in deciding the length of their child's life. Parents recognized an intrinsic value in their child. This value was not defined by length of life nor by traditional developmental milestones and achievements. When they first received the diagnosis and after most were told that their child might not live to be born, their most common hope was that the child would be born alive and that they would meet him. Parents report understanding the implications of the diagnosis and only a few had hopes for a miracle or a cure. These modest hopes reflected acceptance of the serious condition.

Many parents also compared termination with continuing the pregnancy with respect to the impact on their lives, their regrets in the future and concluded the latter was the better option for their personal or family healing. The majority of these parents felt pressure to terminate their pregnancy. This pressure often persisted and came from different providers. Some parents sought another provider because of this pressure or because their provider refused to continue care. The experience of pressure to terminate, abandonment and isolation after choosing to continue pregnancy after

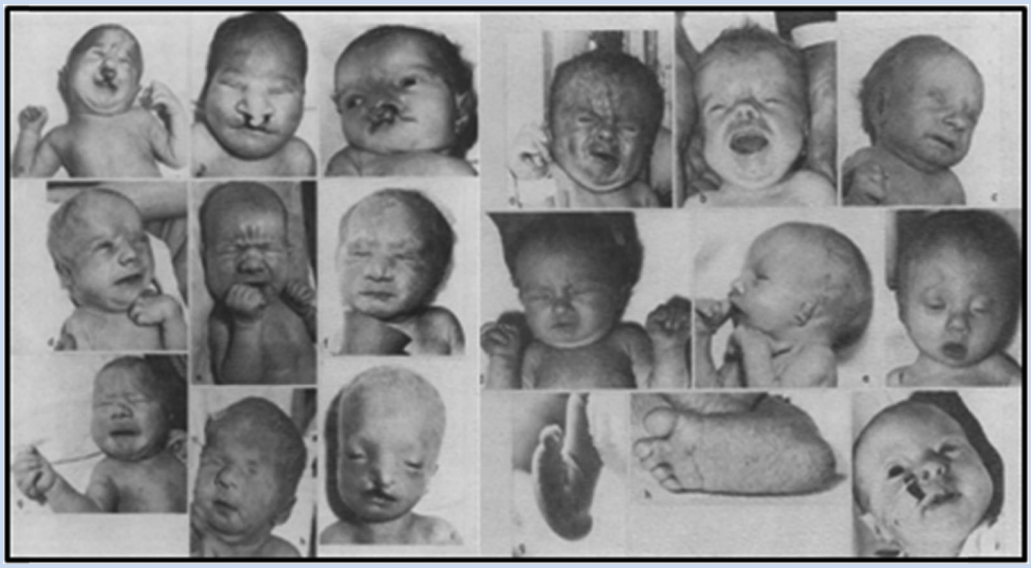

FIG. 2. Typical pictures of children with trisomy 13 and 18 found in the literature (from Taylor, 1968; republished with permission).

FIG. 3. Family pictures of children. From top left to right: Gianna, full T18 (died 1 week), Nolan, full T18 (died 2 years), Beth, full T13 (died 3 months), Guiliana, mosaic T18 (2 years), Emma, full T18 (died 5 years), Joey, full T13 (5 years), Sofia, full T13 (6 years), Allison, full T13 (died 1 day), Annie, full T18 (died 12 years), John, full T13 (died 1 year), Caitlyn (3 tri 18), Cathal, full T18 (died 1 day), Sophee, full T18 (died 6 months), Bristol, full T18 (died 2 months), Devon, full T13 (17 years).

an adverse PND has been reported in other studies and narratives [Redlinger-Grosse et al., 2002; Walker et al., 2008; Farlow, 2009; Thiele, 2010; Côté-Arsenault and Denney-Koelsch, 2011; Farlow, 2011; Berg et al., 2013]. Because of T13-18 outcomes and the assumption of adverse parental and familial outcomes, it is plausible that providers who encourage pregnancy termination act out of a desire to help their patients. Many parents were told that if they terminated pregnancy, they could "go on with their life" or "have another child". However, parents in this study thought of their child as a unique person, not one that could be replaced.

