5-2017

Evaluating FRAME (Faces Redefining the Art of Medical Education) videos: The impact of digital storytelling on medical students' attitudes to disability and their learning preferences

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May 8th, 2017

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ABSTRACT

*FRAME (**F**aces **R**edefining the **A**rt of **M**edical **E**ducation) is a web-based platform created by Positive Exposure, a non-profit organization. FRAME uses digital story telling (DST) by presenting short educational films and image galleries aimed to help healthcare professionals and students gain an understanding of genetic conditions and disability. Families demonstrate hallmark characteristics in the context of a story to make the video educational and true to life. The purpose of this study was to evaluate the effectiveness of FRAME videos for 22q11.2 deletion syndrome (22q11.2 DS) and familial dysautonomia (FD) in improving students' attitudes about disability. Fourth year medical students were recruited by email from three accredited U.S. medical schools. Participants were randomly prompted to read about one of two conditions on Medscape and then complete the Attitudes to Disability Scale (ADS), a validated 16-item survey. They watched the FRAME video on the same condition and completed the ADS again. Participants completed a survey to explore learning preferences and gather feedback on the videos. 48 out of 549 medical students participated in the study (FD n=27, 22q11.2 DS n=21). Significant attitude improvements were found in the Inclusion, Discrimination, Positive Gains and Current/Future Hopes subscales of the ADS for the 22q11.2 DS video and in the Positive Gains subscale for the FD video. The majority of participants preferred the FRAME video for long-term retention (FD 88%, 22q11.2 DS 86%). Most participants (FD 85.7%, 22q11.2 DS 86.4%) agreed or strongly agreed the FRAME video gave them insight into living with a disability. Forty-two students (87.5%) agreed or strongly agreed they learn best with audio, visual and text resources. The results suggest the FRAME videos improved medical students' attitudes related to multiple aspects of disability associated with 22q11.2 DS and a specific aspect of disability associated with FD. Our study shows DST is an effective way to improve medical students' attitudes towards individuals with genetic conditions and disabilities.*

KEYWORDS: *digital storytelling, medical education, Attitudes to Disability scale, genetic conditions*

INTRODUCTION

Patient-centered approaches to health care have been increasingly emphasized with the advent of the medical humanities, a field developed in the 1970s following the realization that medical education may not be adequate enough to teach understanding of humans (Macnaughton, Mbchb, & Drcog, 2015). Patient narratives are stories, literature, movies and other media forms that explore the experience of illness (Kumagai, Murphy, & Ross, 2009). The additional merits of patient narratives are stimulation of critical thinking (Cox, 2001), linkage between theory and practice (Koenig & Zorn, 2002) and development of empathy, cultural sensitivity and tolerance (Kirk et al., 2013)

Positive Exposure, founded in 1997, is a non-profit organization which utilizes photography and video to transform public perceptions of people living with genetic, physical, cognitive and behavioral differences. One component of Positive Exposure is the FRAME project (**F**aces **R**edefining the **A**rt of **M**edical **E**ducation). FRAME is a web-based platform that presents a series of short educational films and photographic galleries designed to help healthcare professionals and students gain an understanding of genetic conditions and disabilities while modeling an attitude of respect for the humanity of the patients (Guidotti, n.d.). Each condition's webpage also includes a separate slide show of the informational text and a link to a nationwide support group. The patients and their families introduce themselves to viewers and demonstrate hallmark characteristics in the context of a story making the video educational and true to life. Another intention of this format is for individuals with genetic conditions to form partnerships with the healthcare community and connect with audiences worldwide. The FRAME films represent a shift in medicine from a traditional curriculum to one in which there is an application of narrative medicine and a utilization of technology in the training of medical professionals.

The Association of American Medical Colleges (AAMC) created a Group of Diversity and Inclusion (GDI) to inform and guide the advancement of diversity and inclusion through academic medicine and the community (Poladian, 2013). There is a "Culture of Disability"

that is subject to stereotypes, prejudiced attitudes and institutional barriers (Eddy & Robey, 2005). Patients with chronic disabilities often perceive a lack of sensitivity among physicians during clinical encounters (Sabharwal, 2001). Moreover, the Office of Surgeon General reports people with disabilities cite a shortage of provider training about the daily realities of living with a disability (Brown, Graham, Richeson, Wu, & McDermott, 2010). Inadequate preparation of doctors may lead to negative perceptions of these patients such as they are time consuming, difficult to relate to, poorly insured and are medically and socially complex (Woodward, L., S.M., Zwygart, & Perkins, 2012).

These findings highlight the importance of disability related training at all levels of health care education and practice (Minihan, Robey, & Long-Bellil, 2011). There is a need to explore alternative strategies for providing students with insight into the lives of people with disabilities so they can be more comfortable providing medical care.

The AAMC has recognized genetics as one of the contemporary issues in medicine (Medical, Society, Genetics, & Genetics, 2008). The American Society of Human Genetics (ASHG) and the Association of Professors of Human and Medical Genetics (APHMG) developed a medical school competency based core curriculum to provide guidance regarding medical genetics knowledge, skills and behaviors all current medical students will need during their careers. (Medical et al., 2008)

Medical students are expected to accomplish a series of knowledge and attitudes based skills that assist in the diagnosis, prevention and treatment of genetic diseases. An underlying theme of the competencies includes the need for psychosocial skills because genetic conditions require family centered care and supportive counseling. Therefore, students must be comfortable maneuvering sensitive relationships in a collaborative manner (Medical et al., 2008). Additionally, genetics presents unique ethical issues in genetic testing, release of information and unique psychological issues. The ability to empathize with patients and understand the need for privacy is crucial to a successful doctor-patient relationship.

