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Randomized Noninferiority Trial of Telephone vs In-Person Genetic Counseling for Hereditary Breast and Ovarian Cancer: A 12-Month Follow-Up

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Abstract

Background: Telephone delivery of genetic counseling is an alternative to in-person genetic counseling because it may extend the reach of genetic counseling. Previous reports have established the noninferiority of telephone counseling on short-term psychosocial and decision-making outcomes. Here we examine the long-term impact of telephone counseling (TC) vs in-person counseling (usual care [UC]).
Methods: We recruited high-risk women for a noninferiority trial comparing TC with UC. Of 1057 potentially eligible women, 669 were randomly assigned to TC ($n = 335$) or UC ($n = 334$), and 512 completed the 12-month follow-up. Primary outcomes were patient-reported satisfaction with genetic testing decision, distress, and quality of life. Secondary outcomes were uptake of cancer risk management strategies.
Results: TC was noninferior to UC on all primary outcomes. Satisfaction with decision ($d = 0.13$, lower bound of 97.5% confidence interval [CI] $= -0.34$) did not cross its one-point noninferiority limit, cancer-specific distress ($d = -2.10$, upper bound of 97.5% CI $= -0.07$) did not cross its four-point noninferiority limit, and genetic testing distress ($d = -0.27$, upper bound of 97.5% CI $= 1.46$), physical function ($d = 0.44$, lower bound of 97.5% CI $= -0.91$) and mental function ($d = -0.04$, lower bound of 97.5% CI $= -1.44$) did not cross their 2.5-point noninferiority limit. Bivariate analyses showed no differences in risk-reducing mastectomy or oophorectomy across groups; however, when combined, TC had significantly more risk-reducing surgeries than UC (17.8% vs 10.5%; $\chi^2 = 4.43$, $P = .04$).
Conclusions: Findings support telephone delivery of genetic counseling to extend the accessibility of this service without long-term adverse outcomes.

Genetic counseling and testing for mutations in the *BRCA1* and *BRCA2* (*BRCA1/2*) genes is integral to the clinical care of women at risk for hereditary breast and ovarian cancer (HBOC) (1). Traditionally, the standard of care for such women has been for them to undergo in-person pre- and post-test genetic counseling with a genetics professional (2,3). However, the increased demand for these services, in conjunction with the limited number and restricted distribution of cancer genetic counselors (4,5), has led to a critical need for alternative strategies to deliver genetic counseling (6–8). Telephone delivery of genetic counseling is an appealing alternative to in-person genetic counseling as it allows a larger and more geographically dispersed population of patients to have access to trained genetic counselors. Although most genetic counselors who provide HBOC services report that they have given *BRCA1/2* test results over the telephone, pretest telephone counseling has not been as widely accepted, with only 41% of counselors reporting that they have provided pretest telephone counseling sessions (6).

To examine the acceptability and impact of telephone genetic counseling, we conducted a multisite, randomized noninferiority trial comparing telephone delivery to standard in-person delivery (ie, usual care [UC]) of pre- and post-test genetic counseling for HBOC (9,10). In short-term analyses, we found that telephone delivery was less expensive and yielded noninferior psychosocial outcomes, including knowledge, satisfaction, decisional conflict, quality of life, and distress. These data are consistent with the only other randomized noninferiority trial of telephone genetic counseling for HBOC conducted by Kinney and colleagues, in which telephone counseling was noninferior to in-person counseling in the short term and at one-year follow-up (11,12).

In this report, we present follow-up analyses focused on one-year outcome data from our trial. In contrast to the trial by Kinney and colleagues, which used a population-based recruitment strategy, we recruited a clinical sample of women who were self- or physician-referred for clinical genetic counseling and testing. As a result of these different recruitment strategies, the Kinney trial had a relatively low rate of genetic testing uptake and thus identified few *BRCA1/2* mutation carriers, while our trial had a much higher uptake rate and more mutation carriers. Thus, the present report provides novel information regarding the potential long-term noninferiority of telephone counseling within a clinical sample of women, most of whom received *BRCA1/2* genetic testing results. Specifically, we focused on testing noninferiority of telephone counseling compared with in-person counseling on psychosocial and quality of life outcomes one year following genetic counseling. We also examined the comparability of the two groups on utilization of breast and ovarian cancer risk management behaviors.

