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Review Paper

Acceptability of childhood screening: a systematic narrative review

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ABSTRACT

Objectives: A systematic narrative literature review was undertaken to assess the acceptability of childhood screening interventions to identify factors to consider when planning or modifying childhood screening programs to maximize participation and uptake.
Study design: This is a systematic narrative literature review.
Methods: Electronic databases were searched (MEDLINE, EMBASE, PsycINFO via Ovid, CINAHL, and Cochrane Library) to identify primary research studies that assessed screening acceptability. Studies were categorized using an existing theoretical framework of acceptability consisting of seven constructs: affective attitude, burden, ethicality, intervention coherence, opportunity costs, perceived effectiveness, and self-efficacy. A protocol was developed and registered with PROSPERO (registration no. CRD42018099763)
Results: The search identified 4529 studies, and 46 studies met the inclusion criteria. Most studies involved neonatal screening. Programs identified included newborn blood spot screening (n = 22), neonatal hearing screening (n = 13), Duchenne muscular dystrophy screening (n = 4), cystic fibrosis screening (n = 3), screening for congenital heart defects (n = 2), and others (n = 2). Most studies assessed more than one construct of acceptability. The most common constructs identified were affective attitude (how a parent feels about the program) and intervention coherence (parental understanding of the program, and/or the potential consequences of a confirmed diagnosis).
Conclusions: The main acceptability component identified related to parental knowledge and understanding of the screening process, the testing procedure(s), and consent. The emotional impact of childhood screening mostly explored maternal anxiety. Further studies are needed to examine the acceptability of childhood screening across the wider family unit. When planning new (or refining existing) childhood screening programs, it is important to assess acceptability before implementation. This should include assessment of important issues such as information needs, timing of information, and when and where the screening should occur.

Introduction

Medical screening is a process whereby individuals undergo tests to determine whether they have, or have an increased risk of, a health condition. During childhood, there are many health conditions that can be screened for, including vision and hearing problems, heart defects, or biochemical genetic disorders. In 1968, Wilson and Jungner[1] defined criteria to be used to guide the selection of health conditions to be screened. Since then, there have been many advances in both diagnostic and therapeutic

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interventions. As such, a modified screening criterion was proposed by Anderman et al.[2] The criteria 'The test should be acceptable to the population,'[1] and 'The overall benefits of screening should outweigh the harm'[2] relate to the acceptability of the screening program. Acceptability of healthcare interventions is a challenging construct. Sekhon et al.[3] acknowledged that there is little guidance on how to define acceptability. They defined acceptability to be 'a multifaceted construct that reflects the extent to which people delivering or receiving a healthcare intervention consider it to be appropriate, based on anticipated or experiential cognitive and emotional responses to the intervention.' They proposed a theoretical framework of acceptability. This includes affective attitude (how an individual feels about the intervention), burden (the perceived amount of effort that is required to participate in the intervention), ethicality (the extent to which the intervention has good fit with an individual's value system), intervention coherence (the extent to which the participant understands the intervention and how it works), opportunity costs (the extent to which benefits, profits, or values must be given up to engage in the intervention), perceived effectiveness (the extent to which the intervention is perceived as likely to achieve its purpose), and self-efficacy (the participant's confidence that he/she can perform the behavior(s) required to participate in the intervention). For childhood screening, there is further complexity as acceptability can be applied to both the individual (i.e., the child) and the caregiver (i.e., the parent or guardian).

Over recent years, there has been increasing demand on healthcare systems.[4,5] Population growth and life expectancy has increased, placing additional stress on existing healthcare systems.[6,7] Advancements in technologies to aid diagnosis and management of health conditions have also contributed to stretched resources.[8,9] Consequently, existing and proposed interventions are examined to ensure that they are both clinically effective and cost-effective.[10] However, the practical and ethical implications on families and children when screening services are planned or reviewed should also be considered.[11] To our knowledge, there has been no review that examines the acceptability of childhood screening interventions. The overall aim of this review was to assess the acceptability of childhood screening interventions with a view to identifying which factors to consider when planning or modifying childhood screening programs to maximize participation and uptake. We applied the framework outlined by Sekhon et al.[3] to establish which aspects of acceptability are most commonly evaluated and which research methodology is used.

Methods

An information specialist was consulted in developing the appropriate search strategy. One researcher (M.P.P.) conducted the searches. Search terms included in the review included the following:

i. Children (and derivatives)
ii. Screening (and derivatives)
iii. Acceptability terms

No restriction on the publication date was applied to the search strategy. Full details of the search strategy are provided in Appendix 1. The electronic databases searched for the systematic review were MEDLINE, EMBASE, PsycINFO via Ovid, CINAHL, and the Cochrane Library. All databases were searched from inception. Searches were conducted between May 1, 2018, and May 5, 2018. An updated search was performed in January 2020 to include publications from January 2018 to January 21, 2020. The following eligibility criteria was applied to the search results: published as a full-text original research article (i.e., not including abstracts, editorials, reviews, opinion pieces, or letters to the editor), inclusion of a postnatal screening program (i.e., not antenatal screening), child health condition screening programs (i.e., not adolescent and/or adult screening or the vaccination program), and child and/or parental perspectives (i.e., not healthcare worker perspectives). Studies that solely included healthcare worker perspectives were excluded. The protocol was registered with PROSPERO.[12]

To apply the eligibility criteria for the selection of articles from the search results, the following steps were performed: (1) two reviewers (M.P.P. and C.J.) undertook 'filtering of titles' independently. Where there was disagreement, articles were retained, and the abstract was scrutinized; (2) two reviewers (M.P.P. and C.J.) undertook 'filtering of abstracts' independently. Where there was disagreement, articles were retained, and the full text was scrutinized; and (3) 'filtering of full-texts' by three reviewers (M.P.P., C.J., and G.H.J.). Discussion and consensus had to be reached for an article to be included within the review.

