

Follow-up Survey of Parents of Children with Major Birth Defects in New York State

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BACKGROUND: One of the objectives of the New York State birth defect surveillance system, the New York State Congenital Malformations Registry (CMR), is to identify children in need of special programs and assist prevention/intervention programs and services with evaluation. In 1999, the CMR started a statewide mass-mailing program to inform parents of children with major birth defects about programs and support groups that might be helpful to them. A follow-up survey by mail was conducted in 2002 to evaluate the usefulness of the mailing program. **METHODS:** The survey was mailed between September and December of 2001 to parents of children born in 2001 and reported to the CMR with selected major birth defects. Non-respondents were followed-up by telephone. **RESULTS:** Of the 226 eligible families, 54% (122) responded to the survey. Approximately 66% of responding parents stated that the information provided by the mailing program was helpful, and 48% had contacted and/or used the programs and services listed in the information sheets. Parents with children younger than 6 months of age at the time of mailing were more likely to find the information helpful, compared to parents with children 6 months of age or older. The majority of responding parents had contacted and/or used the Early Intervention Program. **CONCLUSION:** The findings from the current survey show that the statewide information-mailing program administered by CMR is helpful and useful to the families of children with major birth defects in New York State. *Birth Defects Research (Part A) 70:597–602, 2004.* © 2004 Wiley-Liss, Inc.

Key words: parent survey, birth defects, congenital malformations registry, information sheet, mailing program, children with special needs

INTRODUCTION

Major birth defects are diagnosed in 3 to 4 percent of infants in their first year of life; about 120,000 U.S. babies are born each year with birth defects. The birth of a child with a major birth defect is an unexpected event for most parents and often creates considerable stress within many families. Studies have shown that parents of children with chronic illness or disability may experience greater stress and psychological distress than parents of healthy children (Ayyangar, 2002; Hedov et al., 2002; Lawoko and Soares, 2002; Hauser-Cram et al., 2001; Pelchat et al., 1999; Silver et al., 1998; Black, 1981). Many intervention programs and services have been developed to help children with disabilities receive high quality care. While studies of program effectiveness have been performed, most have focused on relatively small populations (Jeffery and Boorman, 2001; Pelchat et al., 1999; Young et al., 1994; Baker, 1984) and help families receive financial aid and other support services (Bulat and Kantoch, 2003; Starke and Moller 2002; Buran et al., 2002; Young et al., 2001; Pelchat et al., 1998; Hoing and Winger, 1997; Baxter et al.,

1995; Pelchat, 1994). One statewide population-based study by Montgomery et al. examined the Colorado birth defects monitoring program which connects families with services for children with special needs (Montgomery and Miller, 2001). The study surveyed 237 families by telephone to determine whether the families found the contact by a local agency to be helpful. About 50% of parents responded to the survey and 65% indicated that they had learned about services, resources or developmental screening they weren't aware of previously.

One of the objectives of the New York State birth defect surveillance system, the New York State Congenital Malformations Registry (CMR), is to identify children in need of special programs and work with state maternal and

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child health (MCH) programs to assist in evaluating programs and services (Sekhobo and Druschel, 2001). While a strict confidentiality law in New York State prevents the CMR from releasing names to service programs, the CMR has collaborated with MCH to develop ways to inform families about programs and available services. In August of 1999, the CMR started a statewide mass-mailing program to inform parents of children with major birth defects about services and support groups that might be helpful to them. From August of 1999 to August of 2003, about 7,000 information letters have been sent to parents of children who were reported to the CMR as diagnosed with selected major birth defects.

To evaluate the usefulness of the mailing program the CMR initiated a follow-up survey in June of 2002. The objectives of this survey were to determine if parents a) learned about services or resources that they weren't aware of before, b) contacted and/or used services or resources, and c) used and/or needed services that were not listed on the information sheets.

