Family Medicine and Community Health

General practitioners' (GPs) experience, attitudes and needs on clinical genetic services: a systematic review

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ABSTRACT

Objective The proliferation and growing demands of genetic testing are anticipated to revolutionise medical practice. As gatekeepers of healthcare systems, general practitioners (GPs) are expected to play a critical role in the provision of clinical genetic services. This paper aims to review existing literature on GPs' experience, attitudes and needs towards clinical genetic services.

Design A systematic mixed studies review of papers published between 2010 and 2022.

Eligibility criteria The inclusion criterion was peer-reviewed articles in English and related to GPs' experience, views and needs on any genetic testing.

Information sources The PubMed, PsycINFO, Cochrane, EMBASE databases were searched using Mesh terms, Boolean and wildcards combinations to identify peer-reviewed articles published from 2010 to 2022. Study quality was assessed using Mixed Methods Appraisal Tool. Only articles that fulfilled the inclusion criteria were selected. A thematic meta-synthesis was conducted on the final sample of selected articles to identify key themes.

Results A total of 62 articles were included in the review. Uncertainty over GPs' role in providing genetic services were attributed by the lack of confidence and time constraints and rarity of cases may further exacerbate their reluctance to shoulder an expanded role in clinical genetics. Although educational interventions were found to increasing GPs' knowledge and confidence to carry out genetic tasks, varied interest on genetic testing and preference for a shared care model with other genetic health professionals have resulted in minimal translation to clinical adoption.

Conclusion This review highlights the need for deeper exploration of GPs' varied experience and attitudes towards clinical genetic services to better facilitate targeted intervention in the adoption of clinical genetics.

WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ General practitioners (GPs) are well positioned to provide patients with clinical genetic services by screening for potential patients who may benefit from genetic testing. However, GPs are faced with skills, knowledge, time and clinical constraints that hinders the effective adoption of clinical genetic services in primary care setting.

WHAT THIS STUDY ADDS

⇒ This study found that GP's views and attitudes towards adopting clinical genetic services were dependent on their experiences and context. GP's preference for a shared responsibility between them and genetic specialists may help overcome the resistance towards adoption of clinical genetic testing.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ The study highlighted the varied interest among GPs to incorporate genetic services in their clinical practice and the importance of addressing valid concerns and tailoring interventions to overcome barriers for GPs who may wish to adopt genetics in their practice.

INTRODUCTION

Advances in genetic research accompanied by the availability of a wide array of genetic tests is set to revolutionise medical practice worldwide.[1 2] General practitioners (GPs), as the gatekeepers in the healthcare systems, will need to be well informed of the benefits and risks of clinical genetic testing in order to respond to patients' requests for direct-to-consumer (DTC) genetic testing. However, clinical genetics is often regarded by GPs as a specialty arena and not a core component of generalist practice.[3 4] This discrepancy between what GPs should provide and what they perceive as within their role and competency may create confusion for primary care and clinical genetic testing healthcare providers.

Existing reviews mostly examine cancer genetics[5–10] with the most recent review that focused on general clinical genetics published in 2016.[11] Existing reviews have found that GPs experienced a lack of knowledge and confidence in basic genetics and risk assessments in the provision of clinical genetic services.[5 7 8 12 13] In addition, GPs also expressed concerns over ethical, legal and social implications (ELSI),[5 6] time pressures,[5 9] and difficulties in accessing referral

guidelines and genetic tests as barriers in the provision of cancer precision medicine. Despite these barriers, two reviews found that some GPs held positive attitudes as they recognised the importance of their increasing role in the development of primary care genetics.[9 12] Furthermore, in terms of adoption of clinical genetics, the review conducted by Paneque *et al* found that existing genetics educational interventions on patient management and policy for GPs have been poorly assessed. In order to determine the effectiveness of educational initiatives, assessment need to account for the changing primary care practices.[11]

However, little is known about whether existing educational initiatives and clinical interventions has changed GPs' experience and attitudes towards the adoption of clinical genetics. Also, most reviews focusing on primary care mostly included specialists such as oncologists, genetic counsellors, paediatricians and allied health professionals in their study population.[5–8 10 12 13] While alike, specialists would yield different experience as practice styles are heterogeneous.[14] With GPs being positioned in such milieu, it raises the need to better understand their ambivalent attitudes towards adopting clinical genetic testing, and their awareness of an increasingly salient role they could play and in advancing the utility of genetics in their clinical practice.

