

Characteristics and Outcomes of Ptyalism Gravidarum

Moshe Bronshtein MD¹, Ayala Gover MD^{2,4}, Ron Beloosesky MD^{3,4}, Hanin Dabaja MD³, Yuval Ginsberg MD³, Zeev Weiner MD^{3,4} and Nizar Khatib MD³

¹Faculty of Social Welfare & Health Sciences, University of Haifa, Haifa, Israel

²Neonatal Intensive Care Unit, Carmel Medical Center, Haifa, Israel

³Department of Obstetrics and Gynecology, Rambam Health Care Campus, Haifa, Israel

⁴Rappaport Faculty of Medicine, Technion–Israel Institute of Technology, Haifa, Israel

ABSTRACT: **Background:** Ptyalism gravidarum (PG) is a condition of hypersalivation that affects pregnant women early in gestation. Symptoms include massive saliva volumes (up to 2 liters per day), swollen salivary glands, sleep deprivation, significant emotional distress, and social difficulties.

Objectives: To examine maternal and fetal characteristics and pregnancy outcomes of patients with PG.

Methods: Patients diagnosed with PG in our clinic during the years 2001–2016 were identified and contacted. Demographic data were extracted from patient charts and clinical and outcome data was collected via telephone interviews.

Results: The incidence of PG was 1/963 (0.09%) in our sample. Eleven out of 22 women (40%) with PG were also diagnosed with hyperemesis gravidarum. Fetal gender did not increase the risk. Of the mothers presenting with PG, 37% had a positive family history for this condition. There was no associated increase in the rate of fetal or maternal complications. Two women reported a resolution of the symptoms immediately following hypnosis with acupuncture treatment.

Conclusions: Although PG represents an unpleasant mental and physical condition, it does not pose any specific risk to the health of the mother or increase adverse perinatal outcomes for the fetus. Alternative medicine could play a role in the treatment of PG.

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KEY WORDS: hyperemesis gravidarum (HG), hypersalivation, pregnancy, prenatal outcome, ptyalism gravidarum (PG)

Ptyalism gravidarum (PG) is a disorder of excessive salivation affecting pregnant women in early gestation [1,2]. Symptoms include massive saliva volumes (up to 2 liters per day), swollen salivary glands, sleep deprivation, significant emotional distress, and social difficulties. Patients presenting with this condition are compelled to use cups, tissues, and other measures to dispose of redundant saliva. It usually resolves during the second trimester; however, some cases continue throughout the pregnancy.

Pathophysiology is unknown, although beta-human chorionic gonadotropin and estrogen may be involved [3]. Incidence varies significantly worldwide from 0.08%, reported in the United State, to up to 35%, reported in Turkey [2].

Associations with hyperemesis gravidarum (HG), male fetal sex, and small for gestational age have been previously described [4].

The aim of this study was to delineate the characteristics and outcomes of PG.

PATIENTS AND METHODS

We retrospectively reviewed data of 34,372 pregnancies from 22,149 women who attended our clinic (a private clinic) between 2001 and 2016. Using a reliable electronic database, we identified 23 women who complained of excessive salivation. Demographics and patient characteristics were extracted from the database. Pregnant women attending our clinic were asked about their complaints. The diagnosis of PG was concluded based on the women's complaints. They attended our clinic and held a glass for spitting and paper to wipe their mouth due to excessive salivation. HG was defined by vomiting-induced weight loss with or without ketonuria. Pregnancy course and outcome information, including duration of symptoms, treatment and outcome, birth weight, gestational age at birth, and obstetric complications such as preeclampsia, were obtained via telephone interviews.

The study was designed in accordance with the ethics standards of the institutional review board and was approved by the Helsinki Committee (0210-10-RMB).

RESULTS

Twenty-three women who complained of PG were included in the study; 22 women (49 symptomatic pregnancies) responded, and one patient was lost to follow-up. The incidence of the condition was 1:963 pregnant women (0.09%). Maternal age was normally distributed with 67% of women being between 25 and 35 years of age, and matching the age distribution in the general population in our clinic.

We categorized the cases as primary or secondary ptyalism according to the first appearance of this condition. In the primary group (15 women, 37 pregnancies), excessive salivation appeared during the first pregnancy and reappeared in all subsequent pregnancies. In the secondary group (7 women, 9 pregnancies), PG manifested only during the second or subsequent pregnancies.

None of the women in this cohort complained that they required a wet sleeping pad at night; however, the condition reappeared during wakefulness.

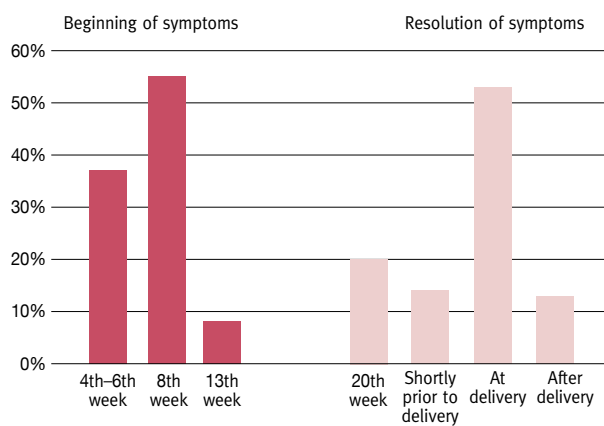
TIMING OF APPEARANCE AND RESOLUTION

For every woman in this cohort, the complaint appeared at the same timing in each pregnancy. In 37% of the pregnancies, ptyalism began early, for one patient excessive salivation started as early as 4 weeks of gestation during each of her three pregnancies, and the others reported symptom onset at gestational week 5–6. In 55% of the pregnancies, symptoms began at 8 weeks of gestation and in 8% they began at 13 weeks [Figure 1].

