Patient Engagement in the Development of HIV-Specific Health Instruments: A Systematic Mixed Studies Review Using Thematic Analysis

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Abstract

Background:

Since the initial spread of HIV infection in the 1980s and the subsequent epidemic, people living with HIV have sought involvement in and helped define the fight against HIV/AIDS. This grassroots HIV activism markedly contributed to the development of patient engagement research methods. Involving patients in research through patient engagement may offer a diverse range of benefits, and is actively supported by funding agencies and governmental bodies. However, there have been few rigorous investigations of the conduct of patient engagement. It is unclear how patient engagement is carried out and described in published HIV health research.

Objectives:

To synthesize current evidence about the role and results of patient engagement in the development of HIV health measures as reported in the scientific literature.

Methods:

This is a mixed studies systematic review that covers scholarly publications from 1993 to 2015. Search for literature describing HIV-specific instrument development was conducted in the following databases: Pubmed, Medline, PsychINFO, Health and Psychosocial Instruments, and Embase. Quality appraisal was conducted using the *Mixed Methods Appraisal Tool*. Then, thematic analysis was performed to meaningfully synthesize knowledge generated about the topic under investigation.

Results:

Our queries generated 4363 records; after screening and verifying eligibility, 39 records were retained for analysis. The quality appraisal highlights poor reporting of engagement

methodology. Thematic analysis results suggest that patient engagement contributes to the development process as researchers report gaining insight into patient concerns. In some instances, the approach is considered essential and recognized as a collaborative approach, involving partnerships with patients. In other instances, patient engagement is considered challenging as the complexity of the research process increases.

Discussion:

Our review provides support for many of the benefits as well as some of the challenges of patient engagement espoused in the literature. Additionally, the reporting of patient engagement in the development of HIV-health measures appears limited, suggesting that the adoption of reporting guidelines should be encouraged. We are in agreement with others who have argued for standardized methods of engagement reporting and offer some suggestions.

Résumé

Problématique :

Depuis l'émergence du VIH dans les années 1980 et l'épidémie qui s'en est suivie, les personnes vivant avec le VIH cherchaient à participer à la lutte contre cette nouvelle maladie. Cet activisme populaire a contribué largement au développement de la méthode de recherche de l'engagement patient. Intégrer les patients dans la recherche peut en effet offrir une diversité de bénéfices, grâce à leur participation, au soutien actif des organismes de financement et entités gouvernementales. Cependant, il existe peu d'enquêtes rigoureuses à propos de l'engagement des patients, comme on ignore de quelle manière cet engagement s'est effectué dans la recherche médicale liée au VIH.

Objectifs :

Synthétiser les données sur le rôle et les résultats de l'engagement des patients dans le développement de mesures cliniques du VIH.

Méthodes :

Nous avons mené une recherche systématique des bases Pubmed, Medline, PsychINFO, Health and Psychosocial Instruments, et Embase, de 1993 à 2015, afin d'identifier la littérature qui décrit le développement d'instruments spécifiques au VIH. Une évaluation de la qualité a été menée à l'aide de l'outil *Mixed Methods Appraisal Tool*. Enfin, nous avons réalisé une analyse thématique pour synthétiser les connaissances générées.

Résultats :

Notre revue a identifié 4363 documents, dont 39 ont été retenus pour une lecture complète. L'évaluation de la qualité souligne une description limitée de la méthodologie d'engagement. Les

résultats de l'analyse thématique suggèrent que l'engagement des patients contribue au processus de développement des mesures, car les chercheurs rapportent avoir pris connaissance de leurs préoccupations. Dans certains cas, l'approche est même considérée comme essentielle et reconnue comme une approche collaborative, impliquant un partenariat avec les patients. Dans d'autres cas, l'engagement des patients est perçu comme difficile car il complexifie davantage le processus de recherche.

Conclusion et perspectives :

Notre étude explicite plusieurs des avantages et certains des défis de l'engagement des patients rapportés par la littérature scientifique. La description de l’engagement patient dans le développement de mesures relatives à la santé des personnes vivant avec les HIV semble limitée, indiquant que malgré certaines exceptions notables, la participation des patients pourrait être davantage symbolique qu’effective et l’adoption de nouvelles directives devraient être encouragées afin d’améliorer cet engagement. Enfin, nous sommes d’accord avec ceux qui argumentent pour des méthodes standardisées et offrons des suggestions.

1. Introduction

Patient engagement is an approach increasingly discussed in health services research as it may confer a diverse array of potential benefits for patients, researchers, and the encompassing healthcare system (1-11). Historically, the emergence and practice of patient engagement in research may largely be traced back to activism in the human immunodeficiency virus (HIV) community at the onset of the HIV epidemic in the 1980s (12-14). During this time, patients and their communities mobilized to successfully influence biomedical research and political motivations (14). Due to their pivotal and precedent setting actions, patient engagement and related practices have arisen at the forefront of biomedical research and healthcare (14).

Patient engagement has been defined as the active and meaningful participation of patients in the research process (1-3, 15, 16). It may enhance the quality of research, can assure relevance of research targets, and may contribute to greater quality and participation in the research process (2-5, 7, 17). More specifically in the development of health-related instruments in the care and research of chronic conditions, patient engagement may importantly lead to more accurate assessments of health by helping to ensure that instruments reflect patient concerns (9, 15, 18-20). However, despite the fact that the literature on patient engagement includes a number of interesting reviews (3, 5, 8-10, 21), it is still unclear how patient engagement has contributed to the development of HIV-specific health measures. In order to fulfil this gap, my purpose in this thesis was to conduct a systematic literature review on patient engagement in the development of HIV-specific health measures.

2. Literature Review: HIV and Patient Engagement

2.1 A brief historical account of HIV

HIV infections were first identified in California and New York in the early 1980s; later, cases were reported in Europe and around the world (22-24). Initially, rare health conditions were detected in previously healthy homosexual men and linked to an unknown underlying critical immune deficiency (22, 23). According to the U.S. Department of Health & Human Services, by the end of 1981, 270 cases of severe immune deficiency were identified among homosexual men, with 121 passing away that year (23, 24). For this reason, researchers first termed the condition Gay-Related Immune Deficiency (GRID), reflecting the original perception that it impacted only homosexuals (23). However, as incidence of HIV in individuals of other groups (e.g. intravenous drug users) were identified, researchers recognized that it was not limited to homosexuals and the condition was termed Acquired Immune Deficiency Syndrome (AIDS) by the Centers for Disease Control and Prevention (23).

Despite this recognition, as the virus' impact was (and remains) felt primarily in the gay community in resource-rich countries, a marginalized group, the issue of HIV/AIDS was not prioritized historically (13). At the onset of the AIDS epidemic, biomedical research about HIV was neglected and slow (13, 25). Medical needs were not being adequately addressed and frustrations grew among those impacted (13, 25). At that time in the 1980s and early 1990s, an HIV infection was a "death sentence"; over several years the virus would progressively impact the afflicted individual, interfering with their immune system. Common causes of mortality were opportunistic infections (e.g. tuberculosis or pneumonia), pathogens rarely harmful to individuals with healthy immune systems, but fatal to a person living with uncontrolled HIV (26, 27). The Mayo Clinic reports up until the mid-1990s, most patients would survive only 6-12 months after seeking medical care for HIV as only the associated opportunistic infections could be treated; without antiretroviral medication, HIV could not be addressed (12).

Per infectious disease specialist Dr. Kent Sepkowitz, in response to the inactivity of politicians and health authorities, those affected by HIV mobilized leading to some of the first examples of participatory action research and patient-oriented research (13). Many innovative approaches ranging from red ribbon campaigns to civil disobedience were attempted to publicize the unmet medical needs of persons living with HIV (PLWHIV) (13, 25). Notable successes resulted: for example, in the mid-1980s, AIDS activists organized protests to advocate for earlier releases of pharmaceutical therapies for HIV (13, 25). The rapid development of antiretroviral therapies (ART) reflects the many contributions of the HIV patient groups who, through grassroots activism, emphasized the necessity to address their medical needs (13).

2.2 Epidemiology

HIV has broad consequences beyond the individuals impacted by infection; the United Nations considers HIV infection as a global epidemic with diffuse and complex implications (13, 28, 29). The condition has a heterogeneous prevalence, with rates among the adult population as high as 27% in Swaziland, and as low as 0.05% in Afghanistan in 2014, per the Central Intelligence Agency (30). Worldwide, according the 2016 UNAIDS report, 36.7 million people were infected with HIV (0.5% of the population); according to the Public Health Agency of Canada, as of 2014, the number of Canadians infected exceeds 75 thousand (0.2% of the Canadian population) (29, 31). Due to social and behavioral factors, HIV disproportionately impacts certain groups. The risk groups and their proportions of the overall sum of Canadians living with HIV, as of 2014, are as follows: 39,630 (53% of people living with HIV) are gay or other men who have sex with men; 6,850 (9%) are aboriginal peoples; 12,960 (19%) are intravenous drug users[1]; and 11,360 (15%) are immigrants from countries where HIV is endemic (31).

2.3 HIV Treatment

Since the mid-1990s, the use of ART has greatly increased the quality of life and health outcomes of PLWHIV. With appropriate treatment and sufficient adherence, the HIV virus may be suppressed. As a result, PLWHIV can face a life expectancy and overall health status which approximates that of the general population (32, 33). If the virus is not sufficiently suppressed, it can continue to replicate and develop resistance to the medication (32, 33). Without ART, the median lifespan of an individual after infection is 9 to 11 years, as the HIV infection may progress to AIDS, a stage of HIV associated with opportunistic infections and weight loss (12, 13, 28, 33, 34). Thus, a high degree of adherence to ART treatment is vital to blocking HIV replication, in order to maximize positive health outcomes and quality of life, and reduce the risk of drug resistance and transmission.

Even with appropriate treatment, HIV infections are life-altering events, leading to salient psychological, social, and physiological ramifications for afflicted individuals. Individuals with HIV may experience considerable interpersonal consequences: HIV is highly stigmatized and can result in social rejection and alienation; events such as disclosure to sexual partners or friends can be emotionally challenging (35-37). Medical treatment of HIV also entails a variety of challenges – adherence to prescribed therapeutic regime may be difficult due to the side-effects as well as pragmatic issues, such as financial expenses and concerns about inadvertent disclosure by conspicuous intake of their medication (33, 36-39). Thus, HIV infection and its

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[1] There is some overlap in these categories, as approximately 2,400 men with HIV are in both categories of men who have sex with men and intravenous drug users.

medical treatment carry with them a considerable burden, both from health and psychosocial standpoints.