For this systematic review, we defined general practice to be 'the medical specialty that manages common and long-term illnesses such as asthma, diabetes and end-of-life care in children and adults, focusing on overall health and well-being.'[15] We defined genetic testing as the use of a laboratory test that comprises a broad range of testing techniques for medical care, ancestry studies or forensics, by detecting variations in an individual's DNA.[16 17] This includes the diagnosis of suspected genetic disease in symptomatic and asymptomatic newborns, children and adults (eg, Huntington's disease); risk assessment where individuals are informed of their increased or decreased risk of developing a condition (*BRCA1/BRCA2*); prediction of drug responses (eg, carbamazepine); and reproductive decision making (eg, thalassaemia).[2 17 18]

The aim of this systematic review was to examine GPs' experience and attitudes towards adopting clinical genetic services, as well as GPs' needs to provide genetic testing in their clinical practice. For this review, we defined experience as any discussion on genetics with patients and subjective experience that includes knowledge, confidence and barriers. Attitudes included views on the utility of genetic testing in their clinical practice and GPs' role in providing such clinical genetics services. Needs included strategies targeted at incorporating clinical genetic services. These included informational resources (eg, education workshops) and institutional system support (eg, practice policies, guidelines and recommendations). This review considered taking family history, recommending and ordering tests, interpreting test results, managing downstream care and referral to clinical genetic centres to fall under the umbrella of potential clinical genetic services for GPs. In this paper, the term 'GP' referred to both family physicians and primary care physicians.

METHOD

This review is reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) 2020 checklist. For this review, a systematic mixed studies review (SMSR) was conducted to identify and synthesise research pertaining to the research questions. The steps taken to conduct the SMSR followed the typical process of a traditional systematic review. However, in contrast to the conventional mixed methods synthesis whereby data set from qualitative and quantitative studies were reported separately, the focal point of SMSR was the consolidation of data sets from a range of quantitative, qualitative and mixed-method studies.[19 20]

Given the complexities surrounding the adoption of clinical genetic practice, an SMSR was considered to be appropriate in synthesising the growing literature from diverse research designs.[21] While, traditional systematic reviews have been given precedence to quantitative evidence,[22] qualitative studies have been concomitantly gaining traction and recognition in healthcare related research.[23] Conducting an SMSR would thus provide a comprehensive understanding of the phenomenon and a highly practical understanding on the complex public health interventions and programmes.[20 22]

Search strategy

Four electronic databases (PubMed, PsycINFO, EMBASE and Cochrane databases) were searched systematically, and studies that fulfilled the eligibility criteria published between 2010 and 2022 were selected. The reviewers used PubMed as one of the databases as it comprises of biomedical literature from MEDLINE, life science journals and online books. In addition, 2010 was chosen as the starting date for this review in view of the emergence of the next-generation sequencing clinical genetic testing, a technology anticipated to reduce the cumulative testing costs and thereby, encouraging mainstream access to genomics.[24 25] Grey literature was not included in this review as we only considered peer-reviewed published studies. Citation search was conducted on a few studies to capture relevant articles. The exact search strategy used by the reviewer is outlined in online supplemental table 1.

Study selection

Studies were included if they addressed genetic testing related to primary care within the scope of family medicine or internal medicine. In addition, worldwide literature were included if they met the following inclusion criteria: (1) peer-reviewed articles; (2) in English and (3) focused on GPs' experience, views and needs on any genetic testing. The inclusion and exclusion criteria are outlined in online supplemental table 2. The title, abstract

and full text of the articles were screened independently by two reviewers (CO and RCAT).

Data extraction and synthesis

A meta-synthesis was conducted to integrate existing studies to identify key themes. A data-based integration approach was used, and two reviewers conducted data transformation by reconstructing quantitative data into categorical themes and narratives for comparison with qualitative data.[20 21] Studies that were included from the full text screening were grouped together if their findings addressed the GPs' experience, attitudes, or needs. Data extracted were then keyed into an Excel spreadsheet independently by two reviewers (CO and RCAT). The data included key findings related to GPs' experience, attitudes and needs in clinical genetics services as defined in the inclusion criteria (online supplemental table 2). Other data sought included authors, publication year, country of study, aims, design methods, participants, genetic type and limitations of the study. Findings from each reviewer were compared for concordance and all discrepancies were adjudicated by a third reviewer (JYYN). Finalised data were tabulated on the Excel sheet and subsequently formatted into a table in Microsoft Word for display. Data extracted from each studies can be found in online supplemental table 5. The findings have been grouped into two categories: (1) GPs' experience, attitudes, views and (2) GPs' needs.

