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Research Letter

Pregnancy and Birth History of Newborns With Trisomy 18 or 13: A Pilot Study

Deborah Bruns*

Department of Educational Psychology and Special Education, Southern Illinois University Carbondale, Carbondale, Illinois

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To the Editor:

The prognosis for infant survival in newborns diagnosed with the trisomy conditions Edward syndrome (trisomy 18) and Patau syndrome (trisomy 13) is poor [Goldstein and Nielsen, 1988; Root and Carey, 1994; Brewer et al., 2002]. Major contributing factors to early mortality include gestational age of less than 32 weeks, limited use of life support measures, apnea and other respiratory difficulties and presence of cardiac anomalies [Root and Carey, 1994; Rasmussen et al., 2003]. Other authors have [Van Dyke and Allen, 1989; Baty et al., 1994; Rasmussen et al., 2003; Carey, 2005] discussed the medical management and overall health needs of survivors. Newborns, surviving past 2 months of age, typically present ongoing medical needs in addition to day-to-day newborn caregiving requirements [Baty et al., 1994; Niedrist et al., 2006; Pont et al., 2006].

Studies published in the 1980s and early 1990s offer an examination of the strengths and needs of newborns with these trisomy syndromes [e.g., Goldstein and Nielsen, 1988; Van Dyke and Allen, 1989; Boss and Broers, 1992; Root and Carey, 1994; Nembhard et al., 2001]. Some recent researchers have reviewed birth registers from the 1980s to the mid 1990s and have found improved outcomes compared to the others [Canfield et al., 2001; Parker et al., 2003]. Further, cardiac surgery has assisted in extending the life expectancy for some infants with rare trisomy conditions [Graham et al., 2004]. Less invasive procedures and interventions are also providing avenues for improved daily management and long term care [Kosho et al., 2006; Lin et al., 2006; Niedrist et al., 2006].

It is imperative to examine the varied courses and outcomes for newborns with these conditions in more detail. Newborns with trisomy 18 (t18) or 13 face an uncertain course in the NICU and after discharge [Baty et al., 1994; Walters, 1998; Rasmussen et al., 2003; McIntosh et al., 2004; Tripp and McGregor, 2006].

In recent years, European investigators have collaborated with clinicians and cytogeneticists throughout the world in the development of international registries of individuals with rare chromosome syndrome [http://decipher.sanger.ac.uk/; www.e-caruca.net]. Additional collaborative endeavors are needed and could include parents and caregivers. The Tracking Rare Incidence Syndromes (TRIS) project can contribute to such efforts through collection of phenotypic data [Bruns, 2006].

The previously described issues highlight the need to investigate maternal pregnancy experiences, childbirth history and presenting needs. This information can also provide useful, parent-completed data for registries, open new avenues for parent-professional advocacy and collaboration, and demonstrate the utility of such databases. This pilot study sought to examine:

(1) What are mothers' pregnancy experiences and their newborns' birth history?
(2) What are the presenting physical characteristics and medical conditions of newborns with t18 or 13?
(3) What is the course of hospital treatment for newborns with t18 or 13?

In 2004, the Tracking Rare Incidence Syndromes (TRIS) project began [Bruns, 2006]. The guiding mission was and continues to be to increase awareness and knowledge for families and professionals

*Correspondence to: Deborah Bruns, Ph.D., Department of Educational Psychology and Special Education Southern Illinois University Carbondale, Mailcode 4618 Carbondale, IL 62901.
E-mail: dabruns@siu.edu
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touched by rare trisomy conditions and aims to facilitate improved decision making for optimal services and support for children and their families [http://web.coehs.siu.edu/grants/tris/]. The overall purpose of the TRIS pilot study was to demonstrate collaboration between support groups and to promote the usefulness of parent-completed survey data to address data registry needs for this clinical subpopulation. Feedback was also solicited on the instrument's content and presentation.

Experiences of parents of living as well as deceased children with rare trisomy conditions [Van Dyke and Allen, 1989; Baty et al., 1994; Root and Carey, 1994] guided the TRIS project and TRIS Survey. Specifically, posts from the tri-family and tri-med listservs (both moderated, members only listservs), and rare trisomy-related websites (e.g., livingwithtrisomy13.org), the existent literature and input from the TRIS Advisory Committee was used to develop the TRIS Survey. The TRIS Advisory Committee is comprised of parents of living or deceased children with rare trisomy syndromes, medical professionals and educational personnel with specialized knowledge of children with these trisomy conditions (e.g., pediatric cardiologist, pediatric neurologist, early interventionist), In addition, the author reviewed key texts in the area of survey development in order to prepare the format and procedures for completing the pilot version of the TRIS Survey [Sudman and Bradburn, 1982; Denzin, 1989; Czaja and Blair, 2004].