Many non-geneticist physicians have minimal direct professional exposure to genetic conditions and thus a limited understanding of its impact on the lives of patients

and their caregivers (Kirklin, 2003). One solution has been a website called *Telling Stories, Understanding Real Life Genetics* (www.tellingstories.nhs.uk) which features over 100 stories in text and video formats. These stories are sorted by 11 themes, which include professional competencies and learning outcomes, genetic conditions, inheritance pattern and genetic intervention. This website is unique because it links the story to a professional practice through a UK genetic-genomics education framework (Kirk et al., 2013).

The resource was evaluated by the network of nurse educators at the NHS National Genetics Education and Development Centre and two e-learning specialists (the delegates). The qualitative feedback was encouraging. Positive aspects of this resource included the use of stories to link theory and practice and that the stories are available in multiple formats. This study demonstrates the value in creating a web-based resource for healthcare professionals to use at their leisure. Additionally, the study highlights the merits in using patients to transmit knowledge (Kirk et al., 2013). The literature suggests the use of patient narrative may be a way to increase exposure to conditions for a range of health care professionals and students.

In this study, we addressed the following questions: 1) Does the FRAME approach to medical education improve students' attitudes so they can cultivate a sense of respect and compassion for individuals with disabilities and/or genetic conditions? 2) Do medical student's learning preferences favor traditional text-based methods or the FRAME approach of digital storytelling? In addition, we asked participants to provide feedback for the FRAME video to determine strengths and areas of improvement for future development of the platform.

MATERIALS AND METHODS

Ethical Considerations

The Sarah Lawrence College Institutional Review Board approved the study for expedited review (#00009775) on December 5th, 2016.
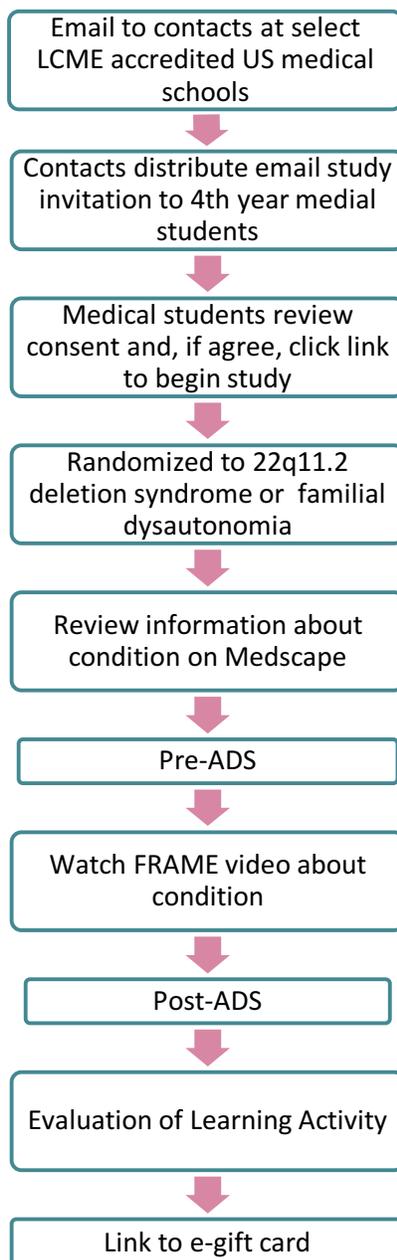
Sample

Fourth year medical students at select medical schools accredited by the Liaison

Committee on Medical Education (LCME) were eligible to participate. The participant pool consists of students from three different medical schools across two states, Iowa and Texas. These schools were selected because they are established contacts of Positive Exposure. Fourth year students were chosen since they have established learning preferences and have completed a majority of their clerkships in the primary specialties.

Methods (figure 1)

Figure 1: Summary of study activities

Email to contacts at select LCME accredited US medical schools

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Contacts distribute email study invitation to 4th year medial students

↓

Medical students review consent and, if agree, click link to begin study

↓

Randomized to 22q11.2 deletion syndrome or familial dysautonomia

↓

Review information about condition on Medscape

↓

Pre-ADS

↓

Watch FRAME video about condition

↓

Post-ADS

↓

Evaluation of Learning Activity

↓

Link to e-gift card

Recruitment began in December 2016 by contacting representatives at seven geographically diverse medical schools to request the distribution of email invitations to fourth year medical students. The recruitment email included a link to the consent and a description of the study. Three medical schools agreed to forward the recruitment email to the fourth year class (N = 549). The first recruitment email was forwarded in January 2017 and the participating medical schools did not send more than two reminders to the class after the initial recruitment email was forwarded. The surveys were active until the end of February 2017.

Medscape Education

After consenting, participants began the study activity by clicking on a link to a Medscape web page about 22q11.2 Deletion Syndrome (22q11.2 DS) or familial dysautonomia (FD). Participants were randomly assigned one of two conditions. The students did not have previous knowledge about which conditions were included in the study. They were prompted to take 5-10 minutes to read about the syndrome in Medscape as if they were preparing to

assist in the care of a patient with that condition the next day. They were given no guidance on which topics to specifically focus on during this study activity. The prompt was intentionally vague to replicate the individual process that medical students use to look up a disease in preparation for a patient encounter. Medscape is the most accessible point-of-care reference available to health care professionals on the internet. It is primarily text based with a few images of the hallmark clinical features.

