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RESEARCH ARTICLE

General population screening for paediatric type 1 diabetes—A qualitative study of UK professional stakeholders

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Abstract

Aims: Identifying children at risk of type 1 diabetes allows education for symptom recognition and monitoring to reduce the risk of diabetic ketoacidosis at presentation. We aimed to explore stakeholder views towards paediatric general population screening for type 1 diabetes in the United Kingdom (UK).

Methods: Qualitative interviews were undertaken with 25 stakeholders, including diabetes specialists, policymakers and community stakeholders who could be involved in a future type 1 diabetes screening programme in the UK. A thematic framework analysis was performed using the National Screening Committee's evaluative criteria as the overarching framework.

Results: Diabetic ketoacidosis prevention was felt to be a priority and proposed benefits of screening included education, monitoring and helping the family to better prepare for a future with type 1 diabetes. However, diabetes specialists were cautious about general population screening because of lack of evidence for public acceptability. Concerns were raised about the harms of living with risk, provoking health anxiety and threatening the child's right to an 'open future'. Support systems that met the clinical and psychological needs of the family living with risk were considered essential. Stakeholders were supportive of research into general population screening and acknowledged this would be a priority if an immunoprevention agent were licensed in the UK.

Conclusions: Although stakeholders suggested the harms of UK paediatric general population screening currently outweigh the benefits, this view would potentially be altered if prevention therapies were licensed. In this case, an evidence-based screening strategy would need to be formulated and public acceptability explored.

KEYWORDS
autoantibodies, paediatric, qualitative research, type 1 diabetes

1 | INTRODUCTION

Symptomatic onset of type 1 diabetes represents the latter stages of its natural history and can be predicted through the measurement of islet-specific autoantibodies.[1] These autoantibodies predate and predict disease such that the presence of two or more islet autoantibodies indicates an almost lifetime certainty of progression to type 1 diabetes.[2] Children with multiple autoantibodies can be further classified with oral glucose tolerance testing as Stage 1 (normoglycaemia), Stage 2 (dysglycaemia) and Stage 3 (clinical diabetes).[3]

The combination of autoantibody and glucose tolerance testing can accurately identify and stage for diabetes risk.[1] General population programmes including Fr1da in Germany[4] and ASK in the United States (US) have already screened over 200,000 children.[5] The benefits of screening include a five-fold reduction in diabetic ketoacidosis[4] and identifying the population who could benefit from preventative treatment or trials to delay onset of Stage 3 disease.[6,7] Screening is also shown to improve glucose control in the early years following clinical onset.[8] However, harms of screening include increased anxiety levels in parents with a child identified at risk.[4] Furthermore, the reduced risk of diabetic ketoacidosis at onset requires monitoring follow-up and this can be burdensome for families.[9–11]

Potential screening programmes in the United Kingdom (UK) are first evaluated by the National Screening Committee (NSC).[12] A recommendation is provided following assessment of the available treatments, cost–benefit and acceptability from the public and professional stakeholders.[12] Much of this assessment occurs against the adopted Wilson and Jungner criteria for screening, one of which is acceptability.[13] Exploring stakeholders' attitudes and concerns towards screening and discussing the practicalities of implementation is important to the success of any future screening programme.[14] Dunne et al. previously conducted interviews with a wide range of clinical and non-clinical US stakeholders.[15] These informed a discrete choice experimental survey, to explore paediatricians' preferences for general population screening.[16] US paediatricians (including 43% non-diabetes specialists) indicated reducing risk of diabetic ketoacidosis was the most important aspect of screening. They felt both education and monitoring follow-up should be offered to children identified at risk. Paediatricians ranked cost savings as the second priority area followed by availability of immunotherapy.[16]

There is currently renewed interest in type 1 diabetes screening because the Food and Drug Administration agency licensed teplizumab in 2022,[17] the first immunotherapy agent to delay onset of Stage 3 type 1 diabetes.[17] Although a licensing decision is awaited in the UK, availability of a treatment for those at risk makes a screening debate particularly timely. As such, it is essential to explore the views of stakeholders in the UK who could be involved in implementing a future screening programme. Here, we undertake a qualitative interview study to explore the barriers and facilitators to implementing a national UK screening programme from the perspective of a diverse group of professional stakeholders.

What's new?

