

**Valuing the benefits and harms of antenatal and newborn screening
programmes in health economic assessments (VALENTIA)**

SUPPLEMENTARY MATERIAL 3

Additional Results from Chapter 7

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Benefits of antenatal and newborn screening programmes as told by individuals who have experienced screening

Benefit Examples	Supporting Quote(s)
Screening is free at point of service	<p>“Well I think we’re pretty lucky that, that where we are we don’t have to pay for it, or like you don’t have to kind of say well I can’t afford all these tests, so I’ll pick which one’s that I’ll do.” - Kelly</p>
Screening allows people learn about conditions	<p>“I don't think there were any negatives - I suppose they made me aware of more things that could be wrong - e.g. I had never heard of Edward's syndrome so without screening I would never have had to consider my baby could have it, but that is far outweighed by the positives - knowledge is power in my opinion!” - Aria</p> <p>“I think as well just sort of learning about different conditions that you know that people have to sort of deal with I think gives you a bit more empathy as well when you’re, yeah, when you’re sort of going through it.” - Emily</p>
Screening tests work and are done for a reason	<p>“I think if anything it just made me realise like the screening tests do work. Like they’re not just a tick box exercise, like they are actually checking for something and the doctors do know what they’re talking about.” - Ada</p> <p>“Like the way that they’re done. Like there’s a real sort of sense of like it being done to support you and you know I can’t, I can’t sort of fault anyone in, in those, you know with the tests that they do.” - Kelly</p> <p>“Also because you trust in the science and the doctors so they can give you and your child the best care.” - Maisie</p>
Screening allows people connect with their pregnancy	<p>“The positive about the screening tests is seeing my baby on the screen. This is my first pregnancy and I don't feel the baby moving yet so it is really nice to be able to see the baby and connect that way.” - Harper</p> <p>“I was glad to have all the scans, it was reassuring at a time when I was frightened. Just seeing him & hearing his heart was everything.” - Lily</p>

Benefit Examples	Supporting Quote(s)
	<p data-bbox="454 315 1374 409">“The fact that we were actually able to find out that we were having a little girl and keep that information to ourselves, and it was just like a nice little secret.”</p> <p data-bbox="454 439 552 472">- Poppy</p> <p data-bbox="454 562 1370 714">“To be honest I think both of us actually looked forward to the scans. Even though they were times we could find out really serious news about the baby, we just saw them as an amazing chance to see an image of our child. Probably a bit naive of us!” - William</p>
Screening may prevent harm	<p data-bbox="454 808 1378 1267">“I mean the, the heel prick test, you know really has saved, I mean maybe not saved their lives but has you know dramatically enhanced their lives. If we didn’t have that test then they would have been eating normally for years and you know they might not have known what it was that caused it, and it could have just kept getting worse and worse, and you know really they could have been very sort of severely, severely handicapped so yeah. No, I think it’s, and you know I think some of the other conditions it is a, it’s a life saver, catching those conditions early. So yeah, no, completely valuable. Yeah.” - Emily</p> <p data-bbox="454 1357 1342 1451">“The diagnosis meant when my baby was born the specialists were there ready if he needed immediate care, fortunately they weren’t needed.” - Lily</p> <p data-bbox="454 1541 1347 1635">“And same with the heel prick if, if you do pick up something you can sort it out, you can’t if you, if you don’t look.” - Owen</p>
Screening may give reassurance	<p data-bbox="454 1666 1362 1760">“Each step through the screening journey helped me to feel that the pregnancy was more real. There was certainly some reassurance from that.”</p> <p data-bbox="454 1789 584 1823">- Elizabeth</p> <p data-bbox="454 1912 1326 2007">“Screening tests felt like milestones in my pregnancies, a step closer to meeting my babies. It was also reassuring to know my babies were doing</p>

Benefit Examples	Supporting Quote(s)
	<p>okay!" - Imogen</p> <p>"So, I guess it gives you reassurance early on in your pregnancy that there's nothing genetic going on, which I think is quite useful. I don't know, I always get a bit nervous when I'm pregnant. I don't consider myself properly pregnant till I've had my 20-week scan and they've confirmed everything's alright." - Isla</p> <p>"So, we knew for certain that baby wouldn't have any chromosomal abnormalities, which is a nice thing to know about your pregnancy, because it's one thing that has been ruled out. So that was good." - Poppy</p> <p>"I think I just naturally worry and I felt quite reassured when I was checked over - especially the scans it was lovely to see baby and to confirm that all was ok. I had concerns re 20 weeks that my baby might develop an issue I had as a child so was relieved to know he was forming perfectly - Sophia</p> <p>"The various different checks you have it definitely makes you feel like there is a safety net there, that like a lot of the investigation, the, feeling constantly worried with newborns that there's something desperately wrong with them, and that they're poorly or something, and I think knowing that there are like this series of tests that have been taken that like basically guarantee like, whether that, I think both pre-birth and post-birth, that you know that there is this regular screening, that things that might go wrong have been screened for and picked up give you that additional sense of sort of security. And sort of calms a lot of worries, I think. At least that did for me." - Thomas</p>
<p>Screening may give people information so they can ask</p>	<p>"I could also reach out to local support groups who were then already supportive by the time my daughter arrive. I had people to ask questions to." - Florence</p>