Baseline scores Attitudes to Disabilities scale (ADS)

After reviewing the Medscape webpage on either FD or 22q11.2 DS, participants completed the Attitudes to Disability Scale (ADS). They were prompted to think about disability and people with either FD or 22q11.2 DS while completing the questions. For most of the questions in the scale, the phrase "people with disabilities" was replaced with "people with familial dysautonomia" or "people with 22q11.2 deletion syndrome" depending on which disease they read about in Medscape. The goal was to assess the medical students' attitudes towards disability only as it relates to the genetic condition they learned about on Medscape.

FRAME Video and post video ADS scores

After completing a baseline ADS, participants watched a FRAME video on the same disease they researched on Medscape. The content for FRAME videos are decided by a team of genetic counselors and medical professionals. There were two FRAME videos chosen for this study, one for 22q11.2 deletion syndrome and one for familial dysautonomia (FD). These genetic disorders were selected because affected individuals exhibit physical and intellectual disabilities. Additionally, to minimize prior knowledge of one of the conditions, the research team chose familial dysautonomia which is a rarer genetic disease. Each video was between 9-10 minutes long to match the time participants were instructed to spend on MEDSCAPE. At minimum, the videos include patients and family members who present information about genetic etiology, clinical findings, pathophysiology and general management of the condition. After watching the video, participants completed a second ADS survey.

Video Evaluation Survey

The final portion of the study included completing the video evaluation questions. Each participant was offered a $10 e-gift card for their participation.

Measures

Information was collected through a self-administered, voluntary and anonymous 46-question survey on Survey Monkey. Items in the survey were based a review of the literature, which identified methods to measure attitudes towards disabilities and to evaluate multimedia learning tools. Participants had the freedom to skip any questions.

1. The Attitudes to Disability Scale (ADS) general version (see Appendix A): This is a 16-item scale used for the assessment of attitudes to disability and quality of life across the adult lifespan. The ADS can be used to assess attitudes in physically and intellectually disabled groups. Compared with existing scales, the ADS includes more aspects, such as prospects and hopes. Additionally, this is the first scale to have cross-cultural validity along with directly drawing on attitudes and experiences of individuals living with disabilities (Power & Green, 2010). ADS used a 5 point Likert Scale for each item (1= strongly disagree and 5 = strongly agree) and focuses on four different aspects of disabilities (Power & Green, 2010)

   I.   Subscale 1: Issues of inclusion and exclusion and burden on families and society.

   II.   Subscale 2: Issues related to discrimination

   III. Subscale 3: Reflects positive gains to self and to others

   IV.   Subscale 4: Current and future hopes and prospects and whether or not disability has an impact on them

2. Video Evaluation Survey (see appendix B): This is a 9-item questionnaire developed by the research team. The purpose of this survey is to evaluate the FRAME video and to understand whether learning preferences favor Medscape or the FRAME approach of digital storytelling.

Sub scale 1: Retention of information and learning styles

Retention of the FRAME video was measured through two questions. The first used a Likert scale to assess likelihood of participants remembering the FRAME video. The second

question asked participants to pick which learning approach (FRAME or Medscape) allows for long term retention about the genetic condition and to explain why. Finally, to measure students' preferences for multimodal learning styles, participants filled out a Likert scale question for the statement: "I learn best when I am exposed to audio, visual and text resources".

Subscale 2: *Perspectives about disability and comfort levels*

The first question used a Likert scale to assess whether the FRAME video gave insight into a patient's perspective of living with a disability. An important goal of the FRAME video is help healthcare professionals improve their comfort level when treating patients with disabilities. Therefore, a Likert scale question prompts participants to select their level of agreement for the statement "I would feel comfortable being involved in the care of someone with (familial dysautonomia or 22q11.2 deletion syndrome)".

Subscale 3: Feedback for the FRAME videos

Participants completed two free response questions that prompted them to explain what they liked the most about the FRAME video and how they think the video can be improved. A yes/no question asks about whether participants would be interested in having access to additional FRAME videos about other disorders.

Data Analysis

Statistical Analyses

Both samples were analyzed using the paired sample t-test to assess change in the ADS scores pre and post FRAME video. For the video evaluation survey, data analysis for each sample included descriptive statistics such as univariate analysis and frequency distribution. All analyses were conducted using SPSS V.24 and the significance level was set at $p < 0.05$.

Qualitative Analyses

Thematic analysis was conducted on the free-response answers in the video evaluation survey. Our data-driven approach used open coding and axial coding to further evolve themes. An initial codebook was developed to capture key thoughts and concepts from the data. The category definitions were reviewed and revised multiple times. The results of the descriptive coding were organized into final themes to better understand participants' perspectives on the FRAME video as a medical education tool.

Results

A. Attitude changes Pre and Post Video

1. 22q11.2 Deletion Syndrome Group

Inclusion Subscale*:* Significant and favorable changes in attitude were found in three out of four items measuring inclusion/exclusion and burden (Table 1). A decrease in the "Mean Post Video" for each statement indicated more disagreement and thus, improved attitudes. No significant change was found for the statement "people with 22q11.1 are a burden on society".