METHODS

Survey Population

The survey population consisted of all families who were sent program mailings between September and December of 2001 and whose children were a) born in 2001, b) alive and not in foster care, and c) reported to the CMR with selected major birth defects. The major birth defects were selected based on the list used by the Colorado birth defects monitoring program (Montgomery and Miller, 2001) with minor modifications. The list excludes conditions which are likely to be less severe, such as certain musculoskeletal conditions, so that families whose children did not need services would not be alarmed by receiving the information package.

Survey Questionnaire

The survey questionnaire (available on request) asked parents 1) if they learned anything new about services and resources from the information sheets, and 2) if they contacted, used and/or found useful any of the programs and services listed. Those who did not use any of the information provided were asked to state their reasons. Parents were also asked if they used services that were not on the information sheets and if there are programs or services that they think should be added. In addition, space was provided in the questionnaire for concerns and comments from the parents. To keep results confidential, a unique case number was assigned to each questionnaire for the purposes of mail tracking and data analysis.

Survey Data Collection

The survey packages sent to the families in the summer of 2002 included a cover letter explaining the survey, the previously mailed information sheets, the survey questionnaire, and a postage-paid return envelope. Two subsequent follow-up mailings were sent to parents who did not respond within three to four weeks of each mailing. If survey questionnaires were returned as not deliverable due to address changes (moved), their new addresses were searched using post office listings, New York State Department of Motor Vehicle files, and Internet search engines.

Questionnaires were resent when new addresses were located.

Parents who did not respond to any of the three mailings were contacted and interviewed by phone to complete the questionnaire. Since the CMR does not have parents' telephone numbers in the database, extensive searches for phone numbers were conducted using the Internet and telephone directories.

Data Analysis

Summary statistics, simple and stratified, were generated using the SAS software package (SAS Institute Inc., Cary, NC). Standard error of estimate (SE) was calculated using 90% confidence level. The SE is the standard deviation of the sampling distribution of a statistic. It reflects how much a sampling fluctuation a statistic will show.

RESULTS

Using the survey selection criteria, 286 families residing in New York State were identified from the CMR's database. A total of 122 (43%) completed the survey questionnaire. Sixteen families (6%) refused to participate and 140 (49%) neither completed the questionnaire nor refused. Out of these 140 families, 52 had moved and could not be contacted either by mail or by phone. The remaining 88 families had valid mailing addresses but did not respond and could not be reached by phone. For the "Other" 8 families (3%), 3 did not complete the survey due to a language barrier and 5 reported that their children did not have the defects.

Response Rate

To calculate response rates only, those known to have received the mailing were included. Those who could not be reached by phone or mail (52) or in the "Other" category (8) were excluded. Thus, the survey response rate is $54 \pm 5\%$ (122 out of 226). Due to relatively low response rate, stratified analyses on selected demographic factors were performed to compare the respondents and non-respondents. The results are summarized in Table 1. The response rates were not significantly different with regard to residential location and child's age at the time of the mailing of the information package. Mothers who were 25 years and older (at the time of birth) had a significantly higher response rate than those who were younger than 25 years and those who did not report their ages. Parents who reported their child's race as white were more likely to respond compared to the parents of children of other races. The distribution by geographic area of residence, mother's age at birth and child's race was significantly different between respondents and non-respondents. The response rate was the highest for the parents who had children with Down syndrome. However, due to small numbers in each category, the effect of specific birth defect on the parents' response rate is difficult to interpret.

