



# Increasing prevalence of gastroschisis in Utah

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## Abstract

**Background:** Recent studies provide conflicting information about gastroschisis prevalence trends. The authors proposed that prevalence of gastroschisis in live births has increased in Utah and that characteristics of these infants would provide clinically useful information about treatment and outcomes.

**Methods:** Primary Children's Medical Center (PCMC) is the sole pediatric surgical referral hospital for Utah. The authors used both pediatric surgical and neonatal databases to identify gastroschisis cases at PCMC from 1971 through 2002. Only infants whose mothers had a primary residence in Utah were included. Individual charts were reviewed for infant characteristics for cases from 1998 through 2002. Utah Vital Statistics Reports were used to determine live birth rates and general infant and maternal characteristics.

**Results:** Gastroschisis prevalence increased from 0.36 to 3.92 cases per 10,000 live births over 31 years ( $P < .001$ ). Young maternal age, primigravida status, and tobacco use were associated risk factors. Using the time required to achieve full enteric feedings at targeted volume and caloric density as a measurement of outcome, we found no association between delivery mode or surgical closure type (primary or secondary) and time to full feedings. Higher birth weight was associated with decreased time to full feedings ( $P = .03$ ).

**Conclusions:** Gastroschisis prevalence has increased 10-fold over the past 3 decades in Utah.

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Malformations of the abdominal wall are among the more common structural developmental defects in the infant and include omphalocele and gastroschisis [1]. Gastroschisis is an abdominal wall defect that occurs at the junction of the umbilicus and the skin and is generally located to the right of

the umbilicus. Although the embryological pathogenesis of gastroschisis is not well understood, it is thought to arise from a weakness or vascular disruption at the site of the rudimentary fetal right umbilical vein. Epidemiologic and clinical studies have identified only young maternal age (<20 years) and primigravida status as factors associated with increased prevalence of gastroschisis [2].

The prevalence of gastroschisis was reported as 1 in 10,000 live births in 1997 [3]. Several reports have described an increase in the birth prevalence of gastroschisis over the past 2 to 3 decades, but retrospective studies have produced conflicting evidence. One study on abdominal

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defects in northern England over the 11-year period from 1986 through 1996 found an increase in prevalence from 1.48 in 1986 to 5.29 per 10,000 births in 1996 [1]. Similar studies from eastern Ireland, Hawaii, and Japan also demonstrated statistically significant increasing prevalence as well [4-6]. A recent study analyzed data from 19 registries and 3073 cases of gastroschisis in the International Clearing House for Birth Defects Monitoring Systems for the period of 1974 through 1988 and found an increased prevalence of gastroschisis in nearly half of the registries studied [7]. However, a study looking at gastroschisis prevalence in Denmark over a similar period (1970-1989) demonstrated no significant change in prevalence with an average point prevalence of 1.3 per 10,000 live and stillbirths [8].

Within the surgical and neonatal groups at PCMC and the University of Utah, Salt Lake City, it was felt that the gastroschisis prevalence in Utah had increased significantly over the past 2 decades, but no formal assessment of this population had been made. This paper examines gastroschisis prevalence among live birth infants in Utah for the 31-year period from 1971 to 2002. We hypothesized that the prevalence of gastroschisis has increased significantly in Utah during this study period. We also characterized selected clinical characteristics for the population born within the most recent 5-year period from 1998 through 2002.

## 1. Methods

Primary Children's Medical Center is the sole pediatric surgical referral hospital for the state of Utah. It also serves as the pediatric referral hospital for much of Wyoming, Idaho, Montana, Nevada, and some of Arizona and Colorado. All infants treated in the PCMC Neonatal Intensive Care Unit (NICU) are transported from outside hospitals, as there are no obstetrics facilities on site. It was decided that studying gastroschisis in Utah infants by reviewing cases at PCMC would provide an accurate estimate of prevalence, because essentially, all live born Utah infants would have undergone repair at PCMC. Potential index cases were identified using 1 of 2 independent databases—the Neonatal Teamup database and/or the PCMC Pediatric Surgical database.

The Neonatal Teamup database includes all infants seen and treated in the PCMC NICU from 1988 through the present. All cases of gastroschisis are admitted to the NICU for surgical evaluation and are logged into this system. Teamup was used to identify all cases of gastroschisis in the NICU from January 1, 1988, through December 31, 2002. The PCMC Pediatric Surgical database is a compilation of all pediatric surgical patients seen at PCMC from 1971 through the present. We also used this data base as a cross reference for the Teamup database for the period from January 1, 1988, through December 31, 2002. Potential cases from either database were confirmed by individual retrospective chart review. All out-of-state transports were

excluded to assess only infants with gastroschisis born in the state of Utah. The PCMC Pediatric Surgical database in addition served as the sole reference for index cases and maternal address for the period from 1971 through 1987. We obtained institutional review board approval for use of these databases and for individual patient chart review.

We reviewed the individual charts of index cases for the 5-year period from January 1, 1998, through December 31, 2002, for more detailed characteristics of maternal history, birth history, and clinical course. We selected this group of infants because they were felt to provide a reasonable approximation of the gastroschisis population for Utah and medical records were readily available for this period. Specifically, sex, race, maternal age, and gravida/para status, maternal tobacco use, prenatal diagnosis, mode of delivery, birth order, Apgar scores, birth weight, gestational age, size for gestational age, presence of meconium staining, fetal abnormalities, method of surgical closure (primary or secondary), need for bowel resection, time of first feedings and time to full feedings, development of cholestasis, length of stay, and survival were all reviewed. Time to full feedings was defined as the point at which an infant was on oral feedings at adequate volume and calories to sustain growth without total parenteral nutrition. Cholestasis was established by discharge code and the administration of pharmaceutical treatment. Results for the gastroschisis cohort were compared with all newborn infant data from the Utah Vital Statistics Report on births and deaths for 2000 [9].

Using the total number of Utah residents born with gastroschisis as the numerator and the number of live births in Utah as the denominator, we calculated the prevalence of gastroschisis in Utah for each of the years from 1971 through 2002. Live births for the past 30 years were obtained from the Utah Vital Statistics Report for 2001. Systat 10, a statistical software package (Systat Software Inc [SSI], Richmond, CA), was used for all statistical analysis of the data. Trends were analyzed using linear regression and correlation coefficients. Dichotomous variables were analyzed by  $\chi^2$  test and continuous variables were tested with Student's *t* test and Pearson correlation.

## 2. Results

### 2.1. Prevalence (1971-2002)

A total of 240 cases of gastroschisis were identified in this study. Over the 32-year period from 1971 through 2002, the prevalence of gastroschisis in live born infants in Utah significantly increased (Fig. 1) from 0.36 cases to 3.92 cases per 10,000, representing more than a 10-fold increase (Pearson coefficient  $r = 0.76$ ,  $P < .001$ ). Because the cases collected from 1988 through 2002 were confirmed using 2 database sources and individual charts, additional statistical analysis (linear regression) was run on this portion of the data. During this 15-year period, there was also a significant

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