Articles to be included in the review were assessed against the seven component constructs proposed by Sekhon et al.[3] by two reviewers (C.J. and G.H.J.). Any disagreement was resolved through discussion. Data were extracted by one reviewer (C.J.) using a piloted data collection form. Studies were examined to determine whether acceptability was assessed prospectively, concurrently, or retrospectively; categorized as to which acceptability construct was assessed; and categorized based on the study methodology. The type of childhood screening, country where screening occurred, and details of the study participants (child or parent/carer) were also noted.

Results

The database searches identified 4529 references. A total of 149 full-text articles were retrieved for further examination. From these, 103 articles were rejected as they failed to meet the inclusion criteria. A total of 46 publications are included in this review (see Fig. 1). The summary of findings for the included studies is shown in Table 1.

Of the 47 studies included in the review, most were conducted in the United States of America (USA) (n = 14), the United Kingdom (UK) (n = 12), the Netherlands (n = 4), Australia (n = 2), Canada (n = 2), and Sweden (n = 2) (Table 1). The majority of studies (55%) were published between 2010 and 2018. The content of the screening programs included is shown in Table 1. These were newborn blood spot screening (to identify biochemical and endocrine genetic disorders) (n = 22), neonatal hearing screening (n = 13), Duchenne muscular dystrophy (DMD) screening (n = 4), cystic fibrosis screening (n = 3), screening for congenital heart defects (n = 2), screening for congenital hypothyroidism (n = 1), and screening for hip dysplasia (n = 1). The details of which biochemical and endocrine genetic disorders were screened as part of newborn blood spot screening programs were not clearly reported, but the program typically included screening for conditions such as phenylketonuria and sickle cell disease, among others. Most of the studies (n = 44) concerned neonatal screening.

Acceptability was assessed quantitatively (n = 30), qualitatively (n = 26), and by a combination of methods (n = 10). Of the studies that adopted quantitative methods, the majority of studies used their own questions or questionnaire, or modified existing questionnaires. Some studies did include validated questionnaires, including the Beck Anxiety Inventory, State-Trait Anxiety Inventory questionnaire, Center for Epidemiological Studies Depression Scale, the depression scale of the Hospital Anxiety and Depression Scale, the Parenting Stress Index, and the General Health Questionnaire. Of the studies that adopted qualitative methods (n = 26), most involved interviews (n = 13) and seven studies undertook focus group sessions (n = 7). Some studies issued questionnaires that

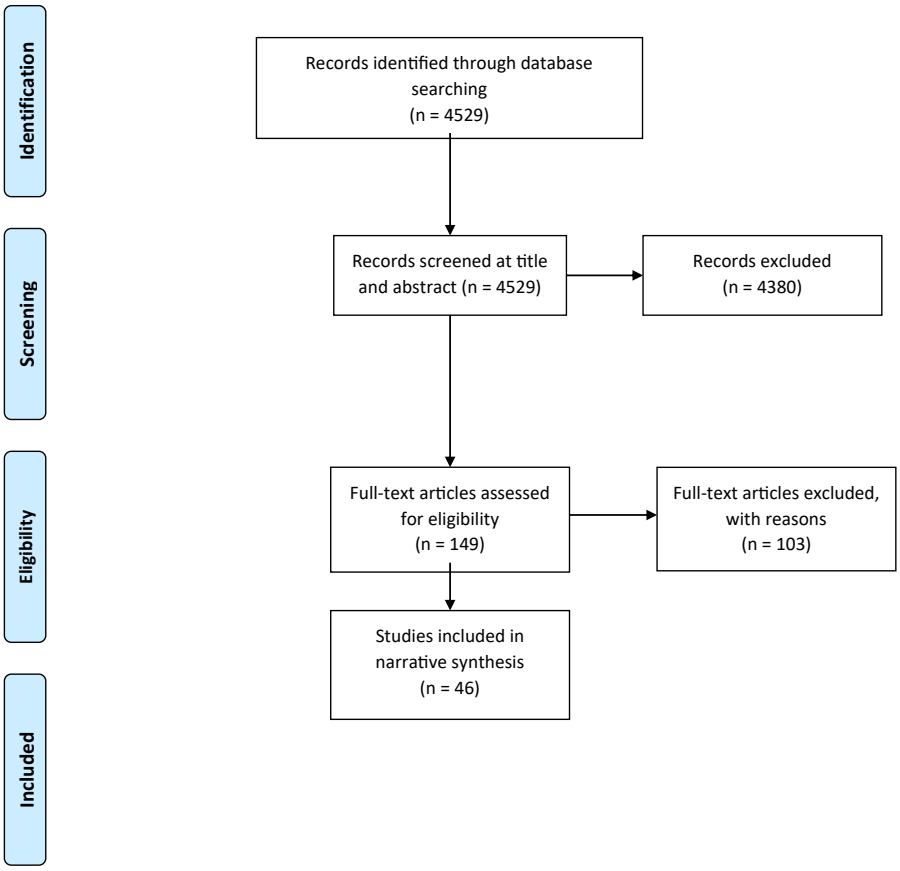

Fig. 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) 2009 Flow Diagram: study identification.

incorporated some open-ended questions, and the free text was analyzed qualitatively (n = 8) (see Table 2).

Table 3 shows only three studies assessed acceptability at the time of screening.[13–15] The majority of studies assessed acceptability retrospectively (n = 40).[13,14,16–52] Ten studies assessed acceptability prospectively.[24,28,31,35,46,53–57] The majority of studies examined acceptability with respect to affective attitude (n = 41) and intervention coherence (n = 31). Other acceptability constructs assessed included burden (n = 9), ethicality (n = 5), perceived effectiveness (n = 9), opportunity costs (n = 6), and self-efficacy (n = 4). Most of the studies assessed more than one construct of acceptability. No study assessed all seven acceptability constructs (Fig. 2).