Many parents reported that taking care of a disabled child was more difficult than they thought it would be and brought about significant challenges. A quarter of parents thought that their child experienced more pain than other children. Despite this, parents of surviving children reported their children were happy and enriched their families. Almost all parents stated that if they received a PND of T13-18 in the future, they would continue the pregnancy or would avoid prenatal testing. As reported by similar studies related to the decision to continue pregnancy for severe and life-limiting conditions, these parents overwhelmingly report a positive experience, even if their child lived only a short time [Calhoun et al., 2003; D'Almeida et al., 2006; Breeze et al., 2007; Janvier et al., 2012]. It is common for parents who receive an adverse PND to receive similar negatively biased information from HCPs for various conditions [Walker et al., 2008; Lathrop and Van de Vusse, 2011]. This information may reflect the common belief that severely disabled children with life-limiting conditions suffer, are a burden to their families, cause divorce and neglect of other children [VanDyke and Allen, 1990; Kopelman, 2010; Merritt et al., 2012]. The assumption of a negative family experience or that surviving children have little or no cognitive ability or worth is often stated in papers without data to support those claims [Bos et al., 1994; Sobsey, 2004; Catlin, 2010; Kumar, 2011; Chervenak and McCullough, 2012; Merritt et al., 2012].

Parental decision-making was not homogenous and reported outcomes were diverse. The majority of parents in this cohort did not choose full interventions for their child. We found that the information these parents describe receiving at the time of PND was similar, independent of the anomalies of the fetus or the variant of T13-18 (full, mosaic, partial, etc.). However, the birth plan chosen by parents was dependent on the major anomalies of the child. For example, few parents chose interventions when their child had both a cardiac and serious brain defect. This pattern suggests that parents consider their child's unique anomalies and make decisions accordingly whereas providers counsel according to the general diagnosis. In our large cohort, the majority of parents who experienced PND and chose any intervention to prolong the life of their child (including tube feedings) felt judged much more so than parents who obtained a post natal diagnosis. Is it possible that HCPs see parents who continue their pregnancy despite this diagnosis as having some responsibility over the poor outcome, as a situation they could control and avoid? While only a minority of parents (35%) had access to perinatal palliative care services, most found it helpful. The provision of this service did not relate exclusively to plan of comfort care.

Ideally, prenatal counseling should be non-directive and informative to allow parents to make decisions consistent with their values. The information given to women and prospective parents

should be balanced and accurate [Carey, 2012]. HCPs involved in PND need to be aware of new data about these conditions as well as the spectrum of outcomes in order to give accurate information [Bruns, 2010, 2011]. These conditions have been traditionally described as lethal. Yet, in Japan, the approach to these conditions has been different for many years and full interventions are often provided. The 1-year survival rates have been reported to be as high as 56% in some Japanese studies [Kaneko et al., 2009; Maeda et al., 2011; Tsukada et al., 2012]. A 12-year review of hospitalizations for American children with T13-18 revealed that more interventions are performed for these conditions than previously thought and concluded "universal application of the term "lethal" to the diagnoses of trisomy 13 and 18 is not appropriate" [Nelson et al., 2012].

The majority of parents met a special HCP. Special HCPs were described as giving accurate and comprehensive information. They did not pressure the parents to terminate the pregnancy and supported them. They provided balanced and personalized counseling; appropriate hope about the uncertainty of the child's birth, life span or about the value of the short time parents might have with their child. These Special HCPs did not focus on the impossibility of curing the trisomy, but rather on the meaning of the child's life and the healing of the family. Our results suggest that information parents are given in prenatal counseling after a diagnosis of trisomy 13 and 18 is commonly disparate from their experience. Parents who discover happy families and surviving children on social networks after being told their child was had a lethal condition or would be a vegetable may develop mistrust, conflict and distress with their HCP [Janvier et al., 2012]. One of the most prevalent messages from parental responses is that the language used by providers matters. Terms to describe one's child such as "lethal anomaly" and statements that the child is "incompatible with life" were often discussed as the least helpful comments made by providers. Comments about lethality may turn normative judgments into clinical ones [Koogler et al., 2003].

Traditionally, recommendations for postnatal management for these patients limit interventions [Bos et al., 1992; Paris et al., 1992; Calhoun et al., 2003; Leuthner, 2004; McGraw and Perlman, 2008; Catlin, 2010, Chervenak and McCullough, 2012]. Newborn resuscitation has been recommended to be withheld (AHA guidelines, NRP statement; ILCOR statement) and care has often been limited to comfort measures to avoid the "burdens and sufferings" of a "genetically doomed child" who would have "unacceptable morbidity" [Paris et al., 1992; Morrison et al., 2010; Merritt et al., 2012]. These postnatal non-personalized guidelines and recommendations may influence prenatal counseling. Is it possible for a provider involved in prenatal counseling to be non-directive when there are explicit directive policies about postnatal care for these conditions?