Assessment of methodological quality

Quality assessments were conducted by two reviewers (CO and RCAT) independently. The quality of all selected studies was assessed using the 2018 version of the Mixed Methods Appraisal Tool (MMAT). MMAT was chosen as it included appraisal of various study types,[26] with five items for each study type that can be found in online supplemental table 4. Studies were first organised into their respective study type – qualitative, quantitative RCT, quantitative descriptive and mixed methods—before they were rated based on the five items specified in the chosen category. For items where the rating 'Can't tell' was given, additional comments were included. As advised by the MMAT developers, the ratings for each study have been presented in the online supplemental table to provide readers with a clearer evaluation of the quality of included studies.[26] Any missing results or data were highlighted.

RESULTS

Characteristics of studies

A total of 871 studies were identified on the database (online supplemental figure 1). After removing 213 duplicates, the titles and abstracts of 658 studies were screened. Full texts were retrieved from 160 studies. A total of 62 studies satisfied the eligibility criteria and were included in the final sample.

Of the 62 final articles included, 36 studies were quantitative studies,[27–62] 13 were qualitative studies,[3 63–74] 9 were randomised controlled trials[75–83] and 4 were mixed-methods study[70 84–86] (online supplemental table 5). The selected studies were conducted in USA (n=20), UK (n=19), Canada (n=11), Australia (n=6), Asia (n=4), New Zealand (n=1) and South Africa (n=1). These studies composed of a range of genetic scope such as general clinical genetics (n=24), oncogenetics (n=18), pharmacogenetics (n=6), cardiovascular (n=6), prenatal/neonatal genetics (n=4) and DTC testing (n=4). Forty studies reported participation of both male and female GPs in their study population. Most articles (82.3%) focused on GPs' knowledge and experiences towards genetic testing (online supplemental table 3).

Risk of bias within studies

All studies were assessed using the MMAT tool. Fifty-five studies achieved a maximum score of compliance for all five assessment items. Seven studies lacked data on non-respondents to properly assess the risk of non-response bias (online supplemental table 4).[27 30 32 40 44 47 53] One common risk across the studies was the low response rate, which was reported in 29 studies.

Uncertainty over GP's role in genetic testing

Out of the 22 studies that reviewed GP's attitudes towards their role in genetic testing, 8 studies reported that GPs felt responsible to perform genetic tasks such as taking family history to identify genetic condition,[51] assisting or counselling patients on genetic testing and results,[32 61 65] referring patients to specialists for advice and follow-up care,[51 69] and to warn families about risks in the family.[53] GPs also found themselves to be well positioned to offer genetic screening such as population-based expanded carrier screening couple test,[66 85] and in the early detection of familial hypercholesterolaemia (FH).[54]

However, 11 studies found GPs to be ambivalent towards their existing role and competency to provide genetic screening in their practice. Hussein *et al* highlighted a mismatch in attitudes and actual practice as GPs took on distinct proactive or reactive approaches. For instance, while 70% of the 271 GPs surveyed considered taking family history as an integral role of general practice,[56] another study comprising of 96 GPs found that a proportion of the GPs surveyed were sceptical if taking family history should be part of their practice due to difficulties to obtain an accurate family history.[76] While genetic concepts are part of their general practice, two qualitative studies found that they are made distinct from genetic practice and thus, not identified as core component of their practice.[3 66] As illustrated by Mathers *et al*, although GPs may appear to be more willing to document family history, the routine use of family history for general disease management was distinguished from those for genetic conceptualisation.