Table 1: Attitude Change toward People with 22q11.2 Deletion Syndrome **(Inclusion)** Pre and Post Video (1 = strongly disagree, 5 = strongly agree)					
Inclusion Subscale	**Mean Pre Video (N=21)**	**Mean Post Video (N=21)**	**Mean Difference**	**t value**	**p level***
People with 22q11.2 find it harder to make new friends	3.85 (SD=.91)	3.05 (SD=1.16)	.80	3.30	p<.01
People with 22q11.2 have problems getting engaged in society	4.09 (SD=.76)	2.62 (SD=1.24)	<u>1.48</u>	5.41	p<.001.
People with 22q11.2 are a burden on society	2.42 (SD=1.21)	2.28 (SD=1.10)	.14	.90	n.s.
People with 22q11.2 are a burden on their family	3.10 (SD=1.17)	2.43 (SD=1.08)	.67	3.16	p<.01
*for one-tailed test					

Discrimination Subscale: Significant and favorable changes in attitude were also found in three out of four items measuring discrimination (table 2). A decrease in the "Mean Post Video" for each statement indicated more disagreement and thus, improved attitudes. No

significant change was found for people with 22q11.1 being a burden on society.

Table 2: Attitude Change toward People with 22q11.1 Deletion Syndrome **(Discrimination)** Pre and Post Video (1 = strongly disagree, 5 = strongly agree)

Discrimination	Mean Pre Video (N=21)	Mean Post Video (N=21)	Mean Difference	t value	p level*
People often make fun of disabilities	3.57 (SD=1.03)	3.33 (SD=1.15)	.24	1.42	n.s.
People with 22q11.2 are easier to take advantage of compared to other people	3.47 (SD=1.08)	3.09 (SD=1.18)	.38	1.79	p<.05.
People tend to become impatient with those with 22q11.1	3.43 (SD=.92)	2.62 (SD=.86)	.81	4.95	p<.001
People tend to treat those with 22q11.1 as if they have no feelings	2.95 (SD=1.20)	2.48 (SD=1.12)	.47	2.68	p<.01
*for one-tailed test					

Positive Gains Subscale: Favorable changes in positive gains of people with 22q11.1 deletion syndrome were found in three out of four areas (table 3). An increase in the "Mean Post Video" for each statement indicated more agreement and thus, improved attitudes. No significant change was found in some people achieving more because of their disabilities.

Table 3: Attitude Change towards People with 22q11.1 Deletion Syndrome **(Positive Gains)** Pre and Post Video (1 = strongly disagree, 5 = strongly agree)

Positive Gains	Mean Pre Video (N=21)	Mean Post Video (N=21)	Mean Difference	t value	**p level***
Having 22q11.1 can make someone a stronger person	3.57 (SD=1.08)	3.95 (SD=.86)	-.38	-2.35	p<.01.
Having 22q11.1 can make someone a wiser person	3.09 (SD=1.13)	3.47 (SD=1.03)	-.38	-2.35	p<.01.
Some people achieve more because of their disabilities	3.24 (SD=1.13)	3.52 (SD=1.03)	-.28	-1.55	n.s.
People with 22q11.1 are more determined than others to reach their goals	2.90 (SD=1.09)	3.38 (SD=.92)	-.48	-2.50	p<.01

Current / Future Hopes: Significant changes in attitude were found in two out of four items measuring current and future hopes (table 4). A decrease in the "Mean Post Video" for each statement indicated more disagreement and thus, improved attitudes. No significant change was found for the two other statements about sex discussions and looking forward to the future.

Table 4: Attitude change towards people with 22q11.2 Deletion Syndrome **(current/future hopes)** Pre and Post Video					
Current/Future Hopes	Mean Pre Video (N=21)	Mean Post Video (N=21)	Mean Difference	t value	p level*
Sex should not be discussed with people with 22q11.1	2.14 (SD=1.12)	1.90 (SD=1.15)	.24	1.31	n.s.
People should not expect too much from people with 22q11.1	2.24 (SD=1.09)	1.90 (SD=89)	.33	2.09	p<.05.
People with 22q11.1 should not be too optimistic about their future	2.19 (SD=1.08)	1.86 (SD=.86)	.33	1.92	p<.05
People with 22q11.1 have less to look forward to than others	2.33 (SD=1.20)	2.00 (SD=.95)	.33	1.67	n.s.
* For one-tailed test					

Overall scores: The greatest mean difference for ADS scores pre and post video was in the "inclusion" subscale (3.09) and the least mean difference was in the "current and future hopes" subscale (1.24). Table 5 shows the mean difference overall and by subscale for the 22q11.2 DS group. The mean difference for total scores pre and post video was 7.76. For all subscales, the p level was significant.

Table 5: Summary of Attitude Changes towards people with 22q11.2 Deletion Syndrome Pre and Post Video				
	Pre video mean	**Post video mean**	**Mean difference**	**p level**
Total score	47.00 (SD = 8.42)	39.23 (SD = 9.14)	7.76	p < .001
Inclusion	13.47 (SD = 3.37)	10.38 (SD = 4.16)	3.09	p < .001
Discrimination	13.47 (SD = 3.22)	11.52 (SD = 3.81)	1.9	p < .001
Positive Gains	12.81 (SD = 4.06)	14.33 (SD = -1.52)	-1.52	p < 0.05
Hopes	8.9 (SD = 4.17)	7.67 (SD = 3.55)	1.24	p < 0.05
Note: The positive gain score was recorded in the same direction as the other 3 subscales to compute a total attitude score (p levels are for a one-tailed test)				

2. F*amilial Dysautonomia Group*

Inclusion Subscale: No significant changes in attitude were found in all four items measuring inclusion, exclusion and burden for people with familial dysautonomia.

