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ORIGINAL ARTICLE

Telehealth for genetic counseling: A systematic evidence review

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Abstract

Telehealth options, such as telephone counseling or videoconferencing, for service delivery in genetic counseling are becoming more widely accepted. However, until now, there has not been a systematic review of the literature focused specifically on genetic counseling outcomes for telehealth. We performed a systematic evidence review to compare telehealth genetic counseling (THGC), including videoconferencing and telephone counseling, across specialties to in-person genetic counseling (IPGC) for a range of outcomes specific to patient and provider experiences and access to care. Several biomedical databases were queried up to January 11, 2021, to identify original research evaluating THGC. Through this search, 42 articles met the inclusion criteria including 13 randomized controlled trials and 29 non-randomized observational studies encompassing 13,901 patients. Most included studies focused only on cancer genetic counseling; however, adult, pediatric, and prenatal specialties were also represented. The majority of studies evaluated patient and/or access to care outcomes. Though most studies reported high patient satisfaction with THGC, as well as comparable rates of trust and rapport, confidence in privacy, health behavior changes, and psychosocial outcomes, few represented diverse populations. Data of provider experiences were limited and varied with more disadvantages noted compared with patient experiences, particularly in studies involving telephone genetic counseling. Studies consistently reported a decrease in the patients' costs and time required for travel when patients are seen via THGC compared to IPGC with a similar reduction in costs to the health system. Overall, results from our evidence synthesis suggest THGC is non-inferior or comparable to IPGC across many domains, even considering that many of the studies included in this review were conducted with telehealth systems, notably videoconferencing, that were less robust and reliable than what is available today. There are notable limitations within this body of literature, leading to potential uncertainty in the generalizability of our analysis. We outline several recommendations for future studies.

KEYWORDS

genetic counseling, service delivery models, systematic review, telegenetics, telehealth, telemedicine, telephone genetic counseling

1 | INTRODUCTION

Traditionally, genetic counselors (GCs) have delivered in-person genetic counseling (IPGC). Even so, GCs have often found alternative and innovative ways to deliver genetic counseling services when needed. Decades ago, GCs adopted telehealth by providing telephone genetic counseling (TGC) as part of local teratogen services (Ormond et al., 2000). With the advent of new technologies, some GCs began pivoting to Web-based and telemedicine genetic counseling, termed telegenetics, to provide comprehensive genetic counseling remotely via videoconferencing (Cohen et al., 2012; Greenberg et al., 2020; Stoll et al., 2018). To reduce confusion in this review between terms TGC, telegenetics, and telemedicine, we use video genetic counseling (VGC) in place of either telegenetics or telemedicine when referring to the practice of genetic counseling via videoconferencing. VGC has now been used across all medical genetics' specialties, including but not limited to prenatal, oncologic, and cardiovascular settings (Greenberg et al., 2020). The use of these alternative service delivery models (SDMs) to IPGC has at times been a necessity and, more recently, to answer the growing call for greater accessibility to GCs as genetic and genomic testing rapidly increases along with consumer demand and interest (Greenberg et al., 2020). Alternative genetic SDMs also help reduce barriers to access to care that include time and geographic location (Stoll et al., 2018). This is especially true for those living in rural areas that are distant from major medical centers.

The pressing question is how effective these alternative models are and, specifically, how patient outcomes compare with IPGC. Individual studies have evaluated outcomes (including but not limited to patient knowledge, patient and provider satisfaction, psychosocial outcomes, test uptake, cost, convenience, and access to care) to compare TGC and/or VGC with IPGC. While several studies concluded that TGC and/or VGC are non-inferior to IPGC, a systematic review focused on genetic counseling outcomes of both TGC and VGC has not been published. Existing systematic reviews of telehealth in genetics have limited use for informing the practice of either TGC or VGC. The earliest review by Hilgart et al. (2012) focused on evaluating genetic services, but not strictly genetic counseling. This was followed a few years later by Vrečar et al. (2017), who reviewed the availability and use of videoconferencing in clinical genetics. However, both Hilgart et al. and Vrečar et al. included studies where the genetic counselor was in-person with the patient and the physician geneticist connected via video. While this is a valuable SDM for pediatric genetics and improves access to genetic services for many in rural communities, this model is different from those in which the patient is interacting with a genetic counselor via either telephone or live interactive video. Bracke et al. (2021) completed a systematic review that focused on TGC; however, their review was limited only to TGC for *BRCA1/BRCA2* genetic testing compared with IPGC. Our review was prompted by the lack of a systematic review of the literature focused on both TGC and VGC compared with IPGC across multiple genetic counseling specialties to inform expert recommendations for the genetic counseling profession.

What is known about this topic

Several primary research studies have compared outcomes related to aspects of telehealth (telephone or videoconferencing) genetic counseling to traditional in-person genetic counseling. Existing systematic reviews of telehealth in genetics either focus on the physician connecting remotely, but not the genetic counselor, or have limited the review to only telephone genetic counseling compared with in-person genetic counseling.

What does this paper add to this topic

This systematic evidence review provides a comprehensive assessment of the existing literature on the effects of telehealth genetic counseling on patient and provider outcomes and access to care.

In addition to the need for this focused review, the data provided by the studies ideally should be synthesized and translated into detailed and practical guidance for GCs who provide these services. This has become more urgent due to the coronavirus disease (COVID-19) pandemic, as many GCs have transitioned to providing care via telehealth without a clear understanding of the impact on patients. Even prior to the COVID-19 pandemic, it was estimated that 12.5% and 6.7% of GCs utilized TGC or VGC 'always or often', respectively, and this number has likely increased (Greenberg et al., 2020). With this rapid shift in practice, it is imperative to have an up-to-date review of the relevant literature addressing genetic counseling outcomes related to alternative modes of service delivery, as telehealth options are quickly becoming 'standard of care'. In this systematic review, we reviewed the literature, paying particular attention to studies published since the systematic review by Hilgart et al. (2012), point out gaps in the published literature, and provide an evidence base for genetic counseling practice and protocol related to telehealth. We chose to focus on the two most common virtual telehealth options currently used by genetic counselors: TGC and VGC. Considering both options together, we refer to these as telehealth genetic counseling (THGC).

2 | METHODS

This systematic review followed applicable principles described in the Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) statement (Page et al., 2021). A detailed methods section, including PICOTS, search strategy for the OVID platform, inclusion/exclusion criteria, detailed outcomes, and information pertaining to data extraction, can be found in the Supplemental Document.

We sought to extract relevant data comparing outcomes of THGC and IPGC. Data were generally categorized as study-specific

information (i.e., study design, years of follow-up, setting, indication for THGC, and population characteristics), patient experience outcomes, provider experience outcomes, and access to care. Where possible, we delineated the THGC method used (i.e., TGC or VGC) (Table 1).

2.1 | Data analysis

Descriptive statistics (e.g., raw counts, means, standard deviations, ranges, and percentiles) were used to describe the general characteristics of the included articles. A random-effects inverse variance method meta-analysis was performed in R (version 3.3.1) using the 'meta' package for uptake of genetic testing and mean patient satisfaction reported in the included randomized control trial (RCT) outcomes. The remaining outcomes were described using descriptive statistics and narratively synthesized. We specifically sought to identify and characterize differences in outcomes between population subgroups, including affected versus unaffected patients, race/ethnicity categories, and socioeconomic levels, and compare and contrast VGC and TGC to each other and to IPGC.

3 | RESULTS

The initial literature search was performed on July 3, 2018, and updated on June 24, 2019, and January 11, 2021. A total of 947 non-duplicated articles were screened for inclusion, and 132 full-text articles were selected to be reviewed in their entirety. After review, 42 articles remained for data extraction and quality assessment (Figure 1). We included 13 RCTs and 29 non-randomized observational studies encompassing 13,901 patients. Where more than one publication reported results using the same population, we preferentially analyzed those reported in the most recent publication or in the study that described the longest term follow-up period. Study populations ranged from 10 to 4,776 participants; 31 of 42 (74%) of the included studies had fewer than 500 participants combined between THGC and IPGC arms, and 21 had fewer than 150 participants. The majority of studies ($n = 27$) focused only on cancer genetic counseling and included predominantly female participants, except a single study with all male participants from the Veterans Health Administration (Voils et al., 2018). Five studies considered the role of genetic counseling for either mixed adult/pediatric or pediatric populations (Burgess et al., 2016; Greenberg et al., 2020; Lea et al., 2005; Shur et al., 2021; Stalker et al., 2006); three studies evaluated the impact of counseling in neurological or psychiatric clinics: Alzheimer's disease (Christensen et al., 2018), Huntington disease (Hawkins et al., 2013), and general psychiatry (Gerrard et al., 2020). Eight studies included participants and patients seen for prenatal/preconception genetic counseling (Arjunan et al., 2020; Burgess et al., 2016; Jeganathan et al., 2020; Lea et al., 2005; Otten et al., 2016a, 2016b; Pagliazzi et al., 2020; Sangha et al., 2003).

A brief summary of all included studies is provided in Table 1, while a more comprehensive summary table is provided in Table S5. Results in studies of TGC and VGC compared with IPGC were substantially similar, and they are presented together as THGC, with specific examples noted where differences were observed or to draw attention to the result.

