

Promoting Testicular Self-Examination and Awareness Amongst Young Men With Intellectual Disabilities: A Parallel Intervention Randomized Study

Abstract

Testicular cancer is the most common type of cancer affecting males between 15 and 34 years of age. Men with intellectual disabilities have an increased risk of testicular cancer, often presenting late. We compared educative teaching (Teaching group) with an educative pictorial leaflet (Leaflet group) using a multi-media questionnaire to assess participants' knowledge, skills, and behaviour. Evaluation included demonstration of self-examination skills using an anatomically correct model. Adults with intellectual disabilities were involved as co-researchers at all stages from planning to writing-up the study. Both educational methods led to significant improvements in knowledge and skills, maintained 6 months after the intervention. At one-week, direct teaching had significantly greater effect on knowledge and skills than being given a leaflet. By six months improvements in knowledge, skills and behaviour were similar, without significant increase in health-related or general anxiety in either group. Young men with intellectual disabilities benefited from health education in group teaching sessions and through information provided pictorially in leaflets to read at home. This has wider implications for health promotion, where information is often withheld to avoid anxiety.

Testicular cancer accounts for around 1% of all male cancers (Le Cornet et al., 2014) and is the most common type of cancer affecting adolescent and young males of European ancestry (Cook et al., 2010). Global testicular cancer incidence has increased since the 1970s, with highest rates in Western and Northern Europe, Australia and New Zealand, and Northern America, although there are variations between countries and between different ethnic and racial groups (Huyghe, Matsuda, & Thonneau, 2003). In the United Kingdom, testicular cancer incidence rates increased from 3.3 per 100,000 person-years in 1973–1977 to 5.4–5.8 per 100,000 person-years in 1998–2002, compared to 4.6 per 100,000 person-years in Canada in 1998–2002 (Chia et al., 2010). Testicular cancer incidence continues to increase worldwide (Znaor, Lortet-Tieulent, Laversanne, Jemal, & Bray, 2015), and is predicted to rise to 10 per 100,000 person-years in the United Kingdom by 2035 (Smittenaar, Petersen, Stewart, & Moitt, 2016).

There appears to be an association with intellectual disability (intellectual developmental disorder) [IDD for short] (Patja, Eero, & Iivanainen, 2001; Patja, Pukkala, Sund, Iivanainen,

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& Kaski, 2006; Thornhill et al., 1988), genetic abnormalities (Dexeus, Logothetis, Chong, Sella, & Ogden, 1988; Sakashita, Koyanagi, Tsuji, Arikado, & Matsuno, 1980) and Down syndrome in particular (Hafeez, Singhera, & Huddart, 2015; Hill et al., 2003; Thornhill et al., 1988; Yang, Rasmussen, & Friedman, 2002). Barr, Gilgunn, Kane, and Moore (1999) reported that 10.8% of men with IDD have some abnormality of the testis. Eleven per cent of testicular cancers develop in men with a history of undescended testis (cryptorchidism) (Braga, Lorenzo, & Romao, 2017); indeed cryptorchidism is the most significant known risk factor for testicular cancer (Cook et al., 2010), and is more common among those with IDD (Cortada & Kousseff, 1984). Testicular cancer prevalence is higher in males with Down syndrome, even after accounting for a higher cryptorchidism rate, (Hasle, Clemmensen, & Mikkelsen, 2000; Miller, 1970). Additional postulated contributory factors include gene hyperexpression and higher levels of follicular stimulating hormone (Lambalk & Boomsma, 1998; Satge et al., 1997), and lifestyle factors including obesity, high fat diets, increased body size and low levels of physical exercise (Lerero, McGlynn, & Cook, 2010; Sasco et al., 2011).

The 5-year net-survival rate for testicular cancer is 96% (Canadian Cancer Society, 2017), with poorer prognosis for those presenting at a later stage of the disease (Cancer Research UK, n.d.). In addition to improving prognosis, early detection may reduce the physical and psychosocial distress associated with prolonged anti-cancer treatment (Shanmugalingam, Soutlati, Chowdhury, Rudman, & Van Hemelrijck, 2013). Approximately 95% of testicular cancers present with a painless testicular lump or swelling (Rudberg, Nilsson, Wikblad, & Carlsson, 2005). However, of the twenty individuals with Down syndrome and testicular cancer reviewed by Dieckmann, Rube and Henke (1997), eight presented with metastatic disease, one had a giant tumour, and two had locally advanced disease, with testicular abnormalities being discovered incidentally by carers or healthcare professionals.

