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Decreasing trend of gastroschisis prevalence in the United States from 2014 through 2022: Is attributed to declining birth rates in young, high-risk gravidae

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ABSTRACT

Objectives: To investigate the prevalence trend of gastroschisis in the United States between 2014 and 2022. *Methods*: A cross-sectional retrospective analysis of the Centers for the United States live births between 2014 and 2022. Pregnancies and neonatal singleton live births with documented isolated gastroschisis were included. Neonates with other major congenital anomalies and known chromosomal abnormalities were excluded. Prevalence per 10,000 live births along with 95 % confidence intervals was estimated.

Results: Among 32,088,301 singleton live births, 6804 cases of isolated gastroschisis were identified (Point prevalence: 2 in 10,000 live births). A significant decline in gastroschisis prevalence was observed, decreasing from 2.86 per 10,000 live births in 2014–1.55 per 10,000 live births in 2022 (P < 0.001). The risk of gastroschisis was significantly higher in teen and nulliparous gravidae, with prepregnancy tobacco use, and among socially vulnerable populations (underweight, < 12th-grade education, Medicaid, non-Hispanic Indigenous Americans). The drop in gastroschisis births from 2014 to 2022, compared to non-gastroschisis births, is more significant in maternal age < 20 years, nulliparous, BMI < 18.5, and in smokers prior to pregnancy than in the overall population (P = 0.02, 0.0008, <0.0001, <0.0001, and 0.01 respectively). All of the associated maternal factors had a significant decline in prevalence (P < 0.001), which may influence the decreasing trend of gastroschisis. There was no perceived considerable impact of the COVID-19 pandemic on gastroschisis trends.

Conclusions: The study highlights a notable decline in gastroschisis prevalence mostly attributable to a declining birth rate in the highest at-risk strata, suggesting recent increases in birth rates among these at-risk gravidae may reverse the trend of declining gastroschisis disease prevalence. These findings support the need for ongoing further research to understand effective means of sustaining this decreasing trend.

Introduction

During the 1990s and 2000s, there was a notable rise in the point prevalence of gastroschisis, which underscored the urgent need for research into the underlying causes, and effective treatment and management strategies [1–5]. Specifically, based on Centers for Disease Control and Prevention (CDC) data, from 1995 to 2012, the prevalence of gastroschisis increased based on, rising from 3.6 to 4.9 per 10,000 live births, with similar trends observed in data from 15 states showing an increase from 2.32 to 4.42 per 10,000 live births during 1995–2005 [6,

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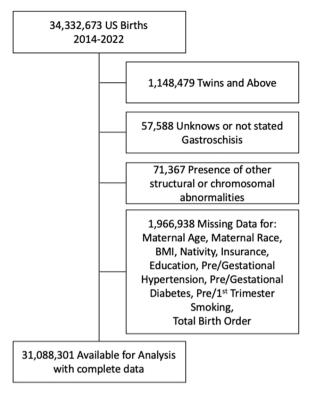


Fig. 1. Population flowchart.

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The association between young maternal age (e.g., under age 20, or "teen pregnancy") and gastroschisis, first described in 1977 [8], has been consistently documented [9–12]. Epidemiologic patterns suggest that health care disparities disproportionately experienced by teen gravidae (incomplete education, Medicaid insurance, poor nutrition, underweight BMI, tobacco and alcohol exposures, and race) may play a role [13,14]. Additionally, several studies suggested an association between genitourinary infections and fetal gastroschisis [11,13,15,16].

Historically, identifying potential risk factors for gastroschisis has been a critical public health objective to devise effective prevention strategies and mitigate its prevalence and health impacts [3]. Moreover, understanding the factors contributing to change in its prevalence is crucial and could establish a predictive framework for managing this condition proactively. Our study aimed to identify factors associated with recently declining trends in the prevalence of gastroschisis using CDC data, to identify attributable factors and persistently at-risk populations with the overarching goal of recalibrating the algorithm of risk factors influencing prevalence patterns, understanding gastroschisis etiology, and informing prevention strategies.

