Parent and Public Interest in Whole-Genome Sequencing

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Key Words

Whole-genome sequencing · Public interest · Parents · Children

Abstract

Objective: The aim of this study was to assess the baseline interest of the public in whole-genome sequencing (WGS) for oneself, parents' interest in WGS for their youngest children, and factors associated with such interest. **Methods:** A random sample of adults from a probability-based nationally representative online panel was surveyed. All participants were provided basic information about WGS and then asked about their interest in WGS for themselves. Those participants who were parents were additionally asked about their interest in WGS for their children. The order in which parents were asked about their interest in WGS for themselves and for their child was randomized. The relationship between parent/child characteristics and interest in WGS was examined. **Results:** The overall response rate was 62% (55% among parents). 58.6% of the total population (parents and nonparents) was interested in WGS for themselves. Similarly, 61.8% of the parents were interested in WGS for themselves and 57.8% were interested in WGS for their youngest children. Of note, 84.7% of the parents showed an identical interest level in WGS for themselves and their youngest children. Mothers as a group and parents whose youngest chil-

dren had ≥ 2 health conditions had significantly more interest in WGS for themselves and their youngest children, while those with conservative political ideologies had considerably less. **Conclusions:** While US adults have varying interest levels in WGS, parents appear to have similar interests in genome testing for themselves and their youngest children. As WGS technology becomes available in the clinic and private market, clinicians should be prepared to discuss WGS risks and benefits with their patients.

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Introduction

Whole genome sequencing (WGS) is the process of identifying nearly every base pair in an individual's DNA or – in the case of whole-exome sequencing – bases that encode proteins. Compared to other genetic testing technologies, WGS generates significantly more information, much of it of uncertain value. Recently, the cost of WGS has fallen to about USD 4,000, and many predict the cost will fall to near USD 1,000 in the near future [1–3] – making its routine use within and outside the clinical arena an impending reality [4, 5]. In addition, the NIH has targeted USD 15 million for research aimed at reducing the cost of WGS to USD 1,000 (RFA-HG-10-012) and USD 6.5 million for clinical research focused on the application of

genomic data to patient care (RFA-HG-12-008, RFA-HG-12-009).

WGS raises numerous ethical challenges [6–11]. These challenges include privacy protection (both for the child and for the child's relatives), availability of data to researchers, how to address incidental findings of limited utility and predictive value, how to address diseases where our clinical interventions are limited, and how to provide effective informed consent. These issues are further complicated in the pediatric setting where the issues of parents' autonomy and children's privacy often compete, especially when disease risk may not be relevant for decades, such as in hypertension or Huntington's disease.

Genome sequencing is now being used in the clinic [4, 12–14], and parents will likely soon be more frequently making decisions about WGS for their children. There are already a number of examples of how WGS has been used with children and their families [15, 16]. These testing platforms may identify unknown genetic causes of childhood diseases or help families better understand their child's predisposition to future disorders.

Previous public opinion studies have explored parental interest in personal genetic testing for their children [17, 18] and looked at qualitative perspectives on ethical issues related to WGS [19, 20]. Levenseller et al. [11] explored such ethical issues in a nonrandom sampling of children with specific health conditions, parents of such children, and health-care providers, and found differences in priorities among these groups. Goldenberg et al. [21] specifically examined interest in WGS in the context of newborn screening and found an interest level of about 70%. In addition, a handful of studies have also assessed public interest in personal genetic testing [17, 22–28]. Despite this interesting work, to the best of our knowledge, no studies have examined a sample of the general population for parents' interest in WGS for themselves or their children in a general clinical setting, or correlated parents' interest in WGS for themselves with their interest in WGS for their children. Such information could both inform the use of next-generation sequencing technology in the clinical setting and also help clinicians better understand the attitudes and concerns of their patients.

Methods

Study Design

In May 2012, we conducted a cross-sectional internet-based survey of a nationally representative sample of the US population regarding the participants' interest in WGS for themselves and, when applicable, for their youngest child. This survey was conducted as

part of the C.S. Mott Children's Hospital National Poll on Children's Health (NPCH), a recurring online survey of parents and nonparents [29], that has served as the platform for several peer-reviewed studies of national attitudes, beliefs, and self-reported behaviors of the US public [30–33]. Prior to conducting our full survey, we ran a pilot study to optimize our survey. The University of Michigan Medical School Institutional Review Board approved this study.