Methods

Participants

From 2005 to 2012, we recruited women for a two-armed, randomized noninferiority trial comparing telephone genetic counseling (TC) to usual care in-person genetic counseling (UC). Participants were recruited from the genetic counseling programs at Lombardi Comprehensive Cancer Center (Washington, DC), Mount Sinai School of Medicine (New York, NY), University of Vermont Cancer Center (Burlington, VT), and Dana-Farber

Cancer Institute (Boston, MA). Eligible participants were women age 21 to 85 years with a minimum 10% risk for carrying a *BRCA1/2* mutation who lived within the catchment area of a study site. We excluded women who had newly diagnosed (less than four weeks) or metastatic cancer, who lacked the cognitive capacity for informed consent, who could not communicate in English, or who were candidates for genetic counseling for another hereditary cancer syndrome.

Details of study accrual are displayed in Figure 1 and described in previous reports (9,13). From 1057 potentially eligible women, we randomly assigned 669 to TC (n = 335) vs UC (n = 334) using computer-generated random numbers in blocks of four. Of the 669 randomly assigned women, 600 completed pretest TC or UC. Of those who completed pretest genetic counseling, 512 (76.5%) completed a 12-month follow-up assessment and are included in the primary per-protocol analysis. For secondary intention-to-treat analyses, we had 12-month outcome data for 514 participants. We utilized a variety of imputation strategies (see the "Statistical Analysis" section) to impute 12-month data for the remaining 154 participants. For analyses of risk management outcomes, we excluded women who received a true negative genetic test result (n = 95) because most risk management options are not relevant to them. There were no differences in attrition across random assignment groups (χ^2 = .11, P = .735).

Procedure

The institutional review boards at all study sites approved this study. After mailing interested women an informed consent document to review, a trained research assistant called participants to administer a verbal consent and complete the baseline (precounseling) telephone survey, during which we collected demographics, cancer history, and psychosocial information. Immediately after the survey, the research assistant randomly assigned participants. After returning the signed informed consent document, UC participants received pretest genetic counseling and result disclosure at one of the four clinic sites, while TC participants received pretest genetic counseling and result disclosure by telephone. Genetic counseling was provided free of charge to participants in both arms of the study. We conducted follow-up surveys two weeks after counseling (pretest disclosure) and three, six, and 12 months after random assignment (post-test disclosure). Here, we report on the 12-month outcomes.

Usual Care

Participants randomly assigned to UC received standard, in-person *BRCA1/2* genetic counseling and result disclosure (13,14) delivered by genetic counselors at the participating study sites. Women were given the option to provide a DNA sample for genetic testing at the conclusion of the counseling session. Women who had BRCA testing were mailed a clinical summary letter with a review of cancer risks and individualized guidelines/recommendations for cancer screening and risk reduction.