The pilot version of the TRIS Survey consisted of three parts: Part I includes 35 items on pregnancy and birth history and collection of demographic information. Part II utilizes 60 items to examine medical and health concerns including, as appropriate, medical procedures, surgery and medications. Part III consists of 46 items concerning available social supports and resource needs. This article describes pilot data from Part I.

Part I, the pregnancy and birth history-related items includes 23 items with multiple parts dependent on parent response to the initial question. The remaining five items comprised demographic information including marital status and highest level of education.

The author developed the pilot version of the TRIS Survey during summer and fall 2005 from the available literature and input from the TRIS Advisory Committee. Concurrently, an Interested Parents database was developed through electronic contacts with listserv members. Website owners, either parents or relatives of living or deceased children with trisomy 18 or 13, were approached to include a link to the TRIS webpage [http://web.coehs.siu.edu/grants/tris/] and the project's email address (tris@siu.edu) for recruitment purposes. In this way, agreements were secured with two website owners (http://livingwithtrisomy13.org/index.htm, www.noahsneverendingrainbow.org). In January 2006, the TRIS project received approval from the University's Human Subject Committee. Data was collected during spring 2006. A graduate assistant maintained contact with parents and sent requested materials. At approximately 2 and 3 weeks after sending materials, the graduate assistant also followed up by email and phone if completed TRIS Survey materials had not been received.

For returned surveys, each was systematically reviewed for completion of all items. All returned surveys were then prepared for entry into the survey database.

Due to the small sample size, only frequencies and percentages were analyzed for quantitative items. In addition, the majority of items in Part I only required a "yes/no" response or selection of one or more variables from a finite list. Means and standard deviations are available for items with continuous variables (e.g., gestational age, birth weight). Results were aggregately as well as examined separately by trisomy diagnosis (t18, 13) and subtype (full, mosaic, partial). Data were also reviewed for child status at the time of survey completion (living or deceased).

Qualitative items were entered into an Excel spreadsheet. The author and the graduate assistant completed multiple readings of the data and developed preliminary themes and categories [Lincoln and Guba, 1985; Huberman and Miles, 1994]. The preliminary themes and categories were then compared as a reliability check of the data. Disagreements were resolved through consensus. Responses to "Other (please specify)" items were compiled in a Word document and examined for patterns [Huberman and Miles, 1994].

Sixty-six parents expressed interest in participating in the pilot of the TRIS Survey. Forty-one percent (N = 27) returned the materials with 22 suitable for analysis. The remaining five surveys were missing large sections of data in one or more parts of the survey. At the time of survey completion, mothers were between 28 and 60 years of age (M = 41 years). Educational levels varied from high school completion (26.4%) to advanced degree (26.4%). All parents were married and 16 (84.2%) reported family income of $50,000 or more. Income data was missing for three parents. Seventeen parents identified themselves as Caucasian (89.5%), one as Hispanic and the remaining parent identified as biracial (see Table I).

Approximately half of the sample had a newborn with t18 (N = 13, 48%) (9 full, 2 mosaic, 2 partial). The remainder had newborns with t13 (N = 14, 52%; 11 full, including one child with a balanced translocation, 2 mosaic, 1 partial). Of these, the majority of children who had passed away were diagnosed with full t13 (N = 8). The oldest survived 53 days. Four of the five children with t18 (4 full, 1 partial) died before 6 months of age. The remaining child lived 21 years, 6 months. Mothers' age at time of conception was 22–43 years.

The sample included 13 males and 14 females. Table II summarizes the chromosome groupings.

TABLE I. Participant Demographics (N = 19)*

	N	%
Age at time of study		
25–29 years	2	10.5
30–34 years	5	26.4
35–39 years	4	21.0
40–44 years	2	10.5
45–49 years	5	26.4
Over 50 years	1	5.3
Ethnicity		
European-American	17	89.5
African-American	0	0.0
Hispanic-American	1	5.3
Native-American	0	0.0
Asian-American	0	0.0
Biracial	1	5.3
Marital status		
Single	0	0.0
Long term relationship	0	0.0
Married	19	100
Separated/divorced	0	0.0
Widowed	0	0.0
Educational level		
High school graduate/GED	5	26.4
Vocational education/trade school	2	10.5
Associate degree	3	15.8
Bachelor degree	4	21.0
Advanced degree	5	26.4
Annual income		
$25,000–$49,999	3	15.8
$50,000–$74,999	4	21.0
$75,000–$99,999	5	26.4
More than $100,000	7	36.8

Totals more or less than 100% due to rounding.
*Data missing for three participants.