- Very little is known about the views of healthcare professionals and stakeholders regarding general population screening for type 1 diabetes in the UK.
- Stakeholders felt the current harms outweigh the benefits for paediatric general population screening for type 1 diabetes. However, availability of a licensed therapy to delay or prevent type 1 diabetes would redress this balance.
- Paediatric general population screening for type 1 diabetes is an important research area that warrants further investigation into public acceptability.

2 | METHODS

2.1 | Recruitment

The ELSA-1 protocol has previously been published.[18] Stakeholders who may potentially facilitate and drive a national screening programme were approached for participation in the study. This included stakeholders who might recruit and screen children in the community, both in general practice (general practitioners (GPs) and community nurses), and in schools (headteachers and school nurses). Stakeholders also included paediatric and adult diabetes consultants, policymakers and commissioners of children's services. A pre-designed sampling grid was used to try and ensure adequate representation of stakeholders across healthcare and community settings (Data S1). Participants were recruited nationwide from direct approaches to relevant organisations, including the Children and Young People's Diabetes Network and the UK's Clinical Research Network. Informed consent was obtained from all stakeholders prior to interview. National research ethics approval was granted (IRAS: 294654).

2.2 | Interviews

Semi-structured interviews were chosen[19] to focus discussions around a potential screening programme, whilst allowing participants freedom to discuss topics important to their role. During the interview, a 3-min video was shown (Video S1) which explained the background to type 1 diabetes, rationale for screening and provided an outline of our future proposed screening programme (ELSA). In brief, ELSA is trialling general population screening in the UK for children aged 3–13 years. ELSA aims to explore the feasibility and acceptability of autoantibody screening.

The topic guide (Data S1) was developed from current literature exploring paediatric type 1 diabetes screening.[6] Questions and prompts were included in the following areas: (1) understanding of type 1 diabetes and experience in their setting, (2) views about paediatric screening for type 1 diabetes including benefits and risks, (3) practicalities and mechanics of a future screening programme and (4) support for research into this area. The topic guide was informed by literature, discussions with the qualitative team and other important stakeholders. Therefore, there was no pilot phase of the interview study and all stakeholder interviews were included for analysis. As the interviews proceeded, the topic guide was revised to make additional enquiries into teplizumab treatment. The revised topic guide received research ethics approval.

All stakeholder interviews were conducted by two qualitative researchers LQ (clinical research fellow [woman]) and IL (senior research fellow [man]), either jointly or individually, by video call.[20] Interviews were conducted in 2022 and each interview lasted for up to 67 min (median duration 58 min).

2.3 | Analysis

Interviews were audio-recorded and transcribed by a third party with whom there was a contractual confidentiality agreement. Transcript analysis began early during data collection, to monitor for data saturation[21] and inform future sampling. The thematic analysis presented here explores the attitudes and views towards paediatric screening for type 1 diabetes (an additional paper has analysed the data for practical learning on the delivery of the screening programme including barriers, facilitators and individual preferences).

A framework was used for analysis with domains drawn deductively from the NSC's criteria for evaluation of a screening programme.[12] This analytic framework comprised five parent domains including (1) The condition, (2) The test, (3) The intervention, (4) The screening programme and (5) Implementation criteria. Data were thematically 'charted' into the framework's overarching domains and the process reviewed in analysis meetings with the research team. The constant comparison method[22] was used. Whilst the data analysis process was largely deductive, emergent themes that did not fit within the framework's domains were also reported, so that in-practice the approach combined elements of inductive and deductive analysis.[23] A final summary of interview themes was shared with participants who had completed a study interview.

Stakeholders were grouped according to their role and resultant experience of managing individuals with type 1 diabetes; views were compared between those with the most experience (diabetes care team and policymakers) to those with less experience (community stakeholders and school staff). Stakeholders' support for general population autoantibody testing was grouped according to those in favour, against or ambivalent towards further research in this area. Finally, responses were grouped according to whether a preventative treatment would affect their support for screening research.

3 | RESULTS

Interviews were conducted with 25 stakeholders, consisting of 19 healthcare professionals (HCP) and six community stakeholders (Table 1). Of the HCPs, six were GPs, four were paediatric diabetes consultants, four were adult diabetes consultants and five were allied HCPs including a diabetes specialist nurse, community diabetes nurse, clinical psychologist and two research nurses. Six HCPs were national policymakers. Of the six community stakeholders, there was a general practice manager, a headteacher, a school nurse, an expert patient, a screening trial administrator and a diabetes services coordinator (national policymaker). Overall, median age was 53 years (range 39–67 years, $n = 20$ stakeholders self-reported age), 56% ($n = 14/25$) were women, and 80% ($n = 20/25$) were White British ethnicity.