3.1 | Patient experience

3.1.1 | THGC compared with IPGC

Thirty-six studies reported outcomes pertaining to the patient experience (Table 2). THGC was generally non-inferior or not significantly different when compared to IPGC for patient satisfaction; trust in privacy and confidentiality; uptake of, or intention to pursue, risk-reducing procedures, magnetic resonance imaging (MRI), screening mammography, or genetic testing; knowledge; psychosocial topics of anxiety, depression, and stress; decision-making; and quality of life. After an experience with one of the modes of delivery, preference for an alternate mode of delivery was 4%–37% for IPGC after THGC, 14%–41% for THGC after IPGC, and 27%–33% for IPGC after VGC (Table 2). Within the limited evidence published since 2020, a majority (73.8%) of patients desired a mix of THGC and in-person appointments for high-risk obstetrical care that included genetic counseling (Jeganathan et al., 2020).

Despite the overall comparability of THGC to IPGC, there were some exceptions. In exploratory analyses by Patrick-Miller et al. (2013) of a cancer population, while overall satisfaction was high in both IPGC and TGC arms, from baseline to post-result disclosure, the improvement in satisfaction scores was lower in African American participants compared with white participants. In a study of patients at high risk for breast or ovarian cancer, patients randomized to TGC felt less supported by their GC than patients randomized to IPGC (52.9% vs. 66%, $p = .002$) (Peshkin et al., 2016). Predictors of feeling supported by the GC included the following: race/ethnicity, age, cancer-specific distress, perceived stress, decisional conflict, and both physical and mental health. In exploratory analyses by Peshkin et al. (2016), non-Hispanic white patients were observed to feel more supported with IPGC, while racial/ethnic minority patients reported feeling more supported with the TGC sessions than with IPGC (58.3% vs. 38.7%; OR = 0.80, 95% CI [0.39, 1.65]); however, this difference was not statistically significant. Further, a significant increase in patient-reported empowerment as measured by Genetic Counseling Outcome Scale (GCOS) scores was identified in IPGC compared with THGC in patients undergoing psychiatric genetic counseling (Gerrard et al., 2020).

Uptake of genetic testing after either TGC or VGC was comparable, though slightly lower than the uptake after IPGC (Figure S1). Individuals with a personal history of cancer and/or those with a higher level of distress and/or higher level of perceived risk were less likely to pursue genetic testing after TGC compared with IPGC (Steffen et al., 2017). Additionally, patients receiving either THGC or IPGC with economic concerns related to the costs associated with genetic testing were less likely to pursue genetic testing compared with patients without cost concerns (Steffen et al., 2017). Similarly, those who faced additional burdens due to travel to a clinic to provide a sample after THGC, such

TABLE 1 Brief summary of all included articles in review ($N = 42$)

Study	Indication for genetic counseling	Appointment type	Service delivery	Sample size	Outcomes		
					Patient	Provider	Access
Arjunan et al. (2020)	Prenatal	Results	TGC	4,776	✓		✓
Baumanis et al. (2009)	Cancer	Results	IPGC TGC	133	✓		
Bradbury et al. (2011)	Cancer	Initial visits, Results, Ongoing	TGC	194	✓	✓	
Bradbury et al. (2016)	Cancer	Initial visits, Results	VGC	61	✓		✓
Bradbury et al. (2018)	Cancer	Results	IPGC TGC	608	✓		
Buchanan et al. (2015)	Cancer	Initial visits	IPGC VGC	130	✓		✓
Burgess et al. (2016)	Adult, Cancer, Pediatrics, Prenatal, Other	Other, NR	IPGC TGC	88[a]		✓	
Butrick et al. (2015)	Cancer	Initial visits, Results	IPGC TGC	669[b]	✓		
Chang et al. (2016)	Cancer	Initial visits, Results	IPGC TGC	771			✓
Christensen et al. (2018)	Adult, Other	Results	IPGC TGC	257	✓	✓	✓
Coelho et al. (2005)	Cancer	Initial visits	IPGC VGC	37	✓		
Cremin et al. (2020)	Cancer	Initial visits, Pretest	IPGC TGC VGC	235	✓		
Fenton et al. (2018)	Cancer	Results	TGC Other	117	✓		
Gerrard et al. (2020)	Psychiatric	Initial visits, Follow-up visits	IPGC TGC VGC	307	✓		
Greenberg et al. (2020)	Adult, Cancer, Pediatrics, Prenatal,	NR	IPGC TGC VGC	517[a]			✓
Hawkins et al. (2013)	Adult, Other	Initial visits, Follow-up visits (ongoing care), Follow-up visit for results	IPGC TGC VGC	28	✓		✓
Helmes et al. (2006)	Cancer	Initial visits	IPGC TGC Other	340	✓		
Interrante et al. (2017)	Cancer	Initial visits, Results	IPGC TGC	512[b]	✓		
Jacobs et al. (2016)	Cancer	Initial visits	IPGC TGC	479		✓	
Jeganathan et al. (2020)	Prenatal	Initial visits, Follow-up visits	TGC VGC	91[a]	✓	✓	✓
Jenkins et al. (2007)	Cancer	Initial visits, Results	IPGC TGC	102	✓		✓
Kinney et al. (2016)	Cancer	Initial visits, Results	IPGC TGC	988	✓		
Lea et al. (2005)	Adult, Cancer, Pediatrics, Prenatal	Initial visits	VGC	105	✓	✓	✓
Meropol et al. (2011)	Cancer	Initial visits	VGC	31	✓		

(Continues)

TABLE 1 (Continued)

Study	Indication for genetic counseling	Appointment type	Service delivery	Sample size	Outcomes		
					Patient	Provider	Access
Mette et al. (2016)	Cancer	Initial visits, Results	IPGC VGC	119	✓		
Otten et al. (2016a)	Cancer, Prenatal, Other	Initial visits, Results	VGC	10[a]		✓	
Otten et al. (2016b)	Cancer, Prenatal, Other	Initial visits, Results	IPGC VGC	128	✓		
Pagliazzi et al. (2020)	Pediatrics, Prenatal	Initial visits, Results, Follow-up visits	IPGC TGC VGC	288	✓		✓
Patrick-Miller et al. (2013)	Cancer	Results	TGC	94	✓		
Patrick-Miller et al. (2014)	Cancer	Results	TGC	86	✓	✓	
Peshkin et al. (2016)	Cancer	Initial visits	IPGC TGC	554	✓		✓
Platten et al. (2012)	Cancer	Initial visits	IPGC TGC	215	✓		
Sangha et al. (2003)	Prenatal	Initial visits	IPGC TGC	24	✓		
Schwartz et al. (2014)	Cancer	Initial visits, Results	IPGC TGC	552[b]	✓		✓
Shanley et al. (2007)	Cancer	Initial visits	TGC	840	✓		✓
Shur et al. (2021)	Pediatrics	Initial visits, Follow-up visits	IPGC VGC	288	✓	✓	✓
Solomons et al. (2018)	Cancer	Initial visits, Results	IPGC VGC	174	✓		✓
Stalker et al. (2006)	Pediatrics	Initial visits	VGC	50	✓		✓
Steffen et al. (2017)	Cancer	Initial visits, Results, Ongoing	IPGC TGC	781	✓		
Sutphen et al. (2010)	Cancer	Initial visits, Results	TGC	22	✓		
Terry et al. (2019)	Adult, Cancer, Neurology, Pediatrics, Preconception, Prenatal, Other	Initial visits, Follow-up visits, Research Result	TGC VGC	51[a]		✓	✓
Voils et al. (2018)	Cancer	Initial visits	TGC VGC	27	✓	✓	✓
Totals				13,901 patients	36	11	19

Abbreviations: IPGC, in-person genetic counseling; NR, not reported; TGC, telephone genetic counseling; VGC, video genetic counseling.

[a]Not included in patient count, *N* reflects number of GCs.

[b]Patient total reflects removal of duplicate populations in Schwartz et al. (2014), Butrick et al. (2015) and Interrante et al. (2017).

as patients in rural settings, were also less likely to pursue genetic testing compared with patients who received IPGC with concomitant sample procurement (Bradbury et al., 2016; Kinney et al., 2016).

3.1.2 | VGC compared with TGC

Two studies reported comparisons between TGC and VGC. In a study of men with cancer receiving genetic counseling comparing VGC ($n = 9$) and TGC ($n = 18$), patients who received VGC had higher mean satisfaction scores and knowledge scores (Voils et al., 2018). VGC was also preferred over TGC in Italy by both patients seen for either prenatal or pediatric indications and their providers (Pagliazzi et al., 2020).