Study Population

The NPCH is conducted using GfK's (Gesellschaft für Konsumforschung, a private market research company) web-enabled KnowledgePanel®, a probability-based panel designed to be representative of the US population [34]. Potential participants are identified by a random selection of telephone numbers and residential addresses and then invited by telephone or by mail to participate in the web-enabled KnowledgePanel. If individuals agree to participate but do not have internet access, GfK provides them a laptop and internet service provider connection at no cost. Panelists are then given unique login information for accessing surveys online and receive emails throughout each month inviting them to participate in research.

Survey Administration

The NPCH was pilot-tested for this study by the GfK in April 2012 with a separate convenience sample of 105 KnowledgePanel members. The descriptions were based on information provided by the National Human Genome Research Institute [35]. Survey background and questions were refined based on pilot testing. A unique KnowledgePanel sample was drawn for the NPCH that contained the survey items for this study. The introductory e-mail invited individuals to participate in a survey about child health. No incentive was offered to prospective participants beyond participation points that GfK panelists receive as part of their routine survey activities. To ensure adequate representation, parents [defined as having child(ren) 0–17 years old living in the household] and particular racial minorities including African-Americans and Hispanics were oversampled.

Survey Items

All survey respondents received the following brief explanation of WGS:

- *Our Health and Our Genes*

Staying healthy and getting sick are affected by many things. Our genes – which we inherit from our parents – can affect our health and illness in many ways.

Genes are made of DNA, and contain the instructions needed for our bodies to grow and function. All of the genes in a person make up that person's genome.

- *What Is Whole-Genome Sequencing?*

It is possible to study a person's entire genome. This process is called whole-genome sequencing. It may give information about a person's risk of having different diseases in the future.

This language was intended to give participants basic information about genetics and sequencing representative of the kinds of information they may find outside of the clinical setting.

We then asked all respondents the following question along with a Likert scale of responses (definitely interested/somewhat interested/not interested/definitely not interested): 'How interest-

Table 1. Characteristics of survey respondents

	Weighted estimates		
	parents (30.4%; n = 1,539)	nonparents (69.6%; n = 605)	overall (100%; n = 2,144)
<i>Respondent characteristics</i>			
Age, years [‡]	38.8 (18–88)	51.8 (18–94)	47.8 (18–94)
18–29 years	17.8 (210)	17.8 (72)	17.8 (282)
30–44 years [‡]	55.6 (872)	14.7 (95)	27.1 (967)
45–59 years	25.3 (433)	30.1 (235)	28.6 (668)
≥60 years [‡]	1.3 (24)	37.4 (203)	26.4 (227)
Race/ethnicity [†]			
White (non-Hispanic) [‡]	62.9 (1,076)	70.6 (447)	68.2 (1,523)
Black (non-Hispanic)	11.4 (150)	11.0 (64)	11.2 (214)
Hispanic [‡]	18.0 (208)	11.4 (62)	13.4 (270)
Other (non-Hispanic)	7.7 (105)	7.0 (32)	7.2 (137)
Female	55.8 (820)	50.8 (316)	52.3 (1,136)
Education			
High school diploma or less	37.7 (486)	42.2 (244)	30.8 (730)
Some college	29.4 (498)	28.6 (170)	28.8 (668)
Bachelor's or greater	32.9 (555)	29.2 (191)	30.4 (746)
Annual income			
USD <30,000	24.6 (287)	29.3 (144)	27.9 (431)
USD 30,000–59,999	28.9 (418)	26.7 (162)	27.4 (580)
USD 60,000–100,000	24.1 (421)	23.7 (144)	23.8 (565)
USD >100,000	22.4 (413)	20.3 (155)	20.9 (568)
Self-identified political ideology			
Liberal	26.3 (350)	30.9 (173)	29.5 (523)
Moderate	39.3 (556)	35.1 (204)	36.4 (760)
Conservative	34.4 (564)	34.0 (210)	34.1 (774)
Respondent in good or better health [‡]	89.0 (1,407)	81.9 (503)	84.1 (1,910)
Planning a child within 5 years [‡]	20.9 (265)	11.2 (51)	24.1 (316)
<i>Child characteristics</i>			
Female youngest child	47.7 (740)	N/A	N/A
Age of youngest child			
<1 year	7.1 (100)	N/A	N/A
1–5 years	34.2 (475)	N/A	N/A
6–10 years	25.2 (399)	N/A	N/A
11–13 years	12.9 (215)	N/A	N/A
14–17 years	20.6 (337)	N/A	N/A
Health of youngest child			
0 health conditions	64.8 (940)	N/A	N/A
1 health conditions	23.5 (337)	N/A	N/A
≥2 health conditions	11.8 (175)	N/A	N/A