The oldest living child was 19 years of age at the time of the pilot (partial t13q and partial t15). The remaining children were between 2 and 11 years of age. In this sample, children with trisomy 13 subtypes lived up to 6 years longer than children diagnosed with a t18 subtype. At the time of data collection, the two oldest children with t18 were a boy with t18 mosaic (7 years, 11 months old) and a boy with partial t18p (8 years, 8 months old).

Data from Part I of the pilot of the TRIS Survey is presented in three parts: (a) prenatal diagnosis and birth history, (b) presenting medical conditions, and (c) hospital treatment postpartum. A number of mothers in the sample did not complete all items in Part I. Therefore, totals are noted as appropriate.

Only two mothers (9%) indicated an awareness of rare trisomy syndromes prior to their child's diagnosis. Confirmation of their child's trisomy condition occurred between 19 and 36 weeks gestational age.

Two parents reported experiencing polyhydraminos during their pregnancy with newborns with full t18. Approximately one quarter of the respondents were diagnosed with preclempsia (N = 5; one full t18, two full t13, one t13 mosaic and one partial t13q).

Six mothers of newborns with full t18 (N = 21; 86%) and 10 mothers of newborns with full t13 (100%) received information about vaginal birth and induction. Approximately half of the mothers in each group were provided with the cesarean option. Based on these options, mothers of newborns with t13 chose vaginal birth (N = 6), induction (N = 2) and cesarean (N = 2). Mothers of newborns with t13 mosaic (N = 2) also chose induction. The one mother with a t18 mosaic newborn as well as the one mother with a partial t18 newborn chose vaginal birth. Sixty percent of the full t13 subgroup (N = 6) also developed birth plans with input from their obstetricians and/or family (N = 21). Four mothers (2 full t13, 2 full t18) looked to Internet sources for information (N = 20).

For newborns with full t13, half of the group (N = 5) had a spontaneous birth (50% planned). Available data for five of the six mothers of newborns with full t18 indicted a planned birth (83%). Of the 13 newborns with a form of t13, results indicate that the actual birth option used were nine vaginal births (8 full, 1 partial) with four requiring induction (all full) and two cesarean sections. For the two newborns with t13 mosaic, induction was selected as the actual birthing method. Time in labor for mothers of 20 newborns ranged between 0 and 25 hr (M = 8.5 hr). Zero hour labors were three emergency cesareans (two full t18. one full t13). The longest labor was reported by a mother of a newborn with full t13. All were live births. Four newborns died on their birth day (one full t18, three full t13). In

TABLE II. Newborn Birth Information (N = 20)

	Gestational age (weeks)			Weight (grams)			Length (inches)		
	Min.	Max.	Mean (SD)	Min.	Max.	Mean. (SD)	Min.	Max.	Mean (SD)
Trisomy 18									
Full (5)	29	41	37.2 (4.76)	1,503	2,155	1,928 (248.69)	16	18	17 (0.82)[a]
Mosaic (1)	41	41	41 (0)	2,722	2,722	2,722 (0)	20	20	20 (0)
Partial (1)	40	40	40 (0)	3,203	3,203	3,203 (0)	19.5	19.5	19.5 (0)
Trisomy 13									
Full (10)	29	40	37.5 (3.24)	1,021	3,629	2,714 (844.37)	14	20.5	18.1 (2.28)[b]
Mosaic (2)	36	40	38 (2.83)	3,105	3,202	3,154 (69.30)	19	20.5	19.8 (1.06)
Partial (1)	36	36	36 (0)	3,175	3,175	3,175 (0)	20	20	20 (0)

[a]N = 4.
[b]N = 8.

addition, one newborn with full t13 survived for 1, 2, and 3 days, respectively.

Gestational age (GA) and birth weight (BW) was provided for 20 newborns. Table II summarizes the growth data.

Results in the remaining sections are based on responses from 20 mothers.

Parents were asked about trisomy related physical characteristics noted at their child's birth. Eighty percent exhibited low set ears (N = 16) and 40% a small jaw. One newborn with full t18 and seven newborns with full or a variant type of t13 presented with extra or webbed fingers or toes. Four newborns with were full t18 and five with full t13 were diagnosed with microcephaly. Several newborns with a form of t13 also had a cleft lip and/or palate (N = 3).