In general, outcomes for patients who underwent THGC were comparable to those who underwent IPGC. Uptake of genetic testing was slightly lower in patients who underwent THGC, and patient satisfaction and trust were reduced in some subgroups, though uptake of health behaviors related to reduction of cancer risk and intent to pursue genetic testing were similar. Regardless of the delivery

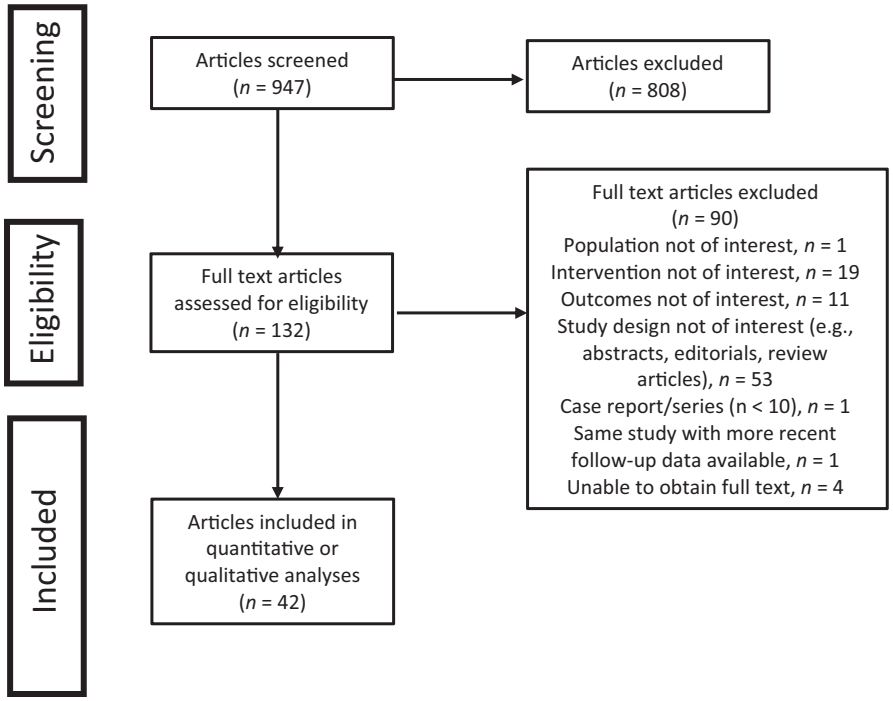

FIGURE 1 PRISMA flowchart. Articles were screened and reviewed for inclusion; exclusion rationales are provided for studies excluded at the full-text review phase

method used, genetic counseling was observed to improve patient knowledge and patient satisfaction was generally high. Patient preference for delivery of genetic counseling by an alternate method (i.e., IPGC for those receiving THGC, and THGC for those receiving IPGC) was similar in the studies that evaluated preference; however, there was significant variability between individual studies. The single study that directly compared TGC with VGC reported better, but not statistically significant results with VGC (Voils et al., 2018).

3.2 | Provider experience

3.2.1 | THGC compared with IPGC

Eleven studies reported outcomes pertaining to the provider experience (Table 3). Genetic counseling providers in diverse clinical settings (i.e., adult and pediatric; neurogenetics/neurology, prenatal/preconception, genetics/metabolic, and cancer) were largely very satisfied with THGC (Jeganathan et al., 2020; Lea et al., 2005; Shur et al., 2021). Despite high provider satisfaction with THGC, provider preference for IPGC was higher than those preferring THGC visits (56% vs. 23%, $p = .024$) (Jeganathan et al., 2020).

Two studies reported positive perceptions of VGC and/or TGC among providers. Lea et al. (2005) noted that decision confidence was high with VGC: It was rated on a 6-point scale and results ranged from 5.29 to 5.81, and all GCs felt the session was successful and addressed the problem correctly. While GCs' perceptions of the counseling session were comparable for TGC and IPGC arms, the participant race/ethnicity was a significant predictor of GC perceptions and counselors' scores on the Genetic Counseling Process Questionnaire (GCQ) were lower with non-white participants in both TGC and IPGC (Jacobs et al., 2016).

Reported and perceived disadvantages of THGC were centered on technical limitations and reduced communication, understanding/knowledge, and delivery of appropriate psychosocial care compared with IPGC (Table 3). Licensure between states was a top concern for GCs who provide cross-state THGC (Terry et al., 2019).

Three studies reported varied outcomes pertaining to workflow (Table 3). GCs in the Patrick-Miller et al. (2014) study noted that with TGC, they observed improvements to patient scheduling, decreased travel, and ease of access to resources for providers during visits. Comparing TGC with IPGC, GCs reported workflow differences with TGC related to coordinating genetic testing (Burgess et al., 2016). Interpretation of these differences may differ based on the specific outcomes desired by the providers. For example, session duration for genetic testing disclosure in an Alzheimer's clinic was 6.6 min shorter for patients receiving TGC compared with those receiving IPGC ($p < .001$) (Christensen et al., 2018). This could be viewed as a positive by some providers or a negative by others.

3.2.2 | VGC compared with TGC

Voils et al. (2018) reported that genetic counselors found both VGC and TGC acceptable but found more advantages to VGC. These included fewer missed appointments, the ability of the GC to more accurately read body language and fewer distractions. No other study directly compared or reported a direct comparison of TGC to VGC for provider-centric outcomes (Table 3).

In general, studies reporting on the provider experience were quite varied, with few reporting on the same outcomes. Surveyed

TABLE 2 Patient-centric outcomes reported in included studies

Outcome	Evidence[a]
Decision-making	
Kinney et al. (2016)	12-month mean score (*SD*): TGC 26.76 (18.1); IPGC 26.88 (18.7); no significant difference
Interrante et al. (2017)	No significant difference in satisfaction with decision-making reported with TGC vs. IPGC
Health behaviors	
Helmes et al. (2006)	Intent to pursue genetic testing (a) TGC vs. IPGC mean difference = 0.09, CI[b] [−0.02, 0.20], $p = .107$ (b) interventions vs. other (no counseling) mean difference = 0.52, CI[b] [0.43, 0.62], $p < .001$
Mette et al. (2016)	Reported for all participants (VGC and IPGC): 34% made change in lifestyle; 38% increased screening frequency
Interrante et al. (2017)	TGC and IPGC groups did not differ on use of self-reported RRM, $p = .10$; self-reported RRSO, $p = .19$; TGC participants more likely to obtain either RRM and/or RRSO compared with IPGC participants, $p = .04$; groups did not differ on use of MRI, $p = .75$
Bradbury et al. (2018)	Difference in change between groups (TGC minus IPGC) with 95% CI in brackets: planning screening mammogram = +0.13, [−0.19, +0.44]; planning screening breast MRI = +0.03, [−0.45, +0.52]; planning prophylactic mastectomy = +0.06, [−0.40, +0.52]; planning prophylactic oophorectomy = −0.04, [−0.54, +0.45]; planning colonoscopy = +0.03, [−0.51, +0.56]; no *p* values provided
Knowledge	
Sangha et al. (2003)	Mean (*SD*) knowledge score (maximum of 9 points): TGC 8.3 (1.2) vs. IPGC 8.3 (0.5); no *p* value provided.
Coelho et al. (2005)	Knowledge score pre-counseling vs. post-counseling: TGC 4.12 to 4.56 vs. IPGC 4.95 to 5.29
Jenkins et al. (2007)	Mean (*SD*) knowledge scores: baseline: TGC 7.83 (2.08) vs. IPGC 8.12 (1.7); post-education/counseling: TGC 9.79 (0.46) vs. IPGC 9.68 (0.71); 12 months: TGC 9.38 (0.91) vs. IPGC 9.39 (0.83)
Sutphen et al. (2010)	Prior to TGC, most rated knowledge as poor/very poor; post-counseling 90%+ self-reported improved understanding about risks, genetic testing knowledge, risk reduction and screening options
Hawkins et al. (2013)	Perceived knowledge of predictive testing for HD (on 5-point Likert scale where 1 = 'strongly disagreed' and 5 = 'strongly agreed'): TGC 3.93 vs. IPGC 4.58, $p = .074$
Patrick-Miller et al. (2013)	Mean score (*SD*) *for* TGC ($n = 86$): baseline 26.5 (3.0) vs. post-telephone disclosure 26.9 (3.2), $p = .25$; IPGC ($n = 57$): baseline 26.3 (3.0) vs. in-person clinic follow-up 27.2 (2.7), $p = .04$
Schwartz et al. (2014)	Mean score (*SD*) for TGC vs. IPGC at baseline 17.3 (4.8) vs. 17.0 (4.8), respectively; at 2 weeks 20.2 (3.9) vs. 20.1 (3.9), respectively; TGC non-inferior to IPGC based on adjusted mean group differences and 97.5% confidence limit.
Buchanan et al. (2015)	BCGKS mean scores (*SD*): TGC ($n = 43$) 13 (5) vs. IPGC ($n = 57$) 15 (4), $p = .08$
Bradbury et al. (2016)	Mean score (*SD*) for VGC: baseline 20.96 (2.74) vs. V1 22.07 (2.99), $p = .005$; baseline 21.20 (3.16) vs. V1 22.14 (3.16) vs. V2 21.61 (3.16), $p = .08$
Bradbury et al. (2018)	Mean change (*SD*) from baseline to post-disclosure: TGC +0.20 (2.24) vs. IPGC +0.52 (2.58); TGC non-inferior to IPGC.
Christensen et al. (2018)	Full recall (all items recalled correctly): TGC 50.1% vs. IPGC 37.3%, *OR* = 1.7, 99% CI [0.8, 3.4]
Fenton et al. (2018)	Recall for all participants: 75% recalled their test result; not significantly related to delivery model.
Solomons et al. (2018)	Remote pre- vs. post-mean score (*SD*): 3.7 (2.25) vs. 6.6 (1.54), $p < .0001$; IPGC pre- vs. post-mean score (*SD*): 4.1 (2.18) vs. 6.9 (1.63), $p < .0001$; VGC vs. IPGC pre- vs. immediate post-difference, $p = .85$; 1 month post-mean score (*SD*) for VGC ($n = 31$) 5.9 (2.08) vs. IPGC ($n = 18$) 6.9 (1.51).
Voils et al. (2018)	Pre-THGC, VGC average correct 5.6/8 questions, TGC average correct 4.4/8 knowledge questions. After THGC, VGC average correct 6.5/8 questions, TGC average correct 4.7/8 questions. Correlation between pre- and post-differences in knowledge and numeracy was $r = 0.3$, $p = .2$
Preference for Service Delivery Model	
Helmes et al. (2006)	TGC: 37.0% preferred IPGC; IPGC: 14.9% preferred TGC