Affective attitude

In the context of screening, this is how a parent feels about the screening program itself. A total of 41 studies that assessed this concept were identified.[13–17,19,21,24–31,33–35,38–47,49–58] Most of the studies also included some form of assessment of parental beliefs on whether screening was thought to be of value.[31,39,41,58] Other studies also reported on parental satisfaction, specifically for screening service, be it in terms of receiving results or the screening test(s) not causing any discomfort to the parents' child.[17,19,33] Not all participants within the studies reported favorably. Tariq et al.[46] reported some parents (n = 10, 4%) to view the test for congenital hypothyroidism to be 'unimportant,' with some parents (n = 8, 3%) considering it to be a painful procedure for their child.

Burden

Nine studies explored the impact of burden.[16,30,33,34,40,43,52,54,57] The burden associated with screening varies from one screening program to another. The amount of effort required for the parent/caregiver to support the intervention (i.e., take the child for testing) can be considered as burdensome. When screening can occur in venues that required minimal effort from the parent (i.e., within the hospital or in the home), the acceptability of the screening is increased.[30,40,52] The burden of attending the appointments owing to work commitments or difficulties with transport can lead to non-attendance.[43] Financial burden may also be a factor as some parents reported their medical insurance did not cover the screening test(s).[16,34,43] Some studies inferred burden by parental observations of discomfort of testing on the child.[33]

Ethicality

Nine studies were categorized as assessing ethicality.[13,19,28,30,40–42,49,57] Some studies included assessments of beliefs with regard to the screening, including moral and religious views.[13,42,49] Parsons et al.[13] reported some mothers consented to screening for DMD as they approved of all screening. In a separate study, Parsons et al.[40] highlighted some mothers felt so positively toward newborn screening that they felt it should be made compulsory.

Intervention coherence

Thirty-one studies identified within this review investigated parental understanding of the screening program itself, and/or the potential consequences of a confirmed diagnosis of the target

Table 1
Summary of findings of the included studies.

Reference	Year	Country in which screening occurred	Condition	Age of the child subjected to screening	N (no. of mothers)	Aim	Summary
Akilan et al.[16]	2014	South India	Hearing	<2 years[a]	83 (83)	To review an existing rural community-based screening project	Community leaders played an important role in facilitating better coverage.
Al-Sulaiman et al.[17]	2015	Saudi Arabia	NBS	Newborn[a]	425 (425)	To assess the attitude and knowledge of mothers toward the NBS program	Positive attitude toward the NBS program; however, better communication is needed to increase awareness.
Araia et al.[18]	2012	Canada	NBS	Newborn, 24 −72 hrs after birth	750 (750)	To identify elements of NBS education and their associations with mothers' knowledge and satisfaction levels	Education and information before screening is important, particularly on the purpose, benefits, process, and possible results of screening.
Christie et al.[53]	2013	Australia	NBS for FXS	Newborn, 24 −72 hrs after birth	1971 (1971)	To determine feasibility and accuracy of two concurrent testing methodologies; to determine postnatal mothers' acceptance and attitudes to screening and reasons for accepting or declining participation; to assess the impact of diagnosis of a child with an abnormal result	Mothers considered an early diagnosis beneficial. Some were anxious about potential test results; others felt their feelings toward their newborn may change if he/she was diagnosed positive. High participation rates and maternal attitudes indicate a high level of maternal acceptance and support for screening.
Crockett et al.[19]	2005	UK	Hearing	Newborn OAE testing within 48 h of birth HVDT at 6–8 months of age	90 (90)	To compare the impact of two screening tests (newborn hearing screening − OAE test and HVDT) and screening recall on maternal anxiety and satisfaction	No significant differences were found (with respect to maternal anxiety, worry, and certainty) between the two tests.
Crockett et al.[20]	2006	UK	Hearing	Newborn within 48 h of birth	344 (344)	To describe the impact of newborn screening on maternal anxiety and to examine the impact of knowledge	Understanding the three screening recall systems may avoid some anxiety.
Cyrus et al.[21]	2012	USA	DMD	12 months	138 (120)	To assess the desirability of DMD screening, the effectiveness of the consent process, and the feasibility of screening in a pediatric office (i.e., after the newborn period)	Parents indicated broad support of screening. Parents understood the risks and benefits of screening. DMD screening is feasible in a pediatric office.
Danhauer and Johnson[22]	2006	USA	Hearing	Newborn[a]	36 (NR)	To assess parents' perceptions of an emerging community-based program in which screening and/or follow-up testing was provided on an 'outpatient' basis through a private practice	Parents were generally positive about all phases of screening. Findings were consistent with those reported from hospital-based programs.
Davis et al.[23]	2006	USA	NBS	Newborn[a]	51 (48)	To gather opinions about the content and timing of newborn screening education to inform recommendations	Parents had limited knowledge and awareness of NBS. Parents wanted concise information on all aspects of screening including benefits, need for retesting, and importance of follow-up (if required). Parents wanted verbal information from the provider and brochures. Parents felt information should be provided in the third trimester of pregnancy.
Detmar et al.[24]	2007	Netherlands	NBS	1st week of life	29 (22)	To investigate the preferences and views of parents and future parents with respect to information about, and consent to, neonatal screening and the possible expansion of the program	Parents were not well informed about what the test involves and viewed it as a routine procedure. If the program was to be expanded, parents would like to be informed earlier, preferably during pregnancy. Most parents preferred an opt-out consent approach.
Din et al.[54]	2011	USA	NBS CMV infection	Newborn[a]	3922 (NR)	To assess attitudes toward newborn screening for CMV	Among most parents, costs, worry, and anxiety associated with newborn screening for CMV would be acceptable. A minority of the parents weakly opposed to newborn screening for CMV.
Etchegary et al.[25]	2016	Canada	NBS	Newborn[a]	32 (30)	To explore parent and HCP experiences of NBS practices	Three themes were identified: offer of consent; content and timing of information; and importance of parental experiences for consent decisions. NBS was viewed as 'routine,' with little evidence of an informed consent process.

