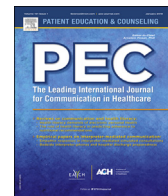


Contents lists available at ScienceDirect

Patient Education and Counseling

journal homepage: www.elsevier.com/locate/pateducou

Animation or leaflet: Does it make a difference when educating young people about genome sequencing?

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ARTICLE INFO

Article history:

Received 8 June 2020

Received in revised form 22 February 2021

Accepted 26 February 2021

Keywords:

Genome sequencing

Education

Young people

Animation

Leaflet

ABSTRACT

Objective: To compare the effectiveness of an animation against two leaflets with and without images, in educating young people about genome sequencing (GS).

Methods: An experimental survey with three assessment points (pre-intervention [T1], post-intervention [T2], 6-week follow-up [T3]). Participants (N = 606) were randomly assigned to receive one of three educational interventions; animation (n = 212); leaflet with images (n = 197); or leaflet with text only (n = 197). Measures of objective and subjective knowledge were completed at T1 (N = 606), T2 (N = 606) and T3 (N = 459). Measures of attitudes, intentions and beliefs towards GS and satisfaction with intervention were completed at T2 only.

Results: The type of educational intervention young people received had no significant impact on their objective or subjective knowledge at both T2 and T3 (all $p > .05$), nor did the educational intervention type affect their attitudes, intentions and beliefs towards GS at T2 ($p > .05$). However, participant satisfaction was significantly higher in the animation group than the leaflet groups ($p < .001$).

Conclusion: Animations and leaflets are both effective ways to deliver genomic education to young people, but the animations lead to higher satisfaction.

Practice implications: Different individuals may find different modes of educational resources more accessible than others. Therefore a range of resources should ideally be made available to patients.

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1. Introduction

Fifty percent of rare diseases affect children, 30% of whom will die before age 5 years [1] and in most cases there is an underlying genetic cause [2]. Genome sequencing (GS), whereby the entire DNA sequence of a person's genome is analysed, is set to have a profound impact for diagnosis of rare diseases in children through improved rates of diagnosis, more accurate prognosis

and provision of better management, surveillance and support [3–5]. GS is increasingly being implemented into mainstream medical practice [5]; in the United Kingdom (UK), GS for some rare diseases (as well as certain cancers) will soon be offered routinely to patients who do not have a diagnosis and for whom a genetic diagnosis will affect the healthcare of a patient or their family members, as part of the new NHS Genomic Medicine Service [6], and healthcare providers who treat genetic and non-genetic disease will be tasked with conducting pre-test counselling. This will include providing information about the benefits, limitations and uncertainties associated with the procedure in a way that enables patients to make informed decisions about testing [7].

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Online educational resources such as animations are increasingly being used in clinical practice to support the patient–clinician interaction, particularly when those consultations involve young people [8,9]. Young people are often referred to as ‘digital natives’ due to the omnipresence of technology in their everyday lives [10]. In 2016, 98% of children and adolescents in the UK had access to the internet, 83% of 12- to 15-year-olds had their own smartphone and 55% had their own tablet [11].

Animations have been shown to be effective for educating children about medical procedures and tests [12–14]. Studies have shown that, particularly in biology, students who learn with animations compared to traditional lectures obtain significantly higher marks [15], and that animations are more effective than static picture-based materials [16]. In contrast, written information can be hindered by young people’s reading skills [17].

Previous research has investigated the impact multimedia resources such as animations can have on patient’s knowledge, prior to undergoing medical procedures. A review of the literature on the use of multimedia educational aids, highlighted how such resources could enhance the way patients and caregivers receive and understand important clinical information [18]. Jeste and colleagues identified 37 randomised controlled trials (RCTs) that compared the effects of multimedia educational aids (video- or computer-based) with those of routine procedures, such as text-based information (leaflets, written documents), to inform patients about medical evaluations or management. Twenty-seven RCT studies reported that multimedia educational aids produced better understanding of information compared to routine methods. Whilst this further adds to the literature supporting the use of multimedia educational resources, the authors also argued that patients with a lower baseline knowledge benefited more from multimedia educational aids than patients with a higher baseline knowledge, suggesting that a ceiling effect for these aids may be possible. The authors also further emphasised that such educational aids should not replace the doctor–patient interaction.

Between 2016 and 2018, our research group developed two animations about GS, each around 2.5 min in length. The animations are aimed at young people including those who may be having GS in the context of rare disease diagnosis, but also young people who do not have a rare disease but who may be learning about GS at school. They were co-designed with multiple stakeholders including school pupils, and young people having GS through the 100,000 Genomes Project [19]. The animations were targeted at and co-developed with young people aged 11–15 years old. We chose this age group as in the 100,000 Genomes Project, 11–15 year olds were encouraged to be active participants in the decision-making process and sign an ‘assent’ form if they wanted to take part [20], and it is likely that this approach will be adopted into clinical practice. In addition, a gap exists in terms of information resources about GS that have been developed specifically for this age group [19].