Out of the 11 studies, 7 studies found that GPs leaned towards having a minor role that focuses more on traditional genetic tasks of identifying, referring and providing psychological support rather than assessing and explaining

genetic risks, benefits and limitations.[28 31 38 51 56 60 86] Two studies found that less than 25% of the 1168 GPs surveyed were willing to discuss genetic testing,[60] and 70% of the 271 GPs did not perceive ordering genetic tests or discussing about testing implications as part of their job scope.[56] Interestingly, one study found that younger GPs were more willing to incorporate genetic tasks into their everyday clinical practice.[61]

Lack of confidence and limited knowledge

GP's uncertainty over their role in genetic testing was compounded by their lack of confidence and knowledge on genetic testing. Out of the 34 studies that reviewed GP's knowledge and experience towards genetic testing, 7 articles shed light on the limited experience and encounters with genetic testing[43 44 63 65] or genomic cases.[28 43 58] Of the 63 GPs surveyed, 77.8% had no experience with referring patients.[36]

Ten articles also highlighted GPs lack of confidence on their genetic knowledge,[40] ability to conduct genetic screening,[66] and to perform genetic tasks[48 51] such as interpreting results[62 65 84] and discussing benefits, risks, limitations and inheritance patterns.[51 62 76] The study by Bernhardt *et al*, found that only 58% of the 481GPs surveyed were confident in interpreting genetic test results. GPs also reported a lack of confidence to educate patients on genetic cardiac disease and answer patients' questions.[35]

Apart from the lack of confidence, 17 studies reported on GPs' lack of knowledge on genetic testing. GPs were found to lack necessary technical expertise and skillsets to convey results[31 67] or had minimal knowledge to interpret results or estimate risks.[50 52 70] GPs were also unfamiliar with genetic concepts,[3 32 34 42 44 46 47 69] evidently revealed through a semi-structured interviews with 15 GPs which indicated a lack of familiarity with genomic terminologies and genomic tests.[73] Rangarajan *et al* also found GP's overall knowledge of FH to be low with only 40.6% of the 133 GPs surveyed being aware of international guidelines, and 13% were cognizant of genetic services available. There was also a lack of awareness on genetic laws among GPs.[41] In addition, 52% of 90 GPs surveyed reported being unsure of how pharmacogenetics could be incorporated into their practice.[33] Insufficient knowledge on referral criteria,[31] referral pathways and appropriate centres for referrals[28 36] have also led to variation in referral patterns.[35] Teng *et al* found a wide discrepancy between GP's self-reported referral rates (87.5%) and actual referral rates (12.5%). Fiederling *et al* similarly reported that only 35% of 35 GPs would refer their patients to specific counselling centres.

Knowledge scores were found to be positively associated with comfort scores to perform genetic tasks and referrals.[86] Henceforth, minimal knowledge and lack of confidence may have discouraged GPs to feel comfortable to order genetic test[42] or adequate to provide genetic counselling. In concurrence, 65% of the 61 GPs surveyed felt that genetic counsellors, medical geneticists, or oncologists were more qualified to perform such tasks.[34] This sentiment was similarly highlighted in another study which reported 74% of the 27 GPs seeing it as others' duty to follow-up on genetic results.[86]

On the contrary, only five studies found GPs to be fairly confident about their ability to determine the need for further evaluation based on family history,[53] with 74% of 271 GPs surveyed having had contact with patients with genetic disease weekly.[61] Two articles also reported that GPs frequently refer patients for cancer genetic testing[37] and conduct cancer family history consultation and risk assessment.[38] However, it is critical to note that while 52.8% of 70 GPs surveyed felt confident to explain risks and benefits only 40% reported being confident in their genetic knowledge.[68]

Genetic education and interventions

There were 13 studies that reported on the effectiveness of clinical interventions. Five studies found an increased in comfort level with genetic testing through a multifaceted educational intervention that comprises of individualised training, supervision and additional resources such as checklists[71 80 84 85] or online genetics modules.[79] A care-based oncogenetics education that includes practical applicability, interactive sessions, small group discussions was also found to achieve a sustained improvement over 3 months after training.[78] Seventy-six per cent of the 1402 GPs surveyed also found 'pushed emails' to be useful for learning about genetics.[55] Another intervention that reported success was the use of an electronic health record coupled with family history tool which helped to increase patient awareness through system prompters that facilitated discussions.[68] Due to the rarity of genetic cases, Lemke *et al* found direct access to pharmacogenetics (PGx) testing was a good approach for GPs to obtain first-hand knowledge although more education was desired.

