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Measuring Patient- and Carer-Reported Outcomes Following Genome Sequencing for Rare Disease Diagnosis: A Psychometric Assessment of Outcome Measurement Instruments

Sally Sansom
sally.sansom@dph.ox.ac.uk

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ISPOR Poster Presentation Handout

PLEASE NOTE

These slides contain preliminary results only and are not for distribution

About me

PhD student within the Health Economics Research Centre (HERC) and Junior Research Fellow within the Centre for Personalised Medicine at the University of Oxford

Bachelor of Biomedicine (genetics major, University of Melbourne) and Master of Public Health (health economics specialism, Monash University)

Worked for 8-years pre-PhD across health economics and strategy consulting, and medical research (including clinical trial) project management

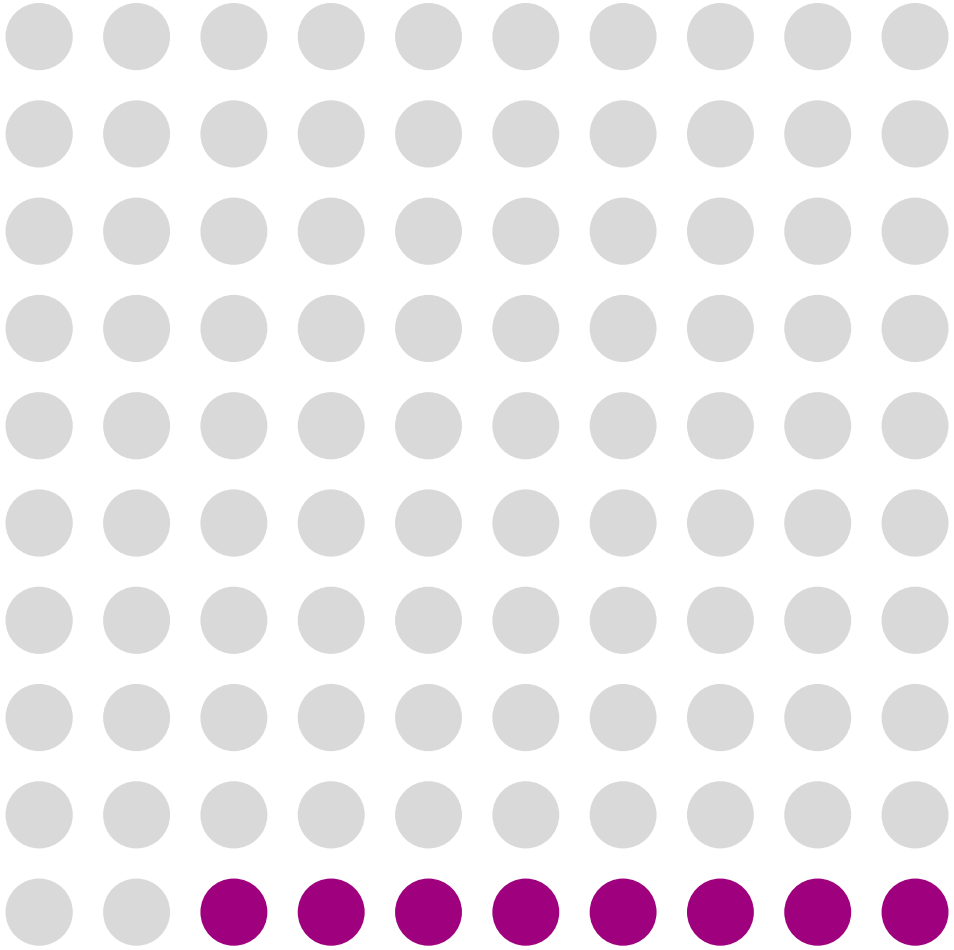
Professional and academic path has been deeply shaped by having a younger sister who has Angelman syndrome (rare, genetic, neuro-developmental disorder)

Background: What are rare diseases?

~80% have a genetic cause

Diagnosis can take several years

~50% remain undiagnosed

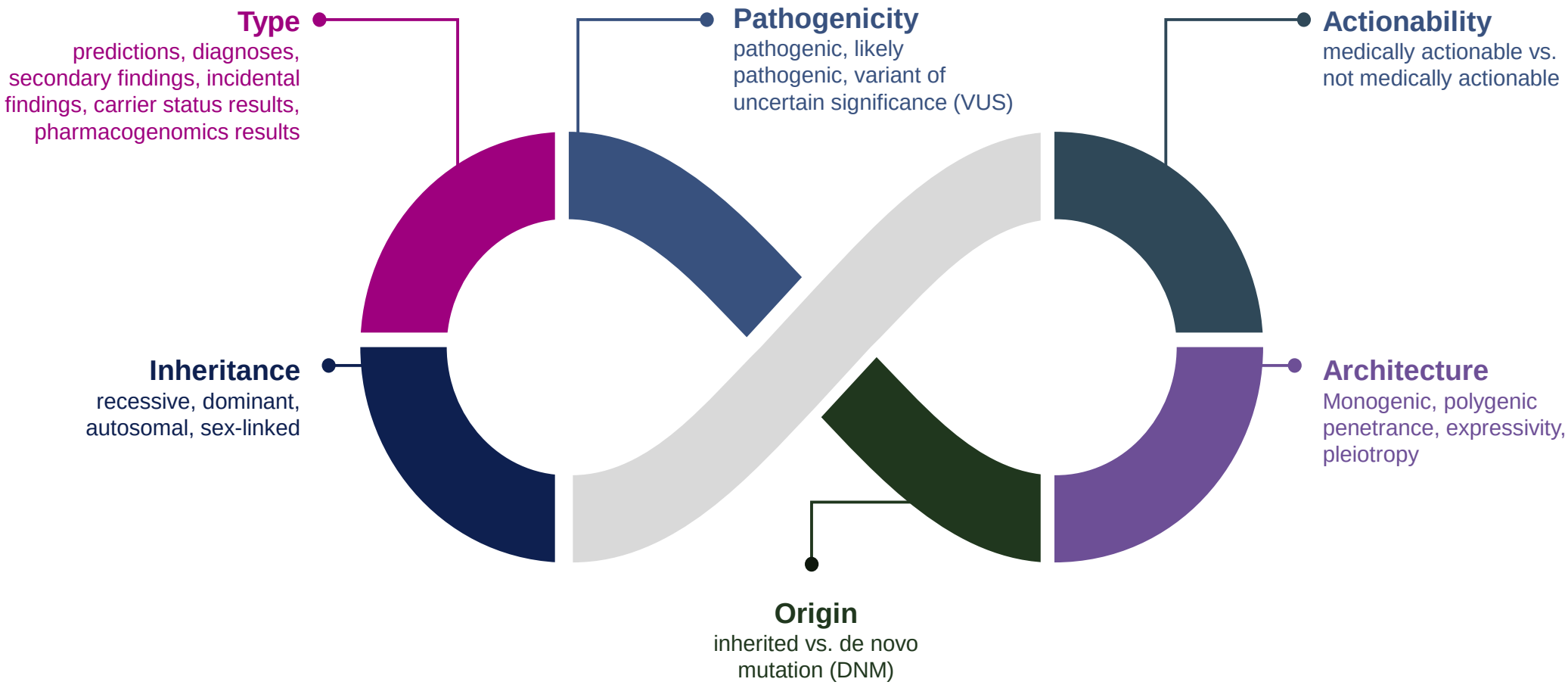


~95% have no approved treatment

50-75% begin in childhood

~30% of children die before their 5th birthday

Background: What is genome sequencing?