Telephone Genetic Counseling

The development and content of the TC intervention are described in detail in a prior report (13). Briefly, prior to the scheduled session, we mailed visual aids to participants randomly assigned to TC. Genetic counselors at all study sites delivered genetic counseling over the telephone, providing comparable educational content as UC counseling sessions. Following counseling, participants could provide a DNA sample for genetic
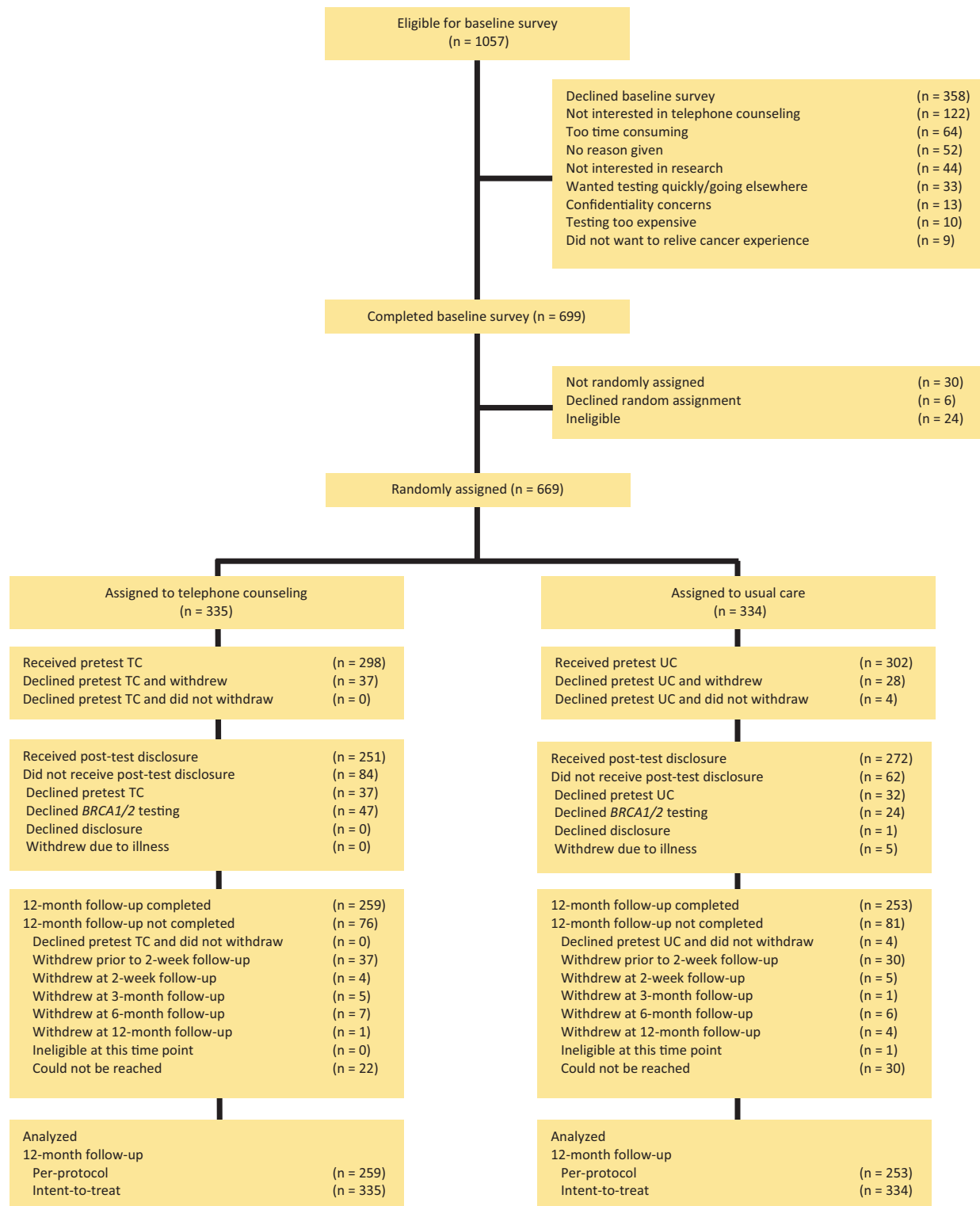
Eligible for baseline survey
(n = 1057)

Declined baseline survey	(n = 358)
Not interested in telephone counseling	(n = 122)
Too time consuming	(n = 64)
No reason given	(n = 52)
Not interested in research	(n = 44)
Wanted testing quickly/going elsewhere	(n = 33)
Confidentiality concerns	(n = 13)
Testing too expensive	(n = 10)
Did not want to relive cancer experience	(n = 9)

Completed baseline survey (n = 699)

Not randomly assigned	(n = 30)
Declined random assignment	(n = 6)
Ineligible	(n = 24)

Randomly assigned (n = 669)

Assigned to telephone counseling (n = 335)

Received pretest TC	(n = 298)
Declined pretest TC and withdrew	(n = 37)
Declined pretest TC and did not withdraw	(n = 0)

Received post-test disclosure	(n = 251)
Did not receive post-test disclosure	(n = 84)
Declined pretest TC	(n = 37)
Declined *BRCA1/2* testing	(n = 47)
Declined disclosure	(n = 0)
Withdrew due to illness	(n = 0)