The importance of genetic education and training were reported in 27 studies. Five studies reported on the interest for more training and information.[28 31 37 40 62] Of which, Yu *et al* reported that 91% of the 409 GPs surveyed saw the importance to keep up with latest information on genetic disorders. Specifically, GPs expressed the need for more guidelines and timely updates on the use of genetic screening, genetic testing, genetic counselling and referrals.[34 39 63 65 70] In addition, clearer guidance that is tailored to their practice and roles as GPs were also coveted.[51 64 65 69] Greater understanding to interpret and communicate test results, care treatment,[64 67 72] and evidence on clinical utility[74] were also sought after by GPs. Of the 13 studies that reported on GPs preference to learn about genetics, 9 studies found Continuing Medical Education (CME), online medical references or journals to be useful for obtaining more information.[27 30 34 36 42 46 52 53 66] Other preferred avenues includes monthly circular on clinical and referral pathways,[35 68] grand rounds, case studies and physical seminars.[32 65]

However, Nippert *et al* reported that only 12.8% of the 1168 GPs surveyed attended CME courses on genetics. Moreover, interactive web-based CMEs were found to have minimal impact on changing clinical practices.[75] While web training has effectively increased self-reported genetic consultation and management skills, the actual number of referrals did not change.[83] Furthermore, interventions such as PGx alerts were reported to be confusing with 52% of 90 GPs surveyed not knowing how to use additional information in their practice.[33]

Varied outlook on responsibility and clinical utility

Discrepancies between interventions and actual practice could be attributed to the lack of consensus on the need for genetic training. An in-depth interview with 21 GPs revealed that the call for education, training and guidelines were not echoed by all.[3] Similarly, Schuurmans *et al* found that training may be more effective for GPs motivated to do so rather than all GPs. Nine other studies found that GPs saw genetic testing to fall under the responsibility of others. For instance, genetic specialists were highlighted to be more appropriate to provide counselling.[35 44] Genetic health professionals or pharmacists were also expected to communicate results and follow-up with patients.[32 77 86] Other than physicians, GPs also highlighted patients and family members' responsibility to follow up and adhere to recommendations,[36 56 86] which aligned with three other studies that call forth the need to educate patients and family members about genetic condition[35] using resources as such patient handouts.[65 68]

On the other hand, nine other articles emphasised on a shared care model with other healthcare professionals. This includes having opportunities to discuss with specialists or pharmacists,[32 34 65] a buddy system with geneticists or contact information on local genetic clinic.[51] GPs also appreciated summary letter, comprehensive report, interpretative comments or telephone call from genetic health professionals to help identify high risk patients and navigate through test results.[54 69 73 82] In addition, GPs placed more emphasis on the responsibility of screening centres.[36]

While GPs may not fully grasp the technicalities of genetics, some recognised the positive clinical utility of genetic results on patient care[86] and believe results would be helpful for patient care management,[58 62 64 70] especially in identifying risk and disease prevention.[40 68 71] Six articles further reported that GPs perceived genetic testing to play a bigger role in future practice.[28 42 51 61 62 65]

Yet, this positive outlook on clinical genetics was not unanimous among GPs. Resistance to integrate genetic testing into clinical practice could be attributed to the additional workload required to discuss recommendations and answer patient's questions amidst their busy practice[68 70 85] and time constraints during clinical consultation.[28 36 44 53 71 73 74 81] Furthermore, the rarity and complexity of genetic cases were perceived to have limited impact on their general practice.[3 44 47 66] Apart from the lack of clinical evidence, GPs also expressed concerns over negative patient attitudes,[43 66] language barrier with patients who did not have English as their first language,[44 71] and confidentiality and discrimination of test results.[36 46 68] Although GPs anticipate a substantial impact of genetic testing on future practice, 78% of the 1404 GPs surveyed felt that genetic testing was less appropriate to inform treatment with 58% expressing beliefs that DTC testing would likely harm patients' general health decisions.[40] Sixty-four GPs surveyed in a separate study also expressed concerns on giving patients a false sense of security or inducing unnecessary anxiety over genetic results.[62]

Moreover, resistance to adopt genetic testing was also exacerbated by organisational barriers such as rigid administration infrastructure,[66] lack of clinical guidelines on genetic practice[43 52 53] and limited access to labs that perform PGx testing.[84] Additional resources could also incur more cost that could overburden the healthcare system.[67 84]