Benefit Examples	Supporting Quote(s)
questions	<p data-bbox="454 315 1366 472">“You’re able to plan or able to, if not plan at least to have that awareness to think well how, what am I going to do from here? Do you know, am I going to seek support from somewhere, seek further information?” - Quinn</p>
Screening may give people information that allows them to consider termination	<p data-bbox="454 506 1374 775">“As far as I’m concerned the more information the better, because as a parent you are forearmed with information that then might be useful for you. You can have a much longer lead time to do what you can prepare for what is to come, or if you don’t want to continue then to stop that pregnancy.” - Alexander</p> <p data-bbox="454 869 1350 1025">“I am pleased that I have had the NIPT test as I mentioned before for me I would have wanted more information and would have to consider if I was to have a child with potential health conditions.” - Elsie</p>
Screening may give people information that allows them to get prepared	<p data-bbox="454 1055 1362 1267">“Even though we had the test and we found out [diagnosis] it would not have changed anything, but we wanted to know. Knowing what the situation was helped us to prepare, so where we, we’re just people who would rather, we felt more in control knowing, knowing what the situation was.” - Donna</p> <p data-bbox="454 1361 1366 1518">“I was glad to have had an early diagnosis. It gave us a chance to make links with local families and become part of a community before our son was born. It was very reassuring.” - Evie</p> <p data-bbox="454 1612 1326 1825">“As the NIPT wasn’t diagnostic and I’d read so much online about it giving inaccurate results, I felt I wanted the diagnosis to be able to prepare and learn as much as I could, and prepare my sons & also my children’s grandparents.” - Florence</p> <p data-bbox="454 1919 1374 2002">“I had NIPT done because my husband and I felt that if the results came back that our baby may have a learning disability or chromosomal problem then we</p>

Benefit Examples	Supporting Quote(s)
	could adapt and get used to the idea before our baby was born.” - Sienna
<p>Screening may give people information that allows them to adjust expectations</p>	<p>“It meant that it was hard in the sense that we’ve been grieving since [getting a diagnosis] essentially in a form because, you know I like to say our hopes for this baby you know changed the minute we heard there was an issue. And we grieved over all that time, the whole, you know the next six months or whatever it was, just to get our heads around what it could mean and I suppose also appreciate the pregnancy maybe in a way we wouldn’t have possibly done so if, if we just thought it was just another regular healthy baby.” - Donna</p> <p>“I was glad that I had all the available scans and antenatal screenings offered to me as it gave me a sense that I was at least in possession of as much knowledge as I could be to manage my expectations and plan for the future.” - Ella</p> <p>“I felt prepared and could prepare those around me. I had the emotional rollercoaster at diagnosis, I don't think I'd have coped as well if I had just given birth & had a postnatal diagnosis, with hormones already all over the place.” - Florence</p>

Harms of antenatal and newborn screening programmes as told by individuals who have experienced screening

Harm Examples	Supporting Quote(s)
<p>Screening rhetoric may not acknowledge potential harms</p>	<p>“Like we weren’t really given that detail of information, we were just told we’d get like a scan, it would be part of the scan, and then we’d get a blood test. And that would be it. So, we just didn’t, it just felt like really light touch and almost like there’s nothing bad’s gonna come out of this, it’s just you know a nice extra thing that we can do for your peace of mind... and you know the more informed you are at least before you even go into it, like the more you can manage your own expectations of what actually you know there are risks that it’s gonna come out negative, and therefore these are like the kinds of things that might happen afterwards.” - Poppy</p> <p>“I’ve definitely seen it framed as... these are things that we’re offering, you should know, and then people who’ve been undecided [HCPs] definitely come down on the side of, ‘Well you should have it done.’ Not necessarily with an argument as to why, other than you need to know sort of thing... And I think actually there is some, as with any screening, there is a potential harm of doing it. I’m not convinced it’s acknowledged consistently at least.” - Owen</p>
<p>Screening for conditions may imply that high chance pregnancies should be terminated</p>	<p>“I also agree that there should be openness about why screening happens. It is done with the language of choice but that immediately infers termination. There should be greater clarity about how it is all optional, which most people seem unaware of.” - Evie</p> <p>“To repeatedly offer termination as an option is not offering choice; it is putting pressure on parents who are already perhaps worried or upset about the diagnosis. The screening is biased towards termination anyway... by providing [this] literature when a diagnosis is given, it is presented as if that is the route it is assumed people are taking.” - Evie</p>
<p>Normative</p>	<p>“Some of the screening I have accepted/requested has made me feel very</p>

Harm Examples	Supporting Quote(s)
<p>screening practices do not suit everyone</p>	<p>uneasy because I feel it has given people, or might potentially give people, reasons or excuses to find something wrong with my pregnancy or my conduct.” - Daisy</p> <p>“I suppose screening in the NHS has to be what is best for most women or most babies but it doesn’t always mean that it is right for each of us.” - Matilda</p>
<p>It may be hard for people to understand what they are consenting to</p>	<p>“I think the language they use to label the tests is confusing. I don't know how they could make it more memorable or friendly but there's so many tests you read about that you've never heard of before when it's your first pregnancy, so when it comes to deciding which to have, or if a HCP says something about it, you have to sort of translate it to yourself and remember 'is that the one where the illness does this'.” - Maya</p> <p>“Yes, I feel like I’m not given enough info about the various tests they offer. I never know what they’re testing me for.” - Sienna</p>
<p>People may be unprepared for unexpected results</p>	<p>“It is difficult not having all the information - which is more of a problem when there is a result that is unexpected or unfamiliar.” - Eva</p> <p>“And I felt that if I had have been a first-time mum I would find myself just going along with everything and not prepared at all for bad news.” - Imogen</p>
<p>Screening may cause emotional distress</p>	<p>“Yes and I think once you have had bad news at a scan, or known someone have bad news, it is hard to ever see them in the same way. I cried my way through all scans until I saw a heartbeat, and then the reassurance they gave was only fleeting. I saw them very much as part of a journey, rather than an end point. I remember even when my babies were in my arms as newborns I was worrying about the cystic fibrosis screening on the heel prick for instance.” - Ella</p>