Background: What do we know about the outcomes from genome sequencing?

Clinical

diagnostic remarkability, appropriateness of follow-up care, informed clinical management, monitoring for early disease detection, referral to clinical trial

Emotional

adverse responses, positive responses

Cognitive

value of knowing information, perceived health risk, satisfaction of curiosity, self-knowledge

Behavioural

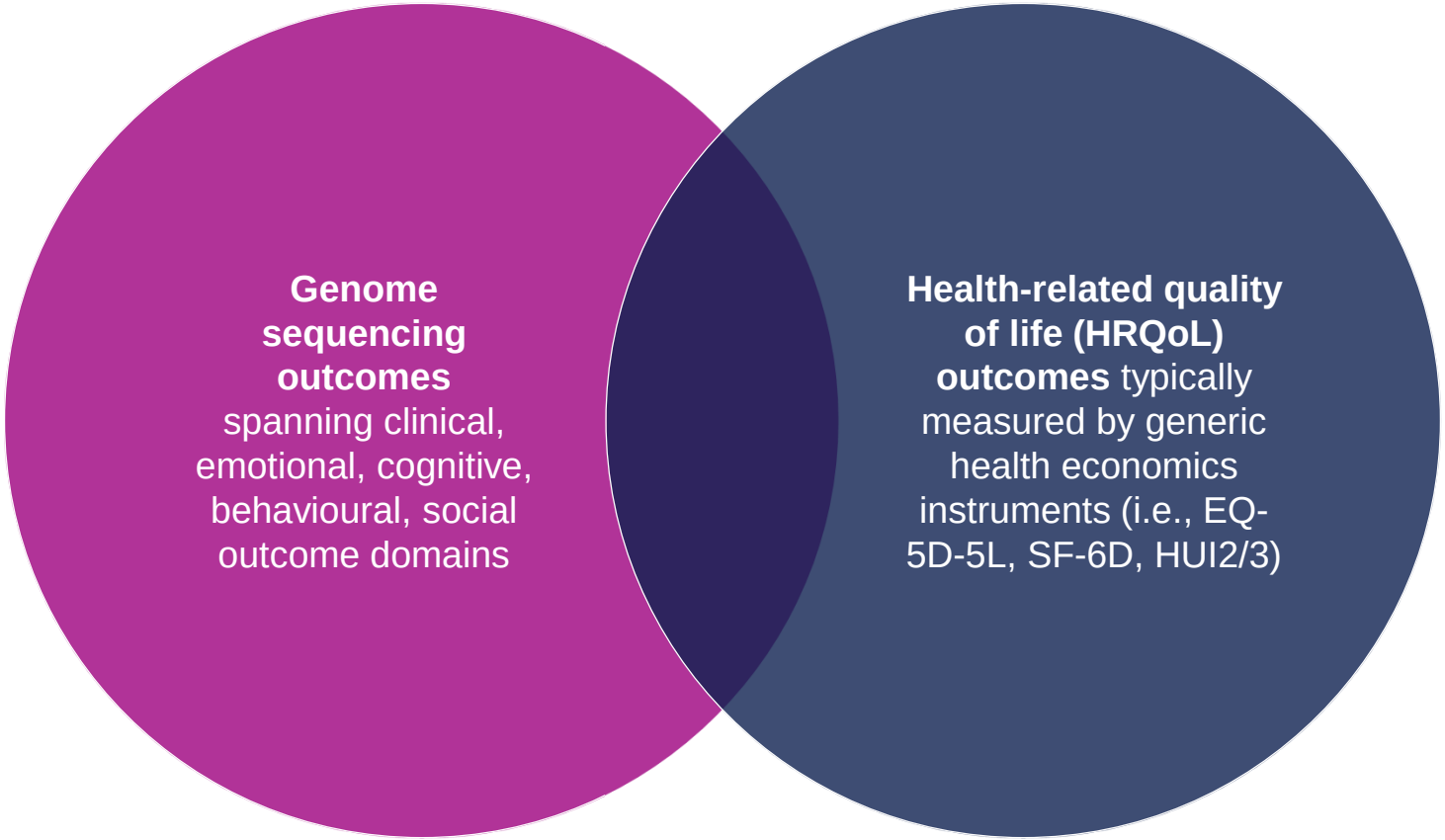
insurance coverage, health habits, information seeking, future planning, parenting decisions, reproductive decision-making, communication with family

Social

advocacy activities, blame, access to support services, degree of social support, discrimination, privacy, social stigma, quality of relationship with care providers, helping others

Primary source: Smith et al., (2021). Supplementary sources: Kohler et al., (2017a;b); Hayeems et al., (2021); Duenas et al., (2023).

Background: What challenges does this pose for health economists?



Background: What are the specific concerns in this context?

"Over the past decade, health economists have repeatedly questioned whether metrics such as the QALY, which focuses on clinical utility, can fully quantify the outcomes that are important to patients when they undergo genomic testing"
- Buchanan & Wordsworth (2019)

"There is widespread recognition that the benefits and harms of genomic medicine extend beyond health-related quality of life and thus are not captured in the conventional quality-adjusted life year (QALY) metric"
- Smith et al., (2025)

Content validity	Construct validity	Responsiveness
The degree to which the content of an instrument is an adequate reflection of the construct to be measured. Includes consideration of an instrument's relevance, comprehensiveness, and comprehensibility.	The degree to which the scores of an instrument are consistent with hypotheses (i.e., with regard to internal relationships, relationships to scores of other instruments, or differences between relevant groups).	The ability of an instrument to detect change over time in the construct to be measured. Also referred to as "longitudinal validity" or "the validity of a change score"