By these facts, it is apparent HIV remains an important public health concern. For this reason, people impacted by HIV continue their activism. Recently there has been a push towards increasing community engagement in HIV prevention and vaccine research (40). Overall, the mobilization and drive for involvement in care and research by people affected by HIV has been exemplary in biomedicine; they were able to successfully engage in research by setting research priorities, by updating regulations and funding priorities, and by increasing awareness of HIV. Some of the precedents for patient engagement arose in this context of HIV activism; contemporary applications reflect this ground-breaking work by those affected. This thesis and its research therefore owe themselves partially to these historical grassroots movements for patient engagement in HIV.

2.4 Understanding Patient Engagement

According to the Canadian Institutes of Health Research (CIHR) (41), patient engagement involves "patients meaningfully and actively [collaborating] in the governance, priority setting, and conduct of research, as well as in summarizing, distributing, sharing, and applying its resulting knowledge." The concept refers to the active involvement of persons affected by a medical condition in the research process and the provision of clinical care (3, 7, 17, 41). The term connotes a higher level of involvement than typically achieved in biomedical research (7, 42, 43). The rationale for patient engagement, fundamentally, is to better reflect the concerns and lived experiences of persons living with a medical condition in the research and clinical care process pertaining to their condition (17, 41, 44, 45).

By a literal definition of “patient engagement,” patients participating in clinical trials are engaged in the research process (46). In these cases, the patient’s participation will depend on the nature of the study; amongst other contributions, it may entail completing questionnaires or offering biological samples (47). These methods of involvement are invaluable to biomedical research, but for our purposes are not considered patient engagement as patients’ implication is passive – they are research subjects providing data points for researchers to analyze and interpret (3, 11, 43). Considered abstractly, the interaction between researchers and patients has traditionally been unidirectional: information is obtained from patients and flows to researchers. Patient engagement constitutes more than a passive participation of patients; it refers to patients collaborating in the research process, engaging in a bi-directional informational exchange (3, 41, 44). Patients can participate in a variety of ways for different purposes: for example, they may offer critical feedback on an early draft of a health measure, or they might participate in focus groups alongside other stakeholder groups to develop content for the measure (1, 4, 44). They might advise the academic research team on selecting appropriate outcomes and interventions, or how to best disseminate or convey the information generated in the research (1, 41, 42, 48). Worth mentioning is that patient engagement exists on a continuum: some studies might engage patients for all the purposes above and more, while other studies may involve patients for only one phase of their study (1, 4, 7, 9, 48). Framed conceptually, during engagement patients are actively contributing to the research; they offer their perspectives derived from their lived experiences with a specific medical condition, which researchers might otherwise not have access to (1, 9, 11, 15, 44, 49). Thus, through patient engagement, researchers explicitly seek patients’ perspectives and work to address them in their projects.

2.5 The Benefits of Patient Engagement

This re-orientation of the research process offers significant benefits that have been documented in the literature. By actively involving patients in health care research, many pragmatic, ethical, and theoretical advantages are expected for patients, researchers, and the broader health care system. It should be noted that some of the literature referenced in this section addresses engagement more broadly, beyond patients (1, 5, 9, 10, 17, 21, 46, 48); studies may refer to community or public participation (more encompassing categories that patients themselves are constituents of), or use different terminology, discussing patient engagement in terms of client or service user engagement.

Firstly, for patients, engagement in research can endow substantial benefits, both theoretical and pragmatic. From an ethical perspective, it has been argued that patient engagement democratizes the research process (3, 21, 46). Patients can be empowered by collaborating with researchers; engagement inherently values a patient's "experiential expertise" and perspectives (11, 44, 46), shifting the emphasis and power from traditional academic or clinical experts (1, 46). Many patients will report feeling valued and listened to, and gain self-confidence and -worth from their research participation (1, 46). Pragmatically, in many cases, patient engagement will serve to better align research and care outcomes with actual patient needs, thus better addressing them (1, 3, 46, 48). Engagement can improve the validity of research, broaden the dissemination of its results, and may enhance the eventual impact of interventions (1, 3).

Secondly, for researchers, many aspects of the research process are potentially enhanced. A study's results may be considered to have greater credibility if patients' have been involved in the research (3), as, for example, the quality and relevance of the outcomes may be improved through the engagement process (1, 11, 46). The planning and execution phases of a study may be improved, facilitating the translation of research outcomes into clinical practice (3). Practically, research may be easier to conduct if patients collaborate with researchers: many research grants emphasize patient engagement, and enrollment and retention of research participants may also be improved as the protocols may be more patient-oriented (1, 3, 46). In a qualitative investigation of researcher perspectives, participants reported benefits of engaging patients: their preconceived assumptions about the research topic were challenged which generated new insight (1, 46). They also suggested involving patients can be invaluable to understanding complex social phenomena that may surround an illness, subsequently helping to better interact with affected communities (1, 46).

Finally, for the health care system, patient engagement can improve health research by fostering greater accountability and transparency in the process (42), and helping to ensure the research appropriately pertains to patient concerns (17). Limited resources are available for research; by integrating the perspectives of patients, engagement provides some level of assurance that actual research needs are prioritized and reflected in the outcomes and interventions generated (17). CIHR guidelines suggest that involving patients improves results of the research process, as novel insights are generated from the integration and consideration of new perspectives (42). The democratization of research and the diffusion of traditional power in professional research additionally serves to better justify public funding for research by involving a more diversified public (21, 46).

To summarize, patient engagement is recognized as a beneficial approach for the different stakeholders involved in health research. Patients are better served as their needs are more likely to be directly reflected in the outcomes of the work (1, 3-5). Researchers gain access to new perspectives, while potentially facilitating their research with access to further funds, easier recruitment and retention of participants, and more effective translation and dissemination of results (3, 5). The health care system can better justify expenditures with increased stakeholder engagement and receives a level of assurance of greater research need-to-funding congruity (1, 3-5, 44).

2.6 Patient Engagement Funding and Policy

Given the many potential benefits of patient engagement, the approach has been increasingly emphasized internationally by research funding agencies and regulatory bodies (1, 3, 7, 9, 17, 21, 44-46). It is reflected in initiatives like the CIHR's Strategy for Patient-Oriented Research (SPOR), which gathers a coalition of stakeholders engaged in health care provision and research. SPOR's objective is to rectify insufficient active patient and other stakeholder involvement in the research process, as well as to better translate research to care (42, 44). Through SPOR, the CIHR offers guidance for researchers and special grants for projects, such as the *Patient Engagement – Collaboration Grants* to facilitate and encourage patient engagement (16, 42). In the United States, the establishment of the Patient-Centered Outcomes Research Institute, and in the United Kingdom, the INVOLVE networks similarly aim to support and increase patient (and other stakeholder) engagement in research (3, 4, 7, 8, 17, 46). Acting in line with a growing body of evidence and ethical imperatives, governmental bodies are working to support further engagement in health research.

2.7 Patient Reported-Outcomes and Patient Engagement

One particular area where patient engagement has been recognized as highly pertinent is in the development of health measures in the care and research of chronic conditions (9, 15, 18-20). Long-term health ailments, such as HIV, are clinically assessed through patient-clinician communication, objective biomedical markers (HIV RNA (viral load) and CD4 T lymphocyte cell count, in the case of HIV) and, increasingly, through patient reported outcomes (PROs) (12, 19). These PROs relay a patient's subjective illness experience to the health care practitioner (15, 18, 19). A patient's experience, as compared to changes in biomarkers, in many cases, is a more sensitive predictor of salient later changes in therapeutic adherence and health outcomes (15). In clinical practice, PROs can offer information about patient outcomes, developments in their quality-of-life and symptoms, and help facilitate patient-clinician communication (9, 15, 19, 20). Inadequate engagement in PRO development may result in validity and accessibility concerns for these instruments (9, 15, 18). In some cases, limitations with numerous PROs have been identified that could have otherwise been avoided had the PROs been developed using a patient engagement approach (18). For these instruments to accurately assess patient concerns and measure aspects of experience that are important to patients, it is vital that researchers integrate the patient's perspective through engagement in the developmental process (9, 15, 18, 19). The utilization of non-PRO health measures differs from that of PROs, but many of these concerns remain applicable (e.g., validity.) Despite these documented benefits, according to some literature, patient engagement in research has not markedly increased (9). This lack of widespread patient engagement implementation may be explained by some of its challenges.

2.8 Challenges Associated with Patient Engagement

Patient engagement is a complex process. Among the primary documented concerns about integrating patient engagement in research are resource and logistical demands. Involving patients may increase the length of the research process for several reasons: the engagement methods themselves can be time-consuming; patients and other lay participants may lack the requisite scientific literacy for integration into the research process, and developing literacy can be time intensive (1, 4, 8, 46). Building a working relationship with participants and overcoming traditional power relationships may also further add to the research timeline (5, 8). With added participants, there is a need to respect more individuals' time constraints, which can further slow the pace of research. Financial expenditures may also increase, both to compensate the additional patient collaborators and to defray the costs associated with the extended timeline of the research (1, 8, 43, 46). The evidence of concrete benefits for patient engagement remains limited, as there have been few systematic efforts to investigate it; thus, researchers may be hesitant to update their practices on the basis of tentative evidence (5, 8, 10, 11, 17, 43, 50).

More philosophically, researchers may have worries in relinquishing power in the research – Thomson et al.'s review cited researchers' concerns that increased lay participation could "de-professionalize" research by deflating the traditional emphasis placed on academic researchers (1, 46). Researchers have also been concerned by "scope creep", i.e. the patients being integrated into the research process may wish to expand the focus of the investigation to reflect a greater diversity of community concerns, negatively impacting the feasibility of the research (3).

Among patients, some have reported negative outlooks on and impacts from the research process (46). They stated it was difficult to express their concerns and perspectives as they assumed researchers would not respect their views because patients lacked scientific literacy or traditional academic expertise (1, 46). Engagement may also lead to uncomfortable disruptions of traditional power dynamics in the patients' communities, as the process typically involves the selection of a few individuals to provide representation for the group (49). Furthermore, in some cases, recalling their experiences for the research process can be emotionally burdensome for patients (46).