12-month follow-up completed	(n = 259)
12-month follow-up not completed	(n = 76)
Declined pretest TC and did not withdraw	(n = 0)
Withdrew prior to 2-week follow-up	(n = 37)
Withdrew at 2-week follow-up	(n = 4)
Withdrew at 3-month follow-up	(n = 5)
Withdrew at 6-month follow-up	(n = 7)
Withdrew at 12-month follow-up	(n = 1)
Ineligible at this time point	(n = 0)
Could not be reached	(n = 22)

Analyzed	
12-month follow-up	
Per-protocol	(n = 259)
Intent-to-treat	(n = 335)

Assigned to usual care (n = 334)

Received pretest UC	(n = 302)
Declined pretest UC and withdrew	(n = 28)
Declined pretest UC and did not withdraw	(n = 4)

Received post-test disclosure	(n = 272)
Did not receive post-test disclosure	(n = 62)
Declined pretest UC	(n = 32)
Declined *BRCA1/2* testing	(n = 24)
Declined disclosure	(n = 1)
Withdrew due to illness	(n = 5)

12-month follow-up completed	(n = 253)
12-month follow-up not completed	(n = 81)
Declined pretest UC and did not withdraw	(n = 4)
Withdrew prior to 2-week follow-up	(n = 30)
Withdrew at 2-week follow-up	(n = 5)
Withdrew at 3-month follow-up	(n = 1)
Withdrew at 6-month follow-up	(n = 6)
Withdrew at 12-month follow-up	(n = 4)
Ineligible at this time point	(n = 1)
Could not be reached	(n = 30)

Analyzed	
12-month follow-up	
Per-protocol	(n = 253)
Intent-to-treat	(n = 334)

Figure 1. Study flow chart. TC = telephone genetic counseling; UC = usual care (in-person genetic counseling).

testing at a physician's office, a local laboratory, or a study site. Results were disclosed over the telephone, and participants were mailed a clinical summary letter with a review of cancer risks and individualized guidelines/recommendations for cancer screening and risk reduction.

Control Variables

At baseline, we assessed sociodemographics, family history, and personal cancer history. By using personal and family cancer history, we calculated a priori risk with the BRCAPRO model (15).

Outcome Variables

Distress

We measured cancer-specific distress at all assessments using the Impact of Event Scale (IES), a 15-item Likert-style scale (16). Reliability in the present study, across all time points, ranged from an α of .88 to .91. Higher scores indicate more distress.

Genetic Testing Distress

We measured genetic testing distress at the 12-month assessment using the Multidimensional Impact of Cancer Risk Assessment (MICRA) (17). This 25-item questionnaire measures distress, uncertainty, and positive experience related to genetic testing for hereditary cancers. In the present study, we used the total score but excluded the final four questions because these questions do not pertain to the whole study population. The excluded questions assessed distress related to participants' children and cancer diagnosis. Cronbach's alpha in the present study was .84.

Quality of Life

We measured mental and physical health-related quality of life at baseline and 12 months using the 12-item Short Form Health Survey (SF-12). Both the Mental Component Summary (MCS) and Physical Component Summary (PCS) are highly reliable (α = .88 to .92), with higher scores indicating better quality of life (18,19).

Satisfaction With Decision

We measured satisfaction with the genetic testing decision using the Satisfaction with Decision Scale (SWD) (20), which had a reliability α of .90 in the present study.

Risk Management Behaviors

We measured the following breast and ovarian cancer risk management outcomes with face valid questions (eg, "Since enrolling in the study, have you had a mammogram?"): risk-reducing mastectomy (RRM), risk-reducing bilateral salpingo oophorectomy (RRSO), recommended mammography, and breast MRI.

Statistical Analyses

After confirming the comparability of the groups at baseline, we tested for noninferiority of TC by calculating the group difference on each outcome (with baseline score on the specific outcome and test result as covariates) and the one-sided 97.5% confidence limit (CI) of this difference. Noninferiority was confirmed when this confidence limit did not cross the noninferiority limit. As in our prior report, we based our noninferiority limits on previous research that defined clinically important differences on outcomes (four points on the IES, 2.5 points on the SF-12 and MICRA) (16,17,21). For outcomes without guidance (SWD), we set the limit as the minimum possible change on the scale (ie, one point) (9).