Methods and materials

Study design

We performed a cross-sectional population study utilizing the natality dataset of the National Center for Health Statistics and Centers for Disease Control and Prevention (CDC) from January 1st, 2014 to December 31st, 2022. The natality dataset includes paternal, maternal, prenatal, labor, and obstetric characteristics. The information is derived from the national birth registry and uses the US Standard Certificate for live birth. This certificate is completed for every newborn upon delivery. This study was considered exempt from oversight by the IRB given that data were de-identified.

Table 1Prevalence (95 % Confidence Interval) of Gastroschisis per 10,000 by Selected Demographics for 2014–2022.

Total	N / Total N 6804 / 31,088,301	Prevalence (95 % Confidence
	31,066,301	Interval) 2.19 (2.14–2.24)
Race/Ethnicity	3835 / 16,214,961	2.37 (2.29–2.44)
Non-Hispanic White	631 / 4374,234	1.44 (1.33–1.56)
Non-Hispanic Black	144 / 239,124	6.02 (5.08–7.09)
Non-Hispanic AIAN Non-Hispanic Asian	122 / 1928,315 12 / 71,934	0.63 (0.53–0.76)
Non-Hispanic NHOPI	243 / 677,614	1.67 (0.86–2.92) 3.59 (3.15–4.07)
Non-Hispanic more than one race	1790 / 7466,387	2.40 (2.29–2.51)
Hispanic	27 / 115,732	2.33 (1.54–3.39)
Origin unknown or not stated		
Maternal Nativity	6120 / 24,175,402	2.53 (2.47–2.60)
Born in U.S. (50 US States)	684 / 6912,899	0.99 (0.92–1.07)
Born outside U.S.	1256 / 1569 050	9.64 (9.10, 0.12)
Maternal Age 15-< 20	1356 / 1568,959	8.64 (8.19–9.12)
20-< 25	2870 / 6141,227 1647 / 8957,063	4.67 (4.50–4.85) 1.84 (1.75–1.93)
25-<30	667 / 8917,239	0.75 (0.69–0.81)
30-< 35	233 / 4499,441	0.52 (0.45–0.59)
35-< 40	31 / 1004,372	0.31 (0.21–0.44)
40 +		
BMI Category	419 / 998,253	4.20 (3.81–4.62)
Underweight (BMI < 18.5)	3812 / 13,144,532	2.90 (2.81–2.99)
Normal weight (BMI 18.5-<25)	1724 / 8265,407	2.09 (1.99–2.19)
Overweight (BMI 25-<30)	600 / 4702,518	1.28 (1.18–1.38)
Obese Class I (BMI 30-<35)	163 / 2321,044	0.70 (0.60–0.82)
Obese Class II (BMI 35-<40) Obese Class III (BMI >=40)	86 / 1656,547	0.52 (0.42–0.64)
Pre-Pregnancy Hypertension, Yes	75 / 650,868	1.15 (0.91–1.44)
No	6729 / 30,437,433	2.21 (2.16–2.26)
Pre-Pregnancy Diabetes, Yes	38 / 293,268	1.30 (0.92–1.78)
No	6766 / 30,795,033	2.20 (2.15-2.25)
Pre-Pregnancy Cigarette Smoking,	1346 / 2543,971	5.29 (5.01-5.58)
Yes	5458 / 28,544,330	1.91 (1.86–1.96)
No		
1 st Trimester Cigarette Smoking, Yes	1025 / 1913,963	5.36 (5.03–5.69)
No Imagenesia	5779 / 29,174,963	1.98 (1.93–2.03) 3
Insurance Medicaid	4275 / 13,119,764 2103 / 15,504,333	.26 (3.16–3.36)
Private Insurance	167 / 1291,770	1.36 (1.30–1.42)
Self-Pay	259 / 1172,434	1.29 (1.10–1.50)
Other		2.21 (1.95–2.50)
Education	1311 / 3972,751	3.30 (3.12–3.48)
< High School Grad	5493 / 27,115,550	2.03 (1.97-2.08)
>=High School Grad		
Interpregnancy Interval for	758 / 3392,656	2.23 (2.08–2.40)
multiparous	385 / 2535,302	1.52 (1.37–1.68)
< 18 m	1606 / 11,837,817	1.36 (1.29–1.42)
18 – $23\mathrm{m}$ $24\mathrm{m}+$	775 / 3396,690	2.28 (2.12–2.45)
Unknown		
Parity	3280 / 9925,836	3.30 (3.19-3.42)
Nulliparous	3524 / 21,162,465	1.67 (1.61–1.72)
Multiparous		
Season of Conception	2027 / 8345,177	2.43 (2.32–2.54)
Spring	1699 / 7641,814	2.22 (2.12–2.33)
Summer	1402 / 7319,781	1.91 (1.82–2.02)
Fall Winter	1675 / 7770,437	2.16 (2.05–2.26)
Winter Year of Birth	975 / 3403,656	2.86 (2.69–3.05)
2014	955 / 3539,061	2.70 (2.53–2.88)
2015	823 / 3601,595	2.29 (2.13–2.45)
2016	818 / 3538,414	2.31 (2.16–2.48)
2017	715 / 3490,951	2.05 (1.90-2.20)
2018	738 / 3443,495	2.14 (1.99–2.30)
2019	647 / 3335,518	1.94 (1.79–2.10)
2020	612 / 3372,689	1.81 (1.67-1.96)
2021 2022	521 / 3362,922	1.55 (1.42–1.69)