Finally, troublesome issues with implementing engagement have been recognized in the literature. The extent of engagement may be tokenistic, where patients do not meaningfully contribute to the research (1, 3, 6, 46). Researchers, especially when faced with limited resources or time, may choose to conveniently sample their participants, resulting in a potential selection bias skewing outcomes (1, 49). In the Canadian context, there are documented difficulties in engaging indigenous groups, a population disproportionately impacted by HIV (49, 51). The sample of patients engaged may thus not be representative of the broader community that the research outcomes are targeted towards (3, 49).

2.9 Patient Engagement Research and Reporting

Aside from the noted challenges of incorporating patient engagement into the research process, further concerns arise when evaluating the process and outcomes of engagement itself. Existing reviews on patient engagement highlight a scarcity of information on the topic as few systematic studies have been conducted (4, 11, 17, 50). The extant reviews have found a highly heterogeneous application of patient engagement in research (21, 52). The term patient engagement may itself be elusive: it has often been used without transparency and its operationalization is frequently poorly described (21, 52-55). Equally, evidence suggests that appropriately detailed documentation of the patient engagement process is generally lacking: studies minimally report their methods, process, and impact of engagement (17, 21, 43). In light of this limited research and being part of a research team that is developing a new patient-reported outcome measure for use in HIV clinical care and research with patient engagement (56), I was interested in examining the contributions of patient engagement to HIV instrument development.

2.10 Aim, Research Question and Objectives

My aim in conducting this literature review was to synthesize current knowledge about the contribution of patient engagement in the development of health measures for PLWHIV. More specifically, I addressed the following research question:

> What does the published scientific literature tell us about the patient engagement approach in the development of HIV-specific health measures?

To respond to the research question, I stated the following explicit objectives (EO) of the review:

OE1. To identify and describe the role of patient engagement in the development of HIV health measures

OE2: To synthesize the results of patients' engagement in the development of HIV health measures, as reported in the scientific literature

2.11 Study Context

To properly contextualize the research presented in this thesis, it is necessary to discuss the broader projects that provided the initial impetus for this literature review.

The umbrella project, within which this literature review may be situated and understood, is the I-Score Study (56-58). This project involves the development and validation of a patient-reported outcome (PRO) intended for use in the clinical care and research of HIV (56-59). The research team, led by the principal investigators Dr. Bertrand Lebouché and Dr. Kim Engler, recognized a lack of PROs in the domain of HIV that were developed in accordance with patient perspectives (56, 58, 59). The I-Score project is the culmination of the research team's concerted efforts to address this clinical and research need, as they seek to develop and implement a measure with a high degree patient and other stakeholder engagement (57, 58).

Given the emphasis on patient engagement in the I-Score Study, a sub-study was developed, eponymously entitled as the “Patient Engagement Project.” This project, co-led by post-doctoral fellow David Lessard, evaluates and considers the engagement of persons living with HIV in the development of the I-Score measure (37, 56, 57). Dr. Lessard collaborates with a committee of patients, the I-Score Consulting Team, to integrate their perspectives into the instrument, as well as to involve them in other elements of the research process, such as knowledge translation and proposal development (37, 56).

3. Methodology

Health measures are frequently developed using mixed qualitative and quantitative methods: the variables integrated into the measure are typically created with the support of qualitative methods, and the resulting tool is then validated through quantitative means (60). To account for this actuality as well as measures developed otherwise, this systematic review is conducted and labeled as a *mixed studies literature review* as cited by Pluye et al. and Grant and Booth (60-62).

3.1 Identification of relevant literature

Five health-related databases where searched for relevant works: Pubmed, Medline, PsychINFO, Health and Psychosocial Instruments (HaPI), and Embase. These databases were chosen by the research team and librarian for their relevance to health research, to allow for a comprehensive review of HIV-specific health measures. All searches were performed on November 17th, 2015. Backward citation tracking was used to check the reference lists of studies included. In the absence of sufficient information on the development of a measure, authors were contacted to provide further details or to check for the availability of additional papers on the measure's development. We did not attempt to identify gray literature as our interest was in published scientific and academic literature on instrument development. No date restrictions were applied in the searches—the entire timeline of the databases was included. No methodological limitations were applied in the search. The records identified through our database searches were compiled in an EndNote reference library and subsequently de-duplicated following guidelines devised by Bramer et al. (63).

3.2 Search Strategy

To construct our search terms for the databases, we consulted a librarian specialized in primary care research. Our search query was customized for each database, combining a variety of terms and medical subject headings, grouped around three primary concepts: (1) HIV infections; (2) health instrument development; and (3) patient engagement/participation. The searches per database are reproduced below in Figure 1.

Medline

(exp HIV Infections/ OR hiv.ti,ab,kw.) and (exp questionnaires/ OR develop*.ti,ab,kw. OR valid*.ti,ab,kw) and ((Community-Based Participatory Research/ or ((Social Participation/ or Patient Participation/ or Advisory Committees/ or Consultants/)) and Patients/) or (((patient$1 or client$1 or communit*) adj5 (perspective* or perception* or involve* or satisfaction or participat*)).ti,ab,kw.))

Embase

(exp Human immunodeficiency virus infection/ or hiv.ti,ab.) and (exp questionnaire/ or exp open ended questionnaire/ or exp structured questionnaire/ or develop*.ti,ab. or valid*.ti,ab.) and ((participatory research/ or ((Social Participation/ or Patient Participation/ or Advisory Committee/ or Consultation/) and Patient/)) or (((patient$1 or client$1 or communit*) adj5 (perspective* or perception* or involve* or satisfaction or participat*)).ti,ab.))

PsychInfo

(exp HIV/ or hiv.ti,ab.) and (develop*.ti,ab or valid*.ti,ab. or exp questionnaires/) and ((Community Involvement/ or ((client participation/ or Advisory Committees/) and Patients/)) or (((patient$1 or client$1 or communit* or participat* or consultant*) adj5 (perspective* or perception* or involve* or satisfaction or participat*)).ti,ab.))

HaPI

1. HIV.mp
2. Develop*.mp

PubMed

("HIV Infections"[Mesh] OR hiv[title]) AND ("Questionnaires"[Mesh] OR develop*[title/abstract] OR valid*[title/abstract]) AND ("Community-Based Participatory Research"[Mesh] OR (("Patient Participation"[Mesh] OR "Advisory Committees"[Mesh] OR "Consultants"[Mesh]) AND "Patients"[Mesh]) OR (((patient[title/abstract] OR patients[title/abstract] OR client*[title/abstract] OR communit*[title/abstract]) AND (perspective*[title/abstract] OR perception*[title/abstract] OR involve*[title/abstract] OR satisfaction[title/abstract] OR empower*[title/abstract] OR participat*[title/abstract])))) AND publisher[sb]

3.3 Selection of the Literature

This review is restricted to empirical research on *patient engagement in the development or adaptation of an HIV-specific health instrument*. These terms were operationalized as follows: a "patient" is a person living with HIV, irrespective of their involvement with the health care system (38); "engagement" is active participation in research, where input is directly solicited from HIV-positive persons on the instrument; "HIV-specific health instruments" include self-, researcher- or provider- administered measures or questionnaires intended for use with people living with HIV that refer to their condition. Studies were to be published in English, French, or Spanish, the languages spoken by the research team. Conference abstracts and non-empirical research, including reviews, commentaries, books and book chapters were excluded.

3.4 Selection of Relevant Studies

Following de-duplication, I screened the title and abstract of all retained records per the inclusion and exclusion criteria for the analysis. To ensure accuracy in screening, another co-author (David Lessard, henceforth DL) independently analyzed 20% of the database. An interrater reliability analysis using a Kappa statistic was conducted to verify rating consistency (64). Records of uncertain eligibility were retained for a full-text reading.

After the exclusions from the initial screening of publication titles and abstracts, the full texts of all retained publications were read through, applying the same inclusion and exclusion criteria. For this phase, DL and I independently screened all the publications. We discussed any discrepancy between us and we reached consensus in every instance.

3.5. Appraisal and Data Extraction

For the next step, I performed quality appraisal of all included studies, applying the *Mixed Methods Appraisal Tool* (MMAT) (61, 65). To ensure rater reliability, DL independently appraised 10% of the studies, performing an additional interrater reliability analysis to ensure appraisal consistency. The tool is intended to evaluate methodological studies of qualitative, quantitative, and mixed method studies during the appraisal phase of systematic reviews. The MMAT separately considers a study's qualitative, quantitative, and mixed methods components, if present.

The qualitative criteria pertain to the relevance of (1.) data sources and (2.) process of data analysis, and if adequate consideration was given for how findings relate to (3.) the context of data collection and (4.) researcher influence (65). The quantitative criteria depend on the specific methodology utilized; it is different for randomized and controlled research, for non-randomized research, and for descriptive studies. When applicable, the criteria accounts for (1.) the study sample (e.g. recruitment and randomization), (2.) the research process, (3.) the appropriateness of measures used, (4.) the outcome data rate, (5.) the participant withdrawal rate, and (6.) the response rate (65). The mixed methods criteria evaluates (1.) the relevance of the research design and (2.) the integration of the qualitative and quantitative data to address the research questions and objectives (65). Finally, (3.) the criteria ask if appropriate reflection was given to the potential limitations of the integration.

Based on appraisal results (discussed later), it was apparent the qualitative and mixed methods sections of most studies lacked clarity as to their methods (e.g. the process of qualitative data analysis and integration) and consideration of the study context and researchers' influence. However, I made the decision to not exclude these studies from the analysis as the aim of my review was to survey and understand what the published scientific literature tells us about the patient engagement process in the domain of HIV-specific health instrument development. Appraisal scores are nonetheless reported in Table 1.

Data extraction from the retained studies was conducted by DL and I, with each performing roughly half and subsequently reviewing the other half. Information extracted included the year of publication, author(s), study design, descriptions of engaged patients (age, number, etc.), descriptions of the engagement method(s), their purpose, and outcome, as well as other comments pertaining to their patient engagement approach. All relevant details of the papers were compiled in a Microsoft Word table and are reported in Table 1, alongside the appraisal scores.