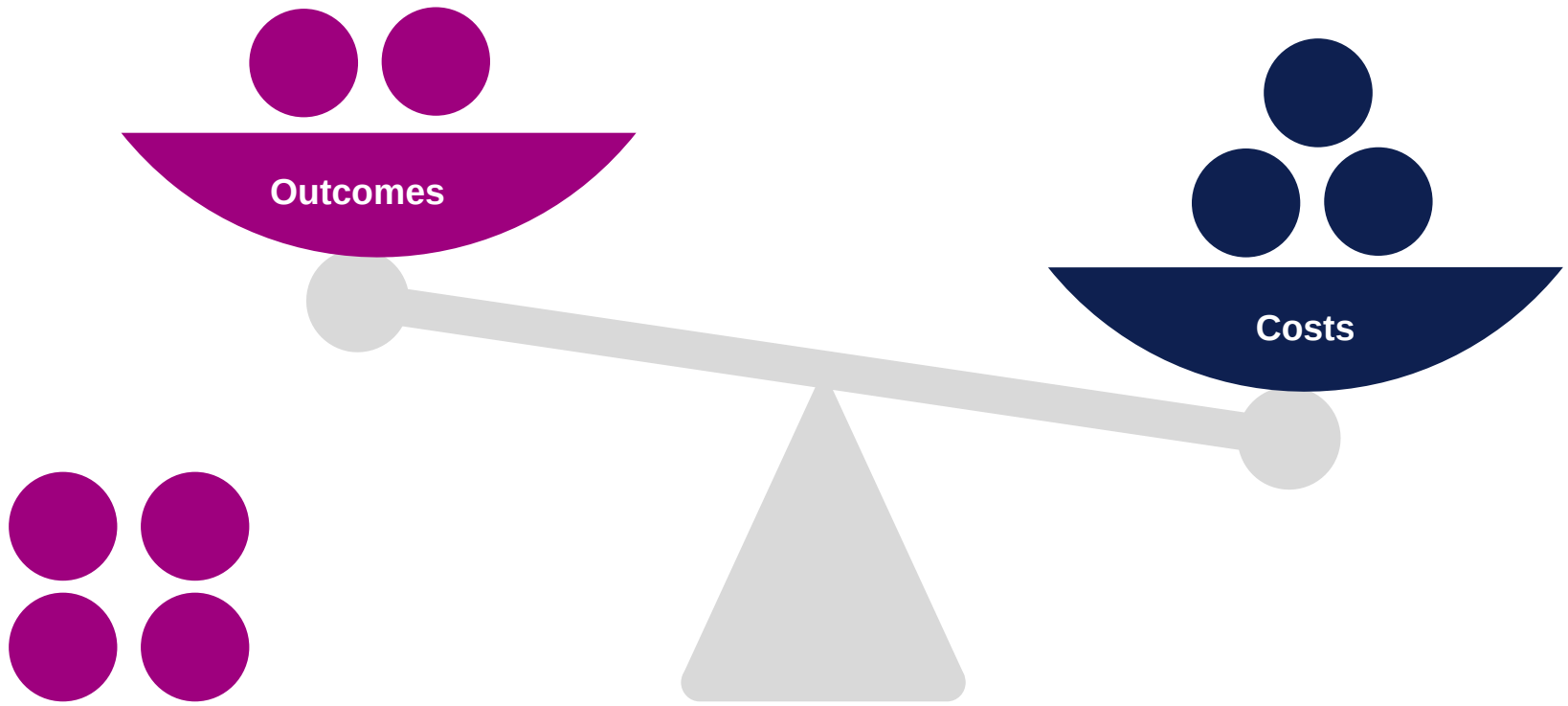
"The tools used to estimate utilities incorporated in the QALY may not be sufficiently sensitive to capture all aspects valued by patients and their families"
- Bouttell et al., (2022)

"Genomic information may have utility regardless of whether it leads to downstream improvements in health and thus make its value difficult to measure in a QALY framework"
- Smith et al., (2025)

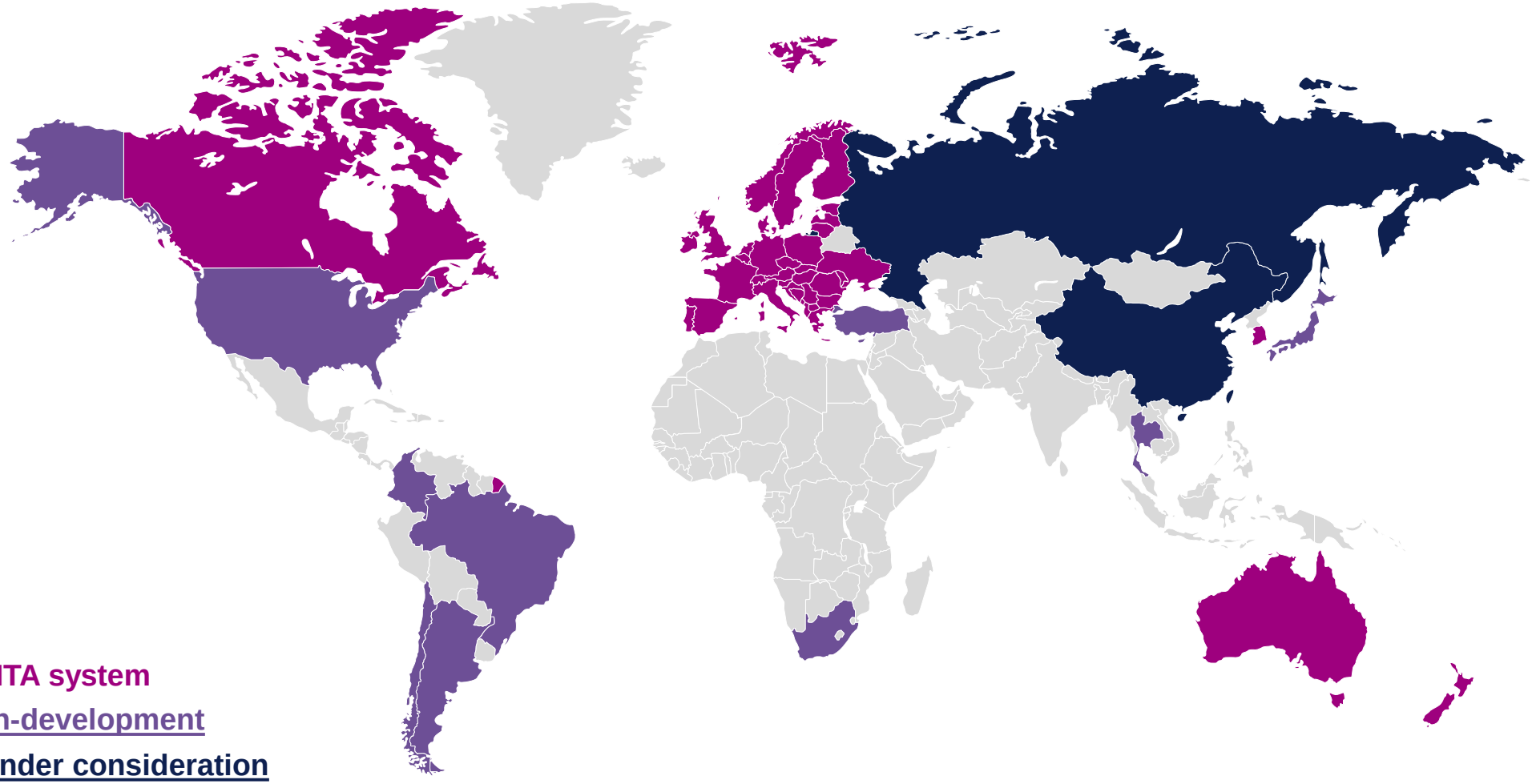
"Given the so-far questionable performance of conventional health economics outcome measures for capturing important value components of GS information to patients and families..."
- Goranitis et al., (2020)

Source: Mokkink et al., (2010).