Discrimination Subscale: No significant changes in attitude were found in all four items measuring discrimination towards people with familial dysautonomia.

Positive Gains: Favorable attitude changes in the positive gains category were found in 3 out of 4 areas (table 6). The statement "having FD can make someone a stronger person" went from a mean of 3.50 or between "agreement" and "uncertain" to 4.00 or "agreement". The statement "Having familial dysautonomia can make a person wiser" went from a mean of 3.44 or between "uncertain" and "agree" to 3.67 or a greater level of "agreement". The statement "People with FD are more determined than others to reach their goals" went from a mean of 2.96 or "uncertain" to 3.33 between "uncertain" and "agreement". No significant change was found in the statement "some people achieving more because of their disabilities".

Table 6: Attitude change towards people with familial dysautonomia (Positive Gains) Pre and Post Video

Positive Gains	Mean Pre Video (N=27)	Mean Post Video (N=27)	Mean Difference	t value	p level*
Having FD can make someone a stronger person	3.50 (SD=.90)	4.00 (SD=.56)	-.50	-2.82	p<.01.
Having FD can make someone a wiser person	3.44 (SD=.85)	3.67 (SD=.68)	-.23	-2.00	p<.05.
Some people achieve more because of their disabilities	3.37 (SD=.97)	3.52 (SD=.88)	-.15	-1.44	n.s.
People with FD are more determined than others to reach their goals	2.96 (SD=.44)	3.33 (SD=.88)	-.37	-2.43	p<.05
* for one-tailed test					

Current and future hopes: No significant changes in attitude were found in the four items

measuring current and future hopes for people with familial dysautonomia.

Overall scores: The only statistically significant mean difference for ADS scores was for the "positive gains" subscale (-1.27). Table 7 shows the mean difference overall and by subscale for the FD group. The mean difference for total ADS scores pre and post video was 2.0.
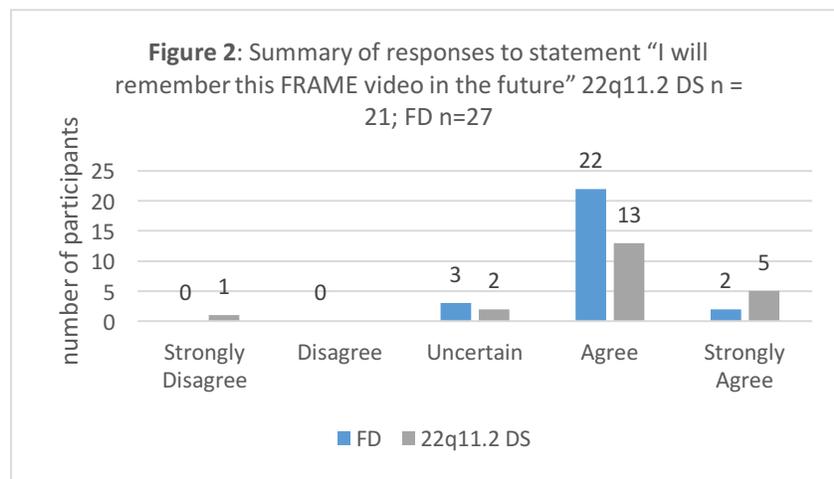
Table 7: Summary of Attitude Changes towards people with familial dysautonomia Pre and Post Video				
	Pre video mean	**Post video mean**	**Mean difference**	**p level**
Total score	41.15 (SD = 6.22)	39.15 (SD = 8.07)	2.00	p < .05
Inclusion	11.00 (SD = 2.66)	10.67 (SD = 3.35)	0.33	n.s.
Discrimination	12.22 (SD = 2.64)	11.67 (SD = 2.88)	0.55	n.s.
Positive Gains	13.27 (SD = 2.32)	14.54 (SD = 2.39)	-1.27	p < .001
Hopes	7.3 (SD = 2.45)	7.48 (SD = 2.71)	-0.18	n.s
Note: The positive gain score was recorded in the same direction as the other 3 subscales to compute a total attitude score (p levels are for a one-tailed test)				