Although testicular cancers, particularly seminomas and teratomas, are highly sensitive to

radiotherapy and chemotherapy, men with IDD do not always receive comprehensive treatment for more advanced disease; for example, Miki, Ohtake, Hasumi and Moriyama (1999) report a case in which “neither chemotherapy nor radiotherapy were performed due to his severe mental retardation.” Evidence of inequitable access to healthcare is recognized in the U.K. 2013 *Confidential Inquiry into Premature Mortality in People with Intellectual Disabilities*, which recommended people with IDD should have access to the same investigations and treatments as others, whilst recognizing that such services may need to be delivered differently (Heslop et al., 2013).

Barriers to accessing healthcare, along with communication difficulties, pose challenges to the detection of early signs and symptoms of testicular cancer in individuals with IDD (Hanna, Taggart, & Cousins, 2011). There is debate about the role of routine testicular self-examination (TSE) in reducing delays in presentation among men in the general population and to a lesser extent among men with IDD. The U.S. Preventative Services Task Force discourages routine screening for testicular cancer in asymptomatic males due to lack of evidence of impact on mortality rates (Lin & Sharangpani, 2010), whereas the European Association of Urology recommends routine TSE for individuals with testicular cancer risk factors (Albers et al., 2010). Men in the general population have poor awareness of testicular cancer and TSE (Saab, Landers, & Hegarty, 2016b), and even opponents of testicular cancer screening by routine TSE accept the importance of educating young men about symptoms and signs to facilitate early presentation (Austoker, 1994; Buetow, 1996; Riley et al., 1998; Westlake & Frank, 1987). Riley et al. (1998) recommend health promotion should occur during childhood and adolescence, with an emphasis on positive health awareness rather than a ritualized “check for cancer.” Given the association of IDD and testicular cancer, and delays in diagnosis, investigations and treatment (Hanna et al., 2011), the case for promoting education about testicular cancer and TSE is particularly strong for men with IDD (Satge et al., 1997).

Overcoming the challenges in providing health promotion and education for people with IDD requires the use of accessible communication methods which are effective from the perspective of the individual with IDD (Bollard, 2002). These include visual representations alternative or supplementary to written or verbal content, and personalized support to optimize the participation of the individual with IDD (Boardman, Bernal, & Hollins, 2014). Hurtado et al. (2014) compared different formats in use with people with IDD and found stand-alone pictures were easier to understand than formats presenting information in more than one medium, such as words illustrated with pictures as used in easy read leaflets.

Research into accessible communication methods around testicular cancer and TSE for men with IDD is scant. One recent systematic review found 11 studies, utilizing a variety of strategies, to increase men's awareness of testicular cancer and their intentions to perform TSE (Saab, Landers, & Hegarty, 2016a; Saab, Landers, & Hegarty, 2016b). Although one included study reported the use of an American Sign Language video to meet the needs of deaf study participants (Folkins et al., 2005), none focused on men with IDD or the adaptations required to make health material accessible to this population.

There is a need to establish whether educating men with IDD can enable them to recognize early symptoms and signs of testicular disease and present to healthcare professionals for further investigation. One concern is whether such education increases participants' anxiety about cancer, leading to increased inappropriate use of healthcare resources. However the identification of benign abnormalities of the genitalia, which may be uncomfortable and distressing and affect sexual and reproductive health, should in itself be beneficial.

The objective of this study was to evaluate and compare, quantitatively and qualitatively, the impact of two different educational interventions about testicular health on knowledge, skills and health-related behaviours in regards to self-reported TSE at 1 week and 6 months post-intervention in male adults with IDD. One

intervention involved direct teaching, the other provision of a leaflet to take away and read. As both interventions utilized educational material specifically adapted for adults with IDD, it was hypothesized that both groups post-intervention would demonstrate increased knowledge of testicular cancer symptoms and signs, and increased skills in TSE (primary outcome). It was also hypothesized that the groups would be similar in secondary outcomes consequent to educational interventions about testicular health, including anxiety, health-related anxiety, health locus of control and self-efficacy. It was further hypothesized that both interventions would inform future approaches to involving young adult males with IDD meaningfully in their own health surveillance.

Methods

Research Design

This was a participatory randomized parallel study of two educational interventions. From the beginning research advisors with IDD were employed in assisting in the design of the study, in creating information, teaching materials and outcome measures accessible to individuals with IDD, in playing an active role in the research and through participating in steering and advisory group meetings. One research assistant with IDD was specifically recruited for the study, with responsibility for co-leading the teaching group with a male community IDD nurse (Butler, Cresswell, Giatras, & Tuffrey-Wijne, 2012).