3.1 | Themes

The four overarching themes which came from the data fitted the NSC's evaluative criteria for screening as follows (Table 2): (1) The condition—prevention of diabetic ketoacidosis, (2) The intervention—preventative treatment versus prevention trials, (3) The screening programme—benefits and burden of screening and (4) Implementation criteria—service impact. The NSC's fifth evaluative domain, 'The test' was fitted into theme four (Implementation criteria).

TABLE 1 Stakeholder characteristics.

Interview number	Age (years, if stated)	Gender (if stated)	Ethnicity	Stakeholder role
1	39	Male	White British	General Practitioner and Practice Partner
2	36	Male	Asian or Asian British	General Practitioner
3	44	Female	White British	General Practitioner
4	60	Female	White British	Research Study Administrator
5		Male	Asian or Asian British	Adult Consultant Diabetologist, Professor of Diabetes Medicine, national policymaker
6		Female	White British	National Screening Committee/General Practitioner, Professor of Clinical Ethics, national policymaker
7	67	Male	White British	Independent, advocate
8		Male	Black, African, Caribbean or Black British	General Practitioner/Royal College of General Practitioners, national policymaker
9		Female	White British	Paediatric Consultant Diabetologist
10	63	Male	White British	Retired Professor in Paediatric Endocrinology
11	57	Male	White British	Adult Consultant Diabetologist, Professor in diabetes and endocrinology
12	52	Female	White British	Practice Business Manager
13		Female	White British	Clinical Psychologist
15	62	Male	White British	General Practitioner, Professor of Primary Care Research, national policymaker
16	51	Male	White British	Head Teacher, School
17	54	Female	Black, African, Caribbean or Black British	Clinical Nurse Specialist in Diabetes
18	53	Female	White British	Research Nurse
19	48	Female	Asian or Asian British	Paediatric Endocrinologist, Associate Professor, national policymaker
20	55	Female	White British	Research Nurse
22	42	Female	White British	School Nurse
23	62	Female	White British	Paediatric Consultant Diabetologist
24	42	Female	White British	Advanced Practitioner, Community Diabetes Specialist Nurse
25	34	Female	White British	Diabetes Transitional Care National Coordinator, national policymaker
26		Male	White British	Adult Consultant Diabetologist, *Professor* of Experimental Diabetes and Metabolism
27	60	Male	White British	Adult Consultant Diabetologist, national policymaker

Note: Stakeholder demographic characteristics and role.

3.2 | The condition—prevention of diabetic ketoacidosis

Diabetes HCPs gave accounts of type 1 diabetes as a condition that has 'life-changing' consequences [interview 17]. All stakeholders described the 'trauma' [interviews 2, 8, 24] of diagnosis, which was exacerbated by an emergency admission with diabetic ketoacidosis [interview 9]:

> 'There are a significant number of people that I see who are traumatised by their diagnosis but not so much the diagnosis but the fact that they have developed diabetic ketoacidosis and ended up in hospital really quite seriously ill at the time'. (11, Adult diabetes consultant).

GPs emphasised the importance of timely diagnosis [interview 15] to prevent diabetic ketoacidosis.[24] A few stakeholders felt education and monitoring for diabetic ketoacidosis prevention 'alone' could justify general population screening [interviews 1, 15]:

> 'So I think that in itself preventing children from suffering harm or sadly dying from DKA, I think is a real rationale for doing this. I think the main pros would be hopefully ideally this type of approach could help to

TABLE 2 Summary of themes.