Values represent mean (range) or % (n). All n values are unweighted. N/A = Not applicable.

[†] $p \leq 0.05$ for parents vs. nonparents; [‡] $p \leq 0.01$ for parents vs. nonparents.

ed would you be in getting your whole genome sequenced?' In addition, respondents who identified themselves as parents received the following question (completed with the age of their youngest child inserted in the brackets): 'How interested would you be in getting your [x-month/year-old] child's whole genome sequenced?' For parents, the order of these two questions was randomized. A 4-point Likert scale was chosen so participants were compelled to choose a position and to avoid ambivalence.

We also collected the following information from respondents about themselves: age, sex, race/ethnicity, education, household income, political ideology, rating of personal health, and if they plan to have a child within 5 years (only asked if respondents were <45 years of age). For parents, we also gathered the following information about all of their children in the household: age, sex, and whether their children have any specific health conditions (see Appendix 1 for a list of health conditions).

Statistical Analysis

The GfK provided the study team with de-identified data, along with census-based post-stratification weights used to match the US population distribution on sex, age, race/ethnicity, education, census region, income, and political ideology. All analyses were conducted with Stata[®] 12 (Stata Corp. LP, College Station, Tex., USA). Results reflect weighted data unless otherwise indicated. Among the completed surveys, the following individual item nonresponse rates were observed: health of the youngest child (6% nonresponse) and political ideology (4% nonresponse overall and 4.5% nonresponse among parents). All other single question nonresponse rates were less than 2%. Cases with nonresponse on specific items were dropped from the analyses on an item-by-item basis.

Respondent Characteristics

We grouped respondents by parent status (parent vs. nonparent) and calculated frequency distributions for age, race/ethnicity, sex, education level, income, political ideology, personal health, and plans to have children in the next 5 years for parents, nonparents, and the overall survey population (table 1). Differences in characteristics between parents and nonparents were tested with Rao-Scott corrected Pearson's χ^2 test for categorical characteristics [36], and with a *t* test of the means for continuous characteristics. For parents, we also calculated the frequencies for sex, age, and number of health conditions in their youngest children.

Overall Adults' Interest in WGS for Themselves

We tabulated the overall interest in WGS for each respondent. We then collapsed the interest level into two categories (interested = definitely interested/somewhat interested vs. not interested = not interested/definitely not interested) and tested the association between the two-category interest level in WGS and the respondent characteristics listed in table 1. Age was tested with binary logistic regression, while categorical variables were tested with Rao-Scott corrected Pearson's χ^2 tests. Tests with a *p* value of ≤ 0.2 were included in a multivariate logistic regression on respondent characteristics versus two-category interest level in WGS for oneself.

Parents' Interest in WGS for Themselves and Their Youngest Children

We tabulated parents' interest in WGS for themselves and for their youngest children and compared individual responses to both questions. Using Rao-Scott corrected Pearson's χ^2 analysis, we found no effect of question order on response to either question. For both questions, we then collapsed the interest level into two categories as we did when analyzing the overall adult cohort. We compared the two-category interest level in WGS for oneself and the interest level in WGS for one's youngest child with an adjusted Wald test. We tested the association between the two-category interest level in WGS and the characteristics of the respondents and youngest children listed in table 1. The age of the respondents was tested with binary logistic regression; all other characteristics were tested with Rao-Scott corrected Pearson's χ^2 tests. Associations with *p* ≤ 0.2 were included in a multivariate logistic regression analysis of respondent characteristics versus two-category interest level in WGS for oneself and for one's youngest child.