B. Video Evaluation Survey Results

Retention of the video and learning styles

Figure 2: Summary of responses to statement "I will remember this FRAME video in the future" 22q11.2 DS n = 21; FD n=27
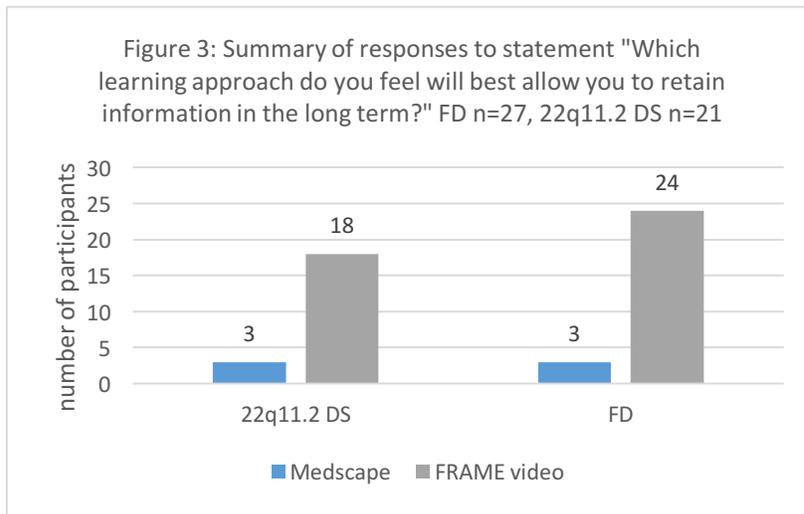
For the group that reviewed the 22q11.2 deletion syndrome video (N=21), all but three people thought the FRAME video, not Medscape, would be best for the retention of information about this disease. All three of these people were uncertain or disagreed they would remember things from the FRAME video, while 100% of those who preferred the FRAME video agreed the FRAME video would be remembered in the future. 86% of participants (n=18) from the 22q11.2 DS group agreed or strongly agreed they would remember the FRAME video in the future (**figure 2**). Finally, 18 participants (82.8%) agreed they learned best when multiple

learning modes (audio, video, text) were utilized.

For the group that was assigned the familial dysautonomia (FD) video (N=27), all but three people thought the FRAME video, not Medscape, would be best for information retention about this disease (**Figure 3**). Two of these three participants (66.7%) were uncertain about

Figure 3: Summary of responses to statement "Which learning approach do you feel will best allow you to retain information in the long term?" FD n=27, 22q11.2 DS n=21
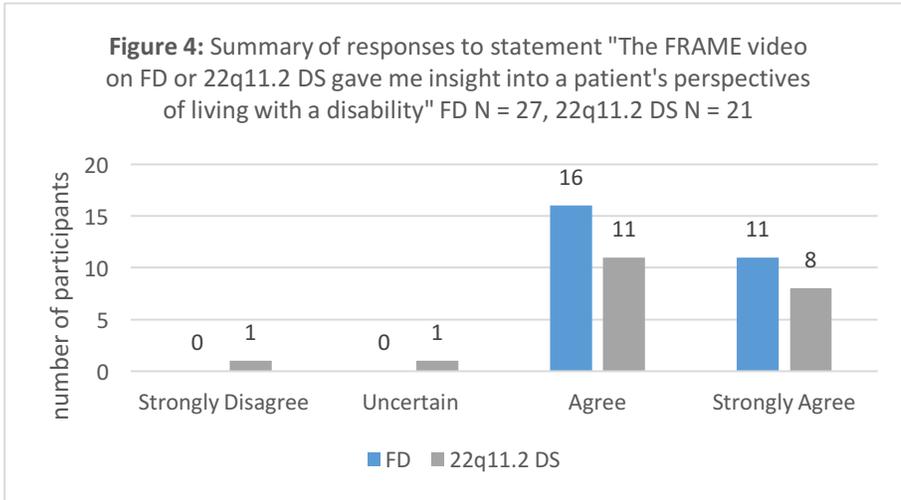
remembering things from the FRAME video, while 93.8% of those who preferred the FRAME video agreed the FRAME video would be remembered in the future. 93% of participants (n=24) in the FD group "agreed" or "strongly agreed" they would remember the FRAME video in the future **(Figure 3).** A total of 24 people (89%) agreed they learned best when multiple learning modes (audio, text, video) were utilized.

Perspectives about disability and comfort levels

For the group reviewing the 22q11.2 deletion syndrome video (N=21), 14 participants (63.6%) agreed or strongly agreed they would feel comfortable being involved in caring for someone with this condition. Nineteen participants (86.4%) agreed or strongly agreed the 22q11.2 deletion syndrome video gave them insight into living with a disability**.** Only one of the three participants who preferred the Medscape information agreed they would be comfortable caring for someone with 22q11.2 deletion syndrome. Similarly, only one of the three agreed it gave them insight into living with the disability (**figure 4)**.

For the the FD group (N=27), 24 participants (85.7%) agreed or strongly agreed they would feel comfortable being involved in caring for someone with FD. Even though three people preferred the Medscape information, all agreed they would be comfortable caring for someone with FD. 100% of participants agreed or strongly agreed the FD video gave them insight into living with a disability (figure 4).

Figure 4: Summary of responses to statement "The FRAME video on FD or 22q11.2 DS gave me insight into a patient's perspectives of living with a disability" FD N = 27, 22q11.2 DS N = 21

Feedback for the FRAME videos

For the 22q11.2 DS sample (N=21), only two people (9.1%) would not be interested in additional FRAME videos about other disorders. For the FD sample, only four people out of 27 (14.8%) would not be interested in additional FRAME videos about other disorders.

C. Thematic Analysis of long term retention of genetic condition

A total of 43 participants answered the open ended question to explain whether FRAME video or Medscape is a better learning approach to retain information in the long term. Three themes emerged from participants that picked the FRAME video. Participants mentioned that seeing a visual (n=24) and hearing stories and experiences (n=16) allowed them to better retain the information. The third theme was organization of the video and how the information was presented (n=3). One participant said:

> "Seeing families talk about how the illness affects their lives and watching them interact with their child with 22q11.2 deletion syndrome provided invaluable information about the day-to-day life of living with this disorder. It takes text and transforms it to real people who want the same thing we all do - to be happy and healthy. I felt more inspired to care for someone with this syndrome after the video

than I did after reading the medscape article."