Domain	Themes	Sub-themes
[1] The condition	Diabetic ketoacidosis prevention to reduce morbidity and mortality	Type 1 diabetes is an important condition
		Importance of early diagnosis to prevent diabetic ketoacidosis
		Diabetic ketoacidosis prevention alone could justify general population screening
[2] The intervention	Preventative treatment	A licensed treatment (e.g. teplizumab) would increase priority for general population screening
		Issues with teplizumab treatment
	Prevention trials	Societal benefit
		Parent and child benefit if treatment effective
		Familiarise the parents and child with research to support future trials
		Minority of first degree relative children (FDR) have enrolled in prevention studies
[3a] The screening programme—benefits	Education	Educating parents for earlier symptom recognition
	Monitoring	Track progression and smoothen transition
	Preparedness and empowerment	Facilitate the 'best start' to the condition
		Improved glycaemic control following diagnosis of Stage 3 type 1 diabetes
[3b] The screening programme—burden of screening	Living with risk—parents	Burden of knowing their child will develop a lifelong condition
		Uncertainty of not knowing when Stage 3 type 1 diabetes will arise
	Living with risk—child	Ruining the innocence of childhood
		Stigma and over-medicalising a healthy child
		Provoking premature health anxiety
		Threatens the child's right to an 'open future'
[4] Implementation criteria—service impact	Identification/ recruitment	FDR versus general population screening—lack of evidence for public acceptability
	Testing process	Convenience and ease of use are priorities
	Implementation	Lack of resources, staff, training and guidelines for management
	Follow-up	Increased service utilisation secondary to health anxiety
		Support for families
	Cost-effectiveness	Lack of evidence for cost-effectiveness in the UK

Note: Domains from the National Screening Committee's evaluative criteria and the themes and sub-themes which fitted into this framework.

almost eliminate death due to DKA in children'. (15, GP, national policymaker).

3.3 | The intervention—preventative treatment versus prevention trials

Most stakeholders agreed that if a preventative treatment, such as teplizumab, were licensed in the UK [interview 27], this would transform the debate in favour of screening (Table 3):

> 'The screening side for me would come into its own if our interventions were more robust. So, if I could sit in front of a family and say it's definitely worth screening because this is what's on offer, and the chances that we can postpone, we can cure etc, are this, then I would be very comfortable'. (23, Paediatric diabetes consultant).

Concerns raised by paediatric diabetes consultants and policymakers about immunotherapies included unknown long-term side effects and infusions not being ideal for children [interviews 19, 24].

> 'It's weighing up what are the long-term problems with the treatment that we're giving to prevent or delay the diabetes from occurring, is it going to ensure that they don't have any other issues later on in life or are we just stopping it for now and bringing something else on?' (17, Diabetes specialist nurse).

Prevention trials were deemed important for societal benefit [interviews 2, 17] and would provide a 'glimmer of hope' [interview 18] for the family. A registry of at-risk children would be useful so that families could be informed about future trials [interview 24]:

TABLE 3 Stakeholder support for childhood autoantibody screening for type 1 diabetes.

Record	Stakeholder role	Supportive of screening research	Ambivalent	More supportive if teplizumab licenced	Could still screen without preventative treatment	Concerns raised about immunoprevention
General practitioners and practice team						
1	GP	X			X	
2	GP	X			X	
3	GP	X		X		
6	NSC/GP/national policymaker		X	X		
8	GP/RCGP/national policymaker	X				
12	Practice manager	X			X	
15	Academic GP/national policymaker	X		X		
Paediatric consultant diabetologists						
9	Paediatric Consultant		X	X		
10	Paediatric Consultant		X	X		
19	Paediatric consultant/national policy maker		X			X
23	Paediatric consultant	X			X	
Adult consultant diabetologists						
5	Adult diabetes consultant/national policy maker		X		X	
11	Adult diabetes consultant	X		X		
26	Adult diabetes consultant	X				X
27	Adult diabetes consultant/national policymaker		X	X		
Members of the diabetes care team/Healthcare professionals						
13	Clinical Psychologist	X		X		
16	School Nurse	X			X	
17	Diabetes Specialist Nurse	X		X		
18	Research Nurse	X		X		
20	Research Nurse		X	X		
24	Community Diabetes Nurse	X				
Other stakeholders—Non-healthcare professionals						
4	Research Administrator	X		X		
7	Independent		X	X		
22	Headteacher, School	X			X	
25	Diabetes Transitional Care Coordinator/national policymaker		X		X	
Total		16/25 (64%)	9/25 (36%)	13/25 (52%)	8/25 (32%)	2/25 (8%)

Note: Stakeholder groups and their level of support for general population screening for type 1 diabetes in children (supportive or ambivalent). Views towards screening if there were a licensed preventative agent. Support for screening without a licensed preventative treatment and concerns raised with immunoprevention.

> 'it was really nice to be able to offer people an intervention study of something to do, rather than just deliver the bad news and say well now we're all just going to sit round and watch and wait kind of thing, and twiddle our thumbs', (18, Research nurse).