Background: What implications does this have for decision makers?



Background: What is the policy and decision-making context?



Established HTA system

HTA system in-development

HTA system under consideration

Source: HTAi, INAHTA.

Aim

My PhD is improving our understanding of **how to measure** the impact of genome sequencing for rare disease diagnosis in economic evaluations

Methods

Phase 1 Systematic literature review

Phase 2 Critical appraisal

Phase 3 Cohort study

Phase 4 Recommendations workshop

Note: ISPOR poster PCR42 only presents the methods and results for the first 3 phases.

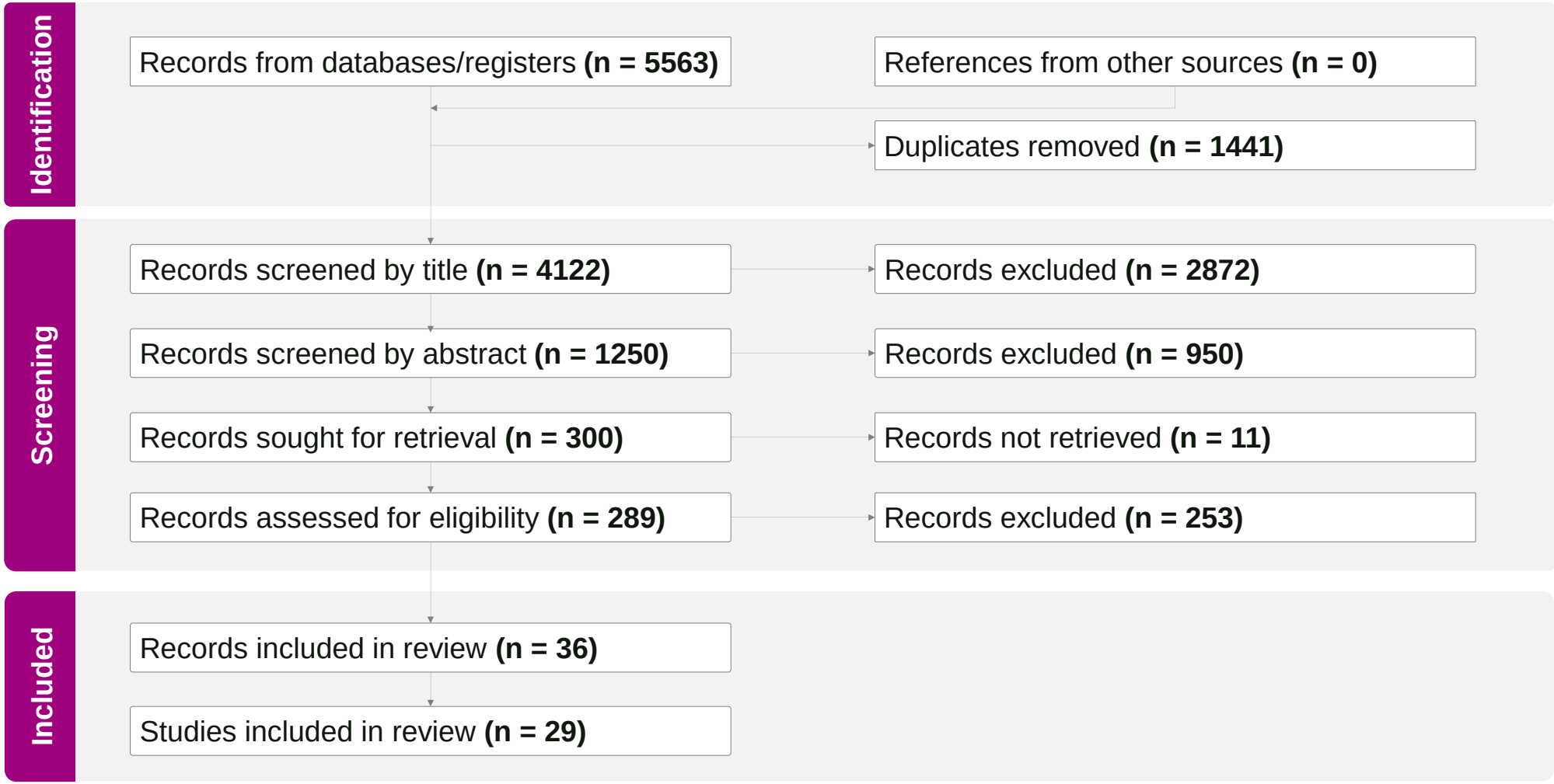
Phase 1: Systematic review

Method: Systematic literature review

Component	Eligibility
Population	• Children and adults undergoing genome sequencing • Unpaid carers (including parents, guardians, and other informal/unpaid carers) • Other family members
Intervention	• Genome sequencing
Control or comparator	• Not required
Outcomes	• Clinical, emotional, behavioural, cognitive and social outcomes
Measurement	• Survey-based • Scorable • Administered at least once post-test • Including unvalidated instruments • Excluding direct preference-based utility instruments/methods
Study design and reporting	• Studies which had used, developed or validated instruments in this context • Peer-reviewed primary research publications

Note: 1) The search strategy was informed by the COnsensus-based Standards for the selection of health Measurement Instruments (COSMIN) “guideline for systematic reviews of Patient-Reported Outcome Measures”, 2) The review was registered in the Prospective Register of Systematic Reviews (PROSPERO) international prospective register of systematic reviews prior to commencement (reference: CRD42023405739).