Harm Examples	Supporting Quote(s)
	<p data-bbox="454 253 1380 409">“Every scan brought days of anxiety and worry and scans without my partner were numb and worrying and any relief was usually washed away with the thought of the next scan to come.” - Layla</p> <p data-bbox="454 499 1380 712">“Some times between results or with the miscarriage potential of a test I got so worked up. When we found out all was okay on the phone I had a mild panic attack couldn't breath and didn't know whether to cry or be happy.” - Maisie</p> <p data-bbox="454 801 1380 1081">“I'm quite an easy going person in general but like I said before I do think the screening made me think about things I wouldn't otherwise have thought about but I also definitely over thought about some of it. My partner found it hard too because he wasn't at appointments in person to ask about things I think he worried more than he usually would.” - Matilda</p>
False-positive screening results may cause residual emotional distress	<p data-bbox="454 1111 1380 1574">“I'm such a worrier like I said it was just always at the back of my mind. And then even for the first few weeks after she was here. I don't have those kind of thoughts anymore, but definitely in the first couple of weeks like, or probably even up to like the first month, definitely kind of did pop into my mind occasionally where I was like, 'Oh my God, could there actually still be something wrong with her that they've just not discovered yet?' Cos like they don't do a head-to-toe body scan when a baby's born, so there could be something wrong and they just don't know it yet.” - Poppy</p> <p data-bbox="454 1664 1380 1877">“There was so much anxiety caused by the sort of perception that [son] was like underweight but they couldn't really under, they couldn't really explain what was going on, and then he was born absolutely fine, and then I do wonder if there was a bit of like measurement error there.” - Thomas</p>
People may not understand that	<p data-bbox="454 1906 1380 2000">“I think it's nice to have reassurance if it's good news. For example, I haven't once worried about our baby having the conditions mentioned in the</p>

Harm Examples	Supporting Quote(s)
screening tests are not diagnostic tests	<p>screening leaflet since we were told they were low risk. Though I appreciate this isn't always accurate, and would feel extra hurt and unprepared if they had one of these conditions despite low result in screening.” - Maya</p> <p>“Really they have to make it clear that as just a screening it’s not diagnostic. They can’t just give you a bit of paper and get you to infer, you know just to know that diagnostic and screening are different. Someone verbally has to say to you just because it says this, it doesn’t mean that you know on the delivery day you may or may not have a child that has these things. Because none of these tests are a hundred percent accurate.” - Mia</p> <p>“Some of the harms - you could end up making a decision based on information that is constantly changing - e.g. our baby was not expected to live to term, and at any point I could have chosen to terminate based on this information, but in the end the baby was born with no health concerns whatsoever. So what was missing was for the medics to inform me that these scans/tests are not always 100% accurate and that babies are constantly growing and changing, so what they see one week might not be there at the next scan.” - Sophie</p>
Communication about results may be unsatisfactory	<p>“I don't think I really spoke to anyone about screening, I got the impression that had anything been wrong I could have had an in-depth chat with my midwife, but because my results were all fine no one ever talked about them.” - Aria</p> <p>“I hate the 'no news is good news' thing they tell you. It would be much better if they just let you know if everything is ok.” - Harper</p> <p>“Communication seems to be lacking throughout the process. As I've said before, they seem to treat it on a need to know basis or just that we don't</p>

Harm Examples	Supporting Quote(s)
	<p>know what's going on." - Scarlett</p> <p>"Also the results to my 12 week scan never made their way back to me... I've been told no news is good news but I haven't actually seen the piece of paper.... Every appointment or communication I have with my midwife I've asked them to chase them up and they still haven't they've just said, 'No news is good news so your results are probably fine'." - Sienna</p> <p>"I felt (re communication of results) that essentially if they were fine no one was really bothered about telling you. I specifically asked my midwife re results of bloods etc as they weren't brought up. I eventually got used to the idea that silence meant all was ok." - Sophia</p>
<p>Communication and support for high-chance results may not be appropriate</p>	<p>"I understand the logic of hearing bad news from a qualified person, but often not knowing/understanding and feeling like people are holding out on you is worse than hearing 'this is something we need to look into more' e.g. I would HATE for a sonographer to notice something and then be unable to tell me why they were referring me on - just tell me what you suspect as it is the uncertainty that causes panic." - Aria</p> <p>"They didn't look after me, or I feel like they didn't look after me during my pregnancy, give us you know had diagnosis sooner, I think you know that, the option of whether we wanted to have a risky pregnancy like that was taken away from us, and I think it's just made the whole experience that much worse that we went into our pregnancy blindfolded if you like and the labour blindfolded, and we put a lot of trust in, in the doctors and the nurses and the medical professionals, and I feel like overall they've let us down hugely." - Erin</p> <p>"I think we had problems around the language that was used to talk about</p>

Harm Examples	Supporting Quote(s)
	<p>risk. We struggled quite a lot with being contacted randomly by phone out of the blue; that was a really hard thing for us to deal with. And I'm not saying this is a bad thing, but we would have benefitted from being able to access information in writing, rather than orally over the phone." - Heather</p> <p>"They wash their hands of it a little bit, that's it. You do the screening and then there's no follow-up or support. That, that was quite hard. Yeah, you do the screening, you come back high risk and then that's it. You have to wait, to get, I mean you have to wait for an appointment for your amniocentesis or you have to just deal with it, there was nothing. They didn't refer me to talk to my own midwife. There was nothing." - Isla</p> <p>"After having to consent so explicitly to the tests, I was blindsided by getting results I didn't even know I was tested for, for a problem I didn't know existed. As someone who likes to be informed, I would have liked to know before what was being tested." - Ruby</p>