3.6 Methods of Synthesis

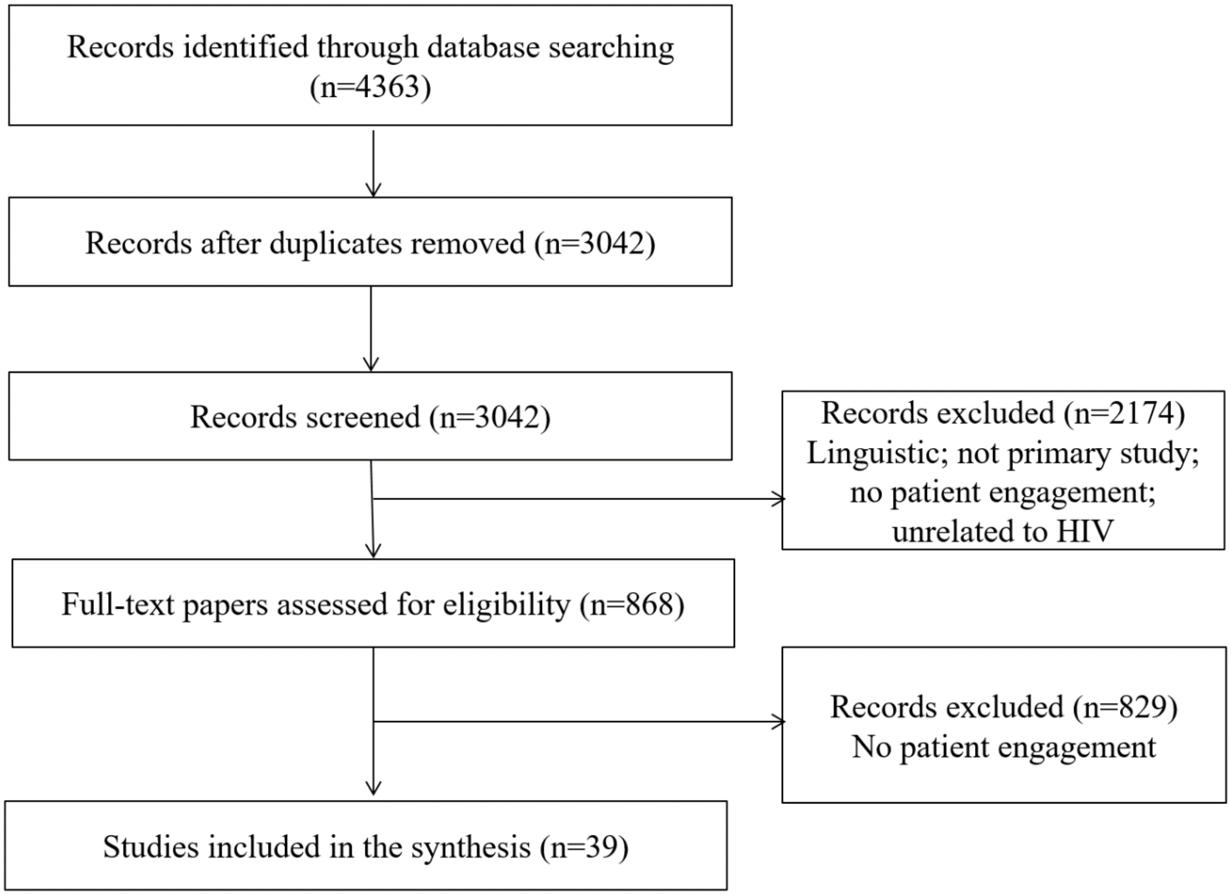
To assess the roles and results of patient engagement, an inductive thematic analysis was chosen as it offers a flexible approach to understanding and interpreting data (66). I adhered to the six steps advised by Braun & Clarke, first (1.) familiarizing myself with the data, then (2.) generating initial codes, and (3.) searching for overarching themes. I followed this by (4.) reviewing and revising the themes, and (5.) further defining and naming the themes. Finally, (6.) I drafted a report. The four themes established in the analysis reflect the roles, outcomes and perspectives reported about the patient engagement approach in the development of HIV-specific health measures in the literature. It serves to highlight gaps in knowledge to focus future research.

4. Results

4.1 Search results

As depicted in the PRISMA flowchart, Figure 2, 4363 records were identified from searching Pubmed, Medline, PsychINFO, HaPI, and Embase (67). The de-duplication process removed 1321 records, leaving 3042. After applying the inclusion and exclusion criteria to the title and abstracts, 2174 records were removed, leaving 868 for a full text review. Of these, 829 were excluded as none actively engaged patients; 39 were retained as fitting the study criteria providing the final sample. Backward citation tracking produced no new studies. For the selection of relevant studies, the Kappa inter-rater reliability score was 0.845, indicating a *Very Good* strength of agreement between reviewers, with a rate of observed agreements at 94.06% (64, 68). For the quality appraisal, the Kappa inter-rater reliability score was 0.818, similarly signifying a *Very Good* strength of agreement at 90.91% (68).

Figure 1 – PRISMA Flowchart



4.2 Study Characteristics

Of the final sample of studies, the year of publication ranged from 1996 to 2015. The studies were conducted in the United States (n=20), the United Kingdom (n=5), Canada (n=4), South Africa (n=1), Romania (n=1), Italy (n=1) Mozambique (n=1), Denmark (n=1), Spain (n=1), Sweden (n=1), India (n=1), Vietnam (n=1), and the Democratic Republic of Congo (n=1). Additional descriptive characteristics, such as the patient sample involved, the methods of data collection and analysis, the main results, and appraisal scores are stated in Table 1.

Table 1: Descriptive Characteristics and Quality Appraisal

Study number	Author(s)	Year/ Country	Study Design	Methodology & Description of instrument(s) (title, type, theme(s))	Sample of patients engaged	Patient Engagement Process: [Method(s) & purpose]	Appraisal Scores and Missing Items	Main results: [Outcomes of engagement or other comments]
#1	Aiken et al.	1997/ US	Cross-sectional study	*Multi-item patient satisfaction scale** (Scale: patient satisfaction with nursing care)	Hospitalized AIDS patients	Focus groups to develop content	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content partially developed from focus groups.
#2	Barford et al.	2005/ Denmark	Cross-sectional study	*Questionnaire assessing psychosocial and behavioral factors and treatment adherence** (Questionnaire: risk factors of poor adherence to HAART)	1. Patients (n=20) 2. Patients (n=10)	1. Qualitative interviews with patients to validate factors of poor HAART adherence 2. Cognitive interviews to examine patients' understanding of the questions	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Descriptive 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	The instrument was revised based on patient engagement. The qualitative interviews validated that the items identified in the literature were relevant for the Danish population. Authors acknowledged that their interpretation of participants' agreement during cognitive interviews may be ambiguous as they did not test in comparison to validated screening instruments.
#3	Beck et al.	1999/ UK	Cross-sectional study	*Self-completion satisfaction questionnaire evaluating standard of care of HIV outpatient services* (Survey: evaluation of standard of care of HIV outpatient services)	1. Client groups 2. Service users from three HIV voluntary organizations 3. Clients of two London HIV clinics	1. Initial workshop to assess the importance of various dimensions of patient satisfaction; 2. Open-ended interviews to define a draft questionnaire	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 Response rate was 54%. QUAN Non-Randomized 3/4 Limits of qual and quan integration is not specified. MIXED 2/3	The workshop and interviews contributed to the content validity of the instrument and the relevance of the question therein. Some of the feedback received from patients (e.g. insufficient space for comments and question; need for minor alterations to wording for some questions and explanations for others) was discussed.

						and formulate relevant questions 2. & 3. Written questionnaire to comment on the applicability, comprehensiveness, design, and wording of the first version of the questionnaire		
#4	Burrage et Vance	2008/ US	Cross-sectional study	*Client Satisfaction Questionnaire and revised version* (Questionnaire: satisfaction with AIDS service organizations (ASOs) services)	1. ASO clients living with HIV (n=3) 2. ASO clients living with HIV (n=2) 3. Clients living with HIV of an ASO in a large metropolitan area	1. Panel discussion to revise items identified through a literature review and draft a first version of the questionnaire. 2. Pilot test and commenting on early version of questionnaire to ensure validity and comprehensiveness 3. Consultation and revision of the questionnaire to conform it to the ASO's objectives and characteristics, and assessment for readability.	Consideration for research context and researchers influence is not specified. QUAL 2/4 Response rate was 40%. QUAN Non-Randomized 3/4 Limits of qual and quan integration is not specified. MIXED 2/3	The instrument was developed and subsequently revised based on patient feedback (e.g. from a 4-point to a 5-point scale.) Authors also developed a five-step guide to developing validated instruments while drawing from multiple sources of stakeholder or researcher expertise.

						Content analysis was applied to qualitative data.		
#5	Cella et al.	1996/ US	Cross-sectional study	*Functional Assessment of HIV Infection (FAHI)* (Questionnaire: physical/functional/ social/family/emotional well-being and relationship with physician, HIV subscale: symptoms and concerns with HIV infection).	HIV-infected individuals (n=15)	Structured interviews to develop HIV subscale item content.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was developed and validated through interviews.
#6.	Coetzee and Kage	2012/ South Africa	Mixed-method design	*Inventory to Assess the Structural Barriers to Clinic Attendance and Pill-taking* (Questionnaire: barriers to clinic attendance and medication adherence)	ART users	In-depth qualitative interviews to gain in-depth understanding of the main structural barriers to ART adherence from different stakeholders' perspectives.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was developed and subsequently revised and adapted based on patient engagement: qualitative interviews provided data to compose a list of the main structural problems experienced by HIV patients in terms of clinic attendance and ART-taking. Authors specified the adaptation and revision of redundant, lengthy, double-barreled and ambiguous terms.
#7	Courtenay-Quirk et al	2006/ US	Cross-sectional study	*Seropositive Urban Men's Study Questionnaire** (Questionnaire: perceived stigma in the gay community)	HIV-positive men who have sex with men (n=250), recruited in New York and San Francisco in AIDS service organizations, mainstream gay-identified venues, and	First version of paper-and-pencil questionnaire and semi-structured interview (58 open-ended questions) to develop items for the questionnaire on perceived	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4 MIXED 3/3	Instrument content was partially developed from information obtained during patient engagement.