Results: Systematic literature review

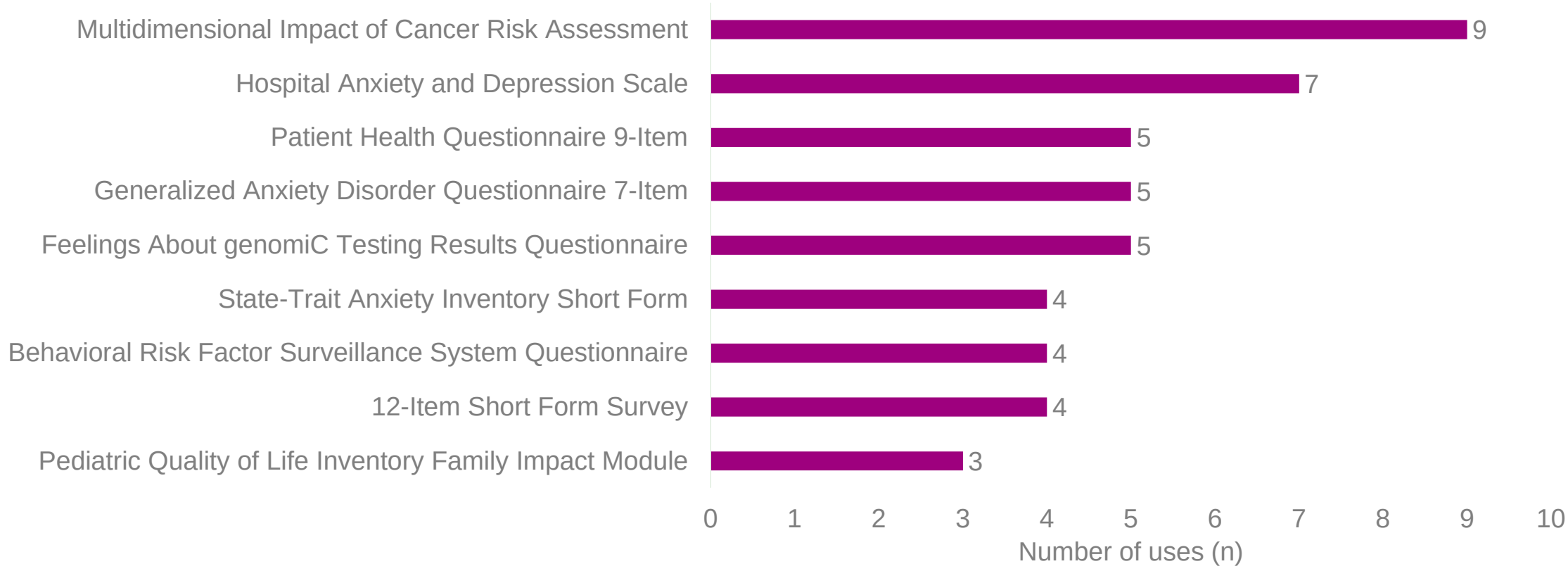


Results: Systematic literature review

Characteristic	By instrument	By study
Total instruments used		
Total	209 instruments (75 validated)	-
Included	63 instruments (49 validated)	-
Excluded	146 instruments (26 validated)	-
Total times used		
Total	278 times	-
Included	106 times	-
Excluded	172 times	-
Range of use		
Total	1-13 per instrument	2-26 instruments per study
Included	1-9 per instrument	1-12 instruments per study
Excluded	1-13 per instrument	1-22 instruments per study
Mean use		
Total	1.3 times per instrument	7.2 instruments per study
Included	1.7 times per instrument	2.2 instruments per study
Excluded	1.2 times per instrument	5.0 instruments per study