AIAN: American Indiana Alaska Native, NHOPI: Native Hawaiian or Other Pacific Islander, BMI: body mass index

Table 2Relative Risk (95 % Confidence Interval) for Gastroschisis.

	Crude	Minimally Adjusted	Fully Adjusted
Year of Birth	0.93 (0.93-0.94)	0.98 (0.97-0.99)	0.97 (0.96–0.98)
Age	0.84 (0.84-0.85)	0.86 (0.85-0.86)	0.86 (0.86-0.87)
Nulliparous	1.98 (1.89–2.08)	1.07 (1.02–1.13)	1.11 (1.05–1.17)
BMI	0.92 (0.92-0.93)	0.94 (0.94-0.94)	0.94 (0.93-0.94)
Smoke Before Pregnancy	2.76 (2.61–2.94)	2.02 (1.80-2.26)	1.73 (1.55–1.94)
Smoke 1st Trimester	2.70 (2.53-2.89)	1.11 (0.98–1.26)	1.07 (0.94–1.21)
Non-Hispanic White (only)	Ref		Ref
Non-Hispanic Black (only)	0.61 (0.56-0.66)		0.50 (0.46-0.55)
Non-Hispanic AIAN (only)	2.55 (2.16-3.01)		1.76 (1.49–2.08)
Non-Hispanic Asian (only)	0.27 (0.22-0.32)		0.61 (0.50-0.74)
Non-Hispanic NHOPI (only)	0.71 (0.40-1.24)		0.97 (0.55–1.71)
Non-Hispanic more than one race	1.52 (1.33–1.73)		1.13 (0.99–1.28)
Hispanic	1.01 (0.96-1.07)		1.04 (0.97-1.10)
Origin unknown or not stated	0.99 (0.68–1.44)		
Born Outside U.S.	0.39 (0.36-0.42)		0.57 (0.52-0.62)
Less than High School Education	1.63 (1.53-1.73)		0.80 (0.75-0.85)
Private Insurance	Ref		Ref
Medicaid	2.40 (2.28-2.53)		1.43 (1.35–1.52)
Self-Pay	0.95 (0.81-1.12)		0.89 (0.75-1.04)
Other	1.63 (1.43–1.85)		1.10 (0.97-1.26)
Pre-pregnancy hypertension	0.52 (0.42-0.65)		1.13 (0.90-1.42)
Pre-pregnancy diabetes	0.59 (0.43–0.81)		1.07 (0.77–1.47)