These results need to be interpreted with caution as we did not conduct a population study and these children are not a representative sample. For example, the survival in this cohort is much higher and longer than that described in the medical literature [Janvier et al., 2012]. We included parents of children with mosaicism and variants. As noted earlier, we have no data on women who chose to terminate their pregnancy or data on couples who experienced a fetal loss. Further, these are self-reported questionnaires with inherent biases, which include recall bias and recruitment bias. Despite these limitations, because of our high response rate and large sample size, we are confident that our data provide a good representation of the experience of parents who decide to continue their pregnancy after a PND of T13 or 18. Additionally, this community of parents participating in online social support networks likely influence parents who face a new T13-18 diagnosis.

Parents who experience a PND of T13-18 may decide to continue their pregnancy for a number of reasons. They may have views, hopes and expectations that are incongruous with those held by some of the clinicians they will encounter. HCPs need to understand parental perspectives and realize that while T13-18, cannot be "cured", the children have value and meaning to their parents regardless of life span and disability. HCPs can provide many positive actions to prepare parents for the life or death of their child. All these interventions result in a measure of "healing" without cure. Pictures are worth a thousand words. The contrast between family pictures and the pictures of children trisomy 13 and 18 found in medical texts is striking and demonstrate the contrasting representations of children with these conditions. Providers should be aware of the experiences of parents represented in this article. These perspectives complement the medical literature and could be incorporated in counseling families with a PND of trisomy 13 or 18.

CLINICAL GUIDELINES AND IMPLICATIONS

Based on the information obtained in this study from parents who continued their pregnancy after a diagnosis of T13-18, we offer suggestions to assist healthcare providers to provide optimal prenatal care.

1. At the time of diagnosis, provide accurate survival figures. Avoid words like "lethal," "incompatible with life" and "vegetable". Avoid value-laden language related to disability.
2. Parents should be informed that most parents who chose to continue pregnancy have reported a positive and enriching experience regardless of the lifespan of their child.
3. Parents who decide to continue their pregnancy need support, not judgement or pressure to change their choice. Parents accept that early death is likely and they have chosen to value the time they have, both before and after birth.
4. Remember that to these parents, their child is a person, not a diagnosis. Refer to the unborn child by name, if possible. Parents expect to receive medical information related to their child, not to the diagnosis.Informing parents of normal organs in addition to anomalies is greatly appreciated. Offer hope when it is reasonable: hope that baby will continue to grow in utero, hope that baby will be born alive and that parents will enjoy some time with baby.
5. Offer to continue prenatal and fetal care as for any pregnancy. Ultrasounds are very special, memorable events and given the high risk of miscarriage, might be the only time parents will see their living baby. Taking a few minutes during the ultrasound to point out normal or "cute" features of the baby can be a lifetime gift to parents.
6. Guide parents to create a birth plan that is best for their child and family. Parents should understand that children with T13 or 18

are unique and some might benefit from life sustaining interventions while some may be harmed by them. Ensure that the birth plan includes collectables for memories such as foot prints and photographs. If indicated, be transparent with parents about any hospital protocol or policy that restricts certain interventions to babies born with T13-18. Parental challenges to these restrictions should be discussed in a multi-disciplinary meeting or ethics consultation.

7. Most parents who choose to continue pregnancy do so because it is the better path according to their personal beliefs. They appreciate empathy and kindness on their extraordinarily difficult journey, especially recognition of and respect for their love for their child.

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Trisomy support groups online: Facebook groups: Trisomy 13; Trisomy 18: Faces of trisomy; trisomy 18 mommies; living with trisomy 13; Support Organization for Trisomy; Patau Syndrome NEWS; Online groups: support for families of babies with Trisomy 13 (Patau's syndrome); www.trisomy18support.org; www.noahsneverendingrainbow.org; www.benotafraid.net; www.trisomy.org; www.leona-ev.de (Europe); www.soft.org.uk; www.trisomyonline.org; www.softireland.com/; www.hopefortrisomy13and18.org; www.prenatalpartnersforlife.org; livingwithtrisomy13.org.

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