The education methods and research evaluation tools were piloted for both teaching ($n = 6$) and leaflet ($n = 6$) interventions and alterations made as necessary. During the pilot study outcome measures were carried out immediately following each intervention; thus the Leaflet group did not have the opportunity to take the leaflet home and discuss the content with a family member, friend or helper prior to the initial post intervention evaluation. This protocol was altered for the main study so that first evaluation post intervention took place one week later for both Teaching and Leaflet groups.

Participants

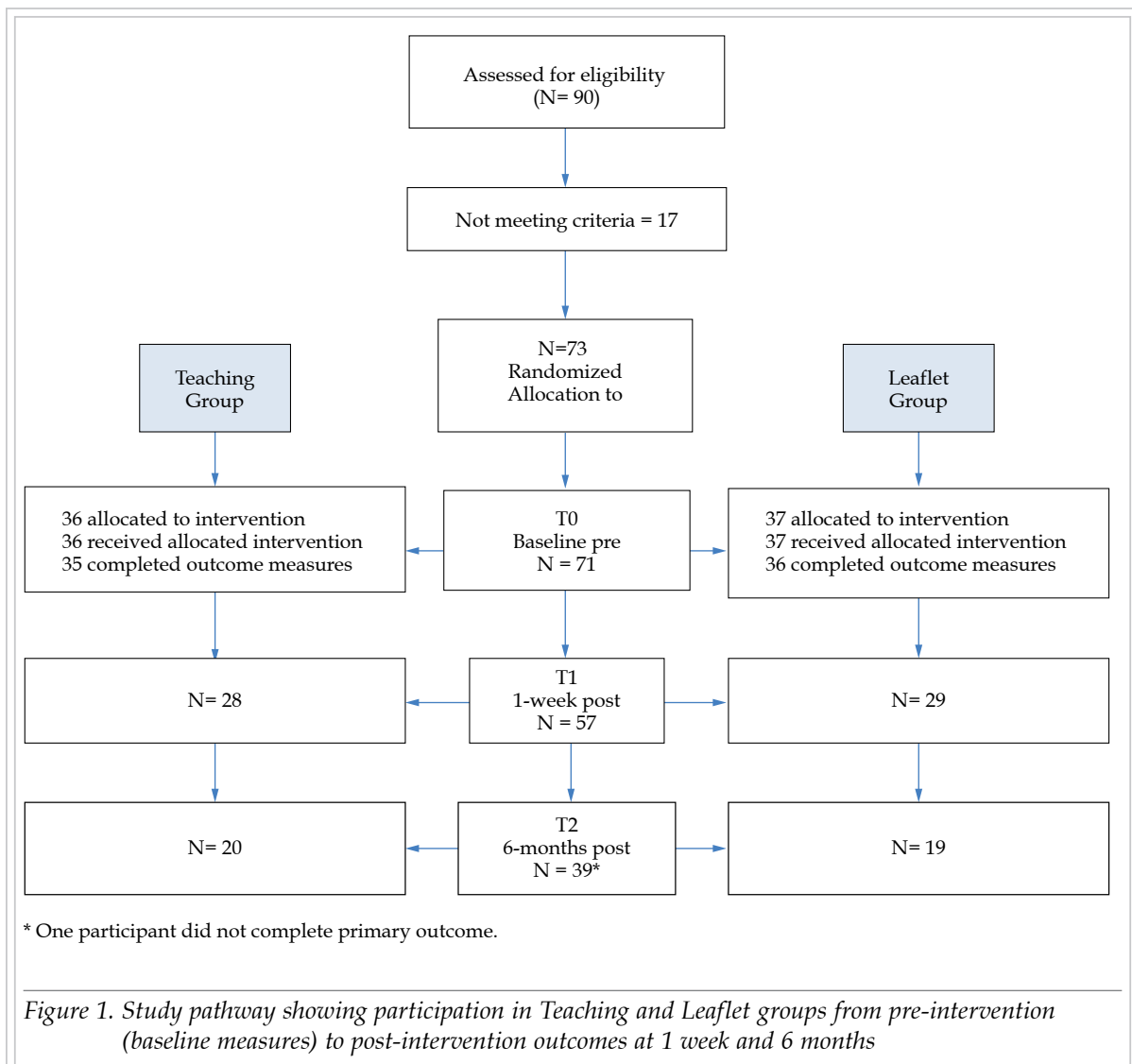
Ninety men with IDD aged 16 years or older were invited to take part in the research, having previously been selected as suitable by teachers and carers as likely to benefit from an educative intervention. Each participant received an information sheet about the study and informed consent was obtained. Seventy-three participants were assessed as meeting the study criteria (Figure 1). Researchers were careful to avoid coercing individuals to commence participation, or to continue as participants if they changed their minds during the study.