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Table 1 (*continued*)

Reference	Year	Country in which screening occurred	Condition	Age of the child subjected to screening	N (no. of mothers)	Aim	Summary
							All participants felt information should be given before birth.
Fitzgerald et al.[55]	2017	Ireland	NBS	Newborn[a]	662 (662)	To determine if antenatal women received information about NBS in the antenatal period and to evaluate their knowledge and attitudes about NBS	Information given about NBS in the antenatal period is inconsistent; consequently, awareness is limited. Mothers require information to be provided in a more structured format.
Hargreaves et al.[26]	2005	UK	NBS	Newborn[a]	47 (42)	To examine parents' and HCPs' views on informed choice in NBS and assess information and communication needs	Parents and HCPs recognize a tension between informed choice in NBS and PH screening in children. Clear, brief, and accurate parent information and effective communication between HCPs and parents, which take into account parents' information needs, are required for informed choice.
Hergils and Hergils[27]	2000	Sweden	Hearing	Newborn[a]	83 (NR)	To assess parental attitudes and concern of relation to universal NHS by OAE testing	Parents wanted early detection of hearing loss and the possibility of early intervention. Screening did not disturb the children. Most parents' experiences of NHS were positive and felt reassured by it.
Jatto et al.[56]	2018	Nigeria	Hearing	Newborn[a]	48 (48)	To determine the knowledge and perceptions of mothers of newborn children on hearing screening	Awareness of newborn screening was poor. Willingness to accept screening increased with increasing levels of education and increasing levels of socio-economic status. Knowledge of what factors are responsible for childhood hearing loss was poor.
Joseph et al.[28]	2016	USA	NBS	Newborn[a]	31 (31)	To examine the perspectives and values of diverse healthy pregnant women and parents of children diagnosed with a primary immunodeficiency disorder about traditional NBS and expanded NBS with the use of whole-genome sequencing.	Four themes emerged: (1) perspectives on traditional NBS, (2) informed consent, (3) return of results, and (4) storage and retrieval of results. Study participants desired greater inclusion in the NBS process. Parents voiced concerns about privacy and control over test results because of limited trust in the medical system and the state-run NBS program.
Khairi et al.[29]	2011	Malaysia	Hearing	Newborn[a]	78 (78)	To investigate maternal anxiety when the child had failed the test in the first stage of the UNHS	FP test results of the UNHS increased maternal anxiety.
Lam et al.[30]	2018	Hong Kong	Hearing	Newborn[a]	102 (102)	To investigate maternal knowledge, attitudes, and satisfaction of the UNHS	Information on the UNHS requires further details, particularly on implications of results and/or infant hearing development. Many did not understand the results.
Lang et al.[31]	2009	USA	NBS (CF and SCD)	Newborn	388 (388)	To examine maternal understanding of NBS for SCD and CF and their knowledge of the genetics, symptoms, and treatments of both conditions.	Poor understanding of NBS, greater familiarity with SCD, and significant knowledge gaps for both SCD and CF were found. There are many missed educational opportunities for educating parents about NBS and specific conditions included in NBS panels in both the obstetric clinics and the nursery.
Lipstein et al.[32]	2010	USA	NBS	Newborn[a]	45 (41)	To describe how parents consider disease and test characteristics while making decisions about newborn screening.	Parents' preferences differed based on experience with genetic conditions. Most parents wanted more detailed information. Some suggested optional testing. Understanding parents' decision-making processes and information needs would support development of screening policies that better address variations in preferences.
	2009	Sweden	Hearing		49 (26)		

Magnuson and Hergils[33]				Newborn (maximum 3 days postpartum)		To evaluate an existing newborn hearing screening program with regard to information and psychological support of parents	A majority of parents were in favor of screening, and screening caused little anxiety. Where more than one retest was required, parental anxiety increased and was linked to information needs.
Mak et al.[34]	2012	China	NBS	Newborn[a]	172 (NR)	To examine parental knowledge and attitudes toward the expanded NBS in Hong Kong	Parents favored having the expanded NBS in Hong Kong. Parental tolerance was high. Parents valued parental autonomy with informed consent and pretest counseling.
Moody and Choudhry[35]	2013	UK	NBS	Newborn <1 week	Survey, 140 (124) FG, 29 (27)	To explore perceptions and attitudes of parents and future parents to an expanded NBS in the UK and the necessary information provision and consent processes.	Parents want guaranteed information provision with clear decision-making powers and an awareness of the choices available to them. The difference between the existing NBS and expanded NBS was not considered to be significant enough by participants to warrant formal written, informed consent for expanded screening.
Narayen et al.[36]	2017	Netherlands	CHD	Newborn 1 hr after birth and at day 2/3	1172 (1172)	To assess the acceptability of PO screening to mothers after screening in the home setting	Overall, mothers were happy with the performance of the test, thought their baby was comfortable during screening, and did not feel stressed while the screening was performed. Most mothers would recommend PO and considered the test important.
Nicholls[37]	2012	UK	NBS	Newborn[a]	18 (16)	To explore whether parents experience the purported tension between compliance and achieving informed consent.	Two themes emerged relating to the voluntariness of choices: the expectation of compliance and presentation of information to promote compliance. In both cases, aspects of provision were noted as negatively impacting on the parents' perceived choice when accepting NBS.
Nicholls and Southern[38]	2013	UK	NBS	Newborn[a]	12 (10)	To understand the factors that influence parental decision-making in accepting NBS	Seven factors were identified: experience, attitudes to medicine, information-seeking behavior, perceived knowledge, attitudes to screening, perceived choice, and perceived decisional quality.
Parsons et al.[39]	2002	UK	DMD	Newborn[a]	97 (NR)	To evaluate the psychosocial implications of newborn screening for DMD	Most families with an affected boy identified through screening were in favor of NBS (to allow choice in future reproductive plans and time to prepare). Anxiety levels in the screened group were higher than in the control group, but were normalized during the study period.
Parsons et al.[58]	2005	Wales (UK)	DMD	Newborn[a]	1347 (NR)	To assess the effect of changing a protocol for DMD (to make the choice more explicit) as part of NBS.	The change in protocol resulted in increased satisfaction, awareness, and choice. No increase in worry was found, and parents indicated they felt a 'greater freedom' to refuse the test.
Parsons et al.[13]	2006	UK	DMD	Newborn[a]	1542 (1542)	To explore the reasons given by women for their decision about an optional newborn screening test for DMD.	Perceptions on screening were related to 3 overarching themes: screening as a routine procedure, screening for reassurance, and screening for disease detection.
Parsons et al.[40]	2007	Wales (UK)	NBS	Newborn[a]	18 (18)	To explore mothers' accounts of NBS and to explore the process of consent	Information about the screening was reported to be varied, and most mothers received this postpartum. Issues of consent were noted, and mothers felt that screening was perceived as routine.
Powell et al.[41]	2013	UK	CHD	Newborn[a]	813 (813)	To assess maternal acceptability of pulse oximetry screening for CHD in newborn infants and to identify factors predictive of participation in screening	Participants were mainly satisfied with screening. Anxiety of mothers given FP results was not significantly higher than of those given the TN result. Different ethnic groups had different participation readiness into the study,