Helpfulness of the Mailing Information

Table 2 presents the responses to the first question in the survey questionnaire, "Did the information sheets help you to learn anything new about services, resources or screening?" About 66% of the respondents answered "Yes", i.e., found the information helpful. A higher percentage of respondents who lived in Upstate New York answered "Yes"

Table 1
Comparison of the Respondents and Non-Respondents (N = 226)

Characteristics	Total	Respondents	Non-respondents	Response rate
		N (% ± SE ^a)	N ^b (% ± SE ^a)	% ± SE ^a
Current Residence				
Upstate New York	145	83 (68 ± 7)	62 (60 ± 8)	57 ± 7
New York City	81	39 (32 ± 7)	42 (40 ± 8)	48 ± 9
Child's age at mailing				
0-5 months	20	12 (10 ± 4)	8 (8 ± 4)	60 ± 18
6-9 months	167	87 (71 ± 7)	80 (77 ± 7)	52 ± 6
10-12 months	39	23 (19 ± 6)	16 (15 ± 6)	59 ± 13
Mother's age at birth				
14-24 years	54	20 (16 ± 6)	34 (33 ± 8)	37 ± 11
25-44 years	148	94 (77 ± 6)	54 (52 ± 8)	64 ± 7
Unknown	24	8 (7 ± 4)	16 (15 ± 6)	33 ± 16
Child's race				
White	159	97 (80 ± 6)	62 (60 ± 8)	61 ± 6
Black	43	16 (13 ± 5)	27 (26 ± 7)	37 ± 12
Other race	24	9 (7 ± 4)	15 (14 ± 6)	38 ± 16
Birth defects (ICD code)				
Down syndrome (758.0)	9	8 (7 ± 4)	1 (1 ± 2)	89 ± 17
Oral cleft (749)	36	15 (12 ± 5)	21 (20 ± 6)	42 ± 14
Atresia/stenosis of intestine (751.1, 751.2)	17	11 (9 ± 4)	6 (6 ± 4)	65 ± 19
Atresia/stenosis of pulmonary valve (746.0)	13	7 (6 ± 3)	6 (6 ± 4)	54 ± 23
Deformities of feet or hip (754.3, 754.5)	50	31 (25 ± 7)	19 (18 ± 6)	62 ± 11
Coarctation of aorta (747.1)	10	7 (6 ± 3)	3 (3 ± 3)	70 ± 24
Tetralogy of Fallot (745.2)	9	4 (3 ± 3)	5 (5 ± 3)	44 ± 27
Transposition of great vessels (745.1)	9	6 (5 ± 3)	3 (3 ± 3)	67 ± 26
Other malformations	73	33 (27 ± 7)	40 (38 ± 8)	45 ± 10
Total	226	122	104	54 ± 5

^aStandard error, calculated using 90% confidence level.

^bParents who refused (16) and those who had valid address but could not be reached by phone (88).

compared to those who lived in New York City. More than 80% of parents with children younger than 6 months at the time of mailing found the information helpful, while only about 64% of parents with older children (6-12 months) found the information helpful (Table 2). However, because

of small numbers in each cell, these differences are not statistically significant.

Table 2
Responses to the Survey Question: "Did the Information Sheets Help You to Learn Anything New About Services, Resources or Screening?" By Some Demographic Variables (N = 122)

Characteristics	Yes	No/Not sure	Total
	N (% ± SE ^a)	N	N
TOTAL	80 (66 ± 7)	42	122
Current residence			
Upstate New York	58 (70 ± 8)	25	83
New York City	22 (56 ± 13)	17	39
Child's age at mailing			
0-6 months	10 (83 ± 18)	2	12
6-9 months	57 (66 ± 8)	30	87
9-12 months	13 (57 ± 17)	10	23
Mother's age at child's birth			
14-24 years	11 (55 ± 18)	9	20
25-44 years	65 (69 ± 8)	29	94
Unknown	4 (50 ± 29)	4	8
Child's race			
White	64 (66 ± 8)	33	97
Other race	16 (64 ± 16)	9	25

^aStandard error, calculated using 90% confidence interval.

Utilization of the Services and Programs Provided on the Information Sheets

About 48% of respondents (58 out of 122) answered "Yes" to the question "Did you call and/or use any of the services listed on the information sheet?" The 64 respondents who did not call or use the services were then asked to indicate why. About 60% indicated that they had no concern or had no need for the services, and 40% indicated other reasons or refused to answer this question.