The first animation explains the genome, genomic variation and GS (“My Genome Sequence”: <http://bit.ly/mygenomesequence>), the second focuses on the limitations and uncertainties of GS (“My Genome Sequence part 2”: <http://bit.ly/mygenomesequence2>). Our primary aim was to develop an educational resource that explained GS for young people accessing diagnostic testing for rare diseases, however, we were also keen that it would be a useful learning resource for young people more generally.

The animations were previously tested with 554 school pupils aged 11–15 in the UK [18]. Using our recently developed measure of knowledge of genome sequencing for young people [21], we found that the mean objective knowledge scores increased significantly after watching the animations for all 10 knowledge

items ($p < 0.001$) and we found that self-rated understanding of the terms gene, genome and GS also increased significantly ($p < 0.001$) [19]. We also found that after watching the animations the majority of participants felt they understood the benefits and limitations of GS (77% and 80% respectively), and there was high overall satisfaction with the animation with the majority of pupils finding the animation very or quite easy to understand (91%) and reporting that it had the right amount of information (81%) [19].

In this current study, our aim was to assess whether the mode of delivery of information impacted young people’s knowledge and attitudes, intentions and beliefs towards GS and satisfaction with information received. The modes of delivery we wanted to compare were the animations compared with the text from the animation delivered as an electronic leaflet with and without images, as previous research yielded encouraging results when comparing the effectiveness of using animation based educational interventions versus information delivered in a traditional static format [18]. Our primary research questions were:

1. Which mode of delivery has the greatest impact on *learning and retention* for objective and self-rated knowledge?
2. Which mode of delivery has the greatest impact on participants’ *attitudes, intentions and beliefs* towards GS?
3. Which mode of delivery has the greatest impact on participants’ *satisfaction* with information received?

Our secondary research questions were:

4. Do participants show significant improvement to, and retention of, objective and subjective knowledge irrespective of mode of delivery?
5. Is there an association between parent and/or participant characteristics on objective knowledge scores?

2. Methods

Ethical approval for the study was granted by West Midlands Black Country Research Ethics Committee (15/WM/0258).

2.1. Participants

An anonymous, online survey (Supplementary Fig. 1) was administered to young people from the UK aged 11–15 years in July 2019. Participants were recruited through the online market research company Panelbase (panelbase.net). Online surveys with children <16 years of age are subject to parental consent. The survey was hosted through the online survey platform Survey Monkey Inc (San Mateo, California, USA).

The Panelbase invitation was randomly sent to members who were registered as having children aged 11–15 years old. The invite described the topic of the survey (‘Genomics’), approximate length (10 min) and the reward for completing it (£1.25). Those that were interested in taking part clicked a link to redirect them to the survey. At the start of the survey, parents were provided with an information sheet about the study. Those that were willing to provide consent for their child to take part were asked to tick a box indicating their consent and were then asked a set of demographic questions (age, ethnicity, household income and education). The young person was then provided with a participant information sheet and if they were willing to take part, asked to tick a box indicating their consent. They were then asked demographic questions (sex, age, school year) and the survey began on the following page.

2.2. Study design

An experimental survey design with three assessment points (pre, post, and follow-up) was implemented, with the follow-up assessment being completed six weeks after completing the intervention. It was important to introduce a time lag to mimic the period between testing and return of results, although we acknowledge that in practice it may take longer than six weeks. The six-week period was chosen as a pragmatic compromise, in discussion with Panelbase, to prevent a large drop-out at follow-up.

2.3. Procedure

At the pre-intervention stage (T1) participants completed questions to measure subjective and objective knowledge. Participants were then automatically randomly assigned to receive one of three interventions through Survey Monkey, either the two animations - from here on referred to as 'the animation'; an electronic 2-page PDF leaflet which included the script and images used in the animation (Supplementary Figs. 2a and b) - from here on referred to as 'the leaflet', or an electronic 2-page PDF leaflet which included the script used in the animation but no images (Supplementary Figs. 3a and b) - from here on referred to as 'the text'.

Immediately after receiving the intervention, participants completed a survey (T2) which included a repeat of the questions measuring subjective and objective knowledge, as well as questions to explore attitudes, intentions and beliefs towards GS, and questions to explore satisfaction with the intervention. The order of objective knowledge questions was randomised to minimise the potential for order bias. At the end of the survey, responders were invited to leave free-text comments about what they thought of the information they were given.

Six weeks after participants had completed the T2 survey, parents of the children who had completed the survey, were sent a link to the T3 follow-up assessment. The survey only included the subjective and objective knowledge questions (order not randomised this time).

2.4. Development of measures

An extensive description of the development of the survey including the measures used is provided in our previous paper [19]. Briefly, *self-rated knowledge* was assessed using composite measures for (1) awareness of genetic terms (five sub-items: DNA, gene, chromosome, genome, and GS) and (2) understanding of genetic terms (same 5 sub-items) as well as a measure for understanding of genetics (3-point scale). *Objective knowledge* was assessed using a new 10-item kids-KOGS scale [21] which includes a series of statements about GS with responders asked to indicate whether the statement is true, false or don't know. The measure is intended to capture 'gist' comprehension of information rather than verbatim recall of language in the intervention. *Attitudes, intentions and beliefs* was the overarching term used to describe: intentions to undergo testing (if they had a health problem and the doctor suggested GS, they would want to have GS), perceived understanding of benefits and limitations of GS, whether the decision to undergo GS would be easy to make, and attitudes towards testing (e.g. good/bad etc). These were measured using seven questions to examine whether responders felt that they understood the benefits and limitations of GS. For each question, three answer options were available (e.g., agree, disagree, not sure). *Satisfaction* with information received was assessed using six questions focusing on how easy the information was to understand, amount, length, look and impact. Each question had

multiple choice answer points e.g. too much, too little, the right amount.