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Table 1 (*continued*)

Reference	Year	Country in which screening occurred	Condition	Age of the child subjected to screening	N (no. of mothers)	Aim	Summary
							which may not reflect upon whether this would be observed in screening itself.
Quinlivan and Suriadi[42]	2006	Australia	NBS	Newborn[a]	200 (200)	To evaluate new mothers' opinions of genetics and newborn screening	Acceptance of screening is high, but mothers consider the need for consent to be mandatory.
Scheepers et al.[43]	2014	South Africa	Hearing	Newborn[a]	50 (NR)	To identify reasons why parents refuse newborn hearing screening and why some default on follow-up rescreening	Most frequent reasons for refusing screening were related to costs and knowledge about the screening process.
Skinner et al.[44]	2011	USA	NBS for FXS	Newborn[a]	1930 (1930)	To document rates of parental consent in a pilot study of screening for FXS, examine demographic characteristics of mothers who consented or declined, and describe the reasons for their decision.	A majority of parents accepted screening, but decision rates and reasons for accepting/declining varied in part as a function of race/ethnicity and in part as a function of what parents most valued or feared in their assessment of risks and benefits.
Stuart et al.[45]	2000	USA	Hearing	Newborn[a]	40 (40)	To determine whether mothers whose infants had failed NHS had more stress than those mothers whose infants had passed NHS	No significant difference was found between the two groups—those mothers whose infant had failed demonstrated equivalent stress levels as those mothers whose infants had passed.
Tariq et al.[46]	2018	Pakistan	Congenital hypothyroidism	Newborn[a]	355 (355)	To determine knowledge of congenital hypothyroidism and to assess the impact of health education on knowledge and attitudes toward screening	Most mothers were unaware of congenital hypothyroidism and its implications. Awareness increased after the intervention survey.
Tluczek et al.[14]	1992	USA	CF (as part of NBS)	Newborn[a]	104 (66, plus 28 responses from both parents)	To examine parental knowledge of (1) the screening program, (2) understanding of negative results, (3) effects of screening-related anxieties, and (4) the effects of FP results	Parents had gaps in knowledge about screening, misconceptions about test results, and high levels of anxiety.
Tluczek et al.[47]	2005	USA	CF (as part of NBS)	Newborn[a]	28 (25)	To investigate the psychosocial effects on parents of infants with abnormal results in CF NBS that uses genetic testing	Most parents experienced high levels of emotional distress waiting for the sweat-test appointment (diagnostic test). Parental uncertainty and emotional distress were influenced by prior knowledge of NBS, CF, their own carrier status, adjustment to having a new baby, and physicians' approach to parents.
Tluczek et al.[47]	2009	USA	CF (as part of NBS)	Newborn[a]	193 (100)	To learn how parents were informed about NBS and obtain their suggestions for improving the process of educating parents about NBS	Parents described much inconsistency in the timing of information and methods used to inform them about NBS. Parents recommended improving communication about NBS at multiple points. Parents suggested that providers take time to explain the purpose and importance of NBS, which diseases are included in testing, and when to expect results.
Ulph et al.[49]	2011	UK	NBS	Newborn[a]	37 (28)	To explore the origins and content of service users' prior knowledge of universal antenatal and newborn screening for hemoglobin disorders.	Families influenced participants' screening knowledge, decisions, and service use. Families were often participants' main source of support.
Vohr et al.[15]	2001	USA	Hearing	Newborn 1st screening before discharge Rescreen 2–8 weeks after discharge	307 (307) 1st screen 40 (40) rescreen	To identify and compare the prevalence and degree of maternal worry about NHS at the time of an initial NHS and rescreening	Maternal worry was greater at the rescreening cf. screening. Those who reported greater worry at the time of the screening were more likely to be socio-economically disadvantaged. Maternal knowledge of screening increased between the two time periods, but the degree of worry was unchanged.

Waisbren et al.[50]	2003	USA	NBS	Newborn[a]	407 (254)	To assess the impact on families of a FP screening result compared with a normal result in the expanded newborn screening program.	Expanded newborn screening may lead to improved health outcomes. FP screening results may place families at risk of increased stress and parent-child dysfunction.
Weichbold et al.[51]	2001	Austria	Hearing	Newborn[a]	90 (90)	To test the hypothesis that there is a positive association between information and positive attitude to NHS	Most mothers who were in favor of screening had a greater knowledge about the hearing test. This included being present at the test and being aware of the result of the test.
Weinreich et al.[57]	2012	Netherlands	NBS for Pompe disease	Newborn[a]	613 (not explicitly reported)	To measure support for neonatal screening for Pompe disease in the general public and to compare it to support among (parents of) patients with this condition	Results suggest a rather high level of support for newborn screening for Pompe disease, not only among those who have personal experience of the disease but also among the general population.
Witting et al.[52]	2013	Netherlands	Hip dysplasia	3 months	703 (NR)	To investigate whether psychosocial factors (attitude, subjective norm, self-efficacy, perceived susceptibility, perceived severity, perceived effectiveness) predicted parental participation in the screening.	Attitude, subjective norm, self-efficacy, perceived susceptibility, and perceived effectiveness predicted parental participation in the screening. Emphasizing positive aspects of the screening, highlighting the effectiveness, removing practical barriers, and being conscious of the influential role of HCPs on decision-making are areas to focus on while organizing the screening.