AIAN: American Indiana Alaska Native, NHOPI: Native Hawaiian or Other Pacific Islander, BMI: body mass index

Definition of variables

The following prenatal characteristics around the time of conception were included from the CDC dataset: year of birth, maternal age, nativity (born in/outside of the U.S.), race (Non-Hispanic (NH) White, NH Black, NH Asian, Hispanic, and others), education, insurance, cigarette smoking before pregnancy and during the first trimester, parity, interval since last pregnancy, pre-pregnancy body mass index (BMI), pre-pregnancy diabetes mellitus (DM), and pre-pregnancy hypertension (HTN). The CDC-Natality datasets record weight at two intervals: once before pregnancy and once before delivery. Height is also recorded for each mother before pregnancy. Hence, pre-pregnancy BMI was calculated for each record. A detailed guide on the definition of each variable for all the included years can be seen in the vital statistics online data portal (https://www.cdc.gov/nchs/data access/vitalstatsonline.htm).

Inclusion and exclusion

We included singleton live births with confirmed isolated fetal and/or neonatal gastroschisis. We excluded pregnancies with twins or higher-order multiple gestations, other major structural anomalies or chromosomal abnormalities, if gastroschisis status was unknown or not stated, and if there were missing data for one of the above-stated prenatal characteristics. Furthermore, the cohort was divided into cases to include patients with gastroschisis and controls to include those without gastroschisis.

Statistical analysis

Frequency distributions, prevalence per 10,000 live births along with 95 % confidence intervals were estimated. Logistic regression and multivariate logistic regression models were to calculate relative risk and 95 % confidence intervals in two models i)a minimally adjusted model to covariates known through previous studies to be associated with gastroschisis including age, smoking, BMI, and parity, and ii) a fully adjusted model to all the covariates in the univariate analysis. To explore the effect of COVID on the declining rate of gastroschisis we chose the 34 months before and after February 2020. We performed a linear regression model using the monthly rate of gastroschisis as the outcome and time, and indicator for pre and post-COVID and interaction of time and pre/post-COVID. The interaction tested the slope of decline

in gastroschisis pre and post-COVID. Slope interaction was also tested between gastroschisis and non-gastroschisis in the high-risk groups. Statistical analysis was performed using SAS (Version 9.4; SAS Institute).

Results

Following exclusion, we identified 32,088,301 singleton live births, among which 6804 (0.02%) were pregnancies with isolated gastroschisis during the study period from 2014 to 2022 (Fig. 1). The overall prevalence of gastroschisis in the population during this eight-year study period was 2.19 per 10,000 live births. A declining trend in the prevalence of gastroschisis was observed over the study period with the year-by-year analysis showing a significant decline from 2.86 per 10,000 births in 2014 to 1.55 per 10,000 in 2022 (Table 1).

The prevalence of gastroschisis was highest among non-Hispanic American Indigenous populations (6.02 per 10,000), those born in the US (2.53 per 10,000), maternal age < 20 years (8.64 per 10,000), prepregnancy underweight BMI group (4.20 per 10,000), pre-pregnancy cigarette smoking (5.29 per 10,000), Medicaid recipients (3.26 per 10,000), and among nulliparous as compared to multiparous gravidae (3.30 per 10,000 vs. 1.67 per 10,000), among gravidae with a short interpregnancy interval < 18 months (2.23 per 10,000), less than high school education (3.3 per 10,000), and more common among pregnancies conceived in the North American spring (2.43 per 10,000) (Table 2).

When adjusting for all the previously mentioned factors in the minimally and the fully adjusted models described in the methods, the risk of gastroschisis still exhibited a significant decreasing trend across all models, with RR of 0.97 in the fully adjusted model (Table 2). Fig. 2 shows the annual prevalence of gastroschisis per 10,000 live births over the study period (2014–2022), respectively.

To understand the etiology behind the decreasing trend of gastroschisis, we investigated the temporal trends by year in demographic characteristics in patients with pregnancies complicated by gastroschisis. There was a significant decrease in the proportion of pregnancies occurring among gravidae < 20 years of age, underweight, and with prepregnancy cigarette smoking over the study period (Table 3). Fig. 3 shows the graphical demonstration of the annual prevalence of gastroschisis per 10,000 births by maternal age, BMI, parity, and prepregnancy smoking.