Benefits of antenatal and newborn screening programmes as told by stakeholders

Benefit Examples	Supporting Quote(s)
<p>Screening may provide information to make informed choices</p>	<p>“I can see that for some women knowing in advance that baby has or may have [condition], some parents do find that of benefit. They like to prepare when they want to know, so I guess for them that works.” - 3</p> <p>“It means you can prepare and best healthcare for pregnancy and afterwards and also you know emotionally and all being ready to celebrate and welcome this new little person to a loving family, yes.” - 5</p> <p>“I suppose for women that want to know and want to get information beforehand then I suppose, you know, information is for many people helpful.” - 5</p> <p>“But I think in our, again in our experience we look at outcomes for people, they would not regret that they were enabled to gain information about the baby, they regret that it happened to them, or you know all that, but not that they were able to, to have the information and make the decisions that they wanted to make, be that to continue or end the pregnancy. And I think research bears that out the majority of women value the opportunity to have that information.” - 7</p> <p>“I think what is beneficial is that it allows people to make what’s known as informed choice, right. Screening is not there to tell you don’t have a baby with [condition]. It’s there to give you all the information so that as parents-to-be you are able to make your own choices about whether if you’re at risk of having a child with [condition], how you will deal with it, how you might need to care for it. So that’s, to me, that’s the benefit.” - 9</p> <p>“So I mean the primary reason for those screenings is around understanding</p>

Benefit Examples	Supporting Quote(s)
	<p>the health of the pregnancy, and that I think is a, that's a primary benefit before one links back to any action. So they understand how healthy a pregnancy is, how the pregnancy is progressing and so on... We believe it should be offered on that basis, so obviously choices can be made or plans changed, or decisions made following the understanding of that information. And those can include making a decision to terminate a pregnancy, but also to gain more information about any kind of signal one is getting from the prenatal tests. And making, hopefully getting time to consider the choice one might want to make, and whether as a couple, as a family, one feels positive towards the idea of continuing a pregnancy that might be affected by a genetic condition, or a fetal anomaly, or whether the best decision for the family or the couple is to terminate the pregnancy." - 10</p> <p>"And then there's information. So I think we should not ignore the fact that just knowing what's going to happen to your child and being able to plan for it and so on, in terms of the trajectory of their condition is enormously valuable." - 10</p> <p>"[Screening for condition] helps the family have a chance to decide on what to do." - 13</p>
Screening may prevent harm	<p>"So from a medical point of view, I think having a high chance or confirmed result does mean that in theory the medical team should be looking at [taking care of baby] so I suppose from that perspective it's good." - 3</p> <p>"It is sensible to maintain health and wellbeing as long as possible and your best chance of doing that is by identifying the condition as early as possible and screening is the best way to do that by a long way." - 4</p> <p>"The ones that are metabolic conditions are clearly of fundamental benefit,</p>

Benefit Examples	Supporting Quote(s)
	<p>that at the very earliest possible stage in a newborn's life we've discovered [it] and we can make adjustments to their diet to almost completely reduce the chances of them suffering negative consequences of that metabolic disease.”</p> <p>- 10</p> <p>“Newborn screening cuts that diagnostic odyssey to a panicky day at the hospital and then a second visit and then you've got your diagnosis. But that comes before you understand, before you've been affected by the condition, but the opportunity to avoid years of concern and, and potentially loss of treatment opportunity. I think [this would be] considered by those that have been on a diagnostic odyssey to be enormously valuable. For those that haven't maybe it's less easy to see the value.” - 10</p>
<p>Screening may allow people to make informed decisions about their future reproductive intentions</p>	<p>“It's helpful for families to know that they have the genetic potential to have a child with [condition] when making other fertility, you know, family-based decisions... The decision whether to have a second child once you've already got a child with [condition] is incredibly difficult.” - 4</p> <p>“Now it's, when screening was introduced nationally there was a big debate as to whether it was a good thing or a bad thing to identify carriers and quite a lot of the clinicians believed it was a good thing to identify carriers because it gave people reproductive choice.” - 4</p> <p>“And we don't believe anyone should kind of have a preference to the way others decide or the choices others make. I mean we should just empower as many people as possible to, to make those decisions, to make the decisions they want to make or need to make as individuals and we know very well that people react very differently to [condition when it's] presented to their families.” - 10</p>

Benefit Examples	Supporting Quote(s)
	<p>“Now a crucial benefit for all of those conditions - that we believe is one that could be considered more widely - is that if you find out that your, an early child in your family, or not the last child in your family is affected by a genetic condition, then you can choose in the future to conceive a child through PIGD or with prenatal testing, and try to avoid a future child being affected by that condition. And that is a tremendous benefit. You know a lot of families would say they can cope with one child affected by a rare genetic condition, but would be very difficult to, to cope with more than one although many do. So that reproductive choice element is, is really important.” - 10</p>
<p>Screening may provide reassurance</p>	<p>“For some women it is you know they, they’re utterly grateful and relieved to know that you know they can have screening and you know once they’ve got you know positive results then they can relax. Then they’re going to be okay.” - 6</p>
<p>Screening is free at point of service</p>	<p>“I think where the current system works particularly well is where you find people with, you know, rare or very serious health conditions and then the NHS generally somewhere has one of the world’s leading experts who is available to help people free of charge and I think that is the, the most amazing thing about our NHS.” - 2</p> <p>“The availability of care is very uniform... [one of the things we have] done very successfully with the clinical community is to write standards of care, service specifications, consensus documents, guidelines, patient information leaflets... a really good job at sharing information through consensus documents... and the implementation of those guidelines and outcomes of treatment.” - 4</p>