2.5. Sample size

Power analyses conducted using G* Power, suggested that under the parameters of small effect size (.25), and with a power of .95, a minimum of 158 participants across each mode of delivery (n = 56 in each mode) was required to draw meaningful conclusions when assessing for meaningful change in objective and subjective knowledge over time between modes of delivery. We aimed to collect 600 completed survey responses in total (approximately 200 per educational mode) as this was feasible with the available budget and would allow for comparisons across participant characteristics.

2.6. Pilot and main survey

To pilot the administration processes, a 'soft launch' was conducted whereby a random selection of 200 Panelbase members were invited to take part in the survey; 38 members aged 11–15 years responded of which 32 completed the survey (16% response rate). The average time taken to complete the survey was 12 min 44 s. No changes were made to the survey. Following the pilot, an invitation to take part in the main survey was administered in batches of between 200 and 500 and sent out every day or every other day by Panelbase. Random assignment to all three experimental conditions was active until participants had completed the study.

2.7. Statistical analysis

Chi-Squared analysis was used to assess if there were significant differences between the three experimental groups in participant characteristics. To determine if mode of delivery had a significant impact on objective knowledge and subjective knowledge (self-rated awareness and understanding of genetic terms) at immediate and 6-week follow-up, as well as self-rated understanding of genetics, a series of repeated-measures ANOVA (RMANOVA) were performed across all three time points.

To assess if there was a significant improvement to, and retention of, objective and subjective knowledge irrespective of intervention type, paired-samples t-tests were performed.

Scores derived from the items used to assess objective knowledge and subjective knowledge (self-rated awareness and self-rated understanding) were summed for the analysis. For objective knowledge, scores were calculated as: 0 = incorrect, 1 = correct. For subjective knowledge, scores were calculated for the questions 'have you heard of these words' and 'do you know what these words mean' as 0 = no, 1 = yes, and for the question 'how would you describe your understanding of genetics' scores were calculated as 1 = none, 2 = some and 3 = good.

Associations between demographic information and objective knowledge scores were analysed using Spearman's Correlation Analyses. Changes in knowledge in relation to demographic information was assessed using Kruskal-Wallis analysis.

2.8. Missing data

Surveys with missing data in the objective knowledge scale at T1 and/or T2 were excluded. Surveys that took less than five minutes to complete were also excluded as this indicated participants hadn't engaged with the intervention - both CL and JH attempted to complete the survey as quickly as possible across all educational modes, and were unable to complete it in under five minutes.

2.9. Content analysis of free-text comments

Participants' free-text comments about the information they received were coded by JH into the themes 'positive', 'negative' or 'mixed' and quantified using a directed content analysis approach [22]. These were then reviewed by a second researcher (CL) and any disagreements discussed.

2.10. Response rate

In total, 3090 invitations were sent out by Panelbase; 2240 didn't click the Panelbase link; 93 participants started the survey but did not continue to the intervention, 120 were excluded as less than five minutes was spent completing the survey (of which 15 received the animation, 46 the text and 59 the leaflet), 17 were excluded due to missing data, 14 were excluded as they did not fit the target age. This left 606 included responses. (19.6% response rate).

Table 1
 Participant characteristics.