					public sex venues.	stigma (in English, 30$ compensation) The content from these interviews was transcribed and coded, using a series of codes developed a priori by the researchers and refined during analysis to reflect emergent themes		
#8	Curtis et al.	1999, 2000/ US	Prospective cohort study	*Measure of quality of patient-clinician communication of end-of-life care* (Questionnaire: quality of patient-clinician communication about end-of-life care)	1. Patients with advanced AIDS (n=57); recruitment through advertisements at community-based organizations, university and private clinics, and an AIDS research clinic, and a hospital registry of patients interested in research. Stated demographics: 9% female;	1. Structured 60-90 minute face-to-face interviews: patients read a copy of the draft questionnaire while the interviewer reads questions aloud and recorded each response to assess the quality of each item. 2. Focus groups to identify four items on the quality of patient-clinician communication about end-of-life care	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was generated and validated in focus groups.

					median age: 39 y.o.; race; HIV risk characteristics (msm; injection drug user); time since AIDS diagnosis 2. Participants having Center for Disease Control and Prevention level C3 HIV disease			
#9	Dang et al.	2012/ US	Cross-sectional study	*Survey Instrument** (Questionnaire: satisfaction, recommendations and trust; scales)	11 English- and 10 Spanish-speaking patients living with HIV (n=21)	One-on-one, face-to-face, audio-taped cognitive interviews for survey pre-test and adaptation, and open-ended questions to verify comprehensiveness (10$ compensation)	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Survey was revised on basis of feedback obtained from cognitive interviews: the wording was modified; items were added; and the response scale was altered. Authors suggest the patient engagement validated the instrument "survey items reflected all aspects of the clinic experience salient to patients."
#10	Davis-Michaud et al.	2004/ US	Multimethod study	*Exploration of patient preferences to aid in the development of quality measures to assess quality of health care for PLHIV.*	PLHIV in two cities (n=29)	Focus groups (method described) to prioritize 18 quality indicators for importance in assessing quality of care and informing consumers	Consideration for research context and researchers influence is not specified. QUAL 2/4 QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was generated and validated in focus groups. Authors reported gaining novel perspectives they considered salient.

				(Questionnaire: patient preferences in health care)		about the performance of health care organization.		
#11	Dima et al.	2013/ Romania	Cross-sectional survey	*CEAT-VIH adaptation** (Questionnaire: adherence-related behavior and satisfaction with treatment)	Convenience sample of service users (n=11), aged 18-22 years old, 5-14 years of education	Face-to-face cognitive interviews to assess item clarity and comprehensibility, and make additional comments.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	The content of the instrument was revised based on patient feedback: wording was simplified or otherwise altered.
#12	Flicker et al.	2004/ Canada	Community-based participatory research model	*Brief Structured Surveys** (Questionnaire: demographics and internet use)	Group of HIV-positive youth	Stakeholder group who collaboratively developed the research design, instruments and protocol	Consideration for research context and researchers influence is not specified. QUAL 2/4	Instrument and research design and protocol were developed by stakeholder group
#13	Garber et al.	2007/ US	Questionnaire development and cross-sectional survey	*Questionnaire on attitudes and concerns about HIV treatment trials* (Questionnaire: experiences with HIV and treatment trials)	HIV-infected African-American patient (n=12) volunteers recruited in a clinical site; 40% female, all aged 25-44, a third had participated in HIV trials.	Focus group for questionnaire revision and critique	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument was validated and revised by focus groups: items were reworded and clarified and their presentation was modified.
#14	Holmes and Shea	1998/ US	Questionnaire development and	*HIV/AIDS-Targeted Quality of Life (HAT-QoL)*	Convenience sample of HIV-positive urban persons (n=42), recruited in	First four groups used nominal group techniques (subjects individually	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4	Focus groups identified and contextualized content from with which to develop items from. The last two focus groups revised the instrument, leading to the removal of items.

			cross-sectional survey	(Questionnaire: Dimensions of well-being)	clinical sites, aged 18 years old and above, with ability to communicate effectively in English divided in six groups depending on their risk factors (injection drug use, blood transfusion, unsafe sex practices.	identified items, and then discussed them in group; each group voted for the 7 most important items) to generate content Last two focus group were used to revise the questionnaire and assess relevance and comprehensibility	Limits of qual and quan integration is not specified. MIXED 2/3	
#15	Holzemer et al.	1998/ US	Descriptive study design	*Living with HIV Scale* (Questionnaire: Quality of life)	Purposive sample of Hispanic, Anglo-American, and African American patient participants (n=38) Grounded theory is used as means of data analysis.	Individual interviews to generate the conceptual framework for the instrument	Consideration for researchers' influence is not specified. QUAL 3/4 Convenience sampling is used. QUAN Non-Randomized 3/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument and its framework was developed partially based on information obtained from Interviews. The wording of items was also revised.
#16	Jallow et al.	2007/ Sweden	Cross-sectional Study	*Pharmacy staff experience with patients receiving ART**	Panel of HIV-infected individuals (n=15) who work as managers or	In-depth semi-structured interviews, lasting 1h to 4h, to develop the questionnaire,	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4	Instrument content generated partially in the interviews.

				*Pharmacy staff and patient questionnaire** (Questionnaire: problems with HIV care in pharmacy)	coordinators at the association level, who were representatives of these 3 specific groups: homo- or bisexuals (4 men and 1 woman), former injecting drug users (2 women and 2 men), and individuals of African origins (3 men and 3 women).	and validation of the readability and relevance of the pre-test questionnaire	Limits of qual and quan integration is not specified. MIXED 2/3	
#17	Justice et al., Wu et al.,	2001/ US	Multistage questionnaire development design	*HIV symptom index* (Questionnaire: symptoms)	Men and women with HIV	Formed part of a committee to advise the research team on the measurement and interpretation of clinical and economic outcomes, and advise on wording, appropriateness, and accessibility.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 Recruitment method and response/outcome rate is not specified. QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was revised and approved by the committee: members reworded content and suggested additional items.

#18	Kalichman and Nachimson	1999/ US	Cross-sectional study	*Self-Efficacy for practicing safer sex and serostatus disclosure* (Scale: disclosure)	HIV-seropositive men (n=16) and HIV-seropositive women (n=6)	Focus groups to validate the relevance and comprehensibility of items	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was generated and validated by focus groups.
#19	Knobel et al.	2002/ Spain	Prospective observational study	*Simplified medication adherence questionnaire (SMAQ)* (Questionnaire: measure of adherence for HIV infected patients)	HIV patients	Formed part of research group to develop and adapt the questionnaire to HIV-infected patients.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 Recruitment method is not specified. QUAN Non-Randomized 3/4 Limits of qual and quan integration is not specified. MIXED 2/3	The instrument was revised by the research group. Based on feedback, content was eliminated, reformulated, or added.
#20	Land et al.	2013/ UK	Cross-sectional study	*Patient satisfaction survey* (Questionnaire: satisfaction with care)	1. Convenience sample of patients/service users in a clinical site: men (n=21) and women (n=20), 37% attending the clinic for the first time, 46% aged under 25 and 40% from black and ethnic minority group. 2. Clinic users (n=10)	1. Semi-structured interviews to identify and refine items. 2. Cognitive interviews to assess comprehensibility, relevance and appropriateness of questionnaire Content analysis was used to identify emerging themes in interview transcripts.	Consideration for research context and researchers influence is not specified. QUAL 1/4 Convenience sampling was used; response rate is 51%. QUAN Non-Randomized 2/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was generated, validated, and revised in interviews. Authors mention the development of the instrument benefited from the patient engagement.

#21	Moore et al.	2010/ US	Cross-sectional study	*“The Quality of Care Through the Patient’s Eyes”—HIV questionnaire (QUOTE-HIV* (Questionnaire: quality of care)	HIV-positive African-American men (n=8) volunteering in AIDS service organizations and community-based organizations; and HIV-positive African-American women (n=8) recruited by a counselor who was HIV-positive; all participants were 18 years old or above and had received health care for at least 1 year.	Two 90-minute sex-specific focus groups to validate the questionnaire and assess its appropriateness; sessions were recorded and transcribed; no remuneration was provided to participants. Thematic analysis was conducted on the qualitative data	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN non-randomized 4/4 MIXED 3/3	Instrument content was validated by focus groups. Authors also specified they received recommendations for revising the instrument.
#22	Newshan et al.	2002/ US	Cross-Sectional study	*HIV Symptom Index (Modified/Updated)* (Questionnaire: symptoms)	Persons with HIV (n=2)	Formed part of a panel of experts who revised the questionnaire to ensure face validity.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 Response/outcome rate is not specified. QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument items were revised and validated by the panel.

#23	Nokes et al.	1997/ US	Clinical tool development	*HIV Needs Assessment Tool** (Questionnaire: patient satisfaction with care)	Clients with HIV disease (n=2) and clients with AIDS (n=2)	Formed part of a group to revise and validated the questionnaire.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 Convenience sampling was used; response/outcome rate is not specified. QUAN Non-Randomized 2/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was validated by patients.
#24	O'Brien et al. #1	2014/ Canada	Cross-sectional and clinical tool development	*HIV Disability Questionnaire (HDQ)* (Questionnaire: symptoms and experiences of disability)	Adults living with HIV (n=2)	Formed part of a Community Advisory Committee: attended two meetings to discuss the content, wording, and order of items. The committee was to provide advice and guidance in all phases of the research (detailed description of recommendations at each meeting) ; consulted with the broader community between meetings	The process of the qualitative data analysis is not specified. QUAL 3/4	Instrument content was generated, revised, and validated by the committee: items were eliminated, modified, or added. Authors mention they explicitly sought multi-stakeholder representation to enhance relevance feasibility, relevance and translation; two members enhanced the geographic and gender diversity on the Committee; two members of the Committee participated in the framework; Committee remained small enough to allow meaningful engagement of members. The authors also report that the committee was integral to generating new items and revising the questionnaire. However, they did express concerns about the tendency to continue generating items outside of their scope and beyond what would be feasible.
#25	O'Brien	2014/ Canada	Cross-sectio	*HIV, Health and Rehabilitation Survey*	Adults living with HIV (n=5)	Formed part of a partnership providing	The process of the qualitative data analysis is not specified; consideration for research context	Authors discuss the used pf equal partnership and collective involvement in all phases of the study, including

	et al. #2		nal study	(Questionnaire: disability and rehabilitation)		advice and guidance throughout all phases of the development and implementation of the survey. The partnership worked as a team on survey development, revision, and pre-testing for content, clarity, and format.	and researchers influence is not specified. QUAL 1/4 Response rate was 53%. QUAN Non-Randomized 3/4 Limits of qual and quan integration is not specified. MIXED 2/3	application for funding, development of the survey, implementation, and interpretation of findings. Participatory approach is mentioned as strength, but the authors acknowledge it may be burdensome for organizations with little time or mandate for engagement.
#26	Owen-Smith et al.	2010/ US	Cross-sectional study	*Complementary and Alternative Medicine (CAM) Use Survey-adapted** (Questionnaire: complementary and alternative medicine use)	5 groups of 6-8 participants (n=35) who had an AIDS Diagnosis, were identified as African-American, were 21 years of age, spoke English, and were not cognitively impaired.	Two focus groups to discuss the strength and limitation of the original CAM survey; two unstructured focus groups to provide critical insight on the operationalization of CAM-related behaviors; and one focus group to discuss and revise the first version of the adapted CAM survey.	QUAL 4/4 QUAN non-randomized 4/4 MIXED 3/3	Instrument content was revised and validated in focus groups.