The inclusion criteria included: (1) being male aged 16 years or older; (2) registered with a GP,

and /or known to IDD services or on the social services IDD register or known to have Down syndrome; and (3) having the capacity to give informed consent. Ethics approval did not permit inclusion of males with moderate/severe and profound IDD.

Procedure

Initially participants from several U.K. London boroughs were recruited through primary care services and IDD registers, but most successful recruitment occurred through colleges, day cen-



tres, voluntary organizations and youth clubs. As previous studies, such as those involving cervical screening for women with IDD, have described difficulties with recruitment for such research, the geographical areas chosen covered what was deemed to be a suitably large population to allow adequate numbers. Each participant was assessed as having capacity to consent to take part in the study by a specialist in IDD psychiatry.

Randomization

Participants were randomly allocated into "Teaching" or "Leaflet" groups. This randomization was coordinated centrally by a statistician such that at each community or clinical site subjects were approximately evenly distributed between Teaching and Leaflet groups. Whilst this had the disadvantage of allowing some potential contamination between the groups, it had the advantage of ensuring that the two arms of the study were similar in terms of age and ethnicity of subjects. This was confirmed on subsequent statistical analysis.

Interventions

Teaching group. An educational programme on testicular awareness was designed, using existing visual aids, video, a computer programme and an anatomically correct model with which to illustrate self-examination. The programme aimed to: (a) promote understanding of cancer risk and prognosis; (b) enhance the recognition of symptoms and signs, including those of more benign abnormalities; (c) instruct on how to seek help and self-refer to the GP; and (d) inform as to what further investigations and treatment might be expected if cancer was suspected. The aim was to design a programme which was understandable and accessible to people with IDD and which motivated the participants to alter their behaviour without provoking excessive levels of anxiety. The teaching methods were developed by a multi-disciplinary steering group incorporating representatives from consumer groups (young men with IDD), speech and language therapy, psychology, psychiatry, general practice, community intellectual disability nursing,

with advice from a urology specialist as needed. The teaching was delivered by a male community IDD nurse and a male co-researcher with IDD through two thirty-minute sessions. The teaching programmes were carried out in small groups no larger than six participants.

Leaflet group. Participants in the leaflet group received an educative leaflet on testicular awareness. The leaflet was designed by members of the steering group and an artist with expertise in publishing materials in accessible format, as part of a development project to create a picture book (on testicular examination) in the Books Beyond Words series (Hollins & Wilson, 2004a; Hollins & Wilson, 2004b). The leaflets were distributed and participants were encouraged to take them away and discuss them with family, friends or carers if they wished. This part of the study was not manualized, and carers/supporters received no training on how to use the leaflets with the men in the study.

Outcome Measures

Members of the steering group were involved in adapting available evaluative tools and designing new tools where necessary.

1: Primary measures. The primary measure was a 10-item questionnaire available in verbal, pictorial and written format (so as to accommodate the communicative preference of the participant) to assess *knowledge and skills* (S&K) and was administered face to face by one of two medically qualified researchers. This S&K questionnaire included demonstration of testicular self-examination skills using an anatomically correct model.

2: Secondary measures. Twelve items were selected from existing inventories of measures of general anxiety, health anxiety, health locus of control and self-efficacy and adapted to the communication needs of participants (to create a 12-item adapted questionnaire – see below). Each item was represented by a statement and a 0 to 10-cm line. The latter was anchored at its ends by descriptive phrases, one end of the line indicating strong disagreement with the statement, and other end indicating strong agree-

ment. The line was then measured in millimetres providing a measure (analogue scale) of the individual's agreement/disagreement with the statement; higher scores indicated greater anxiety, greater self-efficacy and the extent of control (internal, external or chance).

Outline of the 12-item adapted questionnaire (12-item Adpt-Q). Four items measuring *General Anxiety* in the State-Trait Anxiety Inventory (Spielberger, 1989) were adapted and made accessible by using visual analogue scales (e.g., "I am tense" with strongly agree = 10 and strongly disagree = 0). Three items from the Wells Health Anxiety Scale (Wells, 1997), were used to assess *Health Anxiety*, with a higher score on the analogue scale indicating that someone feels in control of their health/not worried about their health. *Locus of control and health care related behaviours* were measured using three items from the Health Locus of Control scale (Wallston, Wallston, Kaplan, & Maides, 1976); this scale describes health related behaviours worded from internal ("if I take care of myself I can avoid illness"), external ("doctors and nurses control my health") and chance ("staying healthy is a matter of luck") perspectives. *Self-Efficacy* with regard to testicular self-examination and seeking help was measured by two questions ("I know how to check my balls/testicles" and "I feel able to tell someone if I find something wrong with my balls/testicles").