Parents	Total (n = 606)	Text (n = 197)	Leaflet (n = 197)	Animation (n = 212)	Significance
Sex					
Female	439 (72.4%)	145 (73.6%)	143 (72.6%)	151 (71.2%)	$\chi^2(2) = .31; p = .86$
Male	166 (27.4%)	52 (26.4%)	53 (26.9%)	61 (28.8%)	
Prefer not to say	1 (0.2%)	0	1 (0.5%)	0	
Age:					
20–30	31 (5.1%)	9 (4.6%)	9 (4.6%)	13 (6.1%)	$\chi^2(8) = 5.18; p = .74$
31–40	223 (36.8%)	70 (35.5%)	79 (40.1%)	74 (34.9%)	
41–50	248 (40.9%)	76 (38.6%)	84 (42.6%)	88 (41.5%)	
51–60	73 (12%)	30 (15.2%)	20 (10.2%)	23 (10.9%)	
61+	4 (0.7%)	1 (0.5%)	2 (1.0%)	1 (0.5%)	
Missing data*	27 (4.5%)	11 (5.6%)	3 (1.5%)	13 (6.1%)	
Ethnicity					
White or White British	545 (89.9%)	177 (89.8%)	183 (92.9%)	185 (87.3%)	$\chi^2(8) = 7.89; p = .45$
Black or Black British	7 (1.2%)	3 (1.5%)	2 (1%)	2 (0.9%)	
Asian or Asian British	32 (5.3%)	8 (4.1%)	8 (4.1%)	16 (7.5%)	
Mixed	16 (2.6%)	5 (2.5%)	3 (1.5%)	8 (3.8%)	
Other	3 (0.5%)	2 (1%)	0 (0.0%)	1 (0.5%)	
Prefer not to say	3 (0.5%)	2 (1%)	1 (0.5%)	0 (0.0%)	
Education Level					
No formal qualifications	30 (5%)	9 (4.6%)	7 (3.6%)	14 (6.6%)	$\chi^2(8) = 4.26; p = .83$
GCSE or equivalent	157 (25.9%)	46 (23.4%)	57 (28.9%)	54 (25.5%)	
A-Level or equivalent	179 (29.5%)	64 (32.5%)	56 (28.4%)	59 (27.8%)	
Degree or equivalent	167 (27.5%)	54 (27.4%)	53 (26.8%)	60 (28.3%)	
Postgraduate qualification	70 (11.6%)	24 (12.2%)	22 (11.2%)	24 (11.3%)	
Prefer not to say	3 (0.5%)	0 (0.0%)	2 (1%)	1 (0.5%)	
Total household income					
Less than £20,000	123 (20.3%)	41 (20.8%)	35 (17.8%)	47 (22.2%)	$\chi^2(10) = 8.72; p = .74$
£20,000 to £34,999	160 (26.4%)	53 (26.9%)	53 (26.9%)	54 (25.5%)	
£35,000 to £49,000	150 (24.8%)	50 (25.4%)	53 (26.9%)	47 (22.2%)	
£50,000–74,999	80 (13.2%)	26 (13.2%)	29 (14.7%)	25 (11.8%)	
£75,000 to £99,999	43 (7.1%)	9 (4.6%)	12 (6.1%)	22 (10.4%)	
Over £100,000	14 (2.3%)	6 (3.0%)	4 (2.0%)	4 (1.9%)	
Prefer not to say	36 (5.9%)	12 (6.1%)	11 (5.6%)	13 (6.1%)	
Children					
	Total (n = 606)	Text (n = 197)	Leaflet (n = 197)	Animation (n = 212)	
Gender					
Female	331 (54.6%)	109 (55.3%)	104 (52.8%)	118 (55.7%)	$\chi^2(2) = 0.40; p = .82$
Male	275 (45.4%)	88 (44.7%)	93 (47.2%)	94 (44.3%)	
Age mean = 12.96; SD=1.39					
11	115 (19%)	37 (18.8%)	30 (15.2%)	48 (22.6%)	$\chi^2(8) = 11.57; p=.17$
12	139 (22.9%)	38 (19.3%)	59 (29.9%)	42 (19.8%)	
13	125 (20.6%)	44 (22.3%)	34 (17.3%)	47 (22.2%)	
14	112 (18.5%)	40 (20.3%)	35 (17.8%)	37 (17.5%)	
15	115 (19.0%)	38 (19.3%)	39 (19.8%)	38 (17.9%)	
School year					
Year 6	101 (16.7%)	32 (16.2%)	23 (11.7%)	46 (21.7%)	$\chi^2(10) = 17.15; p = .071$
Year 7	125 (20.6%)	39 (19.8%)	49 (24.9%)	37 (17.5%)	
Year 8	119 (19.6%)	36 (18.3%)	39 (59.8%)	44 (20.8%)	
Year 9	122 (20.1%)	47 (23.9%)	37 (18.8%)	38 (17.9%)	
Year 10	101 (16.7%)	39 (13.3%)	36 (18.3%)	29 (13.7%)	
Year 11	38 (6.3%)	7 (3.6%)	13 (6.6%)	18 (8.5%)	

Six weeks after completing the survey, all 606 participants were invited to complete the subjective and objective knowledge questions only. In total, 470 participants clicked on the link, however, 11 participants opted out of the survey halfway. This left 459 included responses, with a response rate of 75.7%.

3. Results

3.1. Demographic characteristics

Demographic characteristics for both parents and young people are reported in Table 1. Furthermore, descriptive statistics of objective knowledge, self-rated awareness of genetic terms, understanding of genetic terms, and understanding of genetics at each time point is presented in Table 2. Overall, 54.6% of young person participants were female and the mean age was 12.9 years. Of their parents, nearly half (46.7%) had a total household income of less than £35,000, 60.4% had an educational level of A-level (or

Table 2
 Descriptive statistics of objective and subjective knowledge scores.

		n	Mean	Std. Dev	Median	Range
Time 1	Objective Knowledge	459	4.54	2.52	5	0 - 10
	SR Awareness of Genetic Terms	459	3.28	1.13	3	0 - 5
	SR Understanding of Genetic Terms	457	2.47	1.39	3	0 - 5
	SR Understanding of Genetics	459	1.86	0.50	2	1 - 3
Time 2	Objective Knowledge	459	7.21	2.25	8	0 - 10
	SR Awareness of Genetic Terms	459	4.29	1.11	5	0 - 5
	SR Understanding of Genetic Terms	457	4.01	1.38	5	0 - 5
	SR Understanding of Genetics	459	2.04	0.46	2	1 - 3
Time 3	Objective Knowledge	459	5.07	2.41	6	0 - 9
	SR Awareness of Genetic Terms	459	4.12	1.16	5	0 - 5
	SR Understanding of Genetic Terms	457	3.18	1.52	3	0 - 5
	SR Understanding of Genetics	459	1.96	0.46	2	1 - 3

Note: SR = self-reported.