						Content analysis was used on the qualitative data.		
#27	Page and Mitchell	2006/ Canada	Cross-sectional study	*Patients' opinions on privacy, consent and the disclosure of health information for medical research** (Questionnaire: experience with medical research)	Small number of AIDS patients	Consultations to develop and revise questionnaire	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 Recruitment method is limited; response rate is 49%. QUAN Non-Randomized 2/4 Limits of qual and quan integration is not specified. MIXED 2/3 Only mailed out from one clinic; very low response rate	Patients provided recommendations that were integrated into the questionnaire.
#28	Peipert et al.	2014/ US	Cross-sectional study	*FACIT TS general (G) and patient satisfaction (PS)* (Questionnaire: communication with health care providers and treatment satisfaction)	English-speaking adult undergoing outpatient treatment for cancer or HIV/AIDS at two clinics	Semi-structured interviews to generate and rank items	Consideration for research context and researchers influence is not specified. QUAL 2/4 Recruitment method and response/outcome rate is not specified. QUAN Non-Randomized 2/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was generated in interviews.
#29	Pereira et al.	2003/ US	Prospective study design	*Life Experiences Survey-10 (abbreviated)* (Questionnaire: stressful life events)	HIV-positive women of color in obstetric-gynecology settings, especially postpartum women	Focus groups to select most relevant items and adapt the initial Life Experiences Survey.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 Recruitment method and response/outcome rate is not specified. QUAN Non-Randomized 2/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was identified and generated in focus groups; participants revealed stressors associated with the postpartum period, and other more general stressors.

#30	Posse et al.	2013/ Mozambique	Cross-sectional study	*Food Assistance Program** (Questionnaire: satisfaction with a food assistance program)	Three simple random samples of adult (18+) HIV patients in three of five districts; mixed groups in terms of gender and age; 5 to 10 participants in each group.	Focus groups to generate items. Focus groups transcripts were analyzed using content analysis.	Consideration for research context and researchers influence is not specified. QUAL 2/4 Response/Outcome rate is not specified. QUAN Non-Randomized 3/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was generated in focus groups.
#31	Sheldon et al.	1993/ UK	Cross-sectional study	*HIV Care and General Practitioners ** (Questionnaire: past and current involvement with general practitioners)	1. Interested HIV-positive patients. 2. HIV-positive patients (n=20)	1. Discussions to ascertain the issues of concern regarding the use of general practice for HIV-positive patients and generation of items. 2. Semi-structured interviews to ensure comprehensiveness and comprehensibility of questionnaire.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was generated in discussions with patients; a preliminary version of the instrument was subsequently validated with patients in interviews.
#32	Starace et al.	2002/ Italy	Cross-sectional study	*World Health Organization Quality of Life - HIV*	HIV patients	Focus groups to generate items and profiles suitable for the assessment of	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4	Instrument content was generated and subsequently validated in focus groups.

				(Questionnaire: physical, psychological, level of independence, social relationship, environment, beliefs)		quality of life in HIV-infected subjects.	QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	
#33	Ramanathan et al.; Swendeman et al.	2013; 2015/ US	Longitudinal study	*Mobile Phone HIV Surveys** (Questionnaire: physical and mental health symptoms; medication adherence; consummation; sexual encounters)	PLHIV (n=29) recruited from primary study site: 75% men; 10% transgender; 10% injection drug users; ethnically diverse (African American, Latino, and White); age range from 30 to 60; financially disadvantaged.	Two formative focus groups to inform the design and anticipate challenges with using mobile phone and web surveys. Thematic analysis was performed on the data.	Consideration for research context and researchers influence is not specified. QUAL 2/4 30% of participants discontinued study prior to completion. QUAN Randomized-Controlled 2/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was generated in focus groups; patient also provided information on administration of the instrument.
#34	Tran et al.	2012/ Vietnam	Cross-sectional study	*Satisfaction with HIV/AIDS Treatment Interview Scale (SATIS)* (Questionnaire: satisfaction with care)	HIV/AIDS patients	Three focus groups to incorporate stakeholders' perspectives and improve face validity	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 Outcome data is not specified. QUAN Non-Randomized 3/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was validated in focus groups.

#35	Walsh et al.	2001/ UK	Cross sectional study	*Reasons for non-adherence to antiretroviral therapy Scales** (Self-administered questionnaire: treatment adherence, reasons for missing doses)	Patients taking HAART	Focus groups and informal interviews for item generation on reasons for missing doses	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was generated in interviews and focus groups.
#36	Webb et al.	2001/ UK	Cross-sectional study	*Treatment-related Empowerment Scale (TES)* (Questionnaire/scale: components of communication, treatment choice, decision-making and satisfaction with care)	HIV-positive patients	Unstructured interviews to generate items and select specific empowerment experiences.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 Recruitment method and outcome data is not specified. QUAN Non-Randomized 2/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was generated in interviews.
#37	Wig et al.	2008/ India	Cross-sectional study	*Measuring multiple dimensions of health** (Structured pro forma interview questionnaire: physical, mental, and social life)	1. Patients visiting the HIV clinic. 2. HIV-positive patients in the Outpatient Department of a tertiary care hospital in North India.	1. Focus group discussions to adapt the original version of the questionnaire. 2. Pre-test to validate the translation in Hindi.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 QUAN Non-Randomized 4/4 Limits of qual and quan integration is not specified. MIXED 2/3	Instrument content was adapted and validated by focus groups.
#38	Wu et al.	1996/ US	Cross-sectional study	*Visual Function and Quality of Life in Patients With*	Sample of patients (n=18) followed up in	Semi-structured interviews to ensure inclusion of relevant	The process of the qualitative data analysis is not specified; consideration for research context	Instrument content was generated and validated in interviews.

				*Cytomegalovirus Retinitis Scale** (Questionnaire: visual symptoms, visual function in daily activities, and impact of treatment administration)	two CMV retinitis outpatient clinics	domains in the questionnaire.	and researchers influence is not specified. QUAL 1/4 Recruitment method and outcome data is not specified. QUAN Non-Randomized 2/4 Limits of qual and quan integration is not specified. MIXED 2/3	
#39	Zola et al.	2014/ Democratic Republic of Congo	Cross-sectional study	*Partages Questionnaire (Voluntary Disclosure to one's steady sexual partners)** (Questionnaire: disclosure)	People living with HIV	Formed part of an equitable partnership to develop and pre-test the questionnaire.	The process of the qualitative data analysis is not specified; consideration for research context and researchers influence is not specified. QUAL 1/4 Convenience sample is used. QUAN Non-Randomized 3/4 Limits of qual and quan integration is not specified. MIXED 2/3	Authors developed a 'memorandum of understanding' before the start of the project, defining stakeholder engagement under the principles of mutual respect and understanding, balanced power in decision-making, and control in all phases of the projects The engagement process empowered people living with HIV and other community members involved in the research

*No instrument name provided by authors; instrument name provided by reviewers.

4.3 Quality Appraisal

The *Mixed Method Appraisal Tool* (MMAT) was used to appraise the included 39 studies (61, 65). Overall, considerable heterogeneity was noted in the scores between studies and between subsections of studies. The average score for the qualitative criteria, out of 4, was 1.33 while the mode was 1. The average score for the quantitative evaluation, out of 4, was 3.41 with a mode of 4. Finally, for the mixed methods criteria, out of 3, the average score was 2.08 while the mode was 2. As mentioned, irrespective of the score obtained in the appraisal, studies were not excluded.

4.4 Thematic Analysis

Four themes were extracted from the literature on the role and results of patient engagement approaches. I identified patient engagement (1) as contributing to the development of the instrument, (2) as a challenging approach, (3) as integral to the research process and outcomes, and (4) as a collaborative approach. These themes are summarized in Table 2.

4.5 Theme 1: Patient engagement as contributing to instrument development

All 39 studies discussed their application of a patient engagement approach as pertaining to the development of the health measure in some way. Patient engagement fulfilled this outcome in a variety of ways. Studies reported that engaging patients in the research process helped generate content for the instrument, or to revise or validate the instrument and its content. The data extracts below demonstrate the diverse ways in which patient engagement may contribute to instrument development, depending on research design and need.

4.5.1 Content Generation

Content generation, referring to the creation or adaption of material used in the instrument, was commonly attributed to patient engagement in the development of HIV-health instruments.

This task was either done directly by the patients, or indirectly, as researchers would draw on information contributed by patients. Curtisa et al. describe the former approach, as they state that (69) “focus groups [with patients] were used to develop four generic items assessing quality of communication about end-of-life care.” This suggests patients more directly created the content.

Examples of indirect content generation are reported by Aiken et al. (70). They state “the AIDS care content was developed from focus groups with hospitalized AIDS patients.” Similarly, Kalichman and Nachimson write (71) “[from] information gathered from formative elicitation research with HTV-seropositive men and women, six scenarios were constructed within which potential risk behaviors may occur.”

4.5.2 Validation

Beyond content generation for instruments, studies often reported patient engagement as contributing to validation process of the instrument. Validation helps ensure that the instrument measures what it is intended to, and is therefore of paramount importance for health instruments.