Our primary analyses were based on the available sample at 12 months. In contrast to superiority trials, the use of the available (ie, per-protocol) sample for primary analyses is conventional in noninferiority trials (22–24). We also conducted sensitivity analyses in which we used a conservative mean substitution strategy to impute for missing follow-up data. For UC participants, we substituted the mean score of UC participants. For TC participants, we substituted the mean score for UC plus or minus the noninferiority margin for TC participants (25). We also explored other imputation strategies including multiple regression imputation. Finally, we conducted sensitivity analyses in which we adjusted for multiple comparisons using the Holm-Bonferroni approach (26).

Our sample size calculations for noninferiority at 12 months assumed a two-tailed alpha of .05 (ie, one-tailed 97.5% CI). Based on the final randomized sample of 669 and a 77% retention rate, ad hoc power was greater than 80% to detect noninferiority on each of our primary outcomes: satisfaction with genetic testing decision (SWD), cancer distress (IES), genetic testing distress (MICRA), and quality of life (PCS and MCS).

For secondary analyses of risk management outcomes, power was insufficient to test for noninferiority. Thus, we conducted bivariate analyses comparing TC with UC participants on the use of RRM, RRSO, and breast cancer screening tests (mammography and MRI). In these analyses, we excluded participants who received definitive negative test results (UC n = 45, TC n = 50) because risk-reducing surgery and enhanced surveillance are not recommended as options for them.

All analyses were performed using Statistical Analysis Software (SAS) version 9.4, SAS Inc. (Cary, NC).

Results

Baseline comparisons of randomly assigned participants indicated that participants in both groups were highly similar (Tables 1 and 2).

Noninferiority Outcomes

As displayed in Figure 2, at one year post–random assignment, TC was statistically noninferior to UC on all outcomes. For satisfaction with genetic testing decision, the mean adjusted satisfaction score at one year was slightly higher (0.13 points) for TC than for UC. The lower bound of the one-sided 97.5% confidence interval (–0.34) did not cross the noninferiority limit (–1). TC was also noninferior on the following outcomes: cancer distress (d = –2.10; upper bound one-sided 97.5% CI = –0.07, noninferiority limit = 4), genetic testing distress (d = –0.27; upper bound one-sided 97.5% CI = 1.46, noninferiority limit = 2.5), physical function (d = 0.44; lower bound one-sided 97.5% CI = –0.91, noninferiority limit = –2.5), and mental function (d = –0.04; lower bound one-sided 97.5% CI = –1.44, noninferiority limit = –2.5). Follow-up sensitivity analyses confirmed noninferiority for TC on all outcomes after adjusting for multiple comparisons using Holm-Bonferroni correction (26) and imputing for missing follow-up data.

Risk Management Outcomes

As displayed in Table 3, among participants with at least one intact breast at baseline, TC and UC did not differ on use of self-reported RRM (χ^2 (df = 1, n = 356) = 2.75, P = .10), and among those with intact ovaries, TC and UC did not differ on subsequent self-reported RRSO (χ^2 (df = 1, n = 343) = 1.74, P = .19). However, TC participants were more likely to obtain either RRM and/or RRSO compared with UC participants (17.8% vs 10.5%; χ^2 (df = 1, n = 402) = 4.43 P = .04). Finally, after excluding participants who had an RRM either before or during the study, the groups did not differ on the use of MRI (χ^2 (df = 1, n = 328) = .58, P = .45) or mammography (χ^2 (df = 1, n = 327) = .10, P = .75).

Because we expected differential uptake of risk management strategies based on test results, we repeated the above analyses stratified by test result. Among women who received a positive BRCA1/2 test result, TC and UC groups did not differ on the use of RRM (χ^2 (df = 1, n = 71) = .93, P = .33) or RRSO (χ^2 (df = 1, n = 69) = 1.51, P = .22). However, on the composite measure for uptake of RRM and/or RRSO, 44% of the TC group opted for RRM and/or RRSO compared with 24% of the UC group (χ^2 (df = 1, n = 77) = 3.45, P = .06). The groups did not differ on

mammography (χ^2 (df = 1, n = 63) = 1.21, P = .27) or MRI (χ^2 (df = 1, n = 63) = 2.67, P = .10).