Harms of antenatal and newborn screening programmes as told by stakeholders

Harm Examples	Supporting Quote(s)
<p>Screening may not be implemented presently in the UK</p>	<p>“[Condition] could have, should have been prevented if only they’d been born in France or Germany or America or Singapore. The fact that they’re born here means they don’t get an opportunity to be told about [condition] and they don’t get an opportunity to be tested [for condition].” - 8</p> <p>“I mean at the moment I think the harm from our community’s viewpoint is just where we’re not screening and where the value would lie... So there’s all these avoidable harms happening and we believe there’s a, the way that we’re making decisions in the UK around newborn screening is unfair to people living with [certain] conditions.” - 10</p> <p>“The only answer to how do we change the future prognosis for children with this condition is early intervention. And that early intervention is only going to happen with a timely diagnosis, and sadly most of these children are just diagnosed far too late because by the time something has shown itself, you know, they’ve failed to meet a major milestone... by which point it’s too late, the treatment isn’t going to help them... So, it was just very, very clear to me at that point that newborn screening would be the answer to change the future prognosis for children diagnosed.” - 11</p>
<p>Screening may provide unexpected news</p>	<p>“I think it’s really difficult to really giving difficult news to, to people who are no-where near the position of, of being able to hear that, if that make sense.” - 1</p> <p>“Essentially that person is knocking on the door and introducing themselves to a family who think they’ve got a normal child. So, raising the possibility that that child is affected by a life-long, you know, potentially a lethal condition through a screening test is potentially really, really difficult conversation to have...And particularly bearing in mind the test has its limitations in terms of</p>

Harm Examples	Supporting Quote(s)
	<p>its specificity so there needs to be a really good infrastructure for making sure that that's done well." - 4</p> <p>"There are no, you know that there are no easy decisions, whatever decision, you know to continue with the pregnancy was going to be a difficult decision, clearly to end the pregnancy was going to be a difficult decision. Sometimes I think, if that isn't explained at the beginning of the antenatal screening process, you know having a, having a test then means you're going to get a result and you might assume that the result is just going to give you confirmation that it's everything is, you know, you have a low chance result and then we move onto the next stage. Well, what if the result comes back and actually it isn't what you were expecting?" - 5</p> <p>"While we think it's, it's absolutely crucial that there's good pre-test information and counselling, the reality is that there isn't a lot of time for that in the first trimester, and we would argue from experience and I think research bares it out but psychologically in the first trimester of pregnancy it's very difficult for women to make that imaginative leap to imagine a situation where they get a result that leads to further decisions, or the possibility of things being different. For many women they will opt into screening hoping to be reassured and not really giving it a lot of consideration I think psychologically for many women it's just impossible. It's just impossible for them to tolerate the possibility of things being different, so they just plough on hoping to be reassured, most will. And obviously it does bring them up short when they get the result. And I don't think that we'll ever really get the better of that. We, again that's not us saying you don't try your best with pre-test information, but an awful lot of women, they may have tentative conversations about it, but they really genuinely don't think it's going to happen to them." - 7</p>

Harm Examples	Supporting Quote(s)
<p>Screening may lead to devastating emotional impacts</p>	<p>“It’s absolutely devastating. I think it’s very difficult to explain, explain to people just how devastated parents are when they are faced with this sort of information and, and it leads to the loss of their baby, it completely destroys people’s lives, not only in the short term but also in the very long term. It, it I mean it is astonishing that parents get through it but they do and come to terms with it and they, you know, they do make decisions and they do move on with their lives but it, it’s, as I say, it just devastates people [um] and that requires, when you have to make a choice or when you are, you feel you’re forced into making a choice about whether your baby lives or dies that is a choice you have to live with for the rest of your life and all of the questions and things that surround that and that is, yeah, that is one of the hardest things people I think would ever be faced with.” - 2</p> <p>“But no-one, no-one signs up for the death of a baby and it feels, it feels sort of, it feels that something that shouldn’t happen but of course it does but no-one checks up on people to check whether they are experiencing normal symptoms of grief or if whether they really are suffering a mental illness.” - 2</p> <p>“That’s what I see with, with so many of the families that we work with, they feel almost betrayed sometimes when, when a baby dies. It’s like how, I was not prepared for this, nobody told me that was a possibility.” - 6</p> <p>“This is the biggest, the biggest problem. I think and what we’re working with particularly in, in this specific area around you know testing and decision making, is that women will blame themselves. They fall into a pit of guilt and shame.” - 6</p> <p>“Yeah, if they’ve had a traumatic experience suddenly when they’ve been told something they weren’t expecting, suddenly that environment goes from</p>

Harm Examples	Supporting Quote(s)
	<p>being this exciting environment to go and sit in and wait to see the picture of your baby, to suddenly being a terrifying environment cos this might be the place where they tell us again bad news, you know. And parents remember every detail of that. They will know what room they went in. They will know you know where the sonographer was sitting. They will know the way the sonographer spoke, you know that's the first thing that people say to me, it's like, "Oh God it was the same room," "Oh my God it was the same woman," you know, they know." - 6</p> <p>"I wanted to get across about the parental guilt because that is, and the mental health impact both on the family and on the siblings because I think that is often missed." - 8</p>
<p>People may not understand what screening tests are for</p>	<p>"Well, well of course, of course most, most people don't really realise what these screening tests are even for. So at, you know, the twenty week which is the baby anomaly scan, I mean most people come out of that thing with a picture of their baby and that's pretty much what they think it's all for really. There is very, I don't think there is a huge amount of understanding about what happens at those screening tests to be quite honest." - 2</p> <p>"People contact us because they're then having to, and you know even just interpreting the results and understanding you know the whole false positives and you know the accuracy of the tests, and a low chance doesn't mean no chance... people you know are very confused by the information that they're given... When you are talking about false positives and positive predictive values and you know all those sorts of things, is very, very difficult to understand." - 5</p> <p>"Some people are especially upset that they had low chance, cos they do really, well cos they took that as no chance." - 5</p>