Table 2 shows the mean, standard deviation, median, and range of observed scores for participant's objective and subjective (self-rated (SR) awareness of genetic terms and self-rated understanding of genetic terms) knowledge, as well as self-rated understanding of genetics at each time point.

equivalent) and below and most (89.9%) self-identified as White or White British ethnicity. These are in line with recent census data which show that in England and Wales 86% of the population identify as White and 67% have an educational level of 2 or more A-levels (or equivalent) and below [23].

3.2. Comparison of participants between intervention groups

A total of 606 participants (212 in the animation group, 197 in the leaflet group, and 197 in the text group) completed the T1 and T2 surveys. At T3, 459 participants completed the survey (167 in the animation group, 141 in the leaflet group, 151 in the text group). Results are displayed in Table 3 and Fig. 1 a–d. Power conditions for each analysis were met.

1. Which mode of delivery has the greatest impact on learning and retention for objective and self-rated knowledge?

There was no significant difference in learning and retention of objective knowledge ($F(2) = .400, p > .05$), self-rated awareness of genetic terms ($F(2) = .951, p > .05$), self-rated understanding of genetics ($F(2) = .185, p > .05$), or self-rated understanding of genetic terms ($F(2) = 1.023, p > .05$) based on the mode of delivery participants received (See Supplementary material Tables 1–4). Evaluation of how scoring for each item individually changed from Time 2 to Time 3 using a series of repeated measures ANOVA shows a significant reduction in all items.

2. Which mode of delivery has the greatest impact on participants' attitudes, intentions and beliefs towards GS?

A Kruskal-Wallis analysis was performed to assess for any difference in attitudes, intentions and beliefs towards GS based on the type of intervention received. However, no significant difference was observed ($p > .05$). See Table 3 for average scores at T2 across the three modes of delivery.

Table 3
 Average attitudes, intentions and beliefs scores across each mode of education.

Measure	Total (n = 606)	T2 animation (n = 212)	T2 leaflet (n = 197)	T2 text (n = 197)	Significance
Imagine you had a health problem and your doctor suggested genome sequencing to understand more about the cause of your condition. Would you want to have genome sequencing?					
Yes	368 (60.7%)	135 (63.7%)	121 (61.4%)	112 (56.9%)	$\chi^2 = 3.62, p = .72$
No	7 (1.2%)	1 (0.5%)	3 (1.5%)	3 (1.5%)	
Not sure	81 (13.4%)	25 (11.8%)	28 (14.2%)	28 (14.2%)	
I feel I understand the benefits of genome sequencing					
Agree	457 (75.4%)	162 (75.9%)	149 (75.6%)	146 (74.1%)	$\chi^2 = 4.02, p = .40$
Disagree	22 (3.6%)	4 (1.9%)	7 (3.6%)	1 (5.6%)	
Not sure	127 (21.0%)	46 (21.7%)	41 (20.7%)	40 (20.3%)	
I feel I understand the limitations of genome sequencing (what it can't do)					
Agree	330 (54.5%)	121 (57.1%)	110 (55.8%)	99 (50.3%)	$\chi^2 = 2.49, p = .65$
Disagree	51 (8.4%)	15 (7.1%)	17 (8.6%)	19 (9.6%)	
Not sure	225 (37.1%)	76 (35.8%)	70 (35.5%)	79 (40.1%)	
I feel the decision to have / not have genome sequencing would be easy for me to make					
Agree	340 (56.1%)	125 (59.0%)	109 (55.3%)	106 (53.8%)	$\chi^2 = 4.43, p = .35$
Disagree	72 (11.9%)	18 (8.5%)	29 (14.7%)	25 (12.7%)	
Not sure	194 (32%)	69 (32.5%)	59 (29.9%)	66 (33.5%)	
Genome sequencing is:					
A bad thing	9 (1.5%)	2 (0.9%)	3 (1.5%)	4 (2.0%)	$\chi^2 = 2.24, p = .69$
A good thing	562 (92.7%)	188 (88.7%)	18 (85.3%)	174 (88.3%)	
Neither	35 (5.8%)	22 (10.4%)	26 (13.2%)	19 (9.6%)	
Genome sequencing is:					
Harmful	11 (1.8%)	4 (1.9%)	2 (1.0%)	3 (1.5%)	$\chi^2 = 2.32, p = .68$
Helpful	523 (86.3%)	198 (93.4%)	180 (91.4%)	184 (93.4%)	
Neither	72 (11.9%)	10 (4.7%)	15 (7.6%)	10 (5.1%)	
Genome sequencing is:					
Boring	11 (1.8%)	3 (1.4%)	5 (2.5%)	3 (1.5%)	$\chi^2 = 1.86, p = .76$
Interesting	523 (86.3%)	180 (84.9%)	170 (86.3%)	172 (87.3%)	
Neither	72 (11.9%)	29 (13.7%)	22 (11.2%)	21 (10.7%)	