The decline in gastroschisis births from 2014 to 2022, compared to

Annual Prevalence per 10,000 Births of Gastroschisis for 2014 to 2022 Regression Model for Trend



Fig. 2. Annual Prevalence per 10,000 births of Gastroschisis from 2014 to 2022.

non-gastroschisis births, was significantly more common in nulliparous, underweight (BMI<18.5), young gravidae (<age 20), relative to the overall gravid population (Fig. 4a-d). These trends are further confirmed by a significant interaction between the slopes of the two trends, suggesting that the observed reduction in gastroschisis cases is not simply a reflection of a general decrease in birth rates but is associated with a specific, significant, and unique decline in births among these higherrisk groups. Finally, the rate and trend of gastroschisis did not significantly change between pre-and post-COVID-19 cohorts; notably, in the interval since COVID-19, legislative actions which have resulted in access to reproductive care have also occurred and may lend to the trend in recent increases in gastroschisis previously attributed by others to COVID-19 [17].

Discussion

Principal findings

Our study identified a significant decline in gastroschisis prevalence in a nationwide cohort including over 32 million singleton live births in the US from 2014 to 2022. Notably, this decline correlates with decreases in pregnancy rates among high-risk groups such as individuals under 20 years, those classified as underweight, nulliparous, and smokers.

Result in the context of what is known

Several studies have established a link between gastroschisis and several risk factors including white and Hispanic race [18], low maternal age [19], abnormal BMI [20,21] (mostly low BMI [21–24]), cigarette smoking history [19,23], nulliparity[23], low interpregnancy interval [25], and conception in the spring [26]. Our findings align with this literature, adding a temporal dimension that suggests an effective reduction in these risk factors over time. The declining rate of gastroschisis can be paralleled by the nationwide decline in the maternal age

under 25 years [27,28]. However, the significant decline in gastroschisis cases among higher-risk individuals compared to the general population suggests that targeted improvements in reducing risk factors specific to these groups may be effective [23,29,30]. Additionally, similar to our findings, Medicaid recipients, representing low socioeconomic groups, have been shown to have a higher rate of gastroschisis and worse infant' outcomes [13,31]. Individuals on Medicaid may have limited access to high-quality prenatal care, which can impact early pregnancy health and the management of risk factors known to be associated with gastroschisis, such as nutritional deficiencies and exposure to harmful substances. Lifestyle patterns and environmental habits affecting the teenage population have also been shown to play a role in the association of young age with gastroschisis [16]. It has been suggested that state cigarette taxes are an influential intervention in reducing the rate of cigarette use and therefore, the risk of gastroschisis [32]. Prepregnancy diet and nutrient stores suggested an influence on the etiology of gastroschisis [33,34], potentially explaining the effects related to interpregnancy interval and BMI. National data supported our findings on the trend of decreasing major risk factors for gastroschisis; the maternal age has increased in recent years [35], prepregnancy obesity has risen [36], leading to a decrease in low BMI at pregnancy, and the rate of cigarette smoking during pregnancy has declined in the US [37,38]. In addition, other studies reinforced the lack of relevance of COVID-19 to this decreasing trend in gastroschisis [39].

Clinical and research implications

One interpretation of our findings may be to lessen the urgency for public health research to address the high incidence of gastroschisis, which peaked prior to 2010 [3]. However, our data also notes that the population at highest risk includes young and vulnerable individuals, and occurs in parallel to birth rates within this population [40]. Given that these same young and vulnerable populations may rely on public resources for contraception care and availability of reproductive health care, any state, local, or federal laws that limit that access would be

Table 3Rates of Selected Demographics by Year.