(Continues)

TABLE 2 (Continued)

Outcome	Evidence[a]
Jenkins et al. (2007)	TGC: 4% preferred IPGC; IPGC: 41% preferred TGC
Baumanis et al. (2009)	TGC: 10% would prefer IPG; IPGC: 14% would prefer TGC
Sutphen et al. (2010)	TGC: 13.6% would have preferred IPGC results
Meropol et al. (2011)	VGC: 29% would have preferred IPGC
Hawkins et al. (2013)	Preferred more in-person visits at the HD testing center during the predictive testing process: THGC 2.13 vs. IPGC 2.4 ($p = .548$); preferred more telehealth appointments visits at the HD testing center during the predictive testing process: TGC 2.63 vs. IPGC 2.00 ($p = .186$) (average scores based on 5-point Likert scale, where 1 = 'strongly disagreed' and 5 = 'strongly agreed')
Peshkin et al. (2016)	Overall: 19.1% of all patients would have preferred a different delivery method
Fenton et al. (2018)	TGC: 6% would have liked to receive results in a different way; non-TGC: 18% would have liked to receive their results in a different way
Solomons et al. (2018)	TGC: 32% would prefer IPGC
Jeganathan et al. (2020)	73.8% of patients desired a mixture of both in-person and telehealth
Pagliazzi et al. (2020)	Both patients and providers reported a greater appreciation of VGC over TGC
Psychosocial	
Sangha et al. (2003)	Anxiety: Mean scores (*SD*) based on the Verbal Anxiety Survey for pre-counseling [maximum of 20 points where the highest score is associated with the highest anxiety]: TGC 14.5 (3.1) vs. IPGC 5.8 (2.8); post-counseling [maximum of 40 points where the highest score is associated with the highest anxiety]: TGC 17.7 (3.8) vs. IPGC 20 (5.5); no *p* value provided.
Coelho et al. (2005)	Cancer anxiety: Mean scores (*SD*) VGC pre-counseling 17.17 (4.12) to post-counseling 11.08 (3.08), $p = .00$ vs. IPGC pre-counseling 15.7 (4.28) to post-counseling 12.45 (2.66), $p = .01$
Helmes et al. (2006)	Cancer worry: TGC vs. IPGC, $p = .919$; TGC and IPGC vs. control, $p = .001$
Jenkins et al. (2007)	Anxiety mean scores (*SD*) for TGC vs. IPGC, respectively: Trait anxiety baseline 30.54 (6.81) vs. 30.27 (7.48); 3 months: 28.67 (7.28) vs. 28.77 (7.60), state anxiety baseline: 30.12 (10.84) vs. 27.8 (7.75); 3 months: 29.41 (9.19) vs. 27.81 (9.26), GT distress: IES baseline 11.96 (14.18) vs. 8.14 (8.44); 12 months: 5.83 (8.98) vs. 6.10 (8.60)
Patrick-Miller et al. (2013)	Mean score (*SD*) for: HADS Anxiety: TGC ($n = 86$) baseline 6.6 (4.2) vs. post-telephone disclosure 6.4 (5.0), $p = .60$; IPGC ($n = 57$) 6.1 (3.9) vs. post-in-person clinic follow-up 5.0 (4.3), $p = .01$; state anxiety: TGC ($n = 86$) baseline 35.2 (13.1) vs. post-telephone disclosure 33.0 (12.6), $p = .03$; IPGC ($n = 57$) baseline 34.2 (11.7) vs. post-in-person clinic follow-up 34.1 (10.0), $p = .90$; HADS depression: TGC ($n = 86$) baseline 3.1 (3.0) vs. post-telephone disclosure 3.7 (4.2), $p = .15$; IPGC ($n = 57$) baseline 3.0 (2.7) vs. post-in-person clinic follow-up 2.6 (2.0), $p = .09$; exploratory analysis: women with higher general anxiety and depression scores post-telephone disclosure were less likely to return for in-person clinic follow-up.
Schwartz et al. (2014)[c]	Mean scores (*SD*) for TGC vs. IPGC respectively for cancer distress (IES) at baseline 23.2 (15.1) vs. 20.7 (15.5); 3 months: 14.8 (14.9) vs. 14.0 (14.7); perceived stress at baseline 4.4 (2.4) vs. 4.5 (2.6); 3 months: 3.9 (2.6) vs. 4.0 (2.5). TGC non-inferior to IPGC based on adjusted mean group differences and 97.5% confidence limit.
Bradbury et al. (2016)	Mean scores (*SD*) for general anxiety (range 0–21) completed V1 only: baseline 7.34 (4.00) vs. V1 6.37 (3.99), $p = .003$; completed V1 and V2: baseline 6.67 (3.82) vs. V1 5.59 (3.77) vs. V2 5.54 (3.50), $p = .003$; state anxiety (range 20 – 80) completed V1 only: baseline 37.49 (13.82) vs. V1 36.49 (12.71), $p = .32$; completed V1 and V2: baseline 35.32 (12.97) vs. V1 34.42 (12.26) vs. V2 33.29 (11.10), $p = .27$; general depression (range 0–21) completed V1 only: baseline 3.70 (3.77) vs. V1 3.33 (3.26), $p = .046$; completed V1 and V2: baseline 3.33 (3.43) vs. V1 3.18 (3.22) vs. V2 2.58 (3.23), $p = .01$; cancer worry (range 0–70) completed V1 only: baseline 17.93 (13.06) vs. V1 16.63 (13.21), $p = .36$; completed V1 and V2: baseline 17.10 (13.29) vs. V1 14.76 (12.02) vs. V2 16.88 (13.71), $p = .25$
Kinney et al. (2016)	Anxiety: change from baseline: −0.01, 95% CI [−0.28, 0.24] vs. −0.09, 95% CI [−0.35, 0.18], p = non-inferiority range
Otten et al. (2016b)	Anxiety: STAI change pre to post 0.10 ($SD = 0.43$, $p = .132$) vs. 0.10 ($SD = 0.35$, $p = .033$); no difference between VGC and IPGC

(Continues)

TABLE 2 (Continued)