Results: Systematic literature review

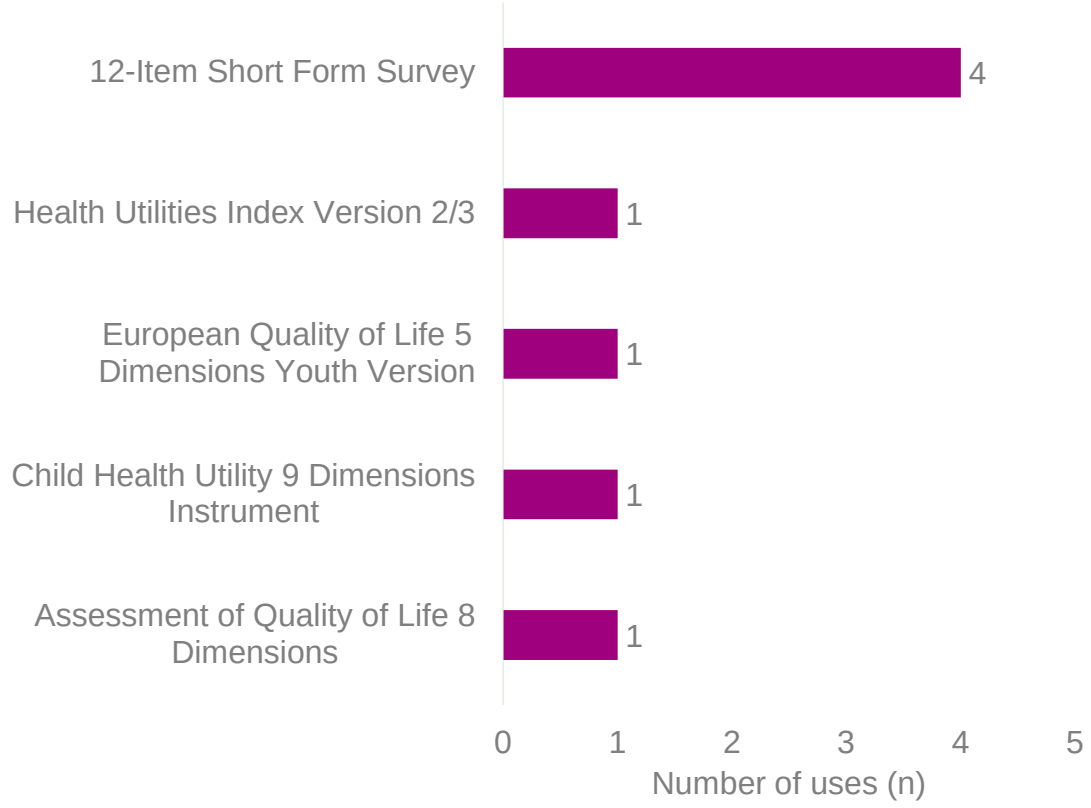
Most commonly used included instruments



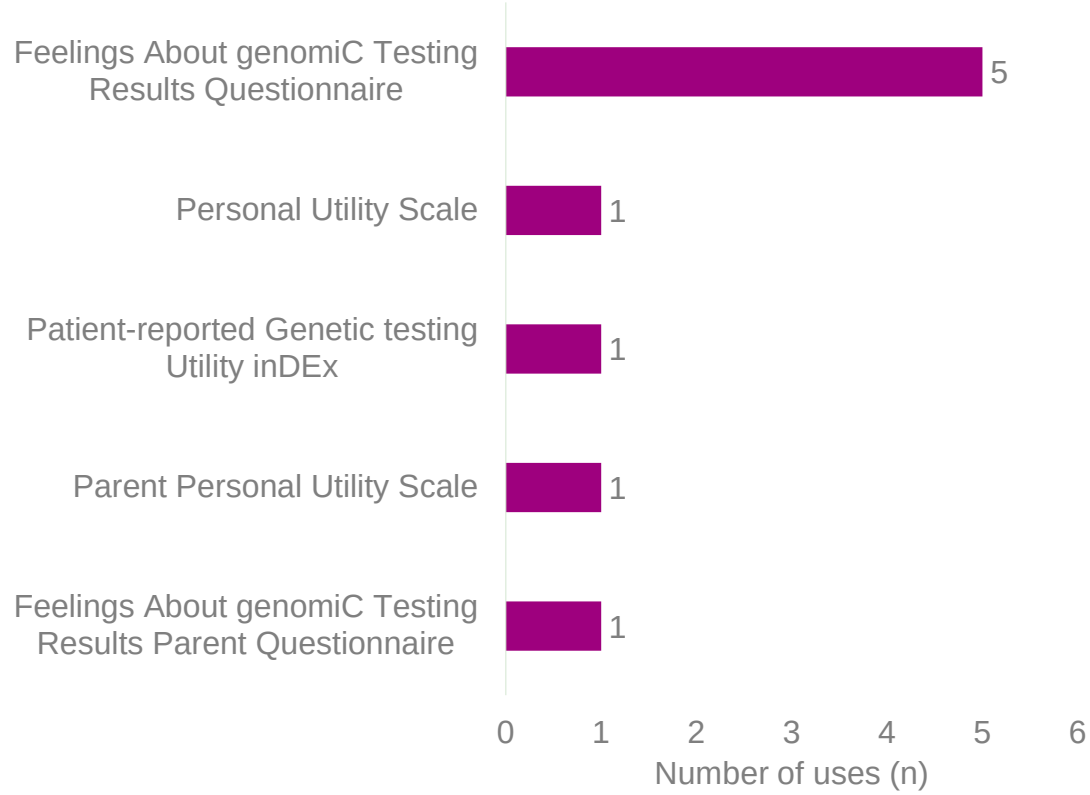
Note: Most frequently used is defined as when n > 2.

Results: Systematic literature review

Generic health economics instruments



Genome sequencing specific instruments



Note: 1) Validated instruments only, 2) Two new instruments have since been published: GENEtic Utility scale for pediatric diagnostic testing and adult screening.

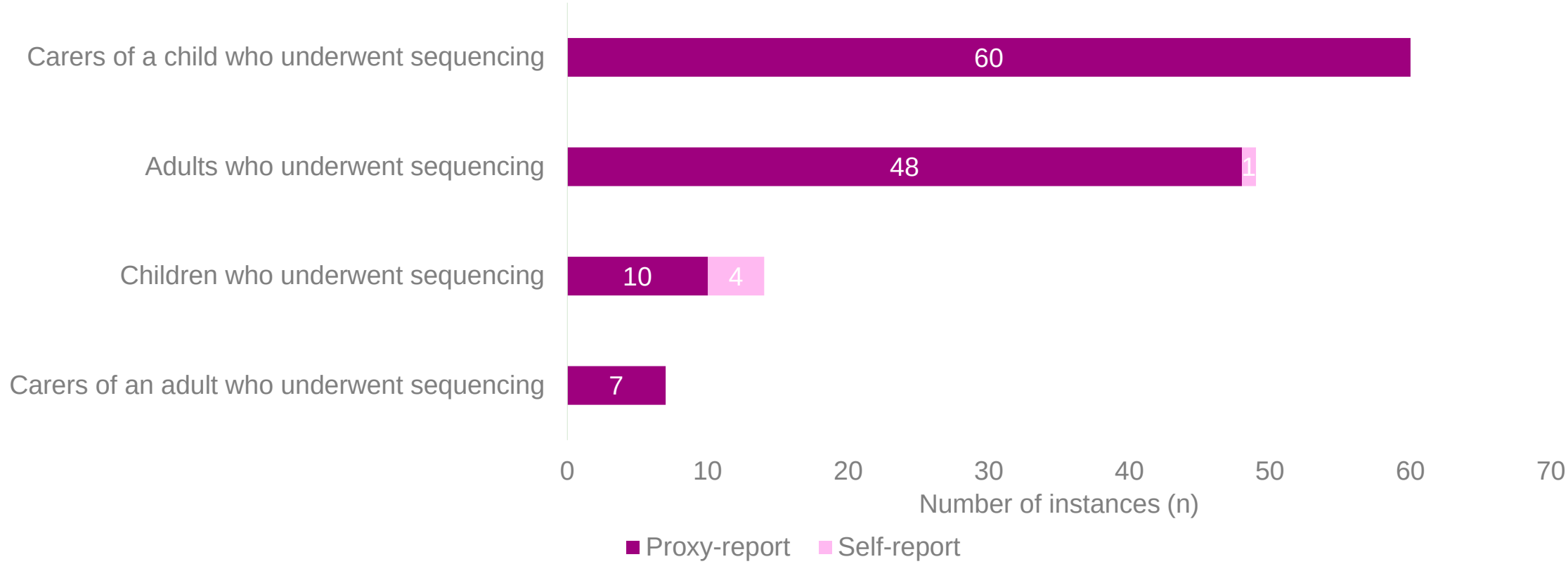
Results: Systematic literature review

Generic health economics instruments

Study	Instrument	Economic evaluation?
Shickh (2019)	SF-12	Describes their plans to assess outcomes, health service use, and economic consequences of using exome sequencing to diagnose cancer and polyposis suggestive of a hereditary cancer syndrome in at-risk adults
Jayasinghe (2019)	SF-12	Describes their plans to conduct an economic evaluation of using genome and exome sequencing to diagnose of genetic kidney disease (GKD) in children and adults
	CHU9D	
Schofield (2020)	AQoL-8D	Outlines the development of a microsimulation model of lifetime economic and social factors in a population of children and adults with moderate to severe familial intellectual disability undergoing genome sequencing, however, results were not reported
	HUI2/3	
Bowman-Smart (2022) & Brett (2020)	SF-12	No
Kelada (2021)	SF-12	No
Lewis (2021)	EQ-5D-Y	No