Impact of healthcare models on GPs' attitudes

Different healthcare models in different regions may also affect GPs' willingness and expectations to adopt clinical genetic services. GPs in the UK and other regions (who were gatekeepers in their healthcare system) recognised their responsibility to provide clinical genetic services and desired greater need for genetic education relevant to their practices.[3 30 47 56 60–62 71 79 83] Within Europe, French GPs ascribed most practice responsibilities to themselves while GPs from the other UK regions assigned most tasks to a genetic specialists.[38] In contrast, GPs in the USA were generally more sceptical of the utility of clinical genetics and saw it as specialists' responsibility to perform genetic tasks.[33 40 42 46 49 52 58 75 77 86] Although Asian GPs are generally more conservative, some advocated the need for education to empower them to take on the role in the early detection of at-risk patients.[54 81]

DISCUSSION

This review highlighted the complex experience GPs face in adopting genetic practices that ranged from taking a genetic family history, recommending and ordering tests, interpreting test results, managing downstream care and referral to clinical genetic centres.

While GPs considered their role in clinical genetics to be salient, they were uncertain about what this role entails. Findings on GPs' uncertainty over their role and responsibility to provide and assess genetic results corroborates with existing literature.[87 88] Findings also concurred with literature that found GPs to perceive genetic tasks to be highly complex that requires specialists' knowledge.[89] While GPs were trained to apply multifactorial clinical risk factors in their practice to inform medication use and patient care management, they were less likely to have adequate experience or exposure to rare genetic diseases.[90] In comparison, specialists may seemed more suited, confident and better prepared to perform and

interpret DTC testing than GPs. Furthermore, a study has found that the use of DTC testing lack standardised laboratory practices and is prone to misclassification in risk assessment and should be used with caution for clinical care decisions.[91] This may pose as a risk for GPs who could be less attuned to the heightened levels of scrutiny towards DTC testing and accept genetic test results at face value.[90]

Yet, GPs remained well positioned to assist their patients in informing their family members about genetic testing options in view of their long-standing relationship. However, help from genetic counsellors is needed to ensure a smooth transition between general and specialist care. A review conducted in 2004 highlighted that GPs played an essential role in the ongoing follow-up with their patients after a genetic diagnosis has been made.[2] Aligned with the principles of generalist–specialist relations, GPs were found to favour a shared care model where specialist assistance is sought to provide comprehensive management in the complex care of clinical genetics.[92] Not just between GPs and geneticists, a national survey of primary care paediatricians also found the majority of them to be actively involved in genetic services through ordering genetic tests and referring patients to geneticists annually.[93] This interdisciplinary approach to genetic testing serves a critical role in ensuring optimal care for complex genetic cases through concerted efforts from paediatricians, genetic counsellors, therapists, nurses, social workers and psychologists to meet the medical and psychological needs of patients.[94] The need for specialists' expertise to craft a follow-up care plan after a genetic diagnosis was further reiterated in a semistructured interview with 15 GPs where they expressed that without proper follow-up actions for GPs to act on, patients may suffer from potential harms that can lead to anxiety and unnecessary investigations.[72]

Even though GPs recognised the benefits of clinical genetic testing, many were reluctant and had concerns about the adoption. First, challenges to document family history have contributed to divided opinions on adopting clinical genetic services in general practice. Time pressures and limited reimbursement for GPs may render a detailed three-generation family history impractical in their general practice.[2] Inaccuracies and gaps in information obtained from patient about their family history may also pose a problem for GPs to make appropriate genetics referral and screening recommendations. Therefore, interventions targeted at GPs' role in taking family history should seek to use family history as a triage tool to catch rare genetic cases in younger patients or potential hereditary cases with a focus on time-efficiency.

Second, clinical barriers such as rarity of cases, patients' psychological well-being and concerns over the accuracy of genetic results may further exacerbate their reluctance to shoulder an expanded role in clinical genetics. In contrast to taking a family history in the usual primary care context, GPs also reported a lack of clinical practice guidelines on how to assess and discuss genetic risks. Consistent with previous reviews, GPs' lack of knowledge and confidence were commonly identified to be barriers in the provision of clinical genetic services.[5 7 8 12 13] Studies have found that GPs often feel unprepared and lacked confidence due to the rarity of genetic cases in their clinical practice[95 96] and time constraints.[97] Despite these barriers, most GPs had expressed interest to further their education in genetics. Education on the ethical and moral issues surrounding genetic testing was an important proposal. As Bathurst *et al* highlighted, litigation was at the forefront of GPs' practices. Thus, education should seek to address ethical and moral issues surrounding the accuracy of tests, ability to interpret results correctly, disclosure and confidentiality in relation to inherited positive or carrier status for genetic diseases.