Outcome	Evidence[a]
Interrante et al. (2017)	Mean scores (*SD*) for TGC vs. IPGC respectively for cancer-specific distress (IES mean scores, range 0–75) at baseline 22.7 (14.9) vs. 19.7 (15.5) and at 12 months 12.6 (14.3) vs. 13.1 (14.3); GT distress (MICRA,[d] mean scores, range 0–105) at 12 months 16.5 (9.2) vs. 17.0 (9.8)
Bradbury et al. (2018)	Mean change from baseline to post-disclosure (*SD*) for TGC vs. IPGC. State anxiety −0.39 (8.92) vs. −0.06 (10.18), non-inferior; general anxiety −0.48 (2.54) vs. −0.26 (2.70), non-inferior; depression: +0.08 (2.29) vs. +0.06 (2.30), non-inferior; cancer-specific distress +0.21 (11.14) vs. +1.48 (10.24), non-inferior; for all outcomes, no difference in patients who received multigene testing vs. those who received standard testing
Christensen et al. (2018)	TGC vs. IPGC scores with 99% CI in brackets: anxiety: BAI 3.5 vs. 3.6; diff −0.2, [−1.5, 1.2]; depression: CES-D: 7.4 vs. 6.2; diff 1.1, [−1.2, 3.4]; GT distress: IGT distress 4.2 vs. 4.4, diff −0.3, [−2.7, 2.2]
Solomons et al. (2018)	Mean scores (*SD*) for anxiety measured on the PHQ−4 (range 0–6): pre-counseling vs. immediate post-counseling, VGC 1.42 (1.84) vs. 1.2 (1.54), $p = .07$; IPGC 1 (1.25) vs. 0.8 (1.14), $p = .004$; VGC vs. IPGC pre- vs. immediate post-change difference: $p = .59$; One-month post-counseling: VGC ($n = 40$) 0.82 (1.37) vs. IPGC ($n = 24$) 0.58 (0.77); depression measured on the PHQ-4 (range 0–6): pre-counseling vs. immediate post-counseling, VGC 0.90 (1.53) vs. 0.67 (1.31), $p = .02$; IPGC 0.42 (0.96) vs. 0.35 (0.84), $p = .13$; VGC vs. IPGC pre- vs. immediate post-change difference: $p = .09$; One-month post-counseling: VGC ($n = 40$) 0.35 (0.73) vs. IPGC ($n = 24$) 0.29 (0.69); overall, no significant difference in anxiety/depression outcomes between methods
Gerrard et al. (2020)	Empowerment measured with GCOS (24 items, 7-point Likert scale). A significant relationship was found between GCOS change scores and mode of GC ($F_{2,304} = 3.067$, $p = .048$).
Quality of Life/Well-being	
Jenkins et al. (2007)	Mean scores (*SD*) for TGC vs. IPGC respectively for general well-being (Dupuy Psychological Well-Being Scale) at baseline: 83.17 (14.73) vs. 83.06 (14.87) and 3 months: 85.33 (12.79) vs. 84.57 (14.16)
Schwartz et al. (2014)	Mean scores (*SD*) for TGC vs. IPGC respectively for QOL (SF-12). Physical function: baseline 51.0 (8.6) vs. 50.7 (9.2) and 3 months 51.6 (8.6) vs. 50.9 (9.2); TGC non-inferior; mental function: baseline 48.9 (10.3) vs. 48.5 (10.6) and 3 months 50.6 (9.3) vs. 51.0 (9.3); TGC non-inferior to IPGC based on adjusted mean group differences and 97.5% confidence limit.
Kinney et al. (2016)	QOL (SF-12, version 2): 1-year change from baseline: (physical health) TGC mean score −0.39, 95% CI [−1.10, 0.32] vs. IPGC mean score 0, 95% CI [−0.62, 0.65]; (mental health) TGC mean score −0.79, 95% CI [−1.58, 0.04] vs. IPGC mean score −1.09, 95% CI [−1.90, −0.23]
Interrante et al. (2017)	Mean scores (*SD*) for TGC vs. IPGC respectively for QOL (SF-12) physical function at baseline 48.8 (10.5) vs. 49.1 (10.4) and 12 months 50.2 (9.0) vs. 50.3 (8.9)
Satisfaction	
Coelho et al. (2005)	No significant difference between TGC and IPGC satisfaction
Lea et al. (2005)	Mean patient satisfaction with VGC using the PPSQ = 3.56 on a 4-point scale, with 4.0 being very satisfied (but only 25% of patients responded)
Helmes et al. (2006)	Not significant, but trend was less satisfaction with TGC than IPGC
Stalker et al. (2006)	Parents agreed/strongly agreed the evaluation was appropriate
Jenkins et al. (2007)	No significant difference in satisfaction between TGC and IPGC
Baumanis et al. (2009)	No significant difference in mean satisfaction between TGC and IPGC
Sutphen et al. (2010)	72.7% (16/22 respondents) indicated satisfaction with TGC; 54.5% (12/22 respondents) would not have pursued GC if not available by telephone
Bradbury et al. (2011)	Reported mean satisfaction rank; no significant difference in mean satisfaction between TGCC and IPGC ($p = .344$)
Meropol et al. (2011)	Satisfied with the overall communication aspects of the study ($M = 4.8$, $SD = 0.4$); highly satisfied with the session ($M = 4.2$, $SD = 0.4$)

(Continues)

TABLE 2 (Continued)

Outcome	Evidence[a]
Platten et al. (2012)	Satisfaction with the genetics provider (Questionnaire 1): 94% ($n = 144$) were completely satisfied; satisfaction with the genetics clinic (Questionnaire 2 and 3): participants in both groups rated contacts as satisfying to a high extent; regarding experience of information and recommendations (Questionnaire 3), participants were satisfied in general but were least satisfied with recommendations on cancer prevention and surveillance (32%, $n = 43$)
Hawkins et al. (2013)	Satisfaction with the support provided during the entire process: THGC 4.63 vs. IPGC 4.55 ($p = .736$) (average scores based on 5-point Likert scale where 1 = 'strongly disagreed' and 5 = 'strongly agreed')
Patrick-Miller et al. (2013)	Mean satisfaction scores (range 9–45) (*SD*): TGC ($n = 86$): baseline 36.8 (4.1) vs. post-telephone disclosure 38.7 (4.1), $p < .01$; IPGC ($n = 57$): baseline 36.6 (3.1) vs. post-in-person clinical follow-up 38.5 (3.0), $p < .01$; exploratory analyses: less improvement in satisfaction in African American participants vs. white participants (mean difference −3.8 points, $SE = 1.74$, $p = .03$) from baseline to post-telephone disclosure.
Patrick-Miller et al. (2014)	Advantages (TGC): for patients: conveniences, setting, and timing. Disadvantages (TGC): for patients: communication (e.g., not being able to see the GC for nonverbal communication), delivery (e.g., interruptions at workplace or home), and patient-specific factors (e.g., receiving positive or uncertain results).
Bradbury et al. (2016)	Satisfaction with GC survey mean scores (*SD*) for V1 vs. V2: 40.36 (3.92) vs. 42.58 (3.25), respectively, $p = .001$; TSQ mean scores (*SD*) for V1 vs. V2: 52.25 (5.26) vs. 53.99 (4.96), respectively, $p = .02$
Mette et al. (2016)	No difference between TGC and IPGC satisfaction
Otten et al. (2016b)	TSQ change pre to post (online only): 0.41 ($SD = 0.57$), $p < .001$; no difference in TSQ between VGC and IPGC
Peshkin et al. (2016)	Not significant but trend was less satisfaction with TGC than IPGC
Interrante et al. (2017)	Mean change from baseline to post-disclosure (*SD*): TGC −0.14 (5.45) vs. IPGC −0.23 (5.70); TGC non-inferior to IPGC
Bradbury et al. (2018)	Mean (*SD*) satisfaction score with GT decision (SWD, range 6–30): TGC 28.6 (2.4) vs. IPGC 28.4 (2.8)
Christensen et al. (2018)	Positive impact 14.3 TGC vs. 13.1 IPGC, diff 1.2, 99% CI [−1.3, 3.8]; subjective impact 63.8% TGC vs. 68.7% IPGC, diff 0.8, 99% CI [0.4, 1.7]
Fenton et al. (2018)	Mean score (*SD*): TGC 3.4 of 4 (0.5); non-TGC 3.4 of 4 (0.5), $p = .08$
Voils et al. (2018)	Mean score (*SD*): TGC 25.2 (2.6) vs. VGC 26.9 (3.0), no *p* value provided.
Arjunan et al. (2020)	Mean satisfaction rating was 4.9/5.0 (range: 1–5). Among individuals with negative screens that provided a satisfaction rating, 93.7% ($n = 943$) rated their satisfaction as 5/5. Of 42 patients with positive screens, 1 rated satisfaction as 2/5, 1 rated satisfaction as 4/5, and 40 rated satisfaction as 5/5
Cremin et al. (2020)	Among 54 (TGC group) completed surveys returned, 53 respondents (97%) stated they agreed or strongly agreed with Statement 1, 'Overall, this appointment was helpful to me'. The same survey was administered to all PDAC patients who attended a 1-on-1 appointment from January 31, 2019, to July 1, 2019. Thirty-one surveys were returned and 30 completed the satisfaction question with 30/30 (100%) stating they agreed or strongly agreed with Statement 1.
Jeganathan et al. (2020)	86.9% of patients were satisfied with the care they received, and 78.3% would recommend THGC visits to others
Pagliazzi et al. (2020)	84 (93%) families responded to the survey, reporting a satisfying level of communication
Shur et al. (2021)	Patients consistently provided verbal and written positive feedback [not otherwise quantified]
Trust and Rapport	
Sutphen et al. (2010)	Most (77.3%) agreed that their insurance company will protect the confidentiality/privacy of their personal health information and confidentiality was maintained during the phone call with the GC

(Continues)

TABLE 2 (Continued)