Results: Systematic literature review

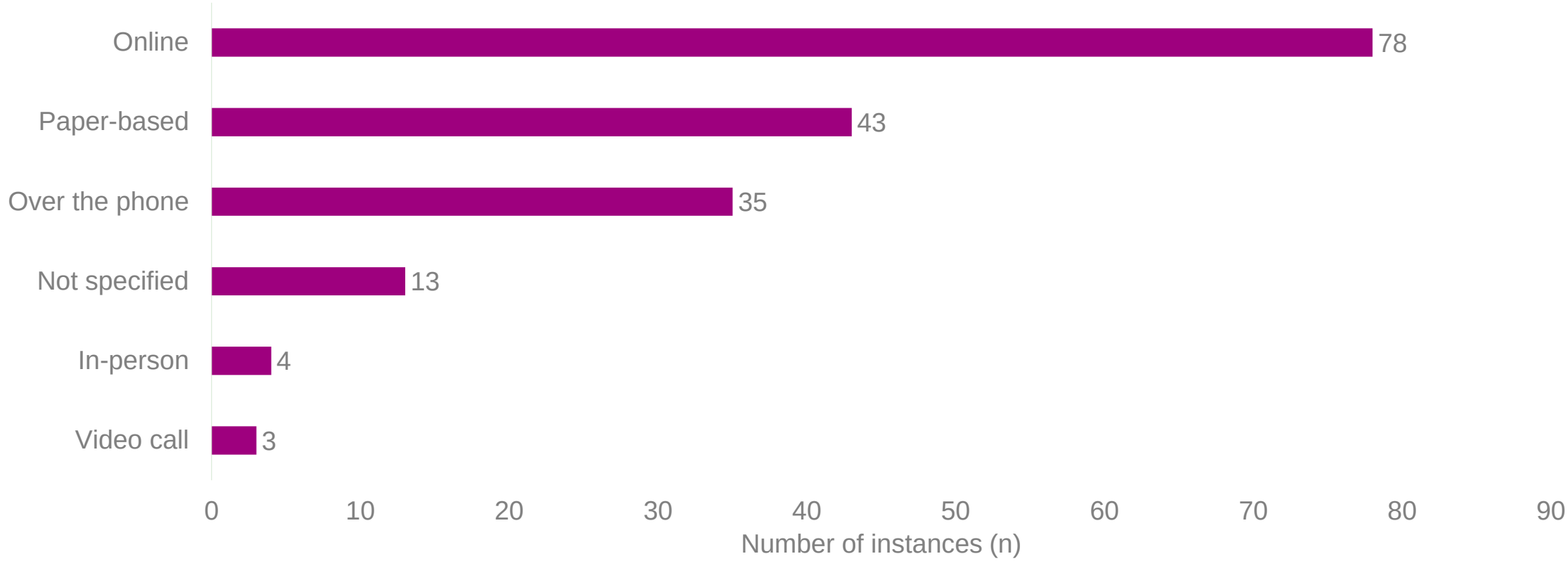
Included instrument subjects and mode of response



Note: Some studies used an instrument to measure outcomes in more than one subject group and/or via more than one mode of response.

Results: Systematic literature review

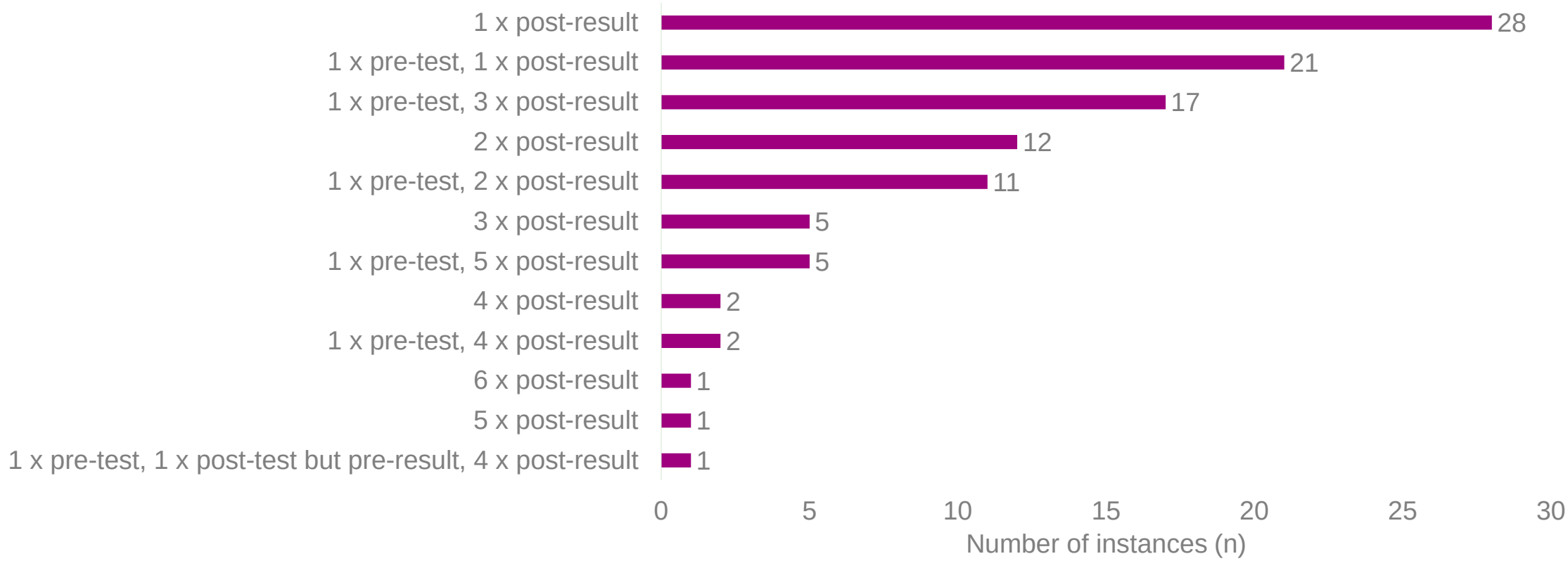
Included instrument administration methods



Note: Some studies used multiple administration methods per instrument.

Results: Systematic literature review

Included instrument administration times and frequencies



Note: Some studies used different administration times per instrument.

Results: Systematic literature review

Instrument selection rationale

- Seventeen (58.6%) of the 29 included studies reported their rationale for selecting at least one of the included instruments
- The most common reasons were that the instrument was widely used; had been used in previous, similar studies; and/or had been validated in a general population sample
  - Other reasons included referring to the efforts of:
    - The Clinical Sequencing Evidence-Generating Research (CSER) Consortium in the US to standardise outcome measurement approaches across their studies
    - The Phenotypes and eXposures (PhenX) Toolkit Genomic Medicine Implementation (GMI) Domain Working Group in the US to harmonise measures
- When provided, rationales were commonly generic statements justifying the use of all or most of the instruments included in the study – very few were tailored to specific instrument selection decisions