The 58 parents who called and/or used the services were further asked "Did you find them helpful?" The responses are summarized in Table 3. A majority of the parents had contacted and/or used the Early Intervention program (69%). Other programs with a high percentage of parents contacting and/or using their services were Child Health Plus (33%) and Internet sources (33%). All parents who had contacted the Physically Handicapped Children's Program, Self Help/Mutual Support Directory and NYS Genetic Services Program, found them helpful. For the other services listed in the information sheets, 60-80% of the respondents who had contacted them found them helpful.

Utilization of the Services and Programs that were not Provided on the Information Sheets

Only 17 respondents answered "Yes" to the question, "Did you use any other services that were not on the information sheet?" As presented in Table 4, when the 17 respondents

Table 3
Responses of Those Who Called/Used the Services (N = 58) to the Survey Question: "If Called/Used Any of the Services Listed on the Information Sheets, Did You Find Them Helpful?"

	Called/used	Find helpful
	N ^b (% ^c ± SE ^d)	N (% ^e ± SE ^d)
Services on the information sheet ^a		
a. Physically Handicapped Children's Program at 800-522-5006	7 (12 ± 7)	7 (100 ± 0)
b. Child Health Plus at 800-698-4543	19 (33 ± 10)	12 (63 ± 18)
c. Early Intervention Program at 800-522-5006	40 (69 ± 10)	30 (75 ± 11)
d. Self Help/Mutual Support Directory at 800-522-5006	5 (9 ± 6)	5 (100 ± 0)
e. NYS Genetics Services Program at 518-474-4148	7 (12 ± 7)	7 (100 ± 0)
f. March of Dimes at 888-663-4637	11 (19 ± 8)	8 (73 ± 22)
g. Parent to Parent of NYS at 800-651-1151	10 (17 ± 8)	6 (60 ± 26)
h. Family Support Services at your local Develop Disabilities Office	9 (16 ± 8)	7 (78 ± 23)
i. Internet Sources	19 (33 ± 10)	15 (79 ± 15)

^aBrief description of the programs listed in this column:

- Provides health insurance/services to ensure access to quality health care for chronically ill and disabled children
- Provides health insurance covering a wide range of benefits for pregnant women and children up to 19 years
- Provides services such as screening and evaluation, family training, counseling, and parent support groups
- Offers people with special needs the opportunity to meet and share their experiences, knowledge, and strengths
- Administers 23 genetics units that provide genetic disease services statewide
- Dedicated to improving the health of babies by preventing birth defects and infant mortality
- Connects and supports families of individuals with special needs in New York State
- Offers a range of services to assist individuals with developmental disabilities and their families.

^bThe numbers are not exclusive since some respondents called/used more than one service.

^cThe percentages were calculated using the total respondents who answered Yes (N = 58) as denominator.

^dStandard error, calculated using 90% confidence level.

^eThe percentages were calculated using the number of respondents who called/used the service in each category as denominator.

were asked to specify these services, about 41% had used services from local doctors, specialists and hospital social workers; 41% had used other programs for children with special needs such as Women, Infants and Children and Care at Home; and 18% had used Internet sources such as medical college websites.

The respondents were further asked if there are services or programs they needed that were not listed on the information sheets. Twenty-one respondents listed additional programs and services. As can be seen in Table 4, the listed programs and services were grouped into two categories: information on specific malformations, doctors and in-house care (48%) and special programs/services and resources (52%).

Parents' Comments

About 40% of respondents (49) provided comments. These were grouped into positive comments (39%), helpful

suggestions (27%) and others (35%). Parents who gave positive comments on the program mostly praised the mailing program and thanked the CMR staff for the useful information provided in the mailing package. The helpful suggestions made by parents included sending the information earlier, providing the information sheets in Spanish, providing more specific information, and making the program/service information sheets available at locations such as doctor's offices.