Harm Examples	Supporting Quote(s)
	<p data-bbox="454 315 1374 958">“I personally feel that you know that there is a responsibility that is being missed here, from health providers, which is to inform parents what this screening is actually about. What they’re going through this process for. Because no-one’s doing that, and, and you see time and time again couples come in for their screening and they, they think they’re coming to find out the sex of their baby. Or, or they think they’re just coming to see, to get a nice scan picture and take it home to show their family. You know they have no awareness of actually the, you know the importance and the significance of, of that procedure. And I think that’s wrong. I think that’s wrong and I think that is why parents experience so much [um] trauma when suddenly they are given some devastating news.” - 6</p> <p data-bbox="454 1048 1374 1263">“Obviously if you go to rural parts of the country they’ve probably never heard of [condition] but what I can say is awareness has really been improved over the last forty years... But they need to know because there’s a lot of stigma around [condition].” - 9</p> <p data-bbox="454 1352 1374 1509">“I’m just a bit concerned that do people really know what their screening for. Do they have an idea in their heads of why this is a good thing and what they might do as a result?” - 12</p>
It is difficult to achieve fully informed consent	<p data-bbox="454 1541 1374 1935">“Many, many times of the year I’ve met, you know, consultants and things, ‘Oh no we don’t tell parents about that.’ Why don’t, why don’t you tell parents about that?’ ‘Oh, because we don’t want them to worry.’ It’s a very difficult thing I think to reconcile. Do you give people the information whether they want it or not? Do you run the risk of creating terrible anxiety in someone who has a completely normal pregnancy? I think these are very, very difficult choices to be, to be made.” - 2</p>

Harm Examples	Supporting Quote(s)
	<p data-bbox="454 259 1380 779">“And then I suppose the other thing, just from having trained lots and lots of midwives and antenatal screening, it’s a really big challenge for them that, with the amount of information and tasks that they have to perform in those initial booking appointments, you know they have probably seventeen or eighteen different things that they have to introduce in a 45 minute, 50 minute appointment.. the intention is that they would have a proper discussion about screening and you know various studies have shown that the average length of time that they spend talking about screening is a few minutes. And that probably isn’t sufficient.” - 5</p> <p data-bbox="454 869 1380 1205">“But we’re never going to get, we’re never going to have every single woman, pregnant woman making a fully informed personal decision around the screening, it’s just feasible. We have to do the best we can to get there of course, of course we do our utmost but the reality is that for a lot of women they do not want to think about it, and you can offer women information but you can’t force them to engage with it.” - 7</p> <p data-bbox="454 1294 1380 1512">“I would worry that women have made decisions because of societal pressure, subtle or otherwise, and I think it’s, it has led to stigma for people with [condition] as well. And you know we know that a lot of money is spent on screening.” - 12</p>
Screening may imply pressure to have a termination	<p data-bbox="454 1547 1380 1937">“We’re going to doctors and we expect doctors to give us medical information and perhaps we’re unrealistic to expect them to also be able to tell us any more than that which is why, you know, they have to collaborate with organisations such as ourselves... women are making life and death decisions, the biggest decision I think a woman can make is to terminate her baby and they’re doing so too often I think without adequate information and support.” - 3</p>

Harm Examples	Supporting Quote(s)
	<p data-bbox="454 259 1380 651">“I’m a feminist and I believe a woman has the right to terminate any pregnancy obviously up to twenty-four weeks, but it’s, you know, it’s not for a doctor, there are no, there’s no medical reason just because a baby has [condition] there is no medical reason for a medical professional to suggest the woman terminates her baby. The only reason is because there’s a genetic difference and the bottom line is actually eugenics and people don’t like it but that’s the fact.” - 3</p> <p data-bbox="454 745 1380 1021">“Women, of course, should be supported both ways [to terminate or continue a pregnancy]. Both options should be equally supported, and there’s still work to be done in better pathways for women continuing their pregnancies, particularly after a diagnosis of [condition], that shouldn’t happen to the detriment of women who choose to end the pregnancy.” - 7</p> <p data-bbox="454 1115 1380 1205">“We’re talking about wanted pregnancies here, so again there’s that sort of question of, you know, which lives matter?” - 12</p> <p data-bbox="454 1299 1380 1693">“It’s the pressure as well for parents. So, women tell doctors, ‘Don’t talk to me about abortion, not considering it. I won’t consider it,’ but the doctor or midwives keep asking and you, you grow up respecting people like doctors and nurses because they, well they’re the people who know about your health and so then if somebody like that is constantly saying to you, “Do you want to abort?” it’s, it immediately makes you think it’s something really bad.” - 14</p> <p data-bbox="454 1787 1380 1877">“I am a bit worried that there’ll be less and less people with [condition]. Their lives are as valuable as anyone else’s.” - 15</p>
Communication about screened-for	<p data-bbox="454 1912 1380 2002">“Flippant but they talk about anomalies, abnormality, disorder, problem, you know, and all of that has negative connotations and, you know, is prejudicial</p>