	2014–2016	2017–2019	2020-2022	P-value
Race/Ethnicity	53.1	51.9	51.4	< 0.0001
Non-Hispanic White (only)	13.8	14.3	14.1	
Non-Hispanic Black (only)	0.8	0.8	0.7	
Non-Hispanic AIAN (only)	6.1	6.5	6.0	
Non-Hispanic Asian (only)	0.2	0.2	0.3	
Non-Hispanic NHOPI (only)	2.0	2.2	2.4	
Non-Hispanic more than one race	23.6	23.8	24.8	
Hispanic	0.4	0.4	0.4	
Origin unknown or not stated				
Maternal Nativity	77.8	77.3	78.2	< 0.0001
Born in U.S.	22.2	22.7	21.8	
Born outside U.S.				
Maternal Age	6.0	4.9	4.2	< 0.0001
15-< 20	21.7	19.5	18.0	,
20-< 25	29.2	29.1	28.1	
25-< 30	27.3	28.6	30.1	
30-< 35	12.9	14.6	15.9	
35-< 40	2.9	3.2	3.6	
40 +	2.9	5.2	3.0	
Mean(SD) Age, Years	28.4	28.9	29.3	< 0.0001
=	3.7	3.2	2.7	< 0.0001
BMI Category	3./ 45.2	3.2 42.3	39.2	< 0.0001
Underweight (BMI < 18.5)				
Normal weight (BMI 18.5-<25)	25.8	26.6	27.4	
Overweight (BMI 25-<30)	13.9	15.1	16.4	
Obese Class I (BMI 30-<35)	6.7	7.5	8.3	
Obese Class II (BMI 35-<40)	4.7	5.3	6.0	
Obese Class III (BMI >=40)	06.7	07.1	07.6	0.0001
Mean(SD), BMI	26.7	27.1	27.6	< 0.0001
Pre-Pregnancy Hypertension, Yes	1.6	2.0	2.7	< 0.0001
No	98.4	98.0	97.3	
Pre-Pregnancy Diabetes, Yes	0.8	0.9	1.1	< 0.0001
No	99.2	99.1	98.9	
Pre-Pregnancy Smoker, Yes	10.2	8.4	5.8	< 0.0001
No	89.8	91.6	94.2	
1 st Trimester Smoker, Yes	7.6	6.3	4.5	< 0.0001
No	92.4	93.7	95.5	
Insurance	43.1	42.3	41.2	< 0.000
Medicaid	48.6	49.7	51.4	
Private Insurance	4.2	4.3	4.0	
Self-Pay	4.1	3.8	3.4	
Other				
Education	14.4	12.6	11.2	< 0.0001
< High School Grad	85.6	87.4	88.8	
> =High School Grad				
Interpregnancy Interval	10.5	10.9	11.5	< 0.0001
< 18 m	8.0	8.1	8.4	
18–23 m	37.8	38.3	38.1	
24 m+	11.4	11.2	10.1	
Unknown				
Parity	32.3	31.5	32.0	< 0.0001
Nulliparous	67.7	68.5	68.0	. 3.000
Multiparous	07.7	00.0	00.0	

AIAN: American Indiana Alaska Native, NHOPI: Native Hawaiian or Other Pacific Islander, BMI: body mass index

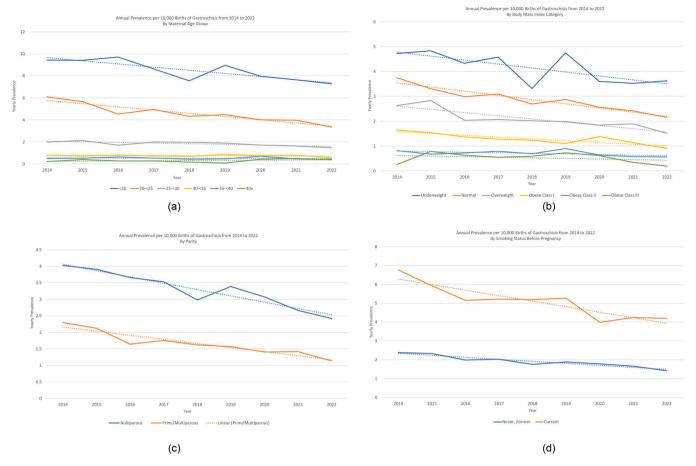


Fig. 3. Graphical demonstration of the annual Prevalence per 10,000 Births of Gastroschisis from 2014 to 2022 by (a) Maternal Age Group, (b) Body Mass Index Category, (c) Parity, and (d) Smoking Status Before Pregnancy.

anticipated to lead to increased occurrence of gastroschisis. Ongoing surveillance is essential to identify these factors, inform public health interventions, and mitigate prevalence.