DISCUSSION

Results from the current survey show that parents who lived in New York City found the information sheets less useful when compared to those who lived in Upstate New York. New York City might provide better access to tertiary care centers and specialists who provide more information to families. Service programs may be more inte-

Table 4
Responses to the Survey Question on Services/Programs That Were Needed But Not Listed on the Information Sheet and Parents' Comments About the Programs

	N	% ± SE ^a
Used other services that were not on the information sheet (N = 17)		
Local doctors/specialists/hospitals (social workers)	7	41 ± 20
Programs for children with special needs (Women, Infant & Children, Care at Home, etc)	7	41 ± 20
Internet sources (medical colleges' websites, clubs, etc)	3	18 ± 15
Total	17	100 ± 0
Used services/programs that you need but not listed on the information sheet (N = 21)		
Specific information (on malformations, doctors, in-house care, etc)	10	48 ± 18
Programs on services and resources	11	52 ± 18
Total	21	100 ± 0

^aStandard error, calculated using 90% confidence level.

grated into the medical system in New York City. It is understandable that parents would be less likely to perceive the information they received as helpful or needed if the child's anomaly was reversed or resolved, if parents were well informed by their physicians or hospital social workers, or if families were already receiving services during the first few months after birth. These factors might be the explanation for the finding that fewer parents whose children were older (aged 6 months or older) at the time the information was received found the information sheets helpful compared to parents with younger children.

The program that the majority of parents had contacted and/or used was the Early Intervention Program. This is a federally funded program that provides many different types of early intervention services to infants and toddlers with disabilities and their families (NYSDOH, 2000). This program is designed to enhance the development of children up to 3 years old so they reach their fullest potential. Early intervention programs have been shown to have a beneficial impact on families of children with major birth defects, especially in the first few months of the child's life (Pelchat et al., 1999; Pelchat, 1994). Previous matching of the CMR database with that of the Early Intervention Program indicated that a considerable number of children reported to the CMR were not receiving needed services (unpublished data, 1995). A re-match of CMR children with the database of the Early Intervention Program is planned to determine if there has been an improvement in the number of children enrolled in the program.

About 33% of responding parents reported that they had used Internet sources provided on the information sheets. The Internet has become a significant resource in the medical field to improve health and health care delivery (Ikemba et al., 2002; Mandl et al., 2000). A recent national survey on Internet use for health care showed that about 40% of respondents with Internet access reported using the Internet to look for advice or information about health or health care in 2001 (Baker et al., 2003). The CMR is continually reviewing and adding useful Internet websites to the information list for the parents of children with special needs.

We learned that parents prefer to receive the information letters earlier, when their children are younger than 6 months of age. Changes have been made to send packages before children reach 6 months of age. In addition, responding to parents' comments and suggestions, additional information and Internet sites specific to various birth defects and social and financial support are being reviewed for possible inclusion in the mailing package.

The study found that about 54% of surveyed parents whose children had major birth defects responded to the follow-up survey, which is consistent with that reported from the Colorado survey (50%) (Montgomery and Miller, 2001). About a 60-70% response rate had been reported previously on parent survey studies using mailed questionnaires (Jeffery and Boorman, 2001; Cetta et al., 1993). The somewhat low response rate in the current survey is mainly due to a considerable number of eligible parents who did not respond to the survey and could not be reached by telephone calls (49%). The CMR does not currently collect telephone numbers. Extensive use of Internet and statewide telephone directories in searching for telephone numbers of non-responding families failed to obtain phone numbers for a majority of these families. Failure in

follow-up might be caused by having non-listed telephone numbers, increased use of cell phones, frequent moves, or by parents discarding mail packages. Follow-up telephone interviews with the parents who did not respond to the mailed questionnaires revealed that some parents were overwhelmed by the care of their child and were too stressed to respond. Since the distributions by geographic area of residence, mother's age and child's race were significantly different for the respondents compared to that for the non-respondents (Table 1), the findings from the current survey might not be representative of all parents of children with major birth defects in New York State. Future studies are needed to target the non-response group to ensure that children with special needs receive high quality care.

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