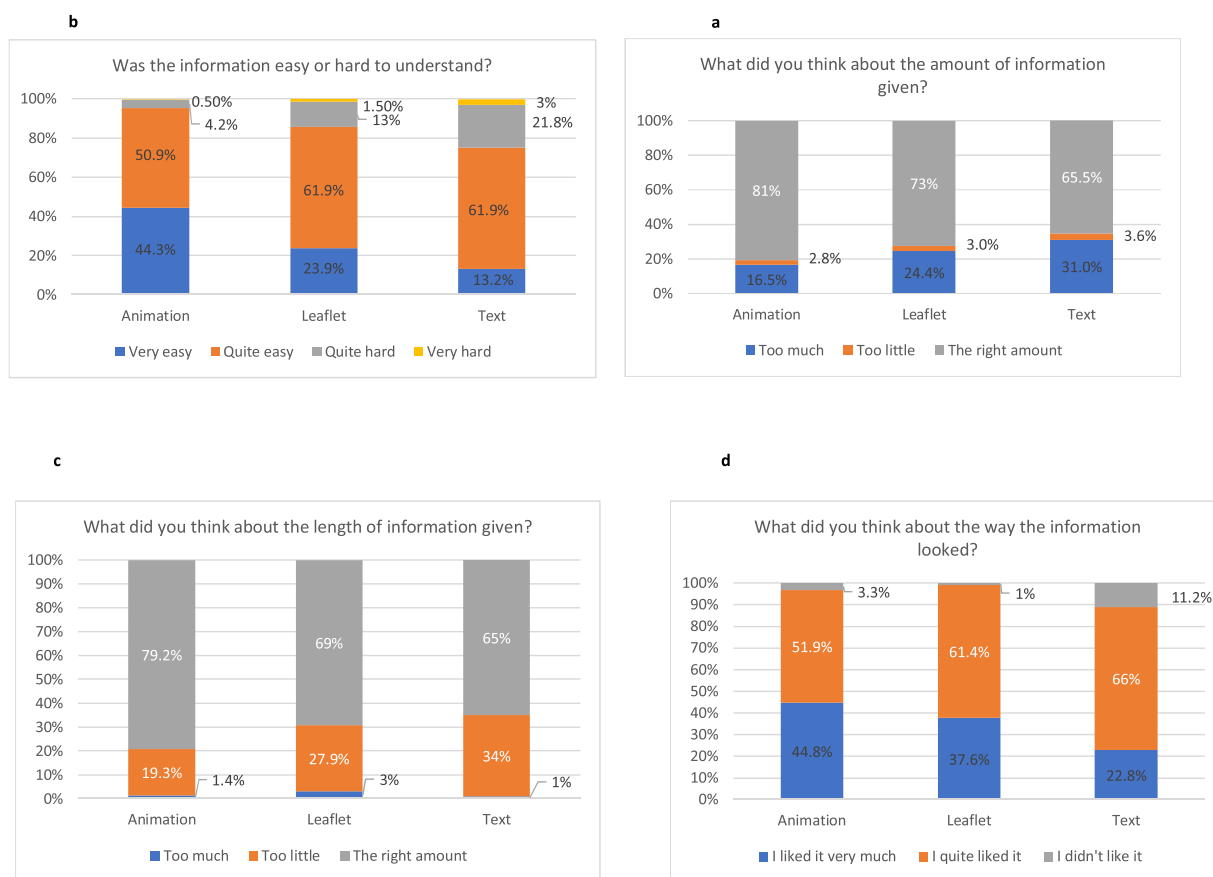


Fig. 1. a–d: Satisfaction and knowledge by mode of delivery during the T2 survey.

3. Which mode of delivery has the greatest impact on participants' satisfaction with information received?

An ANOVA was performed to assess whether the type of intervention received had a significant impact on participants' satisfaction. Results indicate that participants perceived the information was significantly easier to understand when they received the animations compared to the leaflet or text ($F(2) = 14.989$; $p < .001$; partial $\eta^2 = .109$) (Fig. 1a). They perceived the amount ($F(2) = 6.306$; $p = .002$; partial $\eta^2 = .020$) (Fig. 1b) and length ($F(2) = 4.717$; $p = .009$; partial $\eta^2 = .015$) (Fig. 1c) of information to be preferable when received as an animation, and similarly had a greater appreciation for how the information looked ($F(2) = 16.246$; $p < .001$; partial $\eta^2 = .054$) (Fig. 1d) when it was received as an animation. However, no significant difference was found regarding how much participants felt they had learned ($F(2) = .052$; $p > .05$; partial $\eta^2 < .001$) or how useful the information was found to be ($F(2) = 1.722$; $p > .05$; partial $\eta^2 = .006$). See Fig. 1a–d for average 'satisfaction' scores for each intervention, and Supplementary file Table 7 for a breakdown of 'satisfaction' scores for each intervention.