Stakeholders with research backgrounds explained how prevention trials would familiarise the family with research in a non-threatening way, to increase their confidence to take part in future trials[25]:

> 'Whereas I think in people that have come through screening it's like they already know

> okay research is not so scary, I have been there and I have done a bit of it, and it's familiar'. (18, Research nurse).

A paediatric diabetes consultant acknowledged a small proportion of first-degree relatives (FDR) have stepped forward for prevention trials likely due to 'anxiety' about unlicensed treatments [interview 9]:

> 'It is only a minority of families that feel that it's a good thing to do, and they are more often the more educated families that can see through the fact that humans aren't guinea pigs in clinical studies'. (9, Paediatric diabetes consultant).

Overall, 64% of stakeholders supported research into paediatric type 1 diabetes screening (Table 3). GPs and diabetes care team professionals were more likely to support screening research (83%–86%), followed by adult diabetes consultants (50%). In contrast, three in four paediatric diabetes consultants were ambivalent towards screening research. The majority of stakeholders (n = 22/25, 88%) either supported screening research without a licenced treatment (n = 16/25, 64%) or would support if teplizumab was licensed in the UK (n = 6/25, 24%). Licencing of teplizumab in the UK would lead six of the nine (67%) ambivalent stakeholders to support screening research. Only two stakeholders held concerns about immunoprevention treatments, such as teplizumab.

3.4 | The screening programme—benefits of screening

Stakeholders felt education was important for general population families to prevent late presentation [interview 20]. GPs recognised the importance of recording the screening results in a child's medical heath record to raise suspicion of type 1 diabetes for future healthcare contacts:

> 'Once you have realised they have got a predisposition to develop type 1 diabetes, if they were to present with another symptom you would have a higher index of suspicion or a lower threshold for screening at that point'. (3, GP).

Stakeholders felt monitoring was essential to smoothen the transition to Stage 3 disease. Earlier identification of type 1 diabetes would give the family time to prepare and learn to cope:

> 'Being prepared will often put you in a better position to deal with the outcomes and manage it I think, because you have had time to actually understand what the process is and what the treatments are etc, and the impact it may have'. (17, Diabetes specialist nurse).

The knowledge acquired, following identification of risk status could 'empower' the family to gain early access to care services and optimise diabetes treatment [interviews 15, 24]. This would support the 'best start' to clinical management of type 1 diabetes [interview 5] and help facilitate improved glycaemic control in the longer-term[8]:

> 'Because of knowing it's early, we will be able to give them all the technology from the beginning and keep their HbA_{1c} low from the very start'. (5, Adult diabetes consultant and national policymaker).

3.5 | The screening programme—burden of screening

Living with risk was perceived to be a heavy burden for the parent, from knowing their child was going to develop a lifelong condition in the future that could affect their life choices, career decisions and insurance policies [interview 2]:

> 'You have given them not exactly a death sentence, but there's the sword of Damocles hanging over their necks'. (10, Paediatric diabetes consultant).

Not knowing when the diagnosis would 'unmask' [interview 5] itself was raised as an additional burden, with fears parents would continually be seeking out symptoms of type 1 diabetes:

> 'But they are then just aware of the fact that their life is going to change in the future, and that's a really big burden to carry actually for the families who are then watching out for every little sign'. (26, Adult diabetes consultant).

Stakeholders suggested screening could introduce stigma for the child because of (over-) medicalisation:

> 'Yes, I think so you identify a child as high risk. I think there would be the consequences would be what I was just saying I suppose around potential anxiety, this child becomes a protected child or an unusual child in a

family, so the family dynamics might change', (15, GP, national policymaker).

Many stakeholders raised concerns about 'ruining the innocence of childhood' [interview 1] and provoking premature health anxiety in children, threatening the child's right to an 'open future':

> 'You can argue you have just ruined the rest of their childhood by the knowledge they are going to get this pretty unpleasant disease that's going to have massive impacts on their quality of life, day to day living etc'. 'when in fact they could have lived the next 14½/15 years without having to worry about any of this and enjoy the rest of their childhood'. (10, Paediatric diabetes consultant).

