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Adolescent Perspectives on the Return of Individual Results in Genomic Addiction Research

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Abstract

This study surveyed all adolescents who were enrolled in behavioral genomic research and provided DNA to a biobank, including 320 patients undergoing treatment for substance and conduct problems (SCPs) and 109 non-SCP controls. Participants selected from three options on the return of individual genomic results (RIR) and rated eight methods of re-contact. Most individuals with SCPs (77.8%) and non-SCP controls (72.5%) wanted RIR involving health or behavioral implications. The majority of individuals with SCPs (67.2%) and non-SCP controls (69.7%) indicated that phone re-contact was 'best', with e-mail (22.5% SCPs, 33.9% non-SCPs) and social networking websites (21.3% SCPs, 20.2% non-SCPs) being viable options. These results suggest a layered approach for RIR: phone calls, followed by e-mails and a secure message to a social networking account. Data from this special and vulnerable population, which includes youth involved in the criminal justice system and substantial minority participation, bring an essential and missing perspective to the discussion of RIR.

INTRODUCTION

Among the stakeholders in the discussion regarding the return of individual genomic research results (RIR) including incidental findings, research participants have limited input. Attitudes of adult research participants are understudied, and we find no research on adolescent perspectives on RIR. The purpose of this study is to provide data identifying preferences for RIR and preferred means of re-contact from adolescent, including patients in treatment for substance and conduct problems (SCPs) and non-SCP controls who are all enrolled in behavioral genomic research.

Whether and how to RIR are among the most deliberated issues in genomic research (Jarvik et al., 2014; Eckstein et al., 2014; MacArthur et al., 2014; Berg et al., 2013; Biesecker, 201; Koral et al., 2013; Burke et al., 2013; Green et al., 2013; Wolfe et al., 2012). Overviews of strategies for RIR identify areas of consensus including: actionability, participant consent, and referral for follow-up (Jarvik et al., 2014; Berg et al., 2013). The ethical duty to RIR,

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when appropriate, rests upon the principles of respect for persons and protection from harm (Holm et al., 2014; Avard et al., 2011). Clinical utility, misunderstanding of results, potential for emotional harm, limited access to trained clinicians, and potential loss of confidentiality are barriers to RIR (Ziniel et al., 2014; Ramoni et al., 2013).

Studies report the majority of research participants endorse RIR (Allen et al., 2014; Master et al., 2013; Fernandez et al., 2009; Shalowitz and Miller, 2008). A study of parents with children in a biobank reported that most endorsed RIR, even for severe or untreatable diseases, citing reasons of personal utility (Ziniel et al., 2014; Harris et al., 2012). Views of parents of children affected by cancer and other diseases expressed enthusiasm for RIR, irrespective of disease severity (Fernandez et al., 2014; Kleiderman et al., 2014). Adolescents with cancer indicated a right to RIR in a timely manner (Fernandez et al., 2009, 2007).

Studies have also examined the attitudes of the general public (Yu et al., 2014; O'Daniel and Haga, 2011; Roberts et al., 2010; Godard et al., 2007). Those studies and reports in clinical trials literature indicated the general public endorses RIR, frequently despite negative implications (Miller et al., 2010; Murphy et al., 2008; Fernandez et al., 2009). The proliferation of direct-to-consumer genetic testing companies reflects the public's interest in individual genetic information (Evans and Green, 2009; Marietta and McGuire, 2009). Additionally, international surveys are in progress to assess public attitudes toward RIR (e.g., http://www.GenomEthics.org).

There is call in the literature to understand psychiatric research participants' expectations regarding RIR and to clarify researchers' duties (Mathieu et al., 2013). This study surveyed 320 SCP adolescents compared to 109 non-SCP adolescents who provided their genotypic and phenotypic data without name or other identifiers to a National Institutes of Health biobank, which shares information with qualified investigators globally. Since evidence indicates that persons with SCP are prone to be “impulsive” and “risk-takers” when compared to controls (McGue et al., 2001), our hypothesis was that SCPs would be more likely to want RIR with health or behavioral implications because they have a SCP diagnosis and differ demographically from non-SCPs. See Table 1. Although there are no genomic addiction variants to return currently, these adolescents will likely be candidates for RIR in the future. It is important to explore preferences by group in an attempt to develop effective RIR processes for diverse adolescent research participants in genomic addiction research.

The Colorado Institutional Review Board approved this study in accordance with Federal guidelines.

METHODS

Study Participants

Participants were of either sex and any racial or ethnic group. SCPs were recruited from our university-based substance abuse treatment program. Non-SCPs were identified through their participation in an affiliated primary care clinic, online or newspaper advertisements, flyers, etc., and lived in zip code areas that frequently contribute SCPs to the treatment program.

Inclusion criteria for all participants were: (1) age 14–18 years; (2) full-scale IQ ≥ 80; (3) consent from participant or, for minors, assent from participant with consent from a parent. SCPs had serious substance use problems, usually including DSM-IV substance abuse or dependence diagnoses, and serious antisocial problems, usually including symptoms of Conduct Disorder. Exclusion criteria for all participants were: (1) psychosis; (2) obvious intoxication; (3) current risk of suicide, violence, or fire setting sufficiently great to interfere with evaluation or to endanger evaluators; (4) insufficient English skills for consenting/assenting or interviews; (5) parent does not consent for minor child to participate in this study. Non-SCPs were excluded if they had any current or previous treatment for conduct or substance problems.