Outcome	Evidence[a]
Peshkin et al. (2016)	Feeling of support: TGC (n = 144, 52.9%) vs. IPGC (n = 186, 66%) p = .002; predictors of feeling counselor support included non-Hispanic white race/ethnicity (p = .058), older age (p = .039), lower cancer-specific distress (p = .032), lower perceived stress (p < .001), lower decisional conflict (p = .051), and both higher physical (p = .023) and mental quality of life (p = .018); exploratory analysis: Non-Hispanic white patients felt more support with IPGC (58.3%), and Racial/ethnic minority patient felt more support with TGC (38.7%). This was not statistically significant: OR = 0.80, 95% CI [0.39, 1.65]
Jeganathan et al. (2020)	92.9% felt their privacy was secure
Uptake of Genetic Testing	
Sangha et al. (2003)	Amniocentesis uptake: TGC 9/12 vs. IPGC 8/12, no p value provided
Shanley et al. (2007)	Subgroup of 100 patients: urgent blood sample collected, n = 3; tissue blocks collected, n = 5
Sutphen et al. (2010)	Among those eligible, 100% pursued testing
Hawkins et al. (2013)	Tested: THGC 8/15 (53%) vs. IPGC (11/13) 85%, no p value provided
Schwartz et al. (2014)[c]	Tested: TGC 226/268 vs. IPGC 256/284, RR = 0.93, 95% CI [0.88, 0.99]
Buchanan et al. (2015)	Tested: TGC 32/90 vs. IPGC 39/94, no p value provided
Butrick et al. (2015)[c]	Tested: TGC 251/335 vs. IPGC 272/334, no p value provided
Bradbury et al. (2016)	Tested: VGC 41/61
Kinney et al. (2016)	Tested: Rates at 1 year—TGC 138/493 (27.9%) vs. IPGC 185/495 (37.3%), no p value provided
Mette et al. (2016)	Uptake data not reported by intervention group; in total, 79.8% of participants
Otten et al. (2016b)	Tested: TGC 54/57 vs. IPGC 66/71, p = .73
Steffen et al. (2017)	TGC (n = 402) and IPGC (n = 379) Tested: high distress (TGC 26.3% vs. IPGC 44.3%) and high perceived comparative risk (TGC 33.9% vs. IPGC 50.5%).
Interrante et al. (2017)	Tested: TGC: 251/335 vs. IPGC: 272/334, no p value provided
Cremin et al. (2020)	Overall, 59.2% (n = 177/299) of all referred index patients completed genetic testing. The uptake rate with 1-on-1 consultations was slightly higher than for group sessions (88.9% vs. 82.9%), and telehealth resulted in a significantly lower uptake (61.8%, p < .001).

Abbreviations: BAI, Beck Anxiety Inventory; BCGKS, Breast Cancer Genetic Knowledge Scale;CES-D, Center for Epidemiological Studies-Depression Scale; CI, confidence interval; GC, genetic counselor; GCOS, Genetic Counseling Outcome Scale; GT, genetic testing; HADS, Hospital Anxiety and Depression Scale; HD, Huntington disease; IES, Impact of Event Scale; IGT, Impact of Genetic Testing for Alzheimer's disease instrument; IPGC, in-person genetic counseling; MICRA, Multidimensional Impact of Cancer Risk Assessment; MRI, magnetic resonance imaging; NR, non-inferiority range; OR, odds ratio; PDAC, Pancreatic Ductal Adenocarcinoma; PHQ-4, Patient Health Questionnaire for Depression and Anxiety; PPSQ, Patient and Provider Satisfaction Questionnaire; QOL, Quality Of Life; RRM, risk-reducing mastectomy; RRSO, risk-reducing bilateral salpingo-oophorectomy; SF-12, 12-item Short form Health Survey; STAI, State and Trait Anxiety Inventory; SWD, satisfaction with decision; TD, test disclosure; TGC, telephone genetic counseling; THGC, telehealth genetic counseling; TSQ, Telemedicine Satisfaction Questionnaire; V1, visit 1, pretest counseling; V2, visit 2, test disclosure; VGC, video genetic counseling.

[a]Evidence recorded as reported in each study with the exception that terms such as 'usual care' and 'in-person disclosure' were changed to IPGC, and all telephone consultations in lieu of an in-person visit (e.g., genetic counseling, disclosure) were changed to TGC for consistency. Reported scores are based on the metrics used by each study and are not consistent between studies.

[b]The percent for the CI was not provided.

[c]Participants are double-counted in Interrante et al. (2017).

[d]MICRA is reverse-scored.

GCs reported substantial differences in delivering genetic counseling via TGC compared with IPGC (Burgess et al., 2016), which may require specific strategies to address (Patrick-Miller et al., 2014).

3.3 | Access to care

Outcomes related to access to care were presented in 19 studies (Table 4). THGC consistently reduced costs; wait times for initial appointments or result disclosure; and time, travel, and childcare burdens to patients, though the specific reductions compared with IPGC varied by study. Consistent with the provider-reported advantages and patient-reported satisfaction, the convenience of THGC reported in four studies was highly rated by patients and perceived by GCs to improve patient access. Missed appointment/non-attendance rates were observed to improve over time in a 2005 study (Shanley et al., 2007) and improved or remained consistent in two studies documenting response to the COVID-19 pandemic (Jeganathan et al., 2020; Shur et al., 2021). Compared with earlier literature, GCs reported improved ability to bill for services delivered

TABLE 3 Provider-centric outcomes reported in included studies

Outcome	Evidence[a]
Advantages/Disadvantages	
Lea et al. (2005)	Disadvantages (VGC): voice delay ($n = 1$), lack of hands-on examination ($n = 1$)
Bradbury et al. (2011)	Perceived advantages (TGC): for patients: convenience (122/184, 67%), medical benefits (70/184, 38%), psychological (e.g., more control, satisfaction, less anxiety, and more privacy) (64/184, 35%). For GCs: time savings (32/184, 17%) Perceived disadvantages (TGC): for patients: communication and understanding (67/180, 37%); psychological (less emotional support from GC, family/friends; more anxiety) (26/180, 14%); and less compliance with follow-up appointment (22/180, 12%); for GCs: communication difficulties (132/180, 73%), inability to bill for services (24/180, 13%), time to reach patient and follow-up (10/180, 6%); poor experiences that made respondents question utilization of TGC: patient events (32/63, 51%), GC events (24/63, 38%)
Patrick-Miller et al. (2014)	Advantages (TGC): For GCs: greater flexibility for scheduling, setting, and efficiency. Disadvantages (TGC): for GCs: communication (e.g., not being able to see the patient for nonverbal communication), delivery (e.g., interruptions at workplace or home), and patient-specific factors (e.g., delivering positive or uncertain results).
Voils et al. (2018)	Advantages: GCs found both VGC and TGC acceptable but found more advantages with VGC such as fewer missed appointments, less distraction, and ability to read body language
Terry et al. (2019)	Disadvantages/challenges: appropriate psychosocial care and patient education as the most common THGC challenges. Licensure was cited as the most common perceived cross-state THGC challenge among respondents who cited cross-state challenges and was also the most common perceived overall barrier to THGC. TGC programs showed similar patterns in their responses, and challenges including psychosocial care, education, and licensure.
Provider satisfaction and perceptions	
Lea et al. (2005)	Satisfaction (VGC): 4-point scale with 4.0 being very satisfied; satisfaction with all sessions rated with mean of 3.83 (but only 18% of providers responded) Perception (VGC): decision confidence rating on a 6-item Likert-type scale with 6 being strongly agreed: high mean ratings for each statement (range 5.29–5.81); all felt consult was successful and addressed problem correctly
Bradbury et al. (2011)	Perception of patient communication barriers (TGC): 132/180 (73%) perceived communication barriers
Burgess et al. (2016)	Establishing rapport and communication with patients (GCs compared TGC with IPGC): 60.2% (50/83) noted a difference in establishing rapport through verbal and nonverbal interactions; 50% (23/46) noted a difference in establishing rapport through interpreters; 36.7% (29/79) noted a difference in assessing clients' ethnocultural background, health beliefs, values; 28.4% (21/74) noted a difference in identifying family dynamics, emotional responses, and diagnoses requiring confirmation; 32.4% (24/74) noted a difference in assessing client understanding/response; 35.6% (26/73) noted a difference in educating clients about basic genetic concepts/modes of inheritance; 28.4% (21/74) noted a difference in evaluating client risk perception/response; 33.3%–47.8% noted differences in all tasks related to psychosocial assessment
Jacobs et al. (2016)	Perception of counseling session (highest possible composite score of 25 on the GCQ): TGC mean overall score 22.3 ($SD = 2.4$) vs. IPGC mean overall score 22.3 ($SD = 2.5$), $p = .910$; predictor of GCQ score: sessions in which the patient was non-Hispanic white were rated more positive than when patient was not identified as non-Hispanic white [$t(477) = -3.82$, $p = <.001$]; GCQ did not differ between TGC and IPGC.
Otten et al. (2016a)	Provider satisfaction: the mean TSQ item scores (SD) for the average counselor: before the pilot: 3.38 (0.68) vs. after the pilot: 2.95 (0.96). The effect size was 0.52. Individual differences among counselors.
Jeganathan et al. (2020)	Satisfaction: Overall, 87.8% of providers liked using THGC and 90.9% believed that THGC improved patients' access to care. Preference: There was a significantly higher rate of providers who preferred IPGC than THGC visits (56% vs. 23%, respectively, $p = .024$)
Shur et al. (2021)	Satisfaction: Providers expressed satisfaction with this model, and internal polling showed that all providers wanted some form of TM in their practice with the majority targeting around 50%.
Workflow	
Patrick-Miller et al. (2014)	GCs noted greater availability and flexibility for scheduling patient appointments, no travel to and wait in clinic, access to resources in provider's office during appointment.
Burgess et al. (2016)	Coordinating genetic testing (GCs compared TGC with IPGC): 30% (21/70) noted a difference in arranging preliminary diagnostic tests; 40.9% (27/66) noted a difference in facilitating genetic tests.
Christensen et al. (2018)	Session duration: 6–40 min (TGC) vs. 5–50 min (IPGC); TGC sessions ~6.6 min shorter than IPGC sessions, $p < .001$

Abbreviations: GC, genetic counselor; GCQ, Genetic Counseling Process Questionnaire; IPGC, in-person genetic counseling; TGC, telephone genetic counseling; THGC, telehealth genetic counseling; TM, telemedicine; TSQ, Telemedicine Satisfaction Questionnaire;VGC, video genetic counseling.