CF = cystic fibrosis; DMD = Duchenne muscular dystrophy; CHD = congenital heart defect; FP = false-positive; FXS = Fragile X syndrome; HCP = healthcare professional; HVDT = health visitor distraction test; NBS = newborn blood spot screening; NHS = neonatal hearing screening; NR = not reported; OAE = otoacoustic emission; PH = public health; PO = pulse oximetry; RHD = rheumatic heart disease; SCD = sickle cell disease; TN = true negative; UNHS = universal hearing screening program, UK = United Kingdom; USA = United States of America.

[a] Exact age not defined.

condition.[13,14,16–18,20–26,28–31,34,35,38,40,43,46–52,55,56,58] Studies examined the issues of parental knowledge, receipt of information, and previous experience of screening and experiences of friends and/or family members. Some studies explored issues of consent, which also included parents having sufficient information and appropriately timed information to allow for informed consent. Some parents recalled that newborn screening was offered as a choice where active consent was given, whereas other parents were less certain as to whether they did provide consent. Even within the same study cohort, parental accounts with regard to the issue of consent for screening varied.[25] For some parents, the screening process was 'routinized,' and that this can be inadvertently presented as compulsory.

Opportunity costs

Six articles identified issues with regard to opportunity costs.[16,23,26,34,43,54] Some studies discussed the consequences of direct financial costs on attending screening and whether such costs were covered by medical insurance.[23,43] Some parents were not concerned about the costs of testing, and others expressed a willingness to pay.[34,54] However, some parents stated that the expense of additional (screening) tests would result in the refusal of any advised additional testing.[23] One study reported that attendance to screening would come at a cost of missing work and giving up time with other children/family responsibilities.[16]

Perceived effectiveness

Perceived effectiveness was studied in nine studies.[14,16,28,32,38,39,41,43,52] Some studies reported that parents either had doubts in the effectiveness of the test, had doubts in the accuracy of results, or even had distrust of the healthcare system.[14,32,39,41,43,52] Some parents noted that screening would not be offered if it had not already been reviewed or assessed as being acceptable by experts, including medical professionals.[38]

Self-efficacy

Four studies were categorized as assessing for self-efficacy.[16,23,34,52] Parents reported that while they wanted information about the screening process, they noted that the timing the information was received was not appropriate. They felt overwhelmed with information and were 'often exhausted.'[23] The context of exhaustion may be particularly pertinent to screening programs that occur within the first few weeks/months of life. Generally, parents were confident that they were able to arrange other responsibilities to make time to attend for screening (and/or referral) appointments.[16,34,52]

Discussion

Acceptability of the childhood screening program is a relatively under-researched area. A key objective of this review was to identify factors to be considered to encourage participation in childhood screening programs, thereby maximizing the program's cost-effectiveness. Two of the most common constructs identified from the included studies were affective attitude (how the parent feels about the screening program) and intervention coherence (parental understanding of the screening program itself and/or the potential consequences of a confirmed diagnosis of the target condition). Determining how a parent or guardian feels about screening could be considered as an important first step when considering implementing new (or refining existing) childhood screening programs.[59,60] If parents' views are such that they feel

Table 2
Study methodologies of the included studies and types of data collection method(s).

Reference	Both	Quantitative assessment instruments											Qualitative methods		
		Own	EDS	STAI	IoE scale	CHQ-PF28	BAI	GHQ	HADS	PSI	CES-D	CST	Int.	FG	Free-text
Akilan et al.[16]	✗													✓	
Al-Sulaiman et al.[17]	✗	✓													
Araia et al.[18]	✗	✓													
Christie et al.[53]	✓	✓	✓	✓	✓										✓
Crockett et al.[19]	✓	✓		✓											
Crockett et al.[20]	✓	✓		✓											
Cyrus et al.[21]	✓	✓													✓
Danhauer and Johnson[22]	✓	✓													✓
Davis et al.[23]	✗												✓	✓	
Detmar et al.[24]	✗													✓	
Din et al.[54]	✗	✓													
Etchegary et al.[25]	✗												✓		
Fitzgerald et al.[55]	✗	✓													
Hargreaves et al.[26]	✗												✓	✓	
Hergils and Hergils[27]	✗														✓
Jatto et al.[56]	✗	✓													
Joseph et al.[28]	✗													✓	
Khairi et al.[29]	✗						✓								
Lam et al.[30]	✗	✓													
Lang et al.[31]	✗	✓													
Lipstein et al.[32]	✗													✓	
Magnuson and Hergils[33]	✗												✓		
Mak et al.[34]	✗	✓													
Moody and Choudhry[35]	✓	✓												✓	
Narayen et al.[36]	✗	✓													
Nicholls[37]	✗												✓		
Nicholls and Southern[38]	✗												✓		
Parsons et al.[39]	✓	✓		✓				✓					✓		
Parsons et al.[58]	✓	✓													✓
Parsons et al.[13]	✓	✓													✓
Parsons et al.[40]	✗												✓		
Powell et al.[41]	✓	✓		✓					✓						✓
Quinlivan and Suriadi[42]	✗	✓													
Scheepers et al.[43]	✗												✓		
Skinner et al.[44]	✗												✓		
Stuart et al.[45]	✗														
Tariq et al.[46]	✗	✓													
Tluczek et al.[14]	✗	✓													
Tluczek et al.[47]	✓										✓		✓		
Tluczek et al.[47]	✗												✓		
Ulph et al.[49]	✗												✓		
Vohr et al.[15]	✗	✓													
Waisbren et al.[50]	✗									✓		✓			
Weichbold et al.[51]	✗	✓													
Weinreich et al.[57]	✓	✓													✓
Witting et al.[52]	✗	✓													