3.6 | Implementation criteria—service impact

3.6.1 | Identification and recruitment

Stakeholders debated whether FDR testing should be prioritised over general population screening. Most stakeholders saw clear benefits to FDR testing, recognising risk of type 1 diabetes as a 'perennial' worry for these families [interview 5]. Some stakeholders suggested FDR testing was more acceptable because of the lived experience within the family to support the child:

> 'if there are people who have got other family members with diabetes they can, having seen what diabetes means to their other family members, they can perhaps process it a little bit differently for somebody who has no previous or prior experience of diabetes'. (11, Adult diabetes consultant).

Yet, stakeholders were concerned about FDR families having another affected family member [interview 9]. They also recognised FDR testing would not be accepted by all families [interview 19] and suggested screening should be offered on a voluntary basis [interview 26].

> 'The immense guilt around should we really have had children in the first place, are we passing our faulty genetics onto our children, so it's all of that, that can impact on you psychologically'. (17, Diabetes specialist nurse).

For general population families without lived experience of type 1 diabetes, many diabetes specialists felt acceptability data was lacking [interview 10]. There were concerns that a high-risk result in this group could be 'really scary' [interview 25]:

> 'If you were to result to a positive to a family that had no history of diabetes in the family I think I could imagine that being more of a shock, where did that come from, but because there's already a relative it's like oh okay, and occasionally people have said, 'Yeah I am not surprised', or, 'I was expecting it'. (18, Research nurse).

However, other stakeholders suggested screening could help reduce the trauma of diagnosis for these families:

> 'Making the diagnosis a less traumatic experience at the time. I still think it's going to be hard however it's diagnosed, but there are ways of making it less hard, and one of those ways is actually keeping that person out of hospital at the time of their diagnosis'. (11, Adult diabetes consultant).

3.6.2 | The screening test

Almost all stakeholders felt the most important attributes of the screening test were convenience and ease of use [12, 18]. Stakeholders agreed that offering the screening test alongside vaccinations in general practice or schools would ensure it was accessible, particularly to families who otherwise may not come forward for screening:

> 'We have quite a deprived population'. 'I think if it can be incorporated somehow into something that's already being done that's helpful, rather than creating a new visit or a new process', (2, GP).

3.6.3 | Implementation

Stakeholders, particularly national policymakers, worried about the system pressures on the NHS, including lack of staff and resources to effectively implement general population screening [interview 24]:

> 'At the moment, there is very little capacity in either secondary care or primary care, so therefore the, "what were we going to do about

it," I haven't got that answer. The funding at the moment is not there'. (19, Paediatric diabetes consultant and national policymaker).

3.6.4 | Follow-up

HCPs were concerned that health anxiety from screening could translate into increased health-seeking behaviours:

> 'I think it's just this small proportion where there is already a propensity for anxiety either within the family or the child's natural demeanour themselves, I am worried that this could trigger something there', (1, GP).

Stakeholders particularly worried about management of families who opted-out of the monitoring follow-up:

> 'It is the monitoring that we find it's harder to get people to continue with, because once they have the result, they think that's that done, and they are not so keen to go along and have the blood tests'. (4, Screening trial administrator).

Both primary and secondary HCP said appropriate management pathways were needed to guide referrals and treatment:

> 'The fact that your study people would follow them up and give them the education that's great, because otherwise if they turn up at... not necessarily me, but turn up to a GP and are told, 'I've got this', they will be like, 'Well I don't know, who do I refer you to?' (24, Community diabetes nurse).

3.6.5 | Support

Psychological counselling was deemed important for ongoing care of the child to alleviate the burden of living with risk:

> 'Simply giving them a timeline or oral glucose tolerance test and giving them a clinical "this is your stats, this is the statistics," isn't going to be useful for the worried parent. But that's going to be really hard because we barely have enough psychologists'. (19, Paediatric diabetes consultant and national policymaker).

Several stakeholders suggested peer-support was valuable to educate families:

> 'peer support is really invaluable isn't it? It's fine for us to tell them things, but actually having it from people in a similar position is always very valuable'. (23, Paediatric diabetes consultant).

3.6.6 | Cost-effectiveness

National policymakers felt evidence was lacking for cost-effectiveness of screening in the UK. These stakeholders questioned whether cost–benefit would be achieved without a preventative treatment [interview 10] and whether general population screening was a good use of limited resources [interview 19].