Among noncarriers (ie, uninformative negative or variant of uncertain significance results), the groups did not differ on RRM (χ^2 (df = 1, n = 285) = 2.00, P = .16), RRSO (χ^2 (df = 1, n = 274) = .60, P = .44), the composite risk-reducing surgery uptake variable (χ^2 (df = 1, n = 325) = 2.48, P = .12), mammography (χ^2 (df = 1, n = 264) = 1.02, P = .31), or MRI (χ^2 (df = 1, n = 265) = .03, P = .86).

Discussion

We compared the long-term outcomes of patients who were randomly assigned to receive BRCA1/2 genetic counseling by telephone with the outcomes of those who were randomly assigned to receive standard in-person genetic counseling. Consistent with short-term outcomes (9), we found that TC was noninferior to UC on psychosocial, quality of life, and satisfaction outcomes. These data are consistent with the one previous randomized controlled trial reporting on 12-month outcomes of TC (12). These results are also consistent with nonrandomized studies in which women who received telephone counseling reported high satisfaction, low distress, and low regret (27) and studies indicating that mode of counseling delivery is among the least important patient concerns regarding genetic counseling delivery (28).

Taken together, these studies support the use of telegenetics to extend the reach and accessibility of genetic counseling (6–8,29). This is salient given recent evidence indicating that within a community sample the majority of women who received BRCA1/2 gene testing reported that they did not participate in pretest genetic counseling, and the absence of such counseling was associated with lower knowledge and poorer understanding of the implications of BRCA1/2 results (30). Thus, the ability of providers without access to local cancer genetic counselors to refer to telephone counseling could yield improved patient knowledge and outcomes. Access to telephone counseling may be particularly important in settings that are under-resourced. Although this study included few low socioeconomic status or ethic/racial minorities, recent evidence suggests that telegenetic counseling can be effectively extended to low–socioeconomic status patients who are uninsured or receive federal-state health insurance (31).

In our evaluation of the use of self-reported breast and ovarian cancer risk management strategies at 12-month follow-up, we found a trend for higher uptake of risk-reducing surgery among mutation carriers who completed TC compared with UC. Overall, 44% of mutation carriers in the TC group opted for RRM, RRSO, or both compared with 24% in the UC group. These

Table 1. Sample characteristics of participants

Characteristic	Usual care (n = 334) No. (%)	Telephone counseling (n = 335) No. (%)
Age, mean, SD, y	48.4 (14.2)	47.7 (13.1)
BRCA1/2 probability, mean (SD), %	25.7 (24.2)	24.3 (21.6)
Education		
< College	69 (20.7)	67 (20.0)
College or more	265 (79.3)	268 (80.0)
Employment status		
Full time	183 (54.8)	199 (59.4)
< Full time	151 (45.2)	136 (40.6)
Race		
White	289 (87.3)	280 (85.1)
Nonwhite	42 (12.7)	49 (14.9)
Marital status		
Married/partnered	212 (63.5)	205 (61.2)
Single/widowed/divorced	122 (36.5)	130 (38.8)
Jewish ethnicity		
Jewish	100 (29.9)	92 (27.5)
Non-Jewish	234 (70.1)	243 (72.5)
Affected with breast cancer		
Yes	223 (66.8)	214 (63.9)
No	111 (33.2)	121 (36.1)
Affected with ovarian cancer		
Yes	24 (7.2)	9 (2.7)
No	310 (92.8)	326 (97.3)
Proband status		
Proband	215 (64.4)	211 (63.0)
Relative of known BRCA1/2 carrier	119 (35.6)	124 (37.0)
BRCA1/2 test result		
Positive	51 (15.2)	44 (13.1)
True negative	56 (16.8)	57 (17.0)
Uninformative/variant	165 (49.4)	150 (44.8)
Untested	62 (18.6)	84 (25.1)

Table 2. Psychosocial outcomes at baseline and 12-months postcounseling

Outcome	Range of possible scores	Usual care Baseline Mean (SD)	Usual care 12 mo Mean (SD)	Telephone counseling Baseline Mean (SD)	Telephone counseling 12 mo Mean (SD)
Satisfaction with decision*	6–30	–	28.4 (2.8)	–	28.6 (2.4)
Cancer distress	0–75	19.7 (15.5)	13.1 (14.3)	22.7 (14.9)	12.6 (14.3)
Genetic testing distress*	0–105	–	17.0 (9.8)	–	16.5 (9.2)
Physical function	†	50.6 (9.1)	51.6 (9.3)	51.3 (8.6)	52.3 (7.9)
Mental function	†	49.1 (10.4)	50.3 (8.9)	48.8 (10.5)	50.2 (9.0)