Results

Study Population

The survey response rates were 62% overall (parents and nonparents) and 55% among parents. In table 1, characteristics from the survey respondents are presented. The majority of respondents reported themselves to be white, in good or better health, and not planning to have a child in the next 5 years. Education level, income, and political ideology were evenly distributed among parents and nonparents. Parents were more likely to be between ages 30 and 44 years, Hispanic, in at least good health, and planning to have a child in the next 5 years. Nonparents were more likely to be white and >60 years of age.

Two thirds of the youngest children of the respondents had no health conditions. The youngest children were evenly distributed between genders and across the specified age categories.

Overall Interest in WGS among US Adults

Respondents were divided in their interest in having their whole genomes sequenced, with about 58.6% of those surveyed expressing at least some interest (fig. 1). In multivariate models of the interest in WGS, a significantly higher interest in WGS was observed among those planning to have a child in the next 5 years, Hispanics, and those with bachelor's degrees or higher education versus peers (table 2), while ideologically conservative individuals expressed a lower interest rate than ideological moderate ones. Age, sex, income, and health of the respondents were not significantly associated with the population-level interest in WGS.

Parents' Interest in WGS for Themselves and Their Youngest Children

Parents expressed a similar level of interest in WGS for themselves and for their youngest children, with about 61.8% being at least somewhat interested in WGS for themselves, and 57.8% being interested in WGS for their youngest child (fig. 2). Furthermore, 84.7% of the parents gave identical responses to the two questions. However, interest in WGS for their youngest children was slightly less than interest in WGS for themselves (fig. 2). The difference is statistically significant (adjusted Wald's test *F* statistic of 0.0001).

Higher levels of interest in WGS for themselves and their youngest child were seen among women, respondents whose youngest children had 2 or more chronic conditions, and among those educated with bachelor's degrees or more (significant for self-testing only) (ta-

Table 2. Adjusted odds of adults' interest in WGS for themselves (n = 2,037)

Characteristics	Adjusted OR (95% CI)	p value
Age of respondent	1.00 (0.99, 1.01)	0.90
Race/ethnicity		
White (non-Hispanic)	reference	
Black (non-Hispanic)	1.41 (0.85, 2.34)	0.18
Hispanic	1.82 (1.13, 2.94)	0.01
Other (non-Hispanic)	1.48 (0.79, 2.76)	0.22
Education		
High school diploma or less	reference	
Some college	1.14 (0.79, 1.65)	0.49
Bachelor's or greater	1.69 (1.15, 2.50)	0.008
Annual income		
USD <30,000	reference	
USD 30,000–59,999	0.87 (0.58, 1.31)	0.51
USD 60,000–100,000	0.94 (0.61, 1.46)	0.80
>USD 100,000	1.41 (0.88, 2.26)	0.15
Self-identified political ideology		
Liberal	1.09 (0.75, 1.59)	0.65
Moderate	reference	
Conservative	0.53 (0.37, 0.75)	<0.001
Respondent in good or better health	1.40 (0.88, 2.23)	0.15
Planning a child within 5 years	2.01 (1.24, 3.28)	0.005
Being a parent	1.06 (0.78, 1.43)	0.73

Sex of the respondents had a crude p value >0.2 and was therefore not included in the regression.

ble 3). Ideological conservatives expressed significantly less interest in WGS for both themselves and their youngest children than did ideological moderates. Age, race/ethnicity (including being African-American), income, health of the respondents, plans to have a child within 5 years, gender of the youngest child, and age of the youngest child were not significantly associated with WGS interest for either oneself or for one's youngest child. Of note, parent education level had a p value of 0.23 and 0.97 for interest in WGS for oneself and for one's youngest child, respectively, when analyzed alone (above our threshold of 0.2 for inclusion in the final regression). However, because education was significant for the general population's interest in WGS for oneself and became significant for parent interest in WGS for oneself when included in a multivariate regression, we decided to include parent education level in the final multivariate analysis. The inclusion of these data does not significantly change the odds ratios of other characteristics or affect whether other variables are statistically significant.

Fig. 1. Overall interest among US adults in WGS for themselves (n = 2,123).

Fig. 2. Parent interest in WGS for themselves [n = 1,520; solid bars (blue in the online version)] and their youngest children [n = 1,526; open bars (white)].