Resolution of the symptoms occurred at the same timing in each pregnancy for all of the women except two. In one of those patients, ptyalism ended at delivery in the first pregnancy and at 20 weeks of gestation at the second pregnancy, and in another patient the symptoms ceased 1 month after delivery of the first pregnancy and at 20 weeks of gestation in the second pregnancy. Otherwise, the symptoms ceased at the 20–21 gestational week for 20% of the women, shortly prior to delivery in 14%, immediately after delivery for 53, and several days after delivery (the latest was over 1 month after giving birth) in 13% [Figure 1].

Interestingly, one patient experienced extremely excessive salivation during a twin pregnancy compared to a lesser degree of PG during her two other singleton pregnancies. Her symptoms resolved following termination of the twin pregnancy due to the diagnosis of trisomy 21 at 18–19 weeks of gestation.

Figure 1. Beginning and resolution of symptoms



PERINATAL OUTCOMES

One women gave birth at 32 gestational week, the rest of the patients delivered at term, with appropriate weight for gestational age. The average birth weight was 3300 grams. One fetus was diagnosed with unilateral hypoplastic kidney; however, no other fetal congenital malformations were reported.

HYPEREMESIS GRAVIDARUM

Eleven out of 22 women (40%) presented with HG. The rate was higher in the primary group than in the secondary group (69% vs. 14%, respectively) [Table 1]. In our cohort, the prevalence of PG among women with HG was 2.3% compared to 0.09% in women without HG.

FAMILY HISTORY

In 8 of the 22 patients (36.5%) there was a positive family history for PG: 3 of the women reported their sisters had PG, and 5 reported their mothers experienced the same phenomenon. This history was more common in the primary group than in the secondary group [Table 1].

FETAL GENDER

Female/male gender of the fetus was almost identical (23 male, 22 female).

TREATMENT

Some women reported a minor relief in symptoms after eating crackers, dry toast bread, or sour gum and candy. One patient tried homeopathic remedies, which did not mitigate her symptoms. Two women reported that complaints immediately ceased following a combined treatment of hypnosis and acupuncture.

DISCUSSION

The incidence of excessive salivation in our cohort was 1:963 pregnant women. Other international studies reported variable rates of occurrence [1,2,5], possibly due to ethnicity.

Contrary to Suzuki and colleagues [4], there was no gender predominance for PG in our cohort, which may have been due to our small sample size. The etiology of PG is unknown, although hormonal, psychological, and neuronal mechanisms have been proposed [1].

Table 1. Primary vs. secondary PG

	Primary PG	Secondary PG	Total
Number of women	14	5	19
Number of pregnancies with PG	36	7	43
Number of pregnancies with HG	25 (69%)	1 (14%)	26
Positive family history	6 (42%)	1 (20%)	7

HG = hyperemesis gravidarum, PG = ptyalism gravidarum

In our cohort, there was no nocturnal sialorrhea, which raises the possibility of a psychological component. Nevertheless, hormonal involvement cannot be excluded. The occurrence of the phenomenon early in pregnancy, the peak in symptoms after 20 weeks of gestation, and resolution after delivery, suggest hormonal involvement.

All women, except one, gave birth at term to babies with appropriate weight for gestational age. This result is in alignment with several case reports [1,6]. However, Suzuki et al. [4] demonstrated increased rates of infants who were small for gestational age in mothers with PG [4], which may be related to increased maternal weight loss and prematurity [7].

Contrary to HG, in which the symptoms could be alleviated with appropriate treatment [8], in PG such an approach is not available. Several therapies have been previously attempted, including consumption of large amounts of sweets [5,9] or antiemetic medications [10]. Two women from our study group reported complete resolution of the symptoms following hypnosis with acupuncture treatment. Another case of successful treatment of PG using hypnosis was recently described by Beevi and co-authors [11].

CONCLUSIONS

In our population we found no adverse outcomes in pregnant women with PG. Although symptoms are troublesome for the patients, PG does not seem to pose any specific risks for the mother or fetus. Hypnosis and acupuncture may be effective treatments, although further study is needed.

Correspondence

Dr. N. Khatib

Dept. of Obstetrics and Gynecology, Rambam Health Care Campus, Haifa 3109601, Israel

Phone: (972-4) 777-2515

Fax: (972-4) 777-2453

e-mail: khatibnizar@yahoo.com

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Capsule

Travel-associated Zika cases and threat of local transmission during global outbreak, California, USA

Zika and associated microcephaly among newborns were reported in Brazil during 2015. Zika has since spread across the Americas, and travel-associated cases were reported throughout the United States. **Porse** et al. reviewed travel-associated Zika cases in California to assess the potential threat of local Zika virus transmission, given the regional spread of *Aedes aegypti* and *Ae. albopictus* mosquitoes. From November 2015 to September 2017, a total of 588 travel-associated Zika cases were reported in California, including 139 infections in pregnant women, 10 congenital infections, and 8 sexually transmitted infections. Most case-patients

reported travel to Mexico and Central America, and many returned during a period when they could have been viremic. By September 2017, *Ae. aegypti* mosquitoes had spread to 124 locations in California, and *Ae. albopictus* mosquitoes had spread to 53 locations. Continued human and mosquito surveillance and public health education are valuable tools in preventing and detecting Zika virus infections and local transmission in California.

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Eitan Israeli

“Fascism is cured by reading, and racism is cured by traveling”

Miguel de Unamuno y Jugo, (1864–1936), Spanish Basque essayist, novelist, poet, playwright, philosopher, professor of Greek and Classics, and later rector at the University of Salamanca

“What’s right is what’s left if you do everything else wrong”

Robin Williams, (1951–2014), American comedian and actor