*Satisfaction with decision and genetic testing distress were not administered at baseline because they only become relevant following genetic counseling and testing.
†Scores for the Physical and Mental Function subscales of the 12-item Short Form Health Survey were transformed to T-scores (ie, mean = 50 and SD = 10).
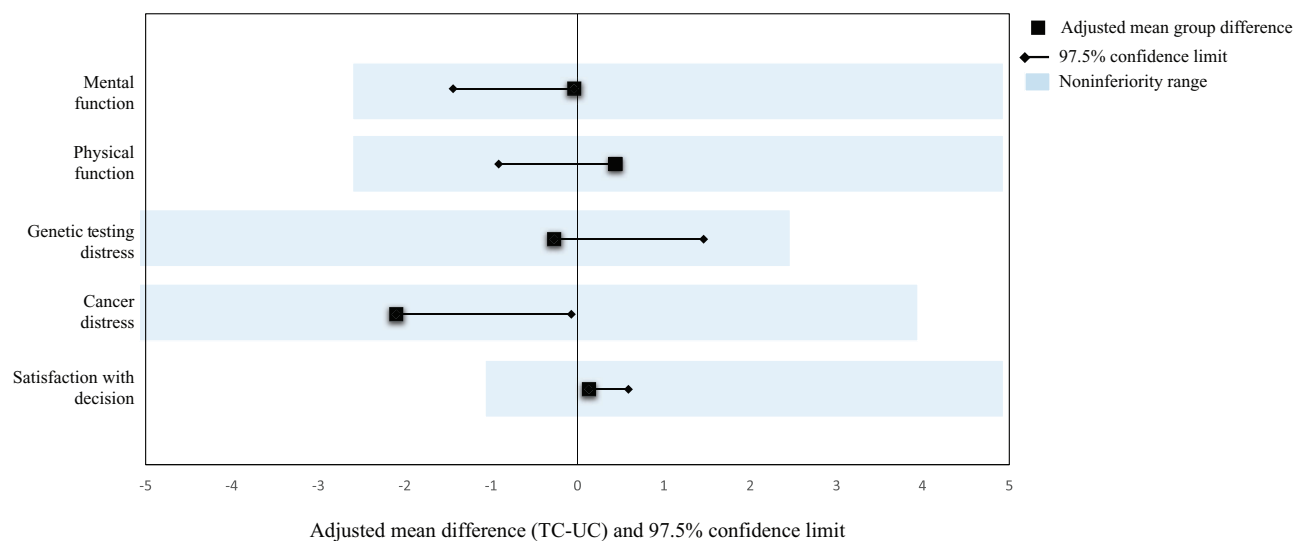
Figure 2. Adjusted noninferiority analysis of telephone genetic counseling vs usual care (in-person genetic counseling) at 12 months postcounseling. Analyses were adjusted for baseline score on the outcome measure and genetic test result. TC = telephone genetic counseling; UC = usual care (in-person genetic counseling).

Table 3. Risk management outcomes at 12 months postcounseling for TC vs UC*

Risk management outcome	Usual care No. (%)	Telephone counseling No. (%)	χ^2	P
RRM†,‡,§			2.75	.10
Yes	9 (5.0)	17 (9.6)		
No	170 (95.0)	160 (90.4)		
Mammogram‖			.10	.75
Yes	146 (86.9)	140 (88.05)		
No	22 (13.1)	19 (11.95)		
MRI§			.58	.45
Yes	55 (32.35)	45 (28.5)		
No	115 (67.65)	113 (71.5)		
RRSO¶,‡,§			1.74	.19
Yes	13 (7.7)	21 (12.0)		
No	155 (92.3)	154 (88.0)		
Risk-reducing surgery¶,‡,§			4.43	.04
Yes	21 (10.5)	36 (17.8)		
No	179 (89.5)	166 (82.2)		