Also recorded were:

3: Participant reported self-examination and

4: Self-referral activities and GP visits. Self-recording diaries (adapted to the communication needs of the participant) were distributed to individuals in each group so that they could monitor their behaviour in terms of self-examination and GP visits over a six-month period.

5: Qualitative evaluation. This was through feedback from participants to the medical researchers using a 5-question semi structured interview. The self-advocate co-researcher participated in this feedback providing opportunity to explore and compare participant personal experiences to each intervention (Tuffrey-Wijne,

Bernal, & Hollins, 2008; Tuffrey-Wijne, Bernal, Butler, Hollins, & Curfs, 2007).

Both the 10-item S&K-Q and 12-item Adpt-Q were completed by the participant with guidance from the medical researchers who read each question out loud, repeating and clarifying the meaning as necessary. The same researchers were used throughout and were blind to the allocation to Teaching and Leaflet group. Each participant completed both questionnaires on three occasions: at baseline (T0); one-week (T1) and six-months (T2) following the interventions (Figure 1).

Analyses of Data

Analyses were undertaken using IBM SPSS v24. Data were analyzed on an 'intention to teach' basis. Differences between the groups with respect to the primary outcome measure (10-item S&K-Q) was analyzed using analysis of covariance including the T0 level of the dependent variable to adjust for regression to the norm. This was done twice, for the one-week (T1) and six-months (T2) follow-ups. Secondary outcome measures, which were normally distributed, were analyzed in the same way. The Mann Whitney U test was used to analyze the data on frequency of self-examination and GP self-referral rates. Categorical variables were analyzed using chi squared analysis. Paired t-tests were used to make within treatment group comparisons to explore change from baseline in all outcomes.

Results

The age range of participants was from 16 to 35 years (mean age = 20.9, *SD* = 4.8). Thirty-six were randomized to the teaching group and 37 to the Leaflet group (Figure 1). The two groups were similar for age (mean teaching = 21.3, *SD* = 5.5; leaflet = 20.4, *SD* = 4.0). At 1-week follow-up (T1), there were 28 participants in the Teaching group and 29 participants in the Leaflet group; at 6-month follow-up (T2) there were 20 participants in the Teaching group and 19 participants in the Leaflet group. Participants who dropped out showed no significant differences in age or scores on baseline measures.

**Primary Outcomes:
Knowledge and Skills**

There was a significant difference between the groups 1 week following the intervention (Teaching group scoring significantly higher ($p = .002$) but this difference between the groups was no longer significant at the 5% level at 6 months ($p = .055$; Table 1). Statistically significant change from baseline in knowledge and skills scores at 1 week and 6 months were seen in both treatment groups ($p \leq .001$) for all hypotheses tested.

Secondary Outcomes

Health anxiety. There was a significant difference between the groups at T1 with respect to health anxiety ($p = .043$) with the Teaching group exhibiting less anxiety, but not at T2 ($p = .283$). There was a significant reduction in health anxiety for the Teaching group at T1 which was maintained at T2 ($p = .044$ and $p = .012$ respectively). Changes in the Leaflet group were similar although not statistically significant.

Table 1. Primary and Secondary Outcomes at Baseline (Pre-Intervention), 1 Week and 6 Months Post-Intervention in Teaching and Leaflet Groups

T0 = baseline (pre-intervention); T1 = 1 week post-intervention; T2 = 6 months post-intervention