[a]Evidence recorded as reported in each study with the exception that terms such as 'usual care' and 'in-person disclosure' were changed to IPGC, and all telephone consultations in lieu of an in-person visit (e.g., genetic counseling, disclosure) were changed to TGC for consistency. Reported scores are based on the metrics used by each study and are not consistent between studies.

both through video and in-person, though GCs in phone-only programs may be less likely to bill insurance (Greenberg et al., 2020; Terry et al., 2019). Costs (to the health system) were reduced with THGC versus IPGC, though the precise reduction varied based on the parameters of the economic analysis (Chang et al., 2016; Schwartz et al., 2014).

Although results were similar across outcomes regardless of the method of THGC, patients undergoing VGC had lower rates of follow-up than those who underwent TGC (Voils et al., 2018), and compared with IPGC, patients undergoing VGC also had lower attendance rates, with non-attenders more likely to be non-white, have a household income <$50,000, and be less comfortable with computers (Buchanan et al., 2015).

3.3.1 | VGC compared with TGC

In the single study that reported a direct comparison between TGC and VGC, fewer patients randomized to VGC completed their session, compared with TGC (66.7% vs. 90.0%, respectively) (Voils et al., 2018). A survey of regional THGC programs found wait times were lower in TGC programs compared with VGC and IPGC; 100% of phone-only programs reported wait times of <2 weeks compared with 56% of VGC programs. In the same survey, convenience was comparable between TGC and VGC programs (Terry et al., 2019).

Overall, the evidence paints a picture of lower costs and improved patient access to genetic counseling with THGC, though regional differences may exist, and specific patient subgroups may not benefit to the same extent.

4 | DISCUSSION

The primary initiative of our systematic review was to compare THGC with IPGC by assessing patient-, provider-, and access-related outcomes. Overall, in our evaluation of the patient experience with THGC, the majority of outcomes were primarily non-inferior or comparable to IPGC. Trust and rapport were lower with THGC compared to IPGC based on a single RCT of TGC for patients at high risk for a *BRCA1/2* pathogenic variant (Peshkin et al., 2016). However, in two observational studies, there was no difference in trust between THGC and IPGC (Hawkins et al., 2013; Sutphen et al., 2010). This may reflect the limitations of TGC for both patients and providers to observe nonverbal cues. However, the limited data suggest that this interpretation could differ with new research in more diverse populations.

When evaluating the provider experience with THGC, the reported disadvantages were focused largely on technological aspects of TGC. Despite stated and perceived disadvantages of THGC compared with IPGC, provider satisfaction remained high and VGC may ameliorate some of the noted disadvantages of TGC. In addition, when considering the body of evidence as a whole, GC experience did not always reflect the patient experience in that some GCs had negative perceptions of THGC sessions. This was more notable in studies involving TGC than VGC, in which the primary negative differences noted by GCs were related to less effective workflow and more limited psychosocial assessment (Bradbury et al., 2011). These negative perceptions stand in contrast to the large high satisfaction with THGC reported by providers. GCs also reported difficulties related to ordering genetic testing and working with interpreters (Bradbury et al., 2011; Burgess et al., 2016). Some of the workflow disadvantages may be the product of early uptake and comfort with a then-novel technology for both providers and patients. Finally, GCs also noted the potential for lower compliance rates for follow-up appointments, evidenced by lower attendance rates for THGC than IPGC (Bradbury et al., 2011). However, it is worth noting that some of these concerns were not reflected in analysis of the patient experience with THGC. Exploratory analysis suggests that lower household incomes, less comfort with computers, and non-white ethnicity predict lower appointment attendance with THGC (Patrick-Miller et al., 2013). It is unclear whether our conclusions regarding the patient or provider experiences with THGC will hold true as both groups become more comfortable with THGC and adapt more to these SDMs over time, and technological deficits are addressed.

Regarding access to care, THGC decreased costs compared with IPGC and had positive impacts on other patient-related burdens such as time off work and childcare and was perceived by providers to increase access to genetic counseling.

Of the 42 articles included in this review, more than half ($n = 26$) were published between 2015 and 2020. The consistency of results from the included studies supports the overall confidence in the key conclusions formed in this systematic review. Nevertheless, there are limitations to this review. First, the majority of the available studies assessed TGC, had small sample sizes and/or small subgroups that were underpowered, focused on cancer genetic counseling, had patient participants who were predominately non-Hispanic white females, had either a moderate or serious risk of bias, or were of poor quality, although this was not an exclusion criterion. Accounting for these limitations, the overall certainty of our data was moderate or high for patient outcomes, low for provider outcomes, and mixed (low and high) for those related to access to care. Second, there were limitations related to the systematic review process in that we only included articles in English and we may have missed articles not captured in our search string.

To improve and standardize research for THGC in the future, we offer several recommendations. First, an improvement in study design is needed. Studies should include a comparison arm to THGC, include statistical analysis of results, and use standardized outcome measures specific to genetic counseling (such as those being developed by the National Society of Genetic Counselors [NSGC] Research Task Force) when possible (Hooker et al., 2017). Continued use of unaltered, validated instruments is recommended. Second, research involving THGC should explicitly state the genetic counseling interventions used, as recommended by Hooker et al. (2017). This is essential to allow accurate comparisons of THGC use across appointment types.

TABLE 4 Access to care outcomes reported in included studies

Outcome	Evidence[a]
Attendance at appointments	
Shanley et al. (2007)	81% required in-person follow-up (screening, genetic testing); 56% required 2–3 phone calls, 1% required 4+; failure to attend rates decreased over time (>20% March 2005 to <5% July 2006)
Buchanan et al. (2015)	Attendance was lower in TGC 79% vs. IPGC 89%, $p = .03$; non-attenders more likely to be non-white, less comfortable with computers, HHI <$50,000
Voils et al. (2018)	Difference in follow-up: VGC 29.6% vs. TGC 70.4%; completed session: VGC 66.7% vs. TGC 90%
Arjunan et al. (2020)	Patients with positive prenatal screening results were most likely to elect genetic consultations, but most consultations were for screen-negative patients. Patients with preexisting risk factors, such as advanced maternal age and family history, even among patients with negative results, were more likely to elect laboratory-provided genetic counseling than patients without these risk factors.
Jeganathan et al. (2020)	In 2020, after the implementation of telehealth, there was a lower rate of attended visits and total canceled appointments compared with 2019. However, in 2020, overall, there was a significantly lower rate of no-show appointments (8.49% vs. 4.61%, $p < .001$), patient-canceled appointments (7.06% vs. 4.96%, $p < .001$), and patient same-day cancelations (2.30% vs. 1.35%, $p < .001$). There was a significantly lower rate of appointments canceled by patients for in-person visits than telehealth visits (5.44% vs. 3.82%, $p = .021$).
Shur et al. (2021)	Pre-COVID-19 TM, $n = 138$; post-COVID-19 TM, $n = 150$. Patient no-show rates from comparable time periods (2018, $n = 860$ and 2019, $n = 1,079$) for in-person visits were 13.6 and 14.4%, respectively. For pre-COVID-19 TM visits ($n = 136$), the no-show rate averaged 9.1% and post-COVID-19 TM ($n = 474$) was 8.9%.
Pagliazzi et al. (2020)	75% of patients reported they would have canceled the appointment [if not for the telehealth option], fearing to contract COVID-19 inside the hospital
Convenience	
Peshkin et al. (2016)	Genetic counseling and testing process rated extremely convenient in TGC 72.4% vs. IPGC 35%, $p < .0001$; predictor of greater convenience: lower BRCA1/2 carrier probability ($p = .035$); lower perceived stress ($p = .071$); lower numeracy ($p = .044$); and higher physical functioning ($p = .046$); TGC vs. IPGC, OR = 4.78, 95% CI [3.32, 6.89]. Participants with a lower objective pathogenic variant risk: OR (0.5 *SD* change) = 0.91, 95% CI [0.82, 0.99] and higher physical functioning: OR (0.5 *SD* change) = 1.10, 95% CI [1.01, 1.21] were also independently associated with higher convenience.
Terry et al. (2019)	Among the active, video-capable programs, respondents endorsed convenience and reduced travel as the biggest benefit of their THGC programs to patients. Similarly, when asked about the biggest benefit THGC offers to providers, respondents cited patient access and convenience most. Responses were similar for video-capable and telephone-only program counselors.
Greenberg et al. (2020)	Although TGC and VGC remain models able to reach patients the furthest away, it appeared that patients who lived nearby were also accessing THGC. This convenience factor may be desired by patients who are unwilling to drive 30–60 min for genetic counseling services. Alternatively, in the day of electronics and the increasing use and comfort with technology, more patients may request or seek out remote services.
Jeganathan et al. (2020)	84.7% of patients found the process of connecting to their appointment easy; patients reported being able to visualize their doctor in 70.3% of cases just and if the appointment were in-person. 90.9% of providers believed that telehealth improved patients' access to care.
Economics	
Lea et al. (2005)	GC/system cost of ISDN lines and consult with telemedicine: $240/month
Schwartz et al. (2014)	GC/system costs; average cost/patient TGC $3,660.00 vs. IPGC $3,774.00; incremental cost of TGC: −$114.40; all sensitivity analyses TGC costs lower than IPGC costs
Chang et al. (2016)	Cost analysis (societal perspective); cost (range) of pretest counseling/patient: TGC $120 ($80–$200) vs. IPGC $270 ($180–$400); cost (range) of genetic testing/patient: TGC $3,400 ($3,396–$3,404) vs. IPGC $3,410 ($3,402–$3,413); cost (range) post-test counseling/patient: TGC: $160 ($100–$250) vs. IPGC: $380 ($240–$610); average cost/patient counseled and tested (range): TGC $3,680 ($3,570–$3,850) vs. IPGC $4,060 ($3,830–$4,420); average cost/pathogenic variant positive (range): TGC: $37,160 ($36,080–$38,920) versus. IPGC: $40,330 ($38,010–$43,870)
Solomons et al. (2018)	Patient economic impact (VGC patients actual vs. if no VGC); missed work: 25% actual vs. 32%; someone else missed work: 12% actual vs. 24%; childcare issues: 5% actual vs. 9%
Terry et al. (2019)	Though not statistically significant, there was also a trend observed where phone-only programs tended to be less likely to bill insurance ($p = .09$). Licensure was the most common perceived overall barrier to THGC and most common challenge for those who cited cross-state challenges.