*We excluded participants who received definitive negative test results from all risk management analyses (n = 95; n = 50 in TC and n = 45 in UC). RRM = risk-reducing mastectomy; RRSO = risk-reducing salpingo oophorectomy; TC = telephone genetic counseling; UC = usual care (in-person genetic counseling).
†We excluded participants who had prior bilateral mastectomy at baseline from RRM analyses (n = 59; n = 31 in TC and n = 28 in UC).
‡Participants who had both bilateral mastectomy and bilateral salpingo oophorectomy prior to baseline were excluded from risk-reducing surgery analysis (N = 11; N = 4 in TC and N = 7 in UC).
§Three participants (n = 2 in TC and n = 1 in UC) obtained both bilateral mastectomy and bilateral oophorectomy following genetic testing. These individuals are included in the analyses for RRM, RRSO, and risk-reducing surgery.
‖We excluded participants who had prior bilateral mastectomy at baseline or who obtained a bilateral mastectomy during the study from mammogram and MRI (n = 59; n = 48 in TC and n = 37 in UC).
¶We excluded participants who had prior bilateral salpingo oophorectomy at baseline from the ovarian cancer risk management analysis (n = 73; n = 33 in TC and n = 40 in UC).

results are in contrast to the lower rates of risk-reducing surgery for TC vs in-person counseling among the extremely small group of carriers in the recent study by Kinney (12) and should ease concerns that telephone delivery yields lower rates of risk-reducing surgery than standard genetic counseling (32). Our finding of increased uptake of risk-reducing surgery among carriers in the TC arm could be related to who chose to be tested within each group. As we reported previously, TC was associated with a slightly lower rate of genetic testing compared with UC (9). This difference is likely due to the fact that while UC

participants could provide DNA in clinic immediately following their counseling session, TC participants had to take additional steps to provide DNA. It is possible that TC participants who intended to use their results to guide risk-reducing surgery decisions were highly motivated to pursue the additional steps needed for testing. In contrast, UC participants may have been more likely to provide DNA regardless of surgical intentions because of the ease of doing so following their in-person session. Thus, the group of women who opted for testing following TC may have been enriched for those who were already

considering risk-reducing surgery. This is consistent with previous research indicating that baseline surgical intentions are a strong predictor of subsequent surgical decisions following a positive genetic test result (33). This explanation is also consistent with the finding that the rate of risk-reducing surgery was slightly, albeit nonsignificantly, higher for noncarriers who received TC compared with UC (12.1% vs 6.9%).

While these results support the use of telegenetics to safely extend the geographic reach and availability of genetic counseling for *BRCA1/2*, there are a few general caveats and study limitations. First, there is the paradoxical finding that telephone delivery, which is designed to enhance access, has been shown to yield slightly lower rates of genetic testing uptake across two randomized controlled trials (9,11,12). In the present study, all participants had ready access to standard genetic counseling and testing. Thus, the full benefit of the telephone approach in reaching individuals without such access was not realized in this trial. It is possible that telephone delivery could increase genetic testing when employed in settings or populations with limited access to traditional genetic counseling and testing resources. However, this remains an open question requiring additional research. Second, the study of telephone genetic counseling in more diverse populations is essential given the low rate of minority participants in this study. Third, we did not collect data on study decliners, preventing us from examining factors related to willingness to participate in TC. Fourth, with increasing use of multigene panels as a firstline genetic testing approach (1,34,35), the complexity of genetic counseling has increased since this trial. Whether telephone delivery is effective in the multiplex testing context is an open question. Finally, our evaluation of risk management outcomes relied on self-report measures and could be improved in future studies with verification of clinical records.

Despite these limitations, there is now strong evidence from multiple studies that patients who receive genetic counseling over the telephone have short- and long-term outcomes that are no worse than patients who receive in-person genetic counseling. Further, there is no evidence that telephone delivery yields lower rates of uptake of recommended risk-reducing surgery or breast cancer screening among mutation carriers. Genetic counselors, referring physicians, payers, and policymakers can be reassured that providing *BRCA1/BRCA2* genetic counseling by telephone is a safe and effective approach to patient care.

Funding

Notes

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