Outcome measure (0-max possible)	Teaching Group		Leaflet Group		Significance F (p-value)	
	Mean (SD)	n	Mean (SD)	n		
Knowledge and Skills (0-20)						
T0	5.8 (2.7)	35	5.7 (2.8)	36		
T1	11.9 (4.1)	28	8.9 (4.5)	29	10.2 (.002)	
T2	12.6 (2.8)	19	9.6 (4.4)	19	4.0 (.055)	
T1-T0 ¹	3.8 (2.8, 4.8)	< .001	26	1.7 (0.7, 2.6)	.001	29
T2-T0 ¹	4.4 (3.2, 5.7)	< .001	19	3.3 (1.7, 4.8)	< .001	19
Health Anxiety (0-30)						
T0	9.8 (6.6)	35	11.9 (6.1)	36		
T1	8.2 (4.2)	26	11.4 (6.4)	29	4.3 (.043)	
T2	8.0 (5.3)	19	10.1 (4.2)	19	1.2 (.283)	
T1-T0 ¹	-2.5 (-4.9,-0.1)	.044	26	-0.7 (-2.5, 1.0)	.382	29
T2-T0 ¹	-3.4 (-5.9, -0.9)	.012	19	-2.8 (-5.7, 0.1)	.057	19
General Anxiety (0-40)						
T0	18.7 (6.7)	35	20.1 (7.0)	36		
T1	17.9 (6.3)	27	20.0 (5.4)	29	0.6 (.436)	
T2	21.0 (2.3)	20	21.5 (3.2)	19	0.5 (.500)	
T1-T0 ¹	0.4 (-1.1, 1.8)	.606	27	0.3 (-2.3, 2.8)	.825	29
T2-T0 ¹	2.1 (-1.4, 5.7)	.224	20	0.3 (-2.8, 3.4)	.839	19

continued on following page.

Table 1. Primary and Secondary Outcomes at Baseline (Pre-Intervention), 1 Week and 6 Months Post-Intervention in Teaching and Leaflet Groups (continued)

All Anxiety (0-70)						
T0	28.6 (10.8)	35	32.1 (11.6)	36		
T1	25.8 (8.2)	27	31.4 (9.9)	29	3.6 (.062)	
T2	28.8 (5.7)	20	31.6 (5.6)	19	1.1 (.311)	
T1-T0 ¹	-2.0 (-5.0, 0.9)	.112	27	-0.5 (-3.9, 3.0)	.787	29
T2-T0 ¹	-1.1 (-4.9, 2.7)	.550	20	-2.5 (-6.8, 1.9)	.248	19
LOC Internal (0-10)						
T0	7.1 (2.4)	35	7.4 (2.3)	36		
T1	8.0 (1.6)	26	7.7 (2.1)	29	0.2 (.883)	
T2	7.4 (2.3)	19	8.2 (1.0)	19	1.5 (.235)	
T1-T0 ¹	0.2 (-0.4, 0.9)	.465	26	0.4 (-0.3, 1.1)	.270	29
T2-T0 ¹	0.7 (-0.9, 2.4)	.376	19	1.5 (0.1, 3.0)	.042	19
LOC External (0-10)						
T0	5.7 (3.0)	35	5.4 (2.5)	36		
T1	6.4 (2.8)	26	5.7 (2.4)	29	0.5 (.502)	
T2	4.3 (2.8)	19	6.4 (1.6)	19	7.1 (.012)	
T1-T0 ¹	0.1 (-0.9, 1.0)	.847	26	0.0 (-1.0, 0.9)	.935	29
T2-T0 ¹	-1.5 (-3.1, 0.1)	.061	19	0.0 (-1.1, 1.2)	.940	19
LOC Chance (0-10)						
T0	5.6 (2.7)	35	5.4 (2.6)	35		
T1	5.7 (2.6)	26	5.2 (2.7)	28	0.1 (.730)	
T2	5.1 (2.8)	19	6.0 (2.4)	18	1.1 (.298)	
T1-T0 ¹	-0.3 (-1.1, 0.6)	.546	26	0.2 (-1.5, 1.1)	.774	28
T2-T0 ¹	-0.8 (-2.2, 0.7)	.272	19	0.1 (-1.5, 1.7)	.906	18
Self-efficacy (0-20)						
T0	12.2 (3.9)	35	13.2 (4.6)	36		
T1	14.5 (4.1)	27	14.3 (3.6)	29	0.1 (.734)	
T2	14.6 (4.7)	20	14.1 (3.5)	19	0.1 (.797)	
T1-T0 ¹	2.3 (0.6, 4.0)	.011	27	1.6 (-0.3, 3.4)	.101	29
T2-T0 ¹	2.3 (-0.2, 4.8)	.066	20	2.3 (0.5, 4.0)	.014	19
	<i>Median (min, max)</i>	<i>n</i>	<i>Median (min, max)</i>	<i>n</i>	<i>MWU (p-value)</i>	
GP visits (over 6 months)						
T2	0.0 (0, 20)	33	0.0 (0, 20)	33	455.0 (.191)	
Frequency of Self-Reported Examination						
T2	1.0 (0, 100)	22	6.0 (0, 100)	19	182.5 (.478)	

¹ Values are mean difference (95% CI around mean difference) p-value resulting from paired t-test, testing for change in outcome between respective time points.