In total, 134 participants (22%) left free text comments of which 45 were about the animation, 45 about the leaflet, and 44 were about the text. The comments were coded as positive or negative. Free text was not coded if participants indicated that they had no comment to make about the intervention. The animation received 26 positive comments including that it was "fun and interesting" and "kept my attention". There were four negative comments, including that the animation was "a bit long", "hard to understand" that it "went over the same thing over and over again" and for one participant, that they would "need to look up the bigger meaning to

give me a good understanding to hold conversation". The leaflet received 24 positive comments. It was found to be "useful", was written in a way "that children could understand" and participants "enjoyed the pictures". Six of the eight negative comments related to the amount of information in the leaflet with one participant reflecting that "I had to read it a few times because there was a lot of info[rmation]." The text received 24 positive comments with participants describing the information as "interesting" and "good and detailed". There were seven negative comments which focused on the aesthetic appearance of the information, for example "some pictures or diagrams would have helped me to understand this better".

4. Do participants show significant improvement to, and retention of, objective and subjective knowledge irrespective of mode of delivery?

Results from the paired samples t-tests show a significant increase in both objective and subjective knowledge from T1 to T2 ($p < .001$), followed by a significant decrease in both objective and subjective knowledge ($p < .001$) between T2 and T3, with a significance of $p < .001$ observed across all educational modes. Nonetheless, the T3 objective and subjective knowledge scores remained significantly higher than scores gained at T1, with all differences significant at the $p < .05$ level (See Supplementary material Tables 5 and 6).

5. Is there an association between parent and/or participant characteristics and objective knowledge scores?

At T1 (pre intervention): There was a significant positive association between the young person's objective knowledge score

and parental education level ($r = .14$, $p < .01$), participant age ($r = .18$, $p = .01$) and school year ($r = .18$, $p < .01$). All other variables (parental ethnicity, parental income, and young person's sex) were not significant ($p > 0.05$). In multivariable analysis, both parental education level and young person's age remained associated with knowledge (the variable school year was excluded because it was highly correlated and deemed too similar to child age [24] ($r = 0.88$; $p = 1.99^{-198}$). We chose to exclude school year as age is more interpretable.

At T2 (post-intervention): There was a significant positive association between knowledge and parental education level ($r = .14$, $p < .01$) and participant age ($r = .10$, $p = .01$). All other variables (parental ethnicity, parental income, school years and participant sex) were not significant ($p > 0.05$). In multivariable analysis, none of the variables remained significant ($p > 0.05$).

Change score: There was a significant association between knowledge and participant age ($H(4) = 19.782$; $p = 0.000551$) and school year ($H(5) = 19.395$; $p = 0.001623$). The greatest change score was for 11 year olds (mean = 3.29, SD = 2.70) followed by 12 year olds (mean = 2.95, SD = 2.71), 13 year olds (mean = 2.79, SD = 2.43), 15 year olds (mean = 2.37, SD = 2.39) and 14 year olds (mean = 2.37, SD = 2.49).

4. Discussion and conclusion

4.1. Discussion

Objective and subjective knowledge of GS increased significantly after receiving an educational intervention, irrespective of the mode of delivery. These results indicate that the script, in whatever format it is delivered is effective. This is important as some families may not have access to the internet. Additionally, some clinics may not have the capacity to show an animation during the clinic appointment and may only be able to distribute written information to patients and their families.

Unlike previous findings whereby animations were found to be more effective than static educational materials [18], we did not find any significant differences according to which intervention participants received other than for the questions assessing satisfaction.

However, similar studies to our which have focused on GS education have been identified elsewhere. Sanderson et al. did not find any differences in adult's knowledge of GS when comparing those who received an animation with those who received a leaflet. However, they did find that the animation scored more highly on many of the satisfaction questions [25]. In a separate study conducted with adolescents but using the same animation as in the Sanderson et al. study, participants' satisfaction with information was higher for those who received the animation even though mode of delivery (animation v leaflet) did not affect overall objective or self-rated knowledge [14]. These results raise an important question. How important is patient satisfaction when weighed against the cost of developing an animation, which can be many thousands of pounds, compared to a written leaflet which can be produced much more cheaply? Another pertinent question is around likely uptake. Are young people as likely to read a leaflet, for example if it is included alongside an appointment letter, compared to watching an animation if the link to that animation was included in the letter? Notably, of those participants who were excluded from the analysis as they spent less than five minutes completing the survey, the majority had been randomly assigned to receive written information (46 received the text and 59 received the leaflet) compared to the animation ($n = 15$). This suggests that young people may be more likely to engage with an animation than a written document. However, we must consider that different individuals may find different modes more accessible than others. For example, families with less reliable Internet

service may favour static educational materials whereas families from limited English-speaking households may favour multimedia materials. Factors such as a learning disability were also not considered, which could undoubtedly play a role in understanding.