Both = the study used both quantitative and qualitative methods; EDS = Edinburgh Depression Scale; STAI = State-Trait Anxiety Inventory; IoE scale = Impact of Event Scale; CHQ-PF28 = Child Health Questionnaire Parent Form 28 items; BAI = Beck Anxiety Inventory; HADS = Hospital Anxiety and Depression Scale; PSI = Parenting Stress Index; CES-D = Center for Epidemiological Studies Depression Scale; CST = client satisfaction tool; Int. = interview; FG = focus group; GHQ = General Health Questionnaire.

negatively about the screening program, this is likely to affect attendance and therefore efficiency of the program itself. Parental beliefs, understanding, and knowledge of the screening program (including what it entails and what the potential consequences may be) are influenced by information. The amount of information and timing of information is important not only to ensure parents understand the screening process but also to ensure that informed consent to participate in the screening program can be obtained.[23–25,35] It is therefore important to fully consider the information needs of parents while planning and implementing childhood screening programs.[13,20,23,25,29,32,38] Information needs may differ between groups and populations.[11] A standardized approach across a whole country may not be appropriate, and localized documents (or other information resources) should be considered. Other issues identified included the burden of the screening program and any costs associated with the screening program.[16,23,26,34,43,54] Acceptability was noted to be influenced by minimal effort in participating in the screening process (i.e., whether the screening was undertaken at a convenient location, such as within the hospital or in their own home [30,40,52] and whether the costs were minimized). When screening exists as part of a suite of health checks, this makes the screening more acceptable to parents.[11] All of these factors may influence screening uptake and attendance. Not all costs were noted to be direct financial costs (such as paying for the screening tests), but could also be related to taking time to travel to the venue where the screening is carried out, how long the screening may take, and any loss of income due to taking time off work.[16] It was difficult to draw any firm conclusions on whether potential financial implications of attending screening could influence the acceptability of such programs. The studies identified varied in the country setting, from low-income countries (India)[16] to upper middle-income countries (China and South Africa)[43,54] to high-income countries (UK and USA).[23,26] Consideration of how healthcare systems are funded is important, particularly if parents

Table 3
Assessment of acceptability and constructs included.

Reference	When acceptability was assessed			Component constructs of acceptability						
	Prospective	Concurrent	Retrospective	Affective attitude	Burden	Ethicality	Intervention coherence	Opportunity costs	Perceived effectiveness	Self-efficacy
Akilan et al.[16]			✓	✓	✓		✓	✓	✓	✓
Al-Sulaiman et al.[17]			✓	✓			✓			
Araia et al.[18]			✓				✓			
Christie et al.[53]	✓			✓						
Crockett et al.[19]			✓	✓		✓				
Crockett et al.[20]			✓				✓			
Cyrus et al.[21]			✓	✓			✓			
Danhauer and Johnson[22]			✓				✓			
Davis et al.[23]			✓				✓	✓		✓
Detmar et al.[24]	✓		✓	✓			✓			
Din et al.[54]	✓			✓	✓			✓		
Etchegary et al.[25]			✓	✓			✓			
Fitzgerald et al.[55]	✓			✓			✓			
Hargreaves et al.[26]			✓	✓			✓	✓		
Hergils and Hergils[27]			✓	✓						
Jatto et al.[56]	✓			✓			✓			
Joseph et al.[28]	✓		✓	✓		✓	✓		✓	
Khairi et al.[29]			✓	✓			✓			
Lam et al.[30]			✓	✓	✓	✓	✓			
Lang et al.[31]	✓		✓	✓			✓			
Lipstein et al.[32]			✓	✓					✓	
Magnuson and Hergils[33]			✓	✓	✓					
Mak et al.[34]			✓	✓	✓		✓	✓		✓
Moody and Choudhry[35]	✓		✓	✓			✓			
Narayen et al.[36]			✓	✓						
Nicholls[37]			✓	✓						
Nicholls and Southern[38]			✓	✓			✓		✓	
Parsons et al.[39]			✓	✓					✓	
Parsons et al.[58]			✓	✓			✓			
Parsons et al.[13]		✓		✓		✓	✓			
Parsons et al.[40]			✓	✓	✓	✓	✓			
Powell et al.[41]			✓	✓		✓			✓	
Quinlivan and Suriadi[42]			✓	✓		✓				
Scheepers et al.[43]			✓	✓	✓		✓	✓	✓	
Skinner et al.[44]			✓	✓						
Stuart et al.[45]			✓	✓						
Tariq et al.[46]	✓		✓	✓			✓			
Tluczek et al.[14]		✓	✓	✓			✓		✓	
Tluczek et al.[47]			✓	✓			✓			
Tluczek et al.[47]			✓				✓			
Ulph et al.[49]			✓	✓		✓	✓			
Vohr et al.[15]		✓		✓						
Waisbren et al.[50]			✓	✓			✓			
Weichbold et al.[51]			✓	✓			✓			
Weinreich et al.[57]	✓			✓	✓	✓				
Witting et al.[52]			✓	✓	✓		✓		✓	✓

are meeting the financial costs of screening. Studies have shown that socio-economic status and risk of having disease (or health condition) do influence screening participation.[11,61–63] Although it can be hypothesized that parents with lower socio-economic status may find screening less acceptable, the studies identified in this review can neither support nor refute this hypothesis. Further studies are required to fully understand the financial burden of screening, either the cost of testing and/or the costs incurred owing to attending screening (such as travel costs and lost income).

The studies identified in this review were conducted on small populations. All the studies assessed acceptability from a parental (often maternal) perspective rather than from the individual's perspective. None of the studies explicitly explored whether acceptability differed within different population groups (such as ethnicity, educational status, and so on). Further research is required to investigate the acceptability of childhood screening programs across the wider family unit, with increased inclusion of modern-day parenting situations and roles.