General anxiety. There was no significant change in generalized anxiety within either group at both stages. There was also no significant difference between groups at T1 and T2. Qualitative feedback identified several participants who confirmed the importance of discussing the subject matter with a parent, career or other supporter. This helped to clarify the information given and reduce subjective anxiety.

Locus of control. Between T0 and T2 there was a significant increase in “internal” locus of control for the Leaflet group ($p = .042$) suggesting that pictorial information empowered people to feel that they had more control over their own health. There was no significant change in the Teaching group. While there was a significant difference between the groups at T2 ($p = .012$) (the Leaflet group scoring higher), changes in locus of control relating to external factors and chance were not statistically significant in either group.

Self-efficacy. There was a statistically significant increase in mean self-efficacy scores from T0 in the Teaching group to T1 ($p = .011$) and in the Leaflet group to T2 ($p = .014$). Subjects felt more confident about their ability to check their testicles and to seek help if they found something wrong. There was no significant difference between the groups.

GP Visits

There was no significant difference between groups in terms of GP visits over the 6-month period ($p = .191$). Qualitative feedback indicated at least two individuals in the Leaflet group successfully sought reassurance and further information from their GP.

Frequency of Testicular Examination

There was no significant difference overall between the groups in terms of frequency of testicular self-examination ($p = .478$; Table 1); seventy-three percent of the Teaching group and 63% of the Leaflet group reported performing testicular self-examination at 6 months ($\chi^2 = 0.43, p = .511$).

Qualitative Outcome

Subjective experience of the interventions. Thirty-six per cent of participants gave suggestions on how education on the topic could be improved. Some suggested that photographs could be used in the leaflets. One participant said that the video used in the Teaching group was particularly helpful. Several participants felt they would benefit from more teaching sessions and that more detail should be included. Two participants wanted to know more about what would happen if you did get testicular cancer (i.e., prognosis and treatment). Some were keen to learn about other ways of keeping healthy. Several participants suggested that the subject should be taught to younger people and be included in sex education at school. Two said a doctor should be involved in the teaching. Others said that discussing the subject with a parent or carer after receiving either the leaflet or teaching was the best way to learn more without getting too worried. Most participants in both groups commented positively (“good”) on the intervention they had participated in and could not think of any suggestions for improvement.

Subjective feedback on both interventions did indicate that many found the content at least “a bit” worrying (28% in the Teaching group and 33% in the Leaflet group). However neither intervention led to a statistically significant increase in measured health-related or general anxiety and indeed the trend was for anxiety levels to decrease between T1 to T3.

Significantly improved confidence in ability to check and seek help was gained initially by Teaching group participants and after six months by those in the Leaflet group, indicating that both interventions had an effect on behaviour as well as on knowledge and skills.

Discussion

This randomized group intervention study involving male adults with IDD, but without prior clinical evidence of testicular cancer, demonstrates that both a formal teaching programme and provision only of a pictorial leaflet in accessible format are effective in improving knowledge and skills and this improvement was maintained 6 months after each intervention. Initially the formal teaching had a significantly greater impact on knowledge and skills

than leaflet provision only but by six months these were similar in each group (Table 1).

Qualitative data suggests that individuals in the Leaflet group tended to seek further information on the subject from family, friends or helpers during the six months after receiving the leaflet to a greater extent than those who received the teaching; it is possible that the leaflet assisted carers/supporters discuss the subject more easily (Jones, Tuffrey-Wijne, Bernal, Butler, & Hollins 2007). In both groups the majority of participants reported performing testicular self-examination 6 months post intervention.

As no testicular pathology was reported and none found at clinic follow-up (not unexpected given the short length of the study, the incidence of testicular pathology and small number of participants), there was not opportunity to evaluate correct identification of pathology by participants. However, the assessment of testicular awareness included the use of hands-on anatomical models which simulated pathology in terms of palpable testicular lumps, and the ability of participants to identify these and explain what they would do if they found such an abnormality was included in the assessment outcomes.

The study findings confirm that young men with IDD can benefit from health education to improve their levels of knowledge, and skills and change behaviour. The subject of testicular self-examination and cancer is potentially embarrassing and anxiety provoking but the participants in both educative approaches, responded well to the content and appeared to benefit without marked detrimental effects. The evidence suggests that when participants receive information in an accessible form they can feel more confident about having influence over their health and seeking help, leading to reduced anxiety levels. This has implications for other areas of health promotion and information provision for people with IDD where information may be withheld with the intent of protecting an individual from anxiety.