Objective knowledge decreased significantly 6 weeks after it was received. This is not unexpected; previous research has demonstrated that newly acquired information is vulnerable and is easily forgotten [26]. Our results highlight a salient point regarding young people's knowledge and understanding immediately after receiving information, and what information they retain on a longer-term basis. Assuming patients were to receive the information, in whichever mode, at the time of their appointment, our results suggest that they will have a better understanding of GS and its limitations and uncertainties at the time of testing than several weeks later when they receive the GS result by which time some information may have been forgotten. One potential solution to address this would be for the animation or leaflet to be watched or read again at the time results are returned. Another solution could be to allow for the animation to be watched at home, after the appointment, so that patients and their families could refer back to animation at a later date. Finally, mode of delivery of the intervention appeared to level the impact of demographic surroundings on knowledge as multivariate analysis at T2 did not find the same advantages for older children and those whose parents had higher educational attainment.

Key strengths of this study include the rigorous objective knowledge measure, the multiple knowledge measures employed, the fact that the study team assessed participant satisfaction, the sample size and the low drop-out rate between T2 and T3. Collecting qualitative data from participants allowed us to gain more in-depth insights about the different modes of education used in the intervention that would not have been captured through quantitative data. The mixed-methods approach used in this study provides a richer and more nuanced picture of how the interventions were received.

There are, however, some limitations to this study. Prior to the intervention, participants were advised to answer the questions on their own and without support from their parents. However, we cannot guarantee that this happened in practice. Additionally, although we excluded data from participants who took less than 5 min to complete the intervention, we are unable to confirm whether the remaining participants fully engaged with the different modes of intervention they received. For example, we don't know whether those assigned the animation watched the animations from start to finish, or clicked through to the second animation before finishing the first. We did not ask participants to complete the attitudes, intentions and beliefs questions prior to receiving the intervention; this would have enabled us to conduct a within-subject comparison and would have provided stronger evidence for how each mode of delivery influenced each of these concepts.

The sample was relatively homogenous (primarily White) and the attitude questions included relatively simple response scales – as such we were unable to gather a nuanced understanding of young people's attitudes, intentions and beliefs from this survey. Our group have recently explored the attitudes and decision-making experiences of young rare disease patients undergoing GS [27], however, further work with young people more generally would be valuable. Finally, we do not know what proportion of the participants in this study were themselves, or knew someone in their family or friendship group, affected by a genetic condition. Literature has indicated that prior experience of a subject can have an impact on how new information about that subject is encoded and remembered [28,29]. Additionally, previous research has suggested that baseline knowledge may affect how beneficial multimedia aids, such as animations, may be for patients [18]. The perceived relevance of information may also contribute to knowledge retention [30]. Thus, our results might be different if

this study were to be replicated with rare disease patients in clinic for whom the information may be more relevant. This is an important area for future research.

4.2. Conclusion

This study highlights that both animations and written leaflets are effective methods of delivering genomic education to young people, however, satisfaction appears to be higher when it is delivered in an animation format, and there is some indication that young people may engage more with this format. To date, the educational resources have not been formally tested with young people with rare diseases making real life decisions about GS. Future research could look at the effectiveness of the interventions with young people who may be eligible to undergo GS through the NHS Genomic Medicine Service. This could follow a similar study design whereby patients are randomly assigned to receive either the animation or leaflet, and complete a before and after survey including measures of knowledge, attitude, satisfaction as well as uptake of GS. As GS for rare diseases becomes more widely used, our animations, which are freely available in a number of foreign languages including Bengali, Chinese and Turkish, (<https://www.gosh.nhs.uk/medical-information/clinical-specialties/clinical-genetics-information-parents-and-visitors/support-and-information>) provide an effective resource to support health professionals discussing this new procedure with young people and their parents.

Practice implications

Different individuals may find different modes of educational resources more accessible than others. Therefore, a range of resources should ideally be made available to patients.

Funding

The animations were funded through a grant (V0416) from the Great Ormond Street Hospital Children's Charity. CL is funded by a Fellowship Award from Health Education England Genomics Programme. MH is partially funded by the NIHR BRC at Great Ormond Street Hospital.

CRediT authorship contribution statement

Jennifer Hammond: Investigation, Formal analysis, Writing - original draft, Writing - review & editing. **Ian Garner:** Formal analysis, Writing - original draft, Writing - review & editing. **Melissa Hill:** Writing - review & editing. **Christine Patch:** Writing - review & editing. **Amy Hunter:** Writing - review & editing. **Beverly Searle:** Writing - review & editing. **Saskia C. Sanderson:** Writing - review & editing. **Celine Lewis:** Conceptualization, Methodology, Investigation, Funding acquisition, Formal analysis, Writing - original draft, Writing - review & editing.

Declaration of Competing Interest

CP has been on a secondment with Genomics England as Clinical Lead for Genetic Counselling since October 2017. The other authors declare no conflicts of interest.

Acknowledgements

The animations were funded through a grant (V0416) from the Great Ormond Street Hospital Children's Charity. All research at Great Ormond Street Hospital NHS Foundation Trust and UCL Great Ormond Street Institute of Child Health is made possible by the

NIHR Great Ormond Street Hospital Biomedical Research Centre (BRC). The views expressed in this publication are those of the author(s) and not necessarily those of the NHS, NIHR or HEE GEP.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:<https://doi.org/10.1016/j.pec.2021.02.048>.

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