Discussion

Public Interest in WGS

We found that 60% of the US adult population expressed at least some interest in WGS, a level consistent with what the study by McGuire et al. [17] found when examining interest in personal genome testing (e.g., sin-

Table 3. Adjusted odds of parents' interest in WGS for themselves (n = 1,375) and their youngest children (n = 1,377)

Characteristics	Interest for themselves, adjusted OR (95% CI)	p value	Interest for youngest child, adjusted OR (95% CI)	p value
Female gender (respondent)	1.54 (1.15, 2.07)	0.003	1.68 (1.26, 2.24)	<0.001
Education				
High school diploma or less	reference		reference	
Some college	1.22 (0.86, 1.77)	0.26	1.19 (0.83, 1.71)	0.33
Bachelor's or greater	1.58 (1.09, 2.29)	0.02	1.28 (0.89, 1.84)	0.18
Self-identified political ideology				
Liberal	1.13 (0.77, 1.66)	0.52	1.09 (0.75, 1.58)	0.65
Moderate	reference		reference	
Conservative	0.59 (0.42, 0.82)	0.002	0.58 (0.42, 0.81)	0.001
Respondent in good or better health	0.73 (0.41, 1.30)	0.29	0.82 (0.47, 1.42)	0.3
Planning a child within 5 years	1.38 (0.92, 2.06)	0.12	1.33 (0.89, 1.98)	0.16
Health of youngest child				
0 health conditions	reference		reference	
1 health condition	0.99 (0.71, 1.39)	0.96	1.05 (0.75, 1.47)	0.77
≥2 health conditions	2.40 (1.45, 3.98)	0.001	2.05 (1.23, 3.42)	0.006

gle-nucleotide polymorphism array testing), and other studies [25–28] probing interest in genetic testing in a variety of contexts and populations. As WGS continues to become more widely available within and outside the clinical setting (e.g., direct-to-consumer), more individuals may begin to express interest in these tests. The challenge will be for clinicians to convey the complexities of this testing (e.g., unanticipated and unrelated findings or results of uncertain clinical significance) to patients and their families in an understandable and meaningful way.

A main objective of our study was to compare parents' interest in WGS for themselves with their interest in WGS for their youngest children. We found that parents expressed very similar levels of overall interest in WGS for themselves (61.8%) and for their youngest children (57.8%), numbers similar to those reported by McGuire et al. [17] who did not specifically poll parents [3]. This level of interest, together with the fact that 84.7% of the parents provided identical responses to both questions (a trend also reported by Tercyak et al. [18] with regards to pediatric genetic testing for newborn screening), suggests that interest in WGS is a decision made for the family. Parents who did not express interest in WGS may be uneasy about such a strong intrusion into their child's privacy or may be more concerned than other parents about possible negative outcomes for their children. However, this similarity may also be due to a poor understanding of WGS and not being able to differentiate between the two

scenarios provided. The effect of more information, or informed consent, on interest in WGS is a notable future area of study.

Factors Associated with Interest in WGS

We also found that parents whose youngest children had two or more medical conditions were significantly more likely to be at least somewhat interested in WGS, both for themselves and for their youngest child. This is consistent with patterns of interest related to newborn screening [18]. Greater interest in WGS in households experiencing more childhood illness may be due to parents' desire to find a reason – either in their own genes or in their children's genes – for those illnesses. These data are notable as WGS continues to grow in use for diagnostic purposes in children with unknown conditions.

We also identified a strong and consistent association between having a conservative ideology and a lower interest in WGS. Although political ideology has been linked to beliefs about health-care policy (e.g., Patient Protection and Affordable Care Act, abortion, stem cell research), to the best of our knowledge, no publication has described a relationship between ideology and genetic testing issues so far. One study found that increased attendance at religious services correlated with more negative attitudes towards genetic testing [37], and another found a complex interplay between knowledge, religion, and attitudes towards genetic testing [38]. We speculate that the patterns seen in these studies and ours may be

due to respondents' association of genetic testing with perinatal genetic testing, pregnancy termination, and stem cell research.

Planning to have a child in the next 5 years was significantly associated with greater interest in WGS among adults overall, but was no longer significant when we focused our analysis on parents. Perhaps having 1 or more (most likely healthy) children puts parents' minds at ease concerning their own genetic makeup compared to new parents who lack this same experience. Finally, we also saw an association between respondents' Hispanic ethnicity and interest in WGS (a complex pattern reported sporadically in previous literature) [33, 39], being educated and interest in WGS for oneself, and being a mother versus being a father and interest in WGS (consistent with data from the study by Tercyak et al. [18]). The reasons for these relationships are unclear and merit future study.