Beck et al. explain the contribution of patient engagement in the development of their health instrument as ascertaining the validity of their other research:

> That the questionnaire has content validity and asks relevant questions was supported by the fact that the focus groups and interviews provided a list of dimensions of the service which were important to service users which was very similar to that obtained from the literature. (72)

After translating and adapting an instrument to a Danish cultural and linguistic context, Barfod et al. explain “[their] own qualitative interviews with 20 patients suggested that these factors were relevant in a Danish population as well.” (73)

4.5.3 Revision

The final commonly cited purpose and outcome for patient engagement in HIV measure development was to revise the instrument, a process that entails reviewing, modifying and improving it.

For example, Holmes and Shea reported:

> These two groups critiqued an early construction of the pilot questionnaire; their task was to discuss the relevance and comprehensibility of items and to identify any gaps in the questionnaire's assessment of health-related quality of life. The 83-item draft was distributed to focus group participants, who then were asked to self-administer it. They were instructed to circle words or whole item stems that they did not understand. They were asked to write in questions that they thought should be included and to strike questions that they thought should not be asked. After this self-administration and critique, the questionnaire was discussed item-by-item by the group as a whole. The two groups agreed that no items should be added, but that seven items should be removed. (74)

A further example is provided in O'Brien et al.'s study:

> The survey instrument was reviewed, revised, and pre-tested three times by our entire team. Pre-testing involved all team members independently reviewing the questionnaire for content, clarity, and format. Members of the team living with HIV completed the questionnaire as potential participants. (75)

These data extracts serve to exemplify the roles played and the outcomes attained by patients engaged in HIV-measure development. As established, studies applying a patient engagement approach will commonly involve patients in the generation of content, and to subsequently validate and revise the instrument under development.

4.6 Theme 2: Patient engagement as a challenging approach

A number of studies (n=4) discussed the difficulties associated with patient engagement. These studies highlighted the potential for disagreement in the context of collaboration and shared decision-making,

Beck et al. reported disagreements between the academic researchers and patients involved:

> One question that had addressed the issue of privacy was found to be confusing as it was nuclear if it was asking about whether the physical environment of the clinic helped maintain privacy or whether it was asking about confidentiality of information. This question was divided into two questions, each focused on one of these issues. There were two questions in particular which were criticized. One was a question asking about occupation; this was felt to be too personal and compromised anonymity and irrelevant to those who were not working. This question was removed from the questionnaire. The other question which respondents criticized asked about whether they felt the staff had prejudiced attitudes to any client groups. This was perhaps the most threatening question as it was asking for direct judgement of the staff members. A number of respondents felt that they were not in a position to judge. The Working Party felt that this was nevertheless an important issue and one that should be addressed in the questionnaire. To accommodate the criticism of the question and yet keep it in the questionnaire, the wording was changed to make it more explicit that clients were being asked for a subjective opinion. (72)

This extract speaks to the compromises that may be necessary when involving more participants in the decision-making process of instrument development. As exemplified, patients may have different perspectives than researchers, which can make working together more difficult in some instances.

This concept is echoed in one of O'Brien et al.'s study, where they state:

> First, we struggled with the tendency to continue to generate items that were important or related to disability, but were beyond the construct. For example, the community advisory committee raised stigma, social support, coping strategies, and causes of disability as concepts to consider including in the HDQ. Health challenges represented in the HDQ can emerge from multiple sources that are difficult to determine. (76)

And:

> Another challenge involved balancing our enthusiasm to immediately implement the HDQ into community-based practice with the need for further measurement property assessment. Since the development of the HDQ, we pilot tested the questionnaire with 22 adults living with HIV and 5 clinicians who work in HIV care. […] Findings from this sensibility assessment were integral in providing feedback on the HDQ content, item wording, terminology, and format. This can be frustrating for community who are eager to implement a new measure into practice. (76)

Furthermore, they discuss logistical challenges in relation to engaging patients in the research, as the approach may necessitate a greater time commitment.

> We surpassed our originally proposed timelines for HDQ development. This may be attributed to the time-intensive nature of community-engaged (or participatory) research that requires ongoing communication and collaborative decision making between community and academic partners. (76)

The authors explained their resolution of these issues:

> [I]n collaboration with the committee advisory committee, we established a clear plan for further measurement property assessment with goals for HDQ implementation in the future. Ongoing knowledge translation and community engagement throughout this process will be integral to ensure the successful next steps of HDQ property assessment and implementation. (76)

As a patient engagement approach increases the number of groups involved in a research, the complexity of the research process increases – more perspectives must be considered and reconciled in the context of shared decision-making. Patients represent their own perspectives – this is the reason they are involved – and these perspectives may not always mesh with those of the primary academic researchers. Thus, patient engagement may contribute to making the research process more elaborate and difficult.

4.7 Theme 3: Patient engagement as integral to the research process and outcomes

Pertaining again to the experiential expertise and community ties of patients, some studies (n=4) explicitly acknowledge the importance of patient engagement to research process and outcomes.

For example, Burrage and Vance state (77) "it was crucial that instrumentation be succinct and tailored to this unique group of ASO clients." In their case, patient engagement ensures the instrument is tailored to their targeted audience, as patients are able to validate the relevance of the instrument or help correct it otherwise.

O'Brien et al. provide further support for the necessity of patient engagement, reporting:

> Our community-engaged approach was integral to the development of the HDQ. Our research adhered to key principles of partnering with the community in research, such as recognizing the community possessed unique contributions to dedicate to the HDQ development, integrating research and experiential knowledge for the benefit of all partners, and ensuring sustainable collaborative partnership throughout all stages of the research. (76)

The potential of patient engagement to provide insight and perspective for the development of the instrument was described in numerous studies (n=16) as well. Pereira et al. mention (78) "[f]ocus groups were convened to determine the most frequent and salient life events."

Implicitly, patient engagement is vital as researchers would not be able to gain a similarly reflective understanding of a PLWHIV's life events. Finally, Owen-Smith et al. reinforces this perception as they explain:

> The purpose of including two additional, unstructured focus groups was to provide a forum within which participants could define CAM on their own terms, thereby providing additional critical insight into the operationalization of CAM-related behaviors. (79)

Researchers, as demonstrated in the text excerpts above, insist on the unique perspectives that PLWHIV have. For this reason, many consider patient engagement an essential component of conducting HIV research.

4.8 Theme 4: Patient Engagement as a collaborative approach

The final theme extracted from the literature was the consideration of patient engagement as a collaborative approach. Studies (n=4) reported partnerships with patients – often presented as equitable in nature – and the approach itself as being beneficial to the broader community, through empowering patients and other members.

Studies might explicitly acknowledge the patient engagement approach as collaborative, as exemplified by Flicker et al. (80) who state "[a] stakeholder group of HIV-positive youth (trained as community researchers) and supporting professionals collaboratively developed the research design, instruments and protocol.“

O'Brien et al. similarly report:

> Our aim was to describe our community–academic partnership in the development of a new self-administered instrument, the HDQ, using a community-engaged approach. […] The committee

> worked with the lead author, who presented suggestions for HDQ content and wording at face-to-face meetings followed by questions for consideration in HDQ development." (76)

In their study, Zola et al. extensively write about their collaborative approach to patient engagement, describing:

> In the "Partages" project, CBPR principles have been adhered to throughout the process. CBOs members, PLHIV and researchers were involved, in an equitable partnership. Tools, like a memorandum of understanding, were developed before the start of the project to ensure mutual respect and understanding, balanced power in the decision-making as well as shared control over all phases of the research process. (81)

Later, they discuss the benefits for the engaged parties:

> Community members were trained to research methods and research ethics. The results of the project were presented to participants and stakeholders in all the countries where data were collected. […] Community mobilization was strong during the whole process. This project gave a voice to the community about a very sensitive issue. […] The CBPR approach empowered community stakeholders and PLHIV in five different countries. Strong partnerships were created throughout the process between researchers and community members. Several workshops were organized with researchers and community members from the seven countries of the consortium, allowing fruitful exchanges of experience and mutual empowerment. And social change, which is a goal in CBPR, was achieved, as the management of the serostatus disclosure issue changed after the project among CBO members and community leaders. (81)

The research process can be mutually beneficial in a broad array of means – for academic researchers and for the additional stakeholders engaged, beyond merely an improved health instrument. The patient engagement approach is often collaborative and serves to empower those members who are engaged, as well as their communities by providing them a voice and even tangible skills to take away.

Table 2. Main Themes of Patient Engagement in in the development of HIV-specific health measures in the published literature

1. **Patient engagement as contributing to the development of the instrument**
The patient engagement approach is commonly used to generate content, revise, and validate HIV-specific health instrument. These applications make use of patients' lived experiences with HIV, to ensure the instrument reflects their concerns.

2. **Patient engagement as a challenging or difficult approach**
Researchers often reported logistical challenges involved with engaging patients in the research process, suggesting that the approach could be more burdensome.

3. **Patient engagement as integral to the research process and outcomes**
The patient engagement approach was recognized as vital to the development of HIV specific health instruments; patients were able to provide insight and perspectives otherwise unavailable to the academic researchers.

4. **Patient engagement as a collaborative approach**
Researchers often referred to patient engagement as a collaborative approach – that patients were partners in the research process.

5. Discussion

This review identified published academic literature in which HIV-specific health measures were developed or adapted with patient engagement. As evinced by scores obtained with the *Mixed Methods Appraisal Tool* (61), few studies thoroughly reported on the qualitative patient engagement components of the research process. However, to comprehensively address my research question, no studies were excluded. This review clearly summarizes that patient engagement in the development of HIV clinical tools was instrumental in content generation, revision, and validation. On one hand, it highlights that the process of patient engagement was sometimes acknowledged as a challenging approach, as it increased logistical or other challenges due to the inherent increase in complexity of the research process. On the other hand, studies generally recognized the approach as being vital to the research – in multiple instances, studies reported gaining vital insight from patients due to their experiential expertise. Finally, and importantly, patient engagement was described as a collaborative approach in which patients worked *with,* not *for,* the academic researchers.

It is important to note that the quality appraisal for this systematic review, conducted per the validated MMAT, found considerable heterogeneity in the scores between studies and between sections of studies in general (61, 65). The quantitative components of studies, almost without exception, were better reported than the qualitative components (where patient engagement is described.) In some instances, the description of the qualitative processes of patient engagement in the development of the health measure were minimal, amounting to a sentence or two in the entire study. Similarly, most studies did not report on the integration of qualitative and quantitative data.