While existing reviews have underscored the need for educational interventions,[12 13] findings from web-based intervention studies revealed that education had minimal impact on changing clinical practices. Although web-based educational initiatives were effective in bridging the knowledge gap, such curricula may pose a challenge for time-constrained GPs.[77] Furthermore, findings on the lack of translation from knowledge to practice were not unique as educational initiatives often fail to meet the demands of the everchanging clinical practice guidelines.[11 75] This may pose a potential risk of GPs conveying genetic information to patients without having updated genetic knowledge. Future educational and training should focus on making content relevant to GPs' current clinical practice, through simple and short presentations codeveloped with GPs.[98] It is also worthwhile noting the increasing importance to include epigenetics in GP training to highlight the impact of environmental and behavioural factors which, is presently underemphasised in most genetic courses,[99] coupled with the limited research on GP's understanding of epigenetics.

Moreover, the need for genetic education and training was not echoed by all. Both Haga *et al* and Carroll *et al* found that interest in genetic testing was associated with higher confidence, a positive outlook on genetic medicine and identifying genetics as GPs' responsibility.[52 80] Concurring with this observation, one GP shared his familiarity and experience with clinical genetics as a result of his personal interest which empowered him to play a very important role in advising patients about the risk of getting inherited conditions and how best to test and manage the risk.[4] Hence, it might be more strategic to target clinical genetic interventions at GPs who have special interest in genetics rather than making it mandatory for all GPs.

Strengths of this systematic review include a broad search strategy on varied terms related to clinical genetics and GPs, which increased the likelihood of capturing relevant literature. A range of study designs were also included to increase the heterogeneity of results. However, there were some limitations to this study. Despite the inclusive approach, a limited number of studies were identified. Furthermore, most studies reported a low response rate

and selection bias of GPs with special interest in clinical genetics. In addition, it is likely that positive responses may not be reflective of all GPs views. Most studies also used quantitative methods, which may not capture the nuances in viewpoints, especially since issues revolving around clinical genetics are highly complex.

Many characteristics of GPs put them in an ideal position to facilitate clinical genetic testing. However, based on our findings, it would be unrealistic to expect GPs to adopt clinical genetic practices without adequate support and training. Moreover, not all GPs were found to incorporate clinical genetic testing into their practice due to the uncertainty of their role. Rather than integrating the entire genetic practice into clinical care, GPs may be more inclined to adopt specific genetic practices that are more aligned to their role, and relevant to their patient profile. GPs alone may not have the sufficient expertise and resources to properly engage or manage potential or diagnosed patients with genetic conditions. As such, future interventions could assess the effectiveness of having a multidisciplinary team model that provides an integrated delivery of services involving GPs, genetic counsellors and specialists to alleviate some pressure for GPs who may be daunted by the cost, time and knowledge required in providing clinical genetic services.

Furthermore, educational interventions were found to have minimal impact on GPs adopting clinical genetic practices. This raised the need for future research on alternative strategies targeted at the clinical integration and application of genetic practice. It is also worth noting that variation in healthcare models coupled with mixed attitudes on the utility of genetic testing suggested that not all GPs were receptive to the adoption of clinical genetic services. Thus, future research should examine GPs' perspectives on providing genetic information and in relation to GPs' concerns on the lack of adequate knowledge, training and other ELSI. Future interventions should also aim to understand and contextualise interventions that fit their respective healthcare models to facilitate the smooth adoption of clinical genetic practices.

CONCLUSION

Genetic testing has the potential to revolutionalise primary healthcare and GPs are expected to play a greater role in the provision of clinical genetic services. Yet, this review found that GPs were hesitant to adopt clinical genetics in their practice due to uncertainty over what their role entails which is exacerbated by their lack of knowledge, confidence and rarity of clinical genetic cases. While existing educational interventions were found to increase GPs' knowledge and confidence, they were insufficient to drive the actual adoption of genetic practices in their clinics. The presence of mixed attitudes towards adopting clinical genetics suggests a need for further in-depth research on GPs' concerns. In addition, future research should also take into consideration the variation in healthcare models across different regions, to propose interventions that are contextualised to fit the respective healthcare models.

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