Notably, we did not find other significant associations between race and WGS interest. A focus group study by Yu et al. [20] suggested differences in interest in WGS between African-Americans and non-African-Americans, though the survey performed was nonrandom, and survey responses were public in the focus groups. However, our results certainly do not mean that attitudes towards, or even interest in, WGS are the same between races. The study by Yu et al. [20] suggests that African-Americans have more distrust of medicine and are more community oriented than non-African-Americans. The relationship between race and WGS continues to merit further investigation.

Study Limitations and Future Research

Our study has the following limitations. First, we acknowledge that the brief introduction of WGS we gave to respondents did not inform the study participants to the level that would occur in the clinical setting. However, we intended for our study to represent a baseline level of understanding that an average citizen would have when they walk into a clinic to speak with their clinician, or when considering the option to pursue WGS should it be available directly to consumers. This survey was not meant to assess public uptake of actual WGS after an extensive educational or consent process. Studies have found that people have an incomplete knowledge of genetics and genetic testing [37, 40–43], and that when low-risk people are faced with the real-life decision to get tested, and undergo preparatory genetic counseling, interest in WGS declines [41, 44]. However, in a real-world 15-min clinic visit, time for true informed consent is limited, and many of the choices parents make will largely be dictated by their previously held beliefs – a phenomenon clinicians

frequently encounter with vaccines. Therefore, we believe an understanding of the baseline reaction of the public to WGS adds significant value to our knowledge base.

Second, although the survey questions were trialed prior to our study and were designed to be simple, the questions may have still been confusing to our audience.

Third, our Likert scale categories of 'not interested' and 'somewhat interested' are not strict inverses and may have biased responses towards interest. However, as mentioned in the Discussion section, our findings on interest in WGS were similar to other study findings of interest in genetic testing. Furthermore, this should not affect the correlation of interest in WGS for oneself and one's child.

Fourth, as with all survey data, nonresponse is a concern, but our response rate is consistent with those of other published nationally representative household surveys from this survey platform [30, 31]. However, if one group of patients (e.g., those whose youngest child had a health condition) were more likely to respond to the survey, this might affect the overall interest level. Nevertheless, our bivariate analysis should be more robust to a possibly nonrepresentative sample.

Fifth, it is likely that there are other demographic and clinical characteristics associated with public and parent interest in WGS. However, our goal in this study was not to generate a fully explanatory model, but rather to generate a launch point for future inquiries.

Finally, our study design did not permit us to probe the rationale behind the respondents' interest levels, nor to understand why certain respondent characteristics are associated with more or less interest in WGS. Further research is necessary to better elucidate these reasons, and by so doing, to help clinicians better inform their patients about WGS.

Conclusion

As the cost and time necessary for WGS continue to decrease, WGS will presumably become more available, both as a clinical test and as a product offered in direct-to-consumer settings. Next-generation sequencing remains a controversial issue, and the indications for testing, utility of testing, and what information should be captured with testing continue to be hotly debated. Our study provides clinicians with a basic understanding of population and parent interest in WGS, highlighting the need to prepare for a potential increase in future requests they may receive from patients. These data also serve as a starting point for future inquiries into the reasons people have for their interest in WGS. These future studies should delve

into the reasons behind the demographic associations we found to provide more informative data on the reasons behind people's attitudes, and thereby give clinicians, both generalists and specialists, data from which they can properly explain WGS to patients and parents and ensure that their patients have the opportunity to make informed decisions about WGS for themselves and their children.

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Disclosure Statement

None of the authors have any conflicts of interest to disclose. The views expressed herein do not necessarily represent those of the University of Michigan or Case Western Reserve University.

Appendix 1

Health conditions of the youngest child. Respondents were asked to endorse all those listed that apply to their children.

Conditions
Asthma
Attention deficit/hyperactivity disorder (ADD/ADHD)
Behavioral disorders
Birth defects
Depression
Hearing problems
Heart problems
Lung disease from prematurity
Overweight or obese
Seizures

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