Three themes emerged from the six participants who chose Medscape over FRAME. These participants emphasized the FRAME videos did not have enough information (n=2) and were less efficient than Medscape (n=1). Two participants also mentioned they learn better by reading and therefore Medscape would allow them to retain information in the long run. One participant said:

> "The FRAME video is more memorable but did not provide adequate information regarding the clinical presentation, diagnosis, and management of the disease. If the participants in the video had a greater discussion regarding those topics, it would have been an excellent means of learning the material…"

D. Thematic Analysis of the strengths of the FRAME video

A total of 45 participants answered the open ended question about what they most liked about the FRAME video. The majority of participants mentioned family involvement (n=14) and patient stories and perspectives (n=16) as their top reasons. Some participants wrote about the wide phenotypic spectrum represented in the videos (n=5) and others said the videos were informative (n=3). One participant wrote about how effective the stories were in creating a memory.

> "The feelings brought about by hearing those who actually have FD and the challenges faced by them from their perspective and their families creates a more vivid memory."

Another participant described how two children with 22q11.2 deletion syndrome could have different clinical features, a genetics concept known as variable expressivity.

> "I liked that it portrayed children on all areas of the spectrum in terms of severity of the condition. I could see that for some parents it took up a significant amount of attention and time to care for the 22q11.2 deletion syndrome child, while in other families it was minor. "

E. Thematic Analysis of improvements to the FRAME video

A total of 44 participants answered the open ended question about how the FRAME video can be improved. Ten participants responded with either "not sure" or "no changes needed". Another major theme was the FRAME video needed more information (n=13),

such as details about prognosis, treatment, diagnostic strategies, pathophysiology and daily challenges. One participant highlighted the difference between the goals of FRAME and Medscape.

> "It all depends on the intended audience. For preparing me as a healthcare provider, it is certainly inadequate, in contrast to knowledge contained within the Medscape pages. However, if it is intended as an overview for the public to understand this condition exists, its sufferers are otherwise "normal" people, and the struggles are significant."

This quote highlights that while the FRAME video is unable to contain as much information as Medscape, it serves as a vehicle for patients to share their experiences living with the condition. Another participant described how the videos did not contain enough stories about family challenges and negative experiences in society.

> "I would have liked to see the full picture of how the syndrome affects families and how these kids are treated in school and society. I understand that the video is intended to be optimistic, but I would have liked to hear more of how much some families struggled to care for the child with this syndrome, especially when there are other children in the family to pay attention to as well. I also wonder how often these children are mistreated in school and in society."

A small number of participants mentioned that the FRAME videos should have included more text slides (n=2), more visual aids for hallmark characteristics (n=3) and the involvement of a health professional (n=5).

DISCUSSION

There is a lack of research comparing the effectiveness of digital storytelling with traditional medical education methods. Our study is one of the first to explore the impact that an online medical education digital story telling tool has on medical students' attitudes towards individuals with disability. The FRAME library of videos and photographic galleries is unique because it is an online, multimedia resource that designates people with genetic conditions and their caregivers as the educators for healthcare students and professionals. Each film is designed to increase understanding of the featured genetic, physical, intellectual and/or behavioral aspects of different conditions. In this study, we used the Attitudes to Disabilities Scale (ADS) to measure medical students' attitudes before and after watching one of two FRAME videos about either 22q11.2 deletion syndrome or familial

dysautonomia. Additionally, we asked participants to complete a video evaluation survey to investigate their preferences for Medscape versus FRAME. Another purpose of the video evaluation was to assess how effective FRAME videos are for helping viewers understand the experiences of living with disability.

Our findings suggest that by watching FRAME videos, medical students attitudes towards individuals with disability are significantly improved. In both the FD and 22q11.2 DS groups, there were favorable attitude changes in three out of the four statements in the "positive gains" category of the ADS pre and post video. These three statements investigate participants' attitudes about whether having 22q11.2 DS or FD makes someone a stronger, wiser and more determined person. This subscale is unique because it explicitly reflects positive gains in relation to self and to others that may be a surprise about disability (Power & Green, 2010). Literature demonstrates that the disabilities community faces stigma, stereotypes, prejudiced attitudes and institutional barriers in health care (Brown et al., 2010; Eddy & Robey, 2005; Woodward, L. et al., 2012).

There were more significant and favorable attitude subscale changes for the 22q11.2 DS group than for the FD group. In the 22q11.2 DS group, there were positive attitude changes for three out of the four statements in the inclusion, discrimination and positive gains subscales. Notably, in the inclusion subscale, participants went from agree to between disagree and uncertain for the statement "people with 22q11.2 deletion syndrome have problems getting engaged in society". This significant improvement may be explained by the video's emphasis on highlighting the story of patients that went on to become successful college students. There were positive changes for the 22q11.2 DS group in two out of the four statements in the subscale which highlights attitudes about current and future hopes. This FRAME video did not discuss topics related to sex which may explain the lack of significant improvement in attitude for the statement "Sex should not be discussed with people with 22q11.2 deletion syndrome".