4 | DISCUSSION

This is the first qualitative interview study to explore the views of diabetes specialists, policymakers and community stakeholders in the UK towards paediatric general population screening for type 1 diabetes. Whilst this study was undertaken in the UK, the findings are potentially applicable more widely. Rich data emerged across the domains of the NSC's evaluative framework.[12]

There was consensus agreement that type 1 diabetes is an important health condition for which emergency presentations in diabetic ketoacidosis should be prevented. This finding corroborates the quantitative study by Dunne et al. showing that reduced risk of diabetic ketoacidosis was the most important aspect of screening for paediatricians in the US.[16] However, for many UK stakeholders, diabetic ketoacidosis prevention alone did not justify general population screening and other primary prevention strategies such as public health campaigns may warrant further exploration.[26] Most stakeholders felt the lack of licensed intervention counted against general population screening. However, a few stakeholders felt participation in trials for immunoprevention addressed the absence of a licensed therapy. A licensed drug for immunoprevention in the UK could transform the debate on general population screening and therefore the outcome of the UK licensing decision for teplizumab is critical.

All stakeholders recognised benefits and harms of screening. Earlier identification offered a smoother transition into this 'devastating' diagnosis, giving the family time to prepare and gather information to give their child the 'best start' to their long-term diabetes care. However, the burdens of screening include living with risk, onus

on parents for symptom recognition and medicalising a healthy child. There were significant concerns that screening could negatively alter the life-course of a child and result in loss of the asymptomatic, care-free, 'golden years' of childhood.[27] Paediatric type 1 diabetes screening appears to open a pandora's box of ethical dilemmas, for which the ripple effects on the parent, child, family unit, their HCP and society as a whole could not foresee when a parent signs-up for screening.

On balance, many stakeholders struggled to justify general population screening and raised concerns about the lack of evidence for public acceptability. Whilst stakeholders felt that risks outweighed the benefits here, they felt differently for FDR testing where benefits outweighed harms but acknowledged this should be opt-in. Importantly, there were universal fears that funding, staffing, resources and system capacity were not established in the UK to support a national screening programme for type 1 diabetes. Paediatric diabetes consultants raised several implementation concerns, which accounted for their ambivalent stance towards paediatric screening research. Nevertheless, general population screening was deemed an important research area by most GPs, community stakeholders and adult diabetes consultants interviewed. Priority research areas included immunoprevention trials, assessing public acceptability for screening and evaluating cost-effectiveness in the UK. Stakeholders felt there should be a follow-up programme that supports the clinical and psychological needs of the family to alleviate the burden of living with risk.

This study contributes important data to the acceptability domain of Wilson and Jungner's screening criteria.[13] Strengths include the diverse cohort of professionals from healthcare and community settings. Although this interview series was conducted prior to the US licensing of teplizumab, enquiries into how licensing would impact views towards screening were gathered and are clearly shown to be relevant to stakeholder views. The data was fitted to the NSC's framework to optimise future evaluations of general population screening.[12] These views are widely applicable to help inform design and delivery of screening programmes around the world. Views towards licencing of teplizumab in the US and impact on screening in the UK will be further explored in focus groups with relevant stakeholders.

Limitations of this study include representativeness of the stakeholder cohort, which was restricted by ethnicity, age and gender, although themes did not differ according to these demographics. Community stakeholders represented a minority of those interviewed, although general population screening could impact these individuals more so than diabetes specialists. However, the interviews revealed consensus agreement of views across the community stakeholder sub-group. Although stakeholder interviews were conducted using video call, use of remote qualitative data collection methods have increased in response to the COVID-19 pandemic and evidence suggests that data quality is high when compared to face-to-face interviews.[28] Unfortunately, the topic guide was not completed for all stakeholders. This was because the stakeholders interviewed had limited time. As a result, interviews were sometimes ended prematurely and it was not possible to arrange a follow-up interview. This further demonstrates the current system pressures stakeholders face in their professional roles.

In conclusion, type 1 diabetes is an important health condition where diabetic ketoacidosis prevention at onset is a national priority. Stakeholders in the UK felt whilst there are some strong arguments in favour, the harms of general population screening currently outweigh the benefits. The licensing of a preventative treatment would allocate higher priority to general population screening. Stakeholders highlighted the need for appropriate support systems for parents and their children identified at risk through a screening programme to mitigate for psychological harm.

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CONFLICT OF INTEREST STATEMENT

LQ, PN, MR, KB, FB, SG and IL have no conflicts of interest to declare.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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