Strengths and limitations

Utilizing national data allowed us to conduct the prevalence trend analysis over a long period. Our subgroup analysis enhances understanding of the associated factors. Limitations are related to utilizing birth certificates to detect gastroschisis occurrences in the United States, which could be subject to collection inaccuracies. The natality dataset includes only live births which could affect true prevalence due to lack of data on pregnancy losses, terminations, and fetal demises. However, that is unanimous over the included years, and the study aimed to track trends rather than period prevalence. Additionally, causality could not be determined from the CDC database due to its cross-sectional design. It is also important to consider variations at more localized levels, which necessitates prospective studies. Furthermore, state-level healthcare policies and variations in data collection practices could influence the reported trends, but these factors could not be analyzed due to the lack of state-specific and policy-related data in the CDC dataset.

Conclusion

Up through 2010, there was an exponential increase in the prevalence of gastroschisis. Our current study is the first to provide an indepth characterization of a significant decline in gastroschisis prevalence among singleton live births in the US from 2014 through 2022. This decline is attributable to overall birth rate declines in the at-risk

population of young/teen gravidae, those who are underweight, and among gravidae who smoke prior to pregnancy. Given recent legislative actions that limit or eliminate reproductive health care access disproportionately, we anticipate the absolute need for continued public health efforts focused on remediating reproductive health care disparities among this population. Future studies should focus on identifying and monitoring potential unidentified risk factors to prevent future peaks in gastroschisis prevalence.

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CRediT authorship contribution statement

Perrone Erin E.: Writing – review & editing, Validation, Supervision. Buchmiller Terry L.: Writing – review & editing, Validation, Supervision. Aagaard Kjersti M.: Writing – review & editing, Supervision, Resources, Methodology. Gray Brian: Writing – review & editing, Visualization, Validation, Supervision. Abiad May: Writing – original draft, Validation, Supervision. Moss Kevin L.: Software, Methodology, Formal analysis, Data curation. Zargarzadeh Nikan: Writing – original draft, Visualization, Validation. Mustafa Hiba J.: Writing – original draft, Supervision, Project administration, Methodology, Investigation, Data curation, Conceptualization. Shamshirsaz Alireza A.: Writing – review & editing, Visualization, Validation, Supervision, Resources, Methodology, Conceptualization.

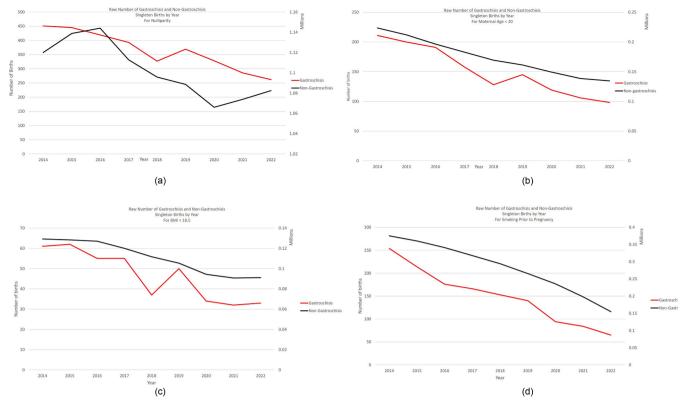


Fig. 4. Numbers of births trend of gastroschisis and non-gastroschisis births for (a) Nulliparity, (b) Maternal Age < 20, (c) BMI < 18.5, and (d) Smoking Prior to Pregnancy.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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