Harm Examples	Supporting Quote(s)
<p>conditions may not be handled well</p>	<p>and it influences a person's perspective, you know, their view. So, you know, as a complete lay person, I entrust my medical practitioners to provide me with information and support that I need and, you know, if they're telling me that this is abnormal and, you know, disorder and all those words, it, it completely influences me as to what I might as what I think." - 3</p> <p>"Where information's coming that one wasn't expecting, it's coming at the wrong time, it's delivered in, in a paternalistic way, one can't find the information one needs to help make a decision. Maybe one googles the name of a condition that you know you just found that your pregnancy is at risk from, and get some information that isn't framed for you at that point in your pathway." - 10</p> <p>"It's just these sort of traumas during pregnancy that women sort of go through and the trauma at birth when you've just given birth, you know your whole life's changed, and the sense, the lack of sensitivity in providing that news to people." - 12</p>
<p>Screening may identify carriers</p>	<p>"Now it's, when screening was introduced nationally there was a big debate as to whether it was a good thing or a bad thing to identify carriers... the geneticists as a general rule were very clear that identifying carriers was a bad thing because you were providing genetic information for somebody who hadn't consented to have that genetic information and also there was the concern that growing up knowing you were a carrier may influence your choices and your, you know, your lifestyle, influence your wellbeing in some, some unknown way, some unknown way. So, there was a feeling generally that it was probably best to minimise the number of carriers identified." - 4</p>
<p>It may be difficult for HCPs to retain specialist</p>	<p>"So it's quite hard I think for people to develop and retain all that sort of information if you're not seeing these, for health professionals, if you're not seeing cases on a sort of regular basis." - 1</p>

Harm Examples	Supporting Quote(s)
knowledge about conditions	

Challenge of information provision as discussed by stakeholders

Theme	Supporting Quote(s)
Variable information needs	<p data-bbox="451 320 1380 835">“I quite understand that, it has to be quite general and quite superficial at that [initial] stage so I think the thing that needs to be worked on is the sort of next stage information so if you get a higher chance result. Then having the right level of information available to people at that stage I think probably would be, would be an area that still needs quite a lot of work... you know, you are starting with different syndromes that the vast majority of people have never heard of. The sort of information that’s available is, is quite limited and people do Google searches and come up with all sorts of things, so I think that’s an area that requires further, sort of further development and support.” - 1</p> <p data-bbox="451 925 1380 1753">“You’re really attempting to do two things. You’re attempting to make sure the information is absolutely correct and up to date but then you also want to make it accessible and so it’s very, it’s quite easy to provide information that’s accurate but it’s much more difficult to provide information that is accessible to the people who probably need it most... So the NHS has always insisted on providing its own information and it does though clearly extremely well but it’s not always the most user-friendly or accessible of information. And they have, I think, failed to grasp the opportunity that they have to work with partners and because working with partners would essentially mean, you know, choosing one partner sometimes over another and there have been really unwilling I think to do that. So, they will link to other people’s information but they, the NHS tries to provide information on absolutely everything and it can’t be good at absolutely everything and they’re not good at absolutely everything.” - 2</p> <p data-bbox="451 1843 1380 2000">“Again, that would range from you know, very little almost shock, just wanting to know the very basics because the vast majority of families actually hadn’t, either knew very little about [condition] or knew nothing at all and had no</p>

Theme	Supporting Quote(s)
	<p data-bbox="454 259 1375 595">family members at all with [condition] and that was always a source of a bit of confusion because on the one hand you were telling them it was a genetic condition but on the other hand they were thinking, 'Well nobody in our family's ever had it, so how can it be a genetic condition?' But some families would want to know everything, you know, with questions like, 'How long will he live? or 'Will he able to go to school?'. " - 4</p> <p data-bbox="454 685 1362 1021">"I think it's really helpful to be guided by the family's questions and I think it's really important to be available for follow up questions very quickly. So, in other words, I think families often need to go away and to digest what has been said, talk between the family and then come back and ask questions once they've had an opportunity to discuss it but very quickly, you know, within, within either hours or days not weeks." - 4</p> <p data-bbox="454 1111 1375 1514">"It depends, you know because people will contact us at various stages. They may be right at the very, you know they may be, they may be thinking about getting pregnant, they might be right at the very beginning having only just found out and going for their initial booking appointment and the looking for information about [condition] generally, or they could have had a high chance result and a, either in the process of making a decision, or have made their decision [and want to prepare]." - 5</p> <p data-bbox="454 1603 1362 2007">"We would like to think that no woman feels that she, by declining a test, or saying I don't want to have a test, that they're opting out, they just may expressing a, a decision or a choice that they don't wish to have a test... It's a programme that will give you information about your developing baby that then you may want to have, you might not want to have, you may want to progress on from having a predictive test or diagnostic test, you might not want to know, you might want to know because you're preparing for the birth</p>

Theme	Supporting Quote(s)
	<p>of your baby, you know it's, it's very personal. And that is where hopefully it would be an advised process where it's present, you know this is the information that's available, you can now make a decision as to what, if any, of those tests you wish to have." - 5</p> <p>"I mean I think our focus has always been on getting the information really up to date and accurate...people, if they have accurate information will make a range of different choices... And so that's really been our focus over the last well throughout the period of time that we've been working in this arena, but particularly over the last five years or so, has been absolutely on making sure that information is accurate and it's up to date, and it's not outdated, and that choice of words are as we would want them to be, as much as possible." - 5</p> <p>"But we have to pragmatic and realistic, for so many women in the first trimester of pregnancy, when their hormones are raging, when they're coping with being pregnant, and thinking about what it means to them, their ability to think through the potential consequences of all the screening they'll have is just not possible. And particularly when we think about scans, if, what we, what, if you wanted a woman to be fully informed you'd have to sit her down and go through a list of all the potential things they're looking for, from anencephaly through to skeletal dysplasia's, and you're going to make it a white-knuckle ride for every single woman. When in reality it's a small percentage that will actually be confronted with that." - 7</p>