A mixture of study methodologies was used to assess childhood screening acceptability. For the studies that used quantitative methods, existing validated questionnaires were administered to parents to measure anxiety associated with the screening process. In these studies, the level of anxiety was used as a proxy for acceptability.[19,20,29,39,41,53] However, it must be acknowledged that anxiety is in itself a multifaceted construct. Many parents find having a new baby a stressful time, even when good support

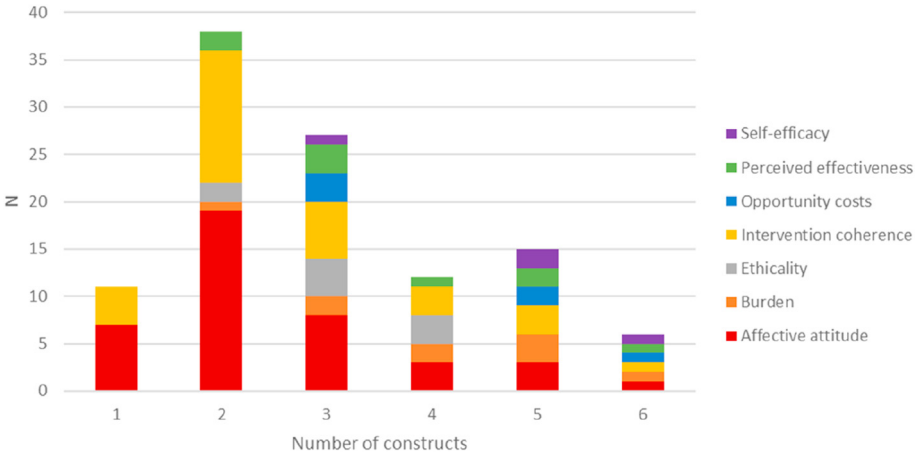

Fig. 2. Number of acceptability constructs reported within identified studies.

networks (such as friends and family) are in place. Increased levels of anxiety with regard to the time of neonatal screening may occur irrespective of whether a parent has anxiety with regard to the screening itself. Therefore, quantifying anxiety using an existing questionnaire(s) may not be the most appropriate method to understand what impact childhood screening has on a parent. To fully understand the individual's behaviors and feelings, qualitative research methods are required. The use of qualitative research methods facilitates an in-depth understanding of behavior and the reasons that govern that behavior[64] and may provide deeper insight into how parents feel about childhood screening programs.

The majority of the studies identified in the review were retrospective in nature, and the results perhaps should be treated with caution. The parental perspective of acceptability may have been influenced by the outcome of the screening program itself, that is, whether the child is found to have (or not) the condition for which the child was screened. This factor was not always disclosed in the included studies. Some may argue that acceptability is linked to satisfaction; however, Sekhon et al.[3] state there is a difference between these concepts. They argue that satisfaction can only be assessed retrospectively, whereas acceptability can be assessed both retrospectively and prospectively. Another important issue relating to retrospective assessment of acceptability is the timing of assessment in relation to the screening episode, i.e., how 'retrospective.' Recall bias is an important consideration when interpreting the results of any study.[65,66] Future studies will need to determine whether any impact of acceptability is present only in the short term (i.e., soon after the screening intervention) or more in the long term (i.e., months or even 1 year after screening). For example, issues of exhaustion and poor timing and information overload[16,23,34,52] may only be apparent or measurable if acceptability is assessed in the short term.

Limitations

This review is not without its limitations. Owing to the limited number of studies identified, no assessment (and therefore restriction) of study quality was performed. It is possible that bias exists within the studies, and the conclusions of individual study findings should be considered against issues such as design bias, sampling bias, measurement bias, interview bias, response bias, and reporting bias. The varied study outcomes and methodologies meant that meta-analysis and synthesis beyond a narrative review was not possible. A further limitation is that of acceptability construct categorization. Some of the constructs within the framework are linked, for example, burden and opportunity costs. The burden of attending a screening program may include time (which may include time off work, which could incur a cost), travel (which will incur a cost), and psychological burden (such as anxiety or worry). Affective attitude and perceived effectiveness are also related. Both constructs are associated with parental knowledge and understanding and information needs. Intervention coherence may relate to parental understanding of what the screening test(s) involves, any risks associated with the test(s), the consequence of a 'positive' screening result, consent for the screening test(s) to take place, and the effect involved in consenting to the screening program. Perceived effectiveness of screening centers on how well/accurate the screening test(s) is in being able to provide an indication of whether a child has the target condition, i.e., is the screening going to work? Similarly, affective attitude and ethicality are also linked. Although studies were assessed by two reviewers, there were inconsistencies with categorizations. Disagreements occurred when the results of the included studies could infer assessment of a construct (i.e., parental feelings of the screening program could infer the ethicality of screening). Most studies concerned neonatal screening. The findings may not apply to screening in older children. The acceptability of screening in older children may include other constructs, and the perspective of the child could also be considered.

Conclusions

Acceptability of childhood screening programs is an under-researched area. The aim of the review was to assess the acceptability of childhood screening interventions with a view to identify which factors to consider when planning or modifying childhood screening programs to maximize participation and uptake. We identified that in the context of childhood screening programs, acceptability was often determined by assessing parental knowledge and understanding of the screening process, the testing procedure(s), and consent. The emotional impact of childhood screening explored maternal anxiety levels associated with the timing of the screening process and the impact of any false referral. There are evidence gaps, and further studies are required to examine the acceptability of childhood screening across the wider family unit, including the child themselves (for screening in older children). While planning new (or refining existing) childhood screening programs, it is important to assess acceptability before

any implementation. The results of such studies can then inform and address issues such as information needs, timing of information, and when and where the screening should occur.

Author statements

Ethical approval

Not applicable.

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Competing interests

The authors have nothing to declare.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.puhe.2021.02.005.

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