The TRIS Survey also requested information about newborns' presenting medical conditions. Seventy-five percent experienced respiratory distress (N = 15; all newborns diagnosed with t18 and N = 10 (77%) with full or a variant of t13). Although representing a different composition of trisomy types, the same percentage were identified with feeding difficulties. Specifically, three newborns with full t18 (60%), 70% of newborns with full t13 (N = 7) and newborns with variants of both trisomy types. Kidney conditions were identified in two newborns with t18 (one full, one mosaic) and four newborns with full t13 (see Table III).

Heart defects commonly identified in newborns with rare trisomy conditions were also represented in this sample [Canfield et al., 2001; Brewer et al., 2002; Graham et al., 2004; Pont et al., 2006]. Specifically, 30% of the newborns were diagnosed with atrial septal defect (ASD), 25% with patent ductus arteriosis (PDA) and 40% with ventricular septal defect (VSD). Table IV provides detailed information regarding specific heart conditions.

For the 20 participants completing items related to their child's stay in the NICU, 12 were taken to the NICU within 24 hr of their birth (60%). The majority of newborns remained on the unit for less than 2 weeks. One newborn with t13 mosaic was on the unit for 5–8 weeks. Discharge weight ranged from 1,899 to 3,065 g.

The newborns in this sample required a variety of equipment during their NICU stay. Specifically, half of the sample (N = 10) needed an apnea monitor (three full t18, six full t13, one t13 mosaic), 25% a nasal canula for oxygen (two full t18, two full t13, one t13 mosaic) and 25% a ventilator or respirator (four full t13, one t13 mosaic). In addition, seven newborns (35%) required a heart monitor. This group encompassed two newborns with full t18, and five with full t13. Ten newborns, including three with full t18, six full t13 and one t13 mosaic also required specialized feeding interventions such as use of a Haberman® feeder or gavage feeding. Parents also noted that their newborns required a pulse oxymeter and/or bililights to treat jaundice.

A quarter of the newborns underwent intubation (four full t13, one t13 mosaic). Half of the sample had an ultrasound performed (90% representing newborns with full or mosaic t13). Forty-five percent (N = 9) had echocardiograms (three full t18, four full t13, one t13 mosaic, one partial t13q). It is important to note that newborns with t18 variants did not require these specialized procedures. Parents also listed several additional procedures not included on the survey item: auditory brainstem response audiometry, bronchoscopy or swallow study and blood tests.

Results from the TRIS Survey pilot study provide maternal and newborn experiences relating to prenatal diagnosis and birth history, presenting medical conditions and hospital treatment postpartum. Approximately one-third of mothers in this sample received a prenatal diagnosis of their newborn's trisomy condition. There was variation in pregnancy course and birth options. Most newborns were spontaneous, vaginal births. Birth weight ranged from 1,021 to 3,629 g. Newborns with mosaic and partial trisomy types were longer in length than infants with full trisomies (range 14–20.5 inches).

Newborns' presenting physical characteristics were the types described in the literature including cleft lip and/or palate and microcephaly [Baty et al., 1994; Lin et al., 2006; Pont et al., 2006]. Respiratory and feeding difficulties were also prominent.

The existent literature offers a bleak outlook for newborns with t18 or 13 [e.g., Boss and Broers, 1992; Brewer et al., 2002; Kosho et al., 2006; Pont et al., 2006]. The results from the pilot study of the TRIS

TABLE III. Presenting Medical Conditions (N = 20)

	Respiratory distress		Feeding difficulties		Kidney conditions	
	N	%	N	%	N	%
Trisomy 18						
Full (5)	5	100%	3	60%	1	20%
Mosaic (1)	1	100%	1	100%	1	100%
Partial (1)	1	100%	1	100%	0	0%
Trisomy 13						
Full (10)	8	80%	7	70%	4	40%
Mosaic (2)	2	100%	2	100%	0	0%
Partial (1)	0	0%	1	100%	0	0%

TABLE IV. Heart Conditions of Newborns With Trisomy 18 or 13 (N = 20)

	ASD		PDA		VSD	
	N	%	N	%	N	%
Trisomy 18						
Full (5)	3	60%	2	40%	4	80%
Mosaic (1)	0	0%	0	0%	0	0%
Partial (1)	0	0%	0	0%	1	100%
Trisomy 13						
Full (10)	2	20%	2	20%	2	20%
Mosaic (2)	1	50%	1	50%	1	50%
Partial (1)	0	0%	0	0%	0	0%

Survey pilot confirm some of these findings. A majority of the newborns in this sample expressed the typical trisomy characteristics and experienced the common trisomy medical conditions described by Baty et al. [1994] and other researchers [e.g., Rasmussen et al., 2003; Lin et al., 2006] such as cleft lip and/or palate and cardiac issues. Results also concurred with previous findings concerning the need for intubation and other types of respiratory assistance [see Canfield et al., 2001].