Conclusion

Both pictorial leaflets and teaching sessions have a place in providing information to men with IDD about testicular awareness. If a leaflet or book is used the researchers recommend

that the man is encouraged to go through the information with a supporter. The use of pictorial leaflets is more efficient in terms of health service resources; additionally, there is no evidence from this study that teaching men with IDD increases demand on health care resources, other than that related to the teaching itself.

The combination of teaching sessions and an accessible information leaflet might be expected to combine the benefits of both approaches although such a conclusion cannot be drawn without further research. There is no evidence that providing accessible information for the carers/supporters to introduce to the person they support, would be an effective alternative. Indeed a previous randomized control trial (RCT) comparing the effectiveness of a bereavement intervention found that carers/supporters were reluctant to use the pictorial material provided on that sensitive subject, whereas bereavement counsellors used the materials effectively (Dowling, Hubert, White, & Hollins, 2006).

Implications for Further Research and for Practice Development

The results of this study suggest that young men with mild IDD can benefit from health education even when the subject is potentially embarrassing and anxiety provoking and that the benefits can outweigh any harmful effects and not lead to inappropriate excessive use of primary care services. This has implications for other areas of health education, including sexual health and cancer information and screening; although similar studies in specific areas e.g. breast awareness for women as well as wider "men's health" education would be beneficial.

It was the intention of the researchers to carry out the study in a way that was as ethically acceptable as possible. To this end we ensured that information on the research was provided in an accessible format and that the process was explained fully using understandable language so that participants were as informed as optimally as possible before considering whether they wished to take part. This effectively maximized the capacity of individuals to give or withhold their consent. Researchers endeavoured to ensure that there was no coer-

cion of the subjects taking part and that their consent and willingness to participate was maintained throughout their participation. The design of the study was such that all subjects had the opportunity to benefit from participation whether in the teaching or leaflet group. Through such measures the study demonstrates that people with IDD, albeit mild in severity in this case, can be ethically included in such research as subjects as well as researchers (Tuffrey-Wijne et al., 2008).

However, it is likely that both the teaching and the leaflet can be used effectively with people with more severe intellectual disabilities, with the involvement of supporters as recommended above. The latter has the added benefit that supporters/carers also learn about the subject together. The wider Books Beyond Words series are designed to be used in this way. While assessing the benefits for people with more severe disabilities was beyond the scope of this study, the pictorial tools involved do help to increase each individual's capacity to understand the issues addressed. The authors believe that the findings of the study are likely to apply to those with more severe disabilities and there is no known reason to exclude them from similar educational interventions.

During the study, the participants scored higher on anxiety measures prior to any intervention. One explanation may be that raising the subject of testicular cancer caused anxiety, which was reduced as more information and education was provided. Alternatively, involvement in the research process itself and being "tested" may have provoked anxiety which reduced as the subjects became more accustomed to the process. Clearly when involving people with intellectual disabilities in research, measures should be taken to explain the process carefully and clearly and to provide full information to minimize anxiety.

The seven-page leaflet used in this project comprised coloured drawn pictures, usually one picture on a page supplemented with a simple phrase or sentence as a descriptor of the picture. The pictures were of the type found in the "Books Beyond Words" series (www.books-beyondwords.co.uk) and were subsequently included in the title in the series "Looking After My Balls." The results indicated that leaflets given directly to a person with IDD by a health

professional, and taken home to read and share with a carer or friend, produced comparable results to face to face group teaching by an IDD nurse and peer. Whilst wordless leaflets are becoming an established method for providing people with IDD with information on difficult and emotive subjects (Hollins, Carpenter, Bradley, & Egerton, 2017), this has not been adopted as the optimal format for health promotional literature for people with IDD. Further research in this area could seek to compare the success of different formats, such as symbols, wordless books, easy read formats, audio and video presentation, for providing such information for people with IDD. Research could also follow participants for longer to determine whether improvements in knowledge and skills from similar health educative interventions can be sustained and whether peer support groups would assist with this objective.

While the focus of this study was on educating adult males with IDD about their testicular health, the study draws attention to the need also to educate health care providers (such as pediatricians, nurses, family doctors) about the increased prevalence of testicular cancer and cryptorchidism in this population. The latter, the most significant known risk factor for testicular cancer, is a condition that should be diagnosed and treated in childhood. This not only allows for easier detection of testicular cancer but also lowers the risk of developing it (Pettersson et al., 2007).

Key Messages From This Article

People with disabilities. Hooray! "Nothing about us without us."

Professionals. Adults with intellectual disabilities are able, and want, to contribute to research about their health.

Policymakers. When adults with intellectual disabilities receive information in pictures, they feel more confident about having influence over their health and in seeking help.

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