(Continues)

TABLE 4 (Continued)

Outcome	Evidence[a]
Greenberg et al. (2020)	More respondents report billing for IPGC and THGC than in the past, and more GCs bill in their own name and NPI, and use the 96040 code across all models
Travel-specific costs/Time for travel	
Lea et al. (2005)	Cost to patient for trip for IPGC: $350
Jenkins et al. (2007)	Patient costs/time; travel costs $0 vs. $120 (range: $5–$550); travel time 0 hr (range 0–0.75) vs. 6 hr (range 0.25–65)
Hawkins et al. (2013)	Patient travel time: THGC 2.73 hr vs. IPGC 6.69 hr, $p = .002$
Schwartz et al. (2014)	Health system/GC costs; travel costs [$0.00 vs. $15.10, no p value]; travel time [0 vs. 70 min, no p value]
Buchanan et al. (2015)	GC travel time: travel time 158.2 hr/71 patients; GC costs: cost of travel $140.98 per patient
Bradbury et al. (2016)	Patient-reported reduced burden of travel: pretest, 61%; post-test 61%
Solomons et al. (2018)	Patient travel burden: (VGC patients' actual travel vs. travel if no VGC available): 13% traveled 50+ miles for VGC appointment vs. 81% would have traveled 50+ miles for IPGC.
Voils et al. (2018)	Patient costs/time; VGC $67.29 median out-of-pocket cost, VGC 2.8 hr travel time, VGC mileage = 40 roundtrip miles. No costs or travel time associated with TGC.
Wait time	
Stalker et al. (2006)	New patient consultation waiting time: start of VGC trial: 16.9 months ($SD = 1.9$) vs. end of VGC trial: 3.0 months ($SD = 1.0$); $p < .0001$
Shanley et al. (2007)	Subgroup of 100 patients; median 6 weeks, range 1–12 weeks
Christensen et al. (2018)	Time to results disclosure: 27.8 days (TGC) vs. 35.2 days (IPGC), $p = .002$
Terry et al. (2019)	Among the currently active, video-capable programs, 56% (18/32) of respondents reported a wait time of less than 2 weeks for consultations through their program; and just 16% (5/32) reported a wait of over 2 months. Among phone-only programs, all eight reported a wait time of less than 2 weeks. Comparison of video to phone programs: phone-only more likely to have a wait time of <2 weeks ($p = .04$) and have a longer program duration ($p = .02$).
Greenberg et al. (2020)	TGC reported shorter average time spent in patient consultation ($p < .01$; $df = 4$) and shorter wait times ($p < .01$; $df = 4$) compared with other methods of service delivery. Wait times appear to be shortest for the telephone genetic counseling and longest for group genetic counseling.

Abbreviations: CI, confidence interval; COVID-19, SARS-CoV-2 (severe acute respiratory syndrome coronavirus 2); GC, genetic counselor; HHI, household income; IPGC, in-person genetic counseling; ISDN, Integrated Services Digital Network; NPI, National provider identification; OR, odds ratio; TD, test disclosure; TGC, telephone genetic counseling; THGC, telehealth genetic counseling; TM, telemedicine; VGC, video genetic counseling.

[a]Evidence recorded as reported in each study with the exception that terms such as 'usual care' and 'in-person disclosure' were changed to IPGC, and all telephone consultations in lieu of an in-person visit (e.g., genetic counseling, disclosure) were changed to TGC for consistency. Reported scores are based on the metrics used by each study and are not consistent between studies.

Areas in which we need more data about potential confounding patient variables include disease state (affected vs. unaffected), presence of comorbid psychiatric conditions, ethnicity, socioeconomic status, location of the patient during the visit (e.g., at home vs. at a medical office), and health literacy. Additional studies using diverse populations that span varied gender, age, ethnicity, socioeconomic, and disease groups are also needed. More studies are needed to evaluate provider satisfaction with THGC. Most of the studies we reviewed did not include information regarding the amount of experience providers had with THGC prior to their involvement in these studies, so it would be helpful to ascertain whether provider experience with THGC influences provider satisfaction.

We recognize that, in the near future, additional telehealth options including Web-based platforms that assist with genetic counseling may become another common mode of service delivery generating a body of literature regarding efficacy (Biesecker et al., 2018). For the purposes of this systematic review, we did not focus on other emerging technologies such as Web-based educational portals, use of recorded videos for pre-appointment/pretest counseling, or chatbots. These and other SDMs may be just as effective, with some possible additional advantages over IPGC, and their implementation and comparisons with IPGC and more commonly used telehealth options for GC should be further explored.

Recently, many GCs have been required to provide remote services in order to ensure patient safety and to remain compliant with state stay-at-home orders during the COVID-19 pandemic. The effects of this rapid transition to the provision of remote services will likely result in more experience and data regarding the use of THGC in the upcoming years, which may ultimately change standards of care. The studies published after 2019, particularly the three that include descriptions of rapid transitions from in-person care to THGC due to COVID-19 pandemic precautions, provide evidence supporting greater acceptance of THGC and reduction of technological limitations that were inherent in early studies (Greenberg et al., 2020; Jeganathan et al., 2020; Pagliazzi et al., 2020; Shur et al., 2021; Terry et al., 2019). Hopefully, future researchers will seize this opportunity

to perform robust studies that include similar data collection tools to existing studies, in order to measure and compare both patient and provider outcomes and further delineate outcomes and best practices. As THGC is further studied in more diverse populations, subgroups may be identified that benefit more from certain types of SDMs. The profession may need to shift toward personalizing the delivery of GC services, when possible, to meet patient needs and preferences.

5 | CONCLUSION

Genetic counselors have been turning to THGC as a way to increase access to genetic services and maximize resources, such as time and cost for patients and providers. Overall, THGC is non-inferior to IPGC across many domains. The perspectives of patients and providers regarding THGC will likely evolve over time. More research into specific technological platforms, appointment types, and diverse patient populations will improve understanding of a variety of nuances that may affect the utility and acceptability of THGC and may enable more personalization of the provision of GC services in the future.

AUTHOR CONTRIBUTIONS

Noelle R. Danylchuk and Dr. Jennifer Malinowski were involved in the supervision of the project; design; data collection and extraction; data analysis and interpretation; and writing, reviewing, and editing the manuscript. Cara N. Cacioppo, Lola Cook, Melanie W. Hardy, Rachel Nusbaum, and Kate P. Shane-Carson were involved in the design; data collection and extraction; data analysis and interpretation; and writing, reviewing, and editing the manuscript. Susan C. Steelman was involved in the methodology; data collection; and writing, reviewing, and editing the manuscript. Authors Noelle R. Danylchuk and Dr. Jennifer Malinowski confirm that they had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. All of the authors (NRD, LC, KPSC, CNC, MWH, RN, SCS, and JM) gave final approval of this version to be published and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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COMPLIANCE WITH ETHICAL STANDARDS

Conflict of interest

Noelle R. Danylchuk, Lola Cook, Kate P. Shane-Carson, Cara N. Cacioppo, Melanie W. Hardy, Rachel Nusbaum, and Susan Steelman declare that they have no conflicts of interest. Dr. Jennifer Malinowski is a contract methodologist for the NSGC with no other conflicts of interest to declare.

Human studies and informed consent

No human studies were carried out by the authors of this article.

Animal studies

No non-human animal studies were carried out by the authors for this article.

Data sharing and data accessibility

The data that support the findings of this study are available in the supplemental material of this article.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

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