This sample included more than the customary 10% of survivors past 12 months of age. This aligns with a shift identified two decades ago by Young et al. [1986] in survival patterns for newborns with t18. Yet, the TRIS Survey collected data from mothers in support groups not a population study [Root and Carey, 1994; Rasmussen et al., 2003]. Importantly, the majority of previous studies did not specifically examine mothers' pregnancy and birth experiences and newborns' NICU course. In this respect, the TRIS pilot study offers new data to begin to increase the knowledge base about rare trisomy conditions.

The data presented here are the results of an initial, pilot study. As such, several limitations are inherent. The sample was small and self-selected from families participating on an online listserv or website. The data presented here is descriptive and generalizations cannot be drawn. Future efforts must encompass a variety of listservs, support organizations, etc. representing parents of living children as well as angels with rare trisomy conditions in their membership. An additional avenue to collect information is through online sources. Skinner and Schaffer [2006] and Zaidman-Zait and Jamieson [2007] describe the availability of support, resources and linkages to services available on the World Wide Web. Specifically, "...the Web has the potential to be a valuable means of enhancing parents' knowledge" [Zaidman-Zait and Jamieson, 2007, p. 21].

It is also critical to expand the reach of the project to a world-wide scale in order to collect data and examine it in light of the available literature [Brewer et al., 2002; Kosho et al., 2006; Lin et al., 2006]. In addition, a larger dataset can be compiled and analyzed to provide both confirming and contrary findings and disseminate them to interested audiences. Demographic data appears to indicate a skewed group in that all parents were married and a majority of their children can be considered long-term survivors.

Survey instruments are inherently prone to bias in their reliance on self-report. While the expectation is that participants answered items truthfully, responses are retrospective and may be affected by, for example, the child's present medical status, parents' interactions with medical professionals providing care to their child and/or available information for their child's daily and specialized care [Bailey and Powell, 2005].

The results presented here offer a number of implications. First, it is critical to continue to examine this population to both confirm previous findings and to add to the literature. Medical professionals, especially those involved in genetics, must have up-to-date information in order to assist parents with decision-making and interventions for their newborns and older children with rare trisomy conditions. Similar to earlier research in this area [Young et al., 1986; Root and Carey, 1994] and more recent efforts [Brewer et al., 2002; Rasmussen et al., 2003], a greater understanding of possible outcomes is necessary to prepare families and professionals to effectively address the varied needs of this population.

It is also vital that data continues to be collected for the purpose of disseminating results to a variety of interested audiences. It is important to highlight various aspects of day-to-day and specialized caregiving so that medical professionals, therapists, etc. are better equipped with knowledge of strengths and needs of newborns and older children with t18 or 13. There is a concomitant need for medical professionals to work closely with early intervention specialists to offer guidance and recommendations for services and supports for the affected child and family [cf. Dunst et al., 1988; Hodapp and Fiddler, 1999; Bailey and Powell, 2005].

The implications described here also point to the need for changes in political and social policy to reflect the value of these children and needs of their families. There must be increased funding appropriations to delve more deeply into the areas examined in this pilot study. The results provided here offer

more data on particular needs of infants compared to prior studies [Young et al., 1986; Brewer et al., 2002; Pont et al., 2006]. It is imperative to continue collecting data about this unique population to learn more about mothers' pregnancy experiences, childbirth history and newborns' presenting physical characteristics and medical needs.

Similar to Lin et al. [2006], collaboration must occur with support groups to collect data on this subgroup. Parent data has demonstrated the potential to provide useful phenotypic data [Baty et al., 1994] and should be further explored and utilized by medical professionals. The TRIS project also offers a model for chromosome condition registries. Specifically, TRIS offers projects such as DECIPHER and ECARUCA [http://decipher.sanger.ac.uk/; www.ecaruca.net;] a model of gathering data from parents to further increase knowledge of rare trisomies.

Efforts have been underway to broaden and expand the TRIS Survey to better encompass these and other areas central to the care and management of newborns with rare trisomy conditions. Beginning February 1, 2007, the revised TRIS Survey (Full and Modified versions) was made available for completion online (http://web.coehs.siu.edu/grants/tris/survey.html).

The data presented here are an initial step of the TRIS project to increase the knowledge base for families with newborns and older children with rare trisomy conditions. The TRIS project also plans to provide professionals and families with data and resources to facilitate decision-making on behalf of surviving children and young adults.

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