Survey Development

The investigators developed a 10-item RIR survey, which was piloted in the study population and revised accordingly. Four hundred twenty nine participants (320 SCPs, 109 non-SCPs) selected from three RIR options: (1) Contact me if researchers find out something about my DNA that affects my health or behavior; (2) Contact me if researchers find out something about my DNA that affects my health or behavior, only if there is a treatment; (3) Do not contact me if researchers find out something about my DNA that affects my health or behavior. All participants were instructed to choose only one of the three options. Participants also rated 8 methods of RIR re-contact separately on a 5-point scale from 1 (best) to 5 (least helpful).

Data Collection

All participants were enrolled in genomic addiction research through the Center on Antisocial Drug Dependence. As part of that study participants completed a standard battery of assessments, including demographic information (age, sex, highest grade completed, ethnicity) and a 10-item RIR survey, which was added near the beginning of the standard assessment battery.

A research assistant verbally introduced the survey and then participants completed them using a paper-pencil, self-report format. Data were entered in Microsoft Access, password-protected, and stored on the secured institutional server.

Data Analysis

Distributions of outcomes were evaluated and analyses were conducted in IBM® SPSS® Statistics 21(IBM Corporation, 2012). SCPs and non-SCPs were compared on demographic characteristics and RIR preferences with independent t-tests and chi-square tests. We considered adjusting for group differences in analyses evaluating associations between each demographic characteristic and RIR preference because our groups were not well-matched demographically. However, since the objective of this study was to understand the preferences of both SCPs and non-SCPs for an effective approach for RIR of genomic addiction results to adolescents, and since very few variables pertaining to RIR differed by group, we determined that more complex models were unnecessary.

We assessed associations of demographic characteristics (age, gender, ethnicity, and highest grade completed) with RIR variables across groups with chi-square tests and independent t-tests. We dichotomized the RIR preference variable into those wanting no RIR and combined those wanting RIR with those wanting RIR *only* if there is a treatment. We then evaluated the adjusted effects of group and demographic characteristics in a multiple logistic regression model.

RESULTS

SCPs and non-SCPs were similar in age but differed significantly in sex, ethnicity and highest grade completed. See Table 1.

Survey Results

RIR was examined across adolescents. RIR was not related to age, gender, ethnicity, or highest grade completed. SCPs and non-SCPs did not differ in their preference for three RIR options ($\chi^2(2)=1.86$, p =NS). Most SCPs (77.8%) and non-SCPs (72.5%) wanted RIR if researchers find out something about DNA that has health or behavioral implications. An additional 16.3% of SCPs and 22.0% of non-SCPs wanted RIR that had health or behavioral implications only A multiple logistic regression determined that adjusted effects of age, gender, ethnicity, highest grade completed and group all remained nonsignificant when combined in a single model to predict those who want RIR versus those who do not.

The majority of SCPs (67.2%) and non-SCPs (69.7%) indicated that phone re-contact for RIR was “best”, but they also considered email (22.5% SCPs, 33.9% non-SCPs) and social networking websites (21.3% SCPs, 20.2% non-SCPs) to be viable options. Because participants could respond favorably to multiple methods of re-contact, no further analyses were done to determine if one method was significantly preferred to others. See Table 2.

LIMITATIONS

This study is potentially limited in that the number of SCPs outnumbered non-SCPs by nearly three to one.

CONCLUSION

The majority of SCPs and non-SCPs alike endorse RIR for findings that have health or behavioral implications. The perspectives of these adolescents stand in contrast to current research regulations, which do not require researchers to RIR with health or behavioral implications. Typically, consent forms state that individual results will not be returned (Ries et al., 2010; Shalowitz and Miller, 2008). The standard practice is to publish non-identifiable aggregate results of a research study in scientific journals and/or websites (Fabsitz et al., 2010).

Most SCPs and non-SCPs alike preferred re-contact for RIR via phone. However, exclusively relying on phone re-contact can be difficult as participants change numbers and may be difficult to locate. These results suggest a layered approach: a phone call followed by an email and a secure message to a social networking account.

The views of this special and vulnerable population, which include youths involved in the criminal justice system and substantial minority participation, bring an essential and missing perspective to the discussion of RIR. This study extends the limited research on RIR to actual adolescent participants who provided their DNA for genomic addiction research.

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Table 1

Demographic Characteristics by Group

	SCPs (n=320)	**Non-SCPs (n=109)**	**Test**	**p-value**
Mean Age in Years (SD=standard deviation)	16.0 (SD=1.1)	16.1 (SD=1.4)	t(159.7) = 0.4	NS
Sex			$c^2(1) = 29.7$	< 0.0005
Male	80.0% (n=256)	53.2% (n=58)		
Female	20.0% (n=64)	46.8% (n=51)		
Ethnicity			$c^2(2) = 13.1$	0.001
White	52.7% (n=169)	58.7% (n=64)		
Hispanic	21.3% (n=68)	6. 4% (n=7)		
Other	26.0% (n=83)	34.9% (n=38)		
Mean Highest Grade (SD)	9.4 (SD=1.2)	9.9 (SD=1.4)	t(427) = 4.0	< 0.0005

Table 2

RIR Preferences by Group

RIR Survey	**SCPs (n=320)**	**Non-SCPs (n=109)**
RIR Preferences:		
1. RIR if researchers find out something about DNA with health/behavioral implications	77.8% (n=249)	72.5% (n=79)
2. RIR only if there is an intervention or treatment	16.3% (n=52)	22.0% (n=24)
3. Do not contact	5.9% (n=19)	5.5% (n=6)
Indicated “Best” Method of Re-Contact:		
Phone	67.2%	69.7%
E-mail	22.5%	33.9%
Social Networking Website	21.3%	20.2%
Letter	13.8%	12.8%
Postcard	7.5%	1.8%
Newsletter	6.9%	2.8%
Post on Research Website	4.4%	3.7%