For the FD group, there were favorable attitude changes in three out of four statements in only one subscale of the ADS. The change in total ADS score was significant indicating overall attitudes towards FD were improved. There may be several explanations

for the difference in subscale scores between the FD and 21q11.2 DS groups. The participants in the FD videos presented with more noticeable physical differences and speech impediments. It is possible the FRAME video in isolation may not be enough to improve ADS scores in all the subscales for more severe disabilities. A previous study suggests that using digital storytelling is a key, but not sole component within a clinical training module designed to improve knowledge, attitudes and comfort levels when caring for patients with disabilities (Woodward, L. et al., 2012). Additional confounding factors when comparing results between the two conditions include differences in video production and prior student exposures to disability training. It is important to note that even though there were less significant changes in the ADS scores for the FD group, 100% of participants agreed this FRAME video provided insight into the lives of individuals with disabilities and 24 participants (85.7%) agreed or strongly agreed that they would feel comfortable being involved in caring for someone with FD. To better understand the different results between the two conditions, a future study could be done in which one individual watches two FRAME videos and the changes in ADS scores for each video are compared. This approach would likely reduce the confounding factors related to different individuals watching different videos.

Another significant finding is 89% of students identified the need for exposure to multiple learning modes (audio, text and visual) for maximum absorption of knowledge. This finding is consistent with literature that suggests digital storytelling and technology prompt deeper learning and knowledge retention (Alessie & Trollip, 2001; Nowaczyk, 2012; Sandars, Murray, & Pellow, 2008; Smeda, Dakich, & Sharda, 2014). Similarly, when asked to explain why participants picked FRAME as their preferred learning approach, the answers were focused around themes of visual imagery, auditory experience of hearing stories and video organization. The majority of participants agreed that the FRAME video would be remembered in the future (86% for 22q11.2 DS, 89% for FD) and there is a majority preference for FRAME over Medscape.

Strengths and Limitations of the Study

We used an established and validated self-report scale for attitude changes and

qualitative surveys. Our mixed method approach can be considered a strength of this study. Additionally, our sample included 4th year medical students from three different schools to create a more representative sample.

One limitation of this study is we only evaluated two FRAME videos on two genetic conditions. Future studies can be done to analyze attitude changes for more FRAME videos on different conditions. Additionally, baseline exposure to the genetic condition and prior disabilities training in medical school were not assessed to avoid excess participant burden. Lastly, our study uses self-reported data to explore the effectiveness of digital storytelling as a supplementary learning tool. A future study could compare FRAME videos and traditional learning methods in medical school.

Conclusion

The results of this study suggest the FRAME videos are unique and effective learning tools in medical education. Overall, the FRAME videos improved attitudes towards individuals with disabilities in both the groups. Additionally, medical students in this study expressed preferences for the FRAME video over a traditional text-based learning method such as Medscape. These videos are a response to the need for healthcare professionals and students to have exposure to genetic conditions and disabilities in the hopes they will be more comfortable providing these patients with medical care. Understanding the benefits and limitations of supplementary medical education tools will facilitate further development and research in digital storytelling as an important addition to health care education and training programs.

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APPENDIX A: ADS Scale (22q11.2 Deletion Syndrome)

		Strongly Disagree	Disagree	Uncertain	Agree	Strongly Agree
1	People with 22q11.2 deletion syndrome find it harder to make new friends					
2	People with 22q11.2 deletion syndrome have problems getting involved in society					
3	People often make fun of disabilities					
4	People with 22q11.2 deletion syndrome are easier to take advantage of (exploit or treat badly) compared with other people					
5	People with 22q11.2 deletion syndrome are a burden on society					
6	People with 22q11.2 deletion syndrome are a burden on their family					
7	Having 22q11.2 deletion syndrome can make someone a stronger person					
8	Having 22q11.2 deletion syndrome can make someone a wiser person					
9	Some people achieve more because of their disability (e.g. they are more successful)					
10	People with 22q11.2 deletion syndrome are more determined than others to reach their goals					
11	People tend to become impatient with those with 22q11.2 deletion syndrome					
12	People tend to treat those with 22q11.2 deletion syndrome as if they have no feelings					
13	Sex should not be discussed with people with 22q11.2 deletion syndrome					
14	People should not expect too much from those with 22q11.2 deletion syndrome					
15	People with 22q11.2 deletion syndrome should not be optimistic (hopeful) about their future					
16	People with 22q11.2 deletion syndrome have less to look forward to than others					

Appendix B: Video Evaluation Survey (22q11.2 deletion syndrome)

		Strongly Disagree	Disagree	Uncertain	Agree	Strongly Agree
1	I will remember this FRAME video in the future.					
2	I would feel comfortable being involved in the care of someone with 22q11.2 deletion syndrome.					
3	The FRAME video on 22q11.2 deletion syndrome gave me insight into a patient's perspective of living with a disability.					
4	I learn best when I am exposed to audio, visual and text resources.					

5. Which learning approach do you feel will best allow you to retain information about 22q11.2 deletion syndrome in the long term?
         A. Medscape
         B. FRAME video
         Please explain your choice _____

6. What do you like most about the FRAME video on 22q11.2 deletion syndrome?

7. How do you think the FRAME video on 22q11.2 deletion syndrome can be improved?

7. Would you be interested in having access to additional videos about other disorders for your resource?
         A. Yes
         B. No

8. What medical specialty are you planning to practice?