This finding of methodological reporting disparity likely reflects an emphasis on the reporting of the quantitative components of the studies. Frequently, studies described these aspects in much greater detail. We cannot conclude much with respect to the patient engagement methodology, simply because its reporting was generally insufficient. This extent of reporting can be problematic in several scenarios: for example, in clinical or primary research settings, where the instruments may be used as a data collection method, it can be more difficult to ascertain for whom the instrument would be pertinent to (3, 21). If the instrument is being considered for the evaluation of health-related quality of life in indigenous populations, but has been developed without consideration or integration of their perspective, it may not adequately reflect their concerns (49, 51). Lived experiences of different groups are unique; adequate representation is necessary to develop an instrument with appropriate validity for people of different life circumstances (49, 51). For example, if studies do not report patient demographics, it is difficult to determine if the instrument possesses specific cultural validity. Furthermore, with minimal reporting on the methods, the process, and the outcomes of engagement, it can be difficult to appraise the results and to develop best practices for patient engagement in health research (3, 5, 21).

Several reasons may account for the low levels of qualitative reporting. Academic journals have word length limitations; thus, studies, in needing to adhere to these limits may need to focus on the discussion or results, in lieu of emphasizing the methods sections (the section in which authors would typically discuss the process and details of patient engagement). Beyond word limitations, the literature suggests there is a lack of standards in reporting on engagement (3, 21). It is likely for these reasons that wide variance in levels of reporting was observed in the studies.

The analysis performed has identified common contributions of patient engagement and researcher perspectives in its regard in the development of an HIV-specific health instrument. In these instances, researchers would acknowledge the benefits or even necessity of patient engagement. These findings reflect the fundamental benefits of the patient engagement approach, that patients uniquely possess experiential expertise (11, 44, 46, 49). Patients can use their experiences living with the medical condition to ensure their concerns are reflected in the instruments' content, helping ensure the pertinence of the instrument. Researchers otherwise do not have access to this lived experience, and cannot easily ascertain what the content should be or if is appropriate, whereas patients can, by virtue of their lived experiences. That study authors will speak of patient engagement in this manner and emphasize the perspectives gained adds to the growing impetus to investigate patient engagement further, to develop best practices, and to reinforce implementation (1, 3, 5, 52).

Another important result of this review has been to put forward the interest of patient engagement as a collaborative approach, that is to say, patients working in conjunction with traditional academic researchers to co-create the research. Patient engagement can provide for positive-sum relationships and outcomes: the approach can be mutually beneficial for all the involved stakeholders. Patients, academics, clinicians, and other parties may have their desired outcomes met and concerns addressed to a greater level. This is an important consideration, as literature has questioned the extent of engagement – that in some instances, patient engagement will be merely symbolic (1-5). Thus, the recognition of patient engagement approaches for leading to equitable research partnerships is an important outcome that is not always achieved, and may fundamentally alter the research process. As discussed, to a degree the validity and applicability of health instruments depends on their reflection of patient concerns, which is more likely with a greater degree of engagement (1-3, 82).

Studies identified in our review reported challenges associated with patient engagement, many of which were described elsewhere in the literature (summarized in section 2.8.). Inherently, the process of patient engagement may entail considerable organizational challenges as it may increase the time and resource expenditure necessary for the research (1, 4, 8, 46). These challenges pertain directly to the type of engagement that occurs; thus, as the number of patients engaged and the extent to which they are involved increases, the organizational, time, and resource demands of researchers increase to reflect this added work (46, 83). Methods of more limited engagement, such as interviews or focus groups, entail fewer or one-off meetings with patients, while methods of more extensive engagement, such as advisory committees, entail multiple meetings over a longer time span, throughout the development of the instrument (5, 48, 83). In some cases, patients will be involved even before or after the development of the measure (1, 48, 83). If patient engagement is to become more prevalent, it is necessary to better understand the specifics of these drawbacks and to develop practices best suited for addressing them.

On balance, our review found that relatively few studies discussed challenges as compared to benefits or outcomes of patient engagement. This finding suggests possible issues with reporting, in accordance with the results of our quality appraisal, as well as literature on the topic (21). A positive reporting bias can have ramifications on research; it compromises the potential for a fair evaluation of patient engagement approaches in general (e.g. when should patient engagement be conducted) and in specific (e.g. what precise engagement methodologies should be applied).

The themes extracted in our review tightly correspond to several of those identified in by Forsythe et al. in their *Patient and Stakeholder Engagement in the PCORI Pilot Projects*, a well-cited thematic analysis of researchers' perspectives on patient engagement (4). Theme #4: *Modifications to interventions* and #5: *Refinement of instruments and interview questions,* extracted by these authors, cohere closely with patient engagement as contributing to the revision of the instrument.

The necessity for patient engagement was echoed verbatim in many of Forsythe et al.'s *Contributions of Patients and Other Stakeholders* themes (4). The authors described the impact that the integration of patient would offer to the research – in many cases, researchers spoke of the many fundamental changes in their research that resulted from patient engagement and that it was "integral" to their outcomes. The PCORI framework identified these patient-researcher partnerships as part of many of their themes, suggesting the validity of our findings (4). For example, Forsythe et al. stated that participants in their survey expressed that the partnership had to be genuine, and that patients had an important role in the development of their project. Finally, challenges with patient engagement were alluded to in Theme #1: *Changes to project outcomes or goals* (4); Forsythe et al. reported an example of a respondent in their survey expressing the possibility of incongruence between researcher and patient goals.

5.1 Limitations

It is necessary to acknowledge several limitations of this review that suggest the findings should be considered cautiously. Firstly, there are limitations associated with the search strategy. This review did not perform hand-searching of pertinent journals, potentially leading to missing literature on the topic. With respect to search strategies, language limitations were used to reflect the linguistic capacities of the research team; studies in languages other than English, French, and Spanish were not considered. Nor were external experts in the field consulted for the review, aside from a specialized librarian for developing the search strategy. However, the review was systematic and comprehensively looked at the identified literature.

With respect to the extraction of the themes, the synthesis was reliant on the quality of reporting within the studies being evaluated, which, as established in the critical appraisal of this review, as well as in other literature on the topic, is poor. Further information could have improved the synthesis of information, but authors were not contacted to obtain further information on the patient engagement process beyond what was available in the manuscripts identified, as the intention of the review was to understand what the published literature tells us about the patient engagement process. However, to triangulate the themes identified, I related them to another widely cited analysis and found them to be congruent.

Various patient engagement frameworks exist in the literature, according to which engagement methodologies may be categorized. As described earlier, patient engagement exists on a continuum; patients' intensity of involvement and contribution to a research project may vary widely (1, 4, 7, 9, 48, 84). In light of this, the International Association for Public Participation classifies engagement from low to high, from information provision to patient empowerment (82, 84). It can be expected that the challenges, processes, and outcomes correspond to the degree of engagement. Thus, more engagement, on balance, would lead to an instrument that is more representative of patient needs and concerns (1, 41, 52, 82). Categorization according to the aforementioned or other similar frameworks may be helpful in stratifying outcomes, and gaining a more nuanced understanding of engagement processes. However, as found in my research, the overall levels of reporting are limited to the extent this type of analysis would not be feasible.

With improved quality of reporting, resulting from the implementation of reporting standards, we expect this type of work to be possible.

5.2 Contributions of research and directions for future research

To our knowledge, this is the first systematic review that has focused on patient engagement in the construction and adaptation of instruments in HIV clinical and research settings. It clearly highlights the importance of this approach of engagement which, not exempted of procedural difficulties, appears to be beneficial for both patients and researchers. At the level of research practice in family medicine, the review identifies the current knowledge gaps regarding best practices in clinical instrument development. Better, more valid instruments can allow for more efficient and comprehensive assessment of a patient's concerns, facilitating better health outcomes and an improved patient-provider alliance. A potential example of this exists in the I-Score study (described in section 2.11), which this review helps inform the development of. Having analyzed a large sample of HIV-measures developed with patient engagement, we are able to better address their shortcomings.

At a more research practice level, one of the most important contributions of this review is that it stresses the need for improved reporting standards in the domain of patient engagement in the development of HIV health instruments. The quality appraisal results suggest poor methodological reporting. As argued by Staniszewska and Barber, evaluations of research are hampered due to the quality of reporting (21). Research has suggested the introduction of reporting standards for clinical trials, such as those established by the EQUATOR Network and CONSORT statements, has improved reporting (15, 21, 53-55). As there are no established standards in the reporting of engagement, the creation and implementation of these could help further the field (3, 21). Doing so may contribute to correcting the problem of substantial variability in reporting on research methods, allowing for better comparisons to be made between methods (3, 21).

As journal word-limits will continue to exist and may offset detailed reporting, a potential solution might be to publish more in-depth descriptions of research methods in online appendices that are published in conjunction with journals articles. This change could facilitate future investigations of patient engagement, as the information becomes more available. This knowledge could help better the application and understanding of engagement, likely resulting in better clinical and research instruments through the elucidation of best practices, in HIV and in other domains. With greater implementation of person-centered care and research, future research and development will encompass a broad spectrum of patient engagement methodologies.

5.3 Conclusion

More and more, health researchers and practitioners are seeking to involve patients in research and clinical encounters. Equally positive is that some funding agencies and governments recognize benefits to patient engagement, emphasizing it and allocating funding based on it (1-5, 7, 16, 49, 85). Instruments developed with patient engagement should be more reflective of patient concerns and valid – when used in clinical practice and research, these should improve primary and infectious disease care. This review has sought to understand patient engagement in the context of HIV measure development by focusing on what the published scientific literature tells us about the process.

Our review suggests patient engagement contributes beneficially to the development of HIV-specific health measures and is generally well-regarded by researchers, although it implies certain challenges at times. Improved documentation in the reporting of patient engagement methodology and outcomes would contribute to the advancement of patient engagement in health research and the many benefits the approach may endow. In considering these results and other literature in the field, we assert improvements can and should be made in the domain of HIV-specific measure development research. We believe these issues may best be rectified, in part, by introducing and applying standards for reporting engagement and for engagement itself (3, 21).

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