Themes from sense check focus group with midwives

Theme	Supporting Quote(s)
<p>Knowing screening can be emotionally challenging</p>	<p>“I’m also counselling women and then obviously sometimes telling them difficult news. Devastating news sometimes. Thankfully, not very often, but it does happen. So lots and lots and lots of difficult communication in the antenatal clinic think” - Midwife 1</p> <p>“Making sure obviously that they know your contact details, because often it you know it's just too much of a shock and they just need to go away, process it, and then maybe they want to come back or they want to call you later or the next day.” - Midwife 2</p> <p>“So I'm thinking about a woman particularly who had already, I think four or five children, and then a baby with a very significant heart defect. And the pregnancy wasn't planned. So there was a lot of complicated feelings around the pregnancy to begin with. And then we were looking at a baby that was going to have significant long term needs when she already had four or five children at home. And there, you know, I think about her a lot.” - Midwife 3</p> <p>“Healthcare professionals... are trying to do the best for women, so they might book them a series of what they would refer to as reassurance scans. But like I said, they're only reassuring at that moment in time and sometimes by arranging these reassurance scans you're almost telling the woman that she needs them, that there's a there's a chance that something could go wrong. So actually I would argue that sometimes reassurance scans can increase anxiety rather than reduce it.” - Midwife 3</p> <p>“That's probably the hardest part of the job. I think it's hard to forget those people. The ones that are most difficult. It's so emotive and you never forgot those people because their lives are changed forever.” -Midwife 4</p>

Theme	Supporting Quote(s)
<p>Acknowledging the difficulty of providing information</p>	<p>“I think most people are more likely to look at something they've got physically. But I always recommend them to have a read through that because we know that, the vast amount of information we're giving them at that appointment. And so hopefully they'll take time to read it, and perhaps discuss it with a partner.” - Midwife 2</p> <p>“I think that's so tricky because it's kind of like giving people the information, but obviously ensuring that you know is there pointing to giving that information. But unfortunately a lot of people will go and Google these things anyway, and actually sometimes it's better for them to hear the facts rationally from a health care professional like. Yeah, the information isn't nice, but it's actually better for it to be put into context by a healthcare professional rather than them going and Googling all these possible things that might happen. And some of them are actually probably quite unlikely, and they you know, may fixate on them and things like that. So I think it just depends how it's delivered really.” - Midwife 2</p> <p>“I don't think that's because the community midwives haven't discussed it with them. I think it is because there's such a vast amount of information at that first appointment that suddenly women feel, you know, a bit bombarded by it and they often they come for the scan and they don't know that they've agreed to this, so they're not quite sure when they get there, what they've agreed to.” - Midwife 3</p> <p>“Sometimes you wish you could just turn off Google for pregnant women... I always say that such a big part of my job is talking to women, like trying to talk them down off the ledge because if they know there's even the slightest possibility of something going wrong their terrified and... there's a million things that I could be, and you'd spend your whole life trying to explain those</p>

Theme	Supporting Quote(s)
	things.” - Midwife 4
Managing the right to informed choice	<p>“Do you want more information? Do you know? Do you want me to tell you what they are? Because by this point most people's eyes have glazed over... I've worked with colleagues in the past, who have gone through each of the conditions. What it means, how it affects the baby, what the treatment is, the incidence. I guess you could argue that that would be more fully informed consent or choice. But is it necessary and does it add anything to it? I don't know.” - Midwife 1</p> <p>“I mean, it's like where do you start and where do you stop?... I think it's difficult because there are so many things I don't think would be able to inform people. And as you were saying you don't wanna entirely scaremonger people. You wanna give them enough information so they're aware there is a potential something could be diagnosed, but you don't want all these women to have chronic anxiety for their entire pregnancy, so I think it's getting the balance.” - Midwife 2</p> <p>“Yes, you don't want to cause women a lot of anxiety, but we're talking about informed consent here. So a woman should know what she's consenting to.” - Midwife 3</p> <p>“Typical postnatal mothers, they've got like a million things to think about. They're probably not really thinking too much about [the heel prick]. You know, it's just something that was out of their heads and you know if the result comes back [positive] they're probably going to be very shocked by that because it's just one of those things that have gone out of their heads. But I don't know if that's anything really that we can change.” - Midwife 4</p>
Seeing the pressure to have	“I don't have very many people decline the neonatal screening but once in a blue moon. But again, it's just okay, if that's the case, you need to understand

Theme	Supporting Quote(s)
screening tests	<p>why, and these are the things that you need to look for, so it's kind of taking the responsibility of saying, okay, you are an autonomous person and you have the right to make this choice. And it's my responsibility to make sure that you understand the implications and what to do if something happens. So then I can go away feeling like, okay, I've done the best I can for them." - Midwife 1</p> <p>"I have had women that have declined, say for example a blood test or the anomaly scan, that have then been constantly asked throughout their pregnancy. And when they've gone to consultant appointments, you know, being told they should have done the 20 week scan...and they are pressured quite a lot." - Midwife 2</p> <p>"I think sometimes women when you tell them the worst case scenario, they can perceive that as a as a pressure or as a scare tactic when it's not. It's literally just trying to explain to them what could actually happen." - Midwife 3</p>