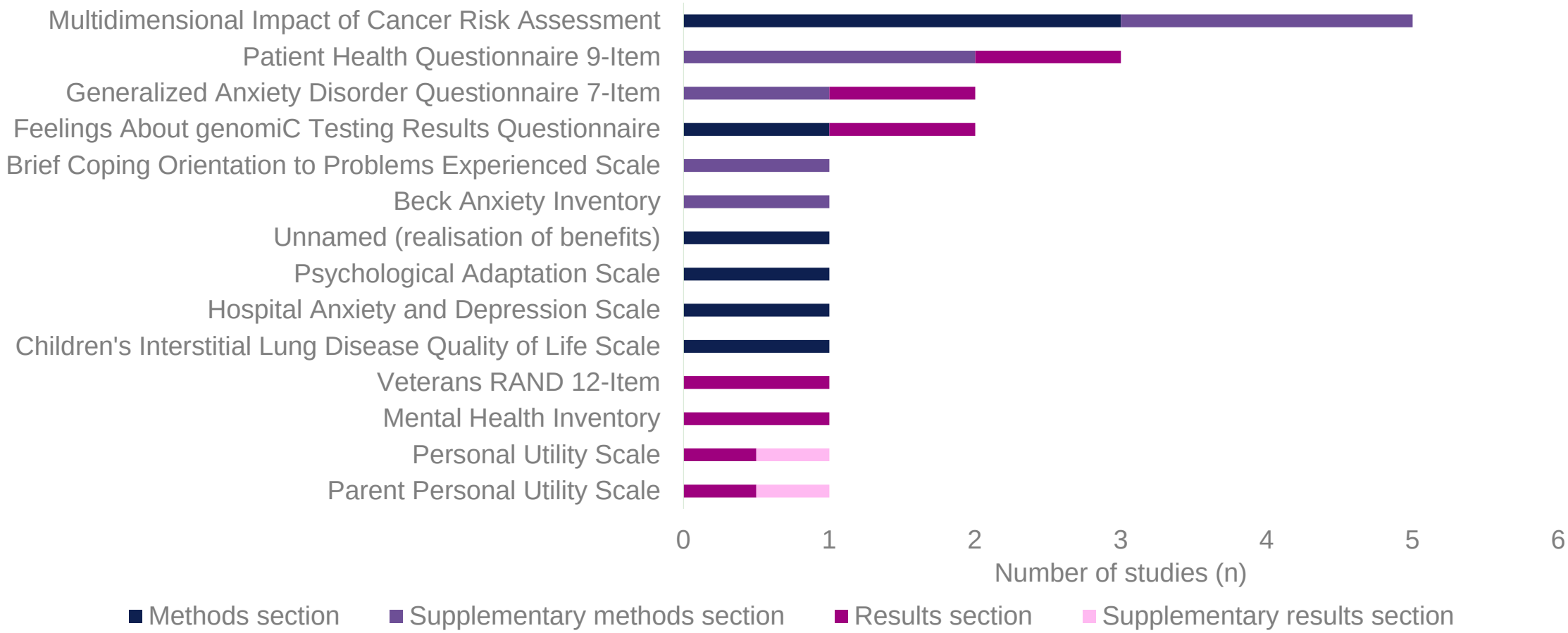
Results: Systematic literature review

Instrument selection and outcome interpretation challenges

1. Determining what outcomes to measure
2. Determining whose outcomes to measure
3. Identifying suitable, validated outcome measurement instruments
4. Distinguishing between types of results
5. Determining whether meaningful change has occurred
6. Comparing results across studies

Results: Systematic literature review

Studies reporting psychometric property results by instrument and reporting location



Results: Systematic literature review

Risk of bias assessment (i.e., quality of the study for each psychometric result reported)

Psychometric property		PrU	P-PrU	FACToR	GAD-7	MHI-5	PHQ-9	VR-12
Development	Concept elicitation	A	A	D	I	I	I	I
	Pilot study	D	D	D	I	I	I	I
	Overall	D	D	D	I	I	I	I
Content validity	Relevance	NA	NA	NA	NA	NA	NA	NA
	Comprehensiveness	NA	NA	NA	NA	NA	NA	NA
	Comprehensibility	NA	NA	NA	NA	NA	NA	NA
	Overall	NA	NA	NA	NA	NA	NA	NA
Structural validity		A	A	VG	NA	NA	NA	NA
Internal consistency		VG	VG	VG	VG	VG	VG	VG
Cross-cultural validity		NA	NA	NA	NA	NA	NA	NA
Reliability		NA	NA	A	NA	NA	NA	NA
Measurement error		NA	NA	NA	NA	NA	NA	NA
Criterion validity		NA	NA	NA	NA	NA	NA	NA
Hypotheses testing for construct validity		VG	I	VG	VG	VG	VG	VG
Responsiveness		NA	NA	NA	NA	NA	NA	NA

Note: 1) VG = very good (green), A = adequate (yellow), D = doubtful (orange), I = inadequate (red), NA = not applicable (white), 2) Studies providing further context regarding instrument development were also included in the concept elicitation and pilot study assessment .

Results: Systematic literature review

Study result assessment (i.e., quality of each psychometric result reported by each study)

Psychometric property		PrU	P-PrU	FACToR	GAD-7	MHI-5	PHQ-9	VR-12
Content validity	Relevance	+	+	+	+	+	+	+
	Comprehensiveness	±	±	±	-	-	-	-
	Comprehensibility	?	?	+	?	?	?	?
	Overall	±	±	±	?	?	?	?
Structural validity		+	+	?	NA	NA	NA	NA
Internal consistency		+	+	+	+	+	+	+
Cross-cultural validity		NA	NA	NA	NA	NA	NA	NA
Reliability		NA	NA	+	NA	NA	NA	NA
Measurement error		NA	NA	NA	NA	NA	NA	NA
Criterion validity		NA	NA	NA	NA	NA	NA	NA
Hypotheses testing for construct validity		+	+	±	+	+	+	+
Responsiveness		NA	NA	NA	NA	NA	NA	NA

Note: 1) + = sufficient (green), - = insufficient (yellow), ? = indeterminate (orange), ± = inconsistent (red), NA = not applicable (white), 2) Studies providing further context regarding instrument development were also included in the assessment of content validity.

Results: Systematic literature review

Certainty of evidence assessment (i.e., quality of the summarised evidence per property)

Psychometric property		PrU	P-PrU	FACToR	GAD-7	MHI-5	PHQ-9	VR-12
Content validity	Relevance	L	L	L	VL	VL	VL	VL
	Comprehensiveness	L	L	L	VL	VL	VL	VL
	Comprehensibility	NA	NA	L	NA	NA	NA	NA
	Overall	L	L	L	VL	VL	VL	VL
Structural validity		M	M	NA	NA	NA	NA	NA
Internal consistency		H	H	H	H	H	H	H
Cross-cultural validity		NA	NA	NA	NA	NA	NA	NA
Reliability		NA	NA	VL	NA	NA	NA	NA
Measurement error		NA	NA	NA	NA	NA	NA	NA
Criterion validity		NA	NA	NA	NA	NA	NA	NA
Hypotheses testing for construct validity		H	VL	M	H	H	H	H
Responsiveness		NA	NA	NA	NA	NA	NA	NA

Note: 1) H = high (green), M = medium (yellow), L = low (orange), VL = very low (red), NA = not applicable, 2) Studies providing further context regarding instrument development were also included in the assessment of content validity.

Phase 2: Critical appraisal

Method: Critical appraisal

Relevance: The proportion of the total instrument items that represent an outcome model sub-domain

Comprehensiveness: The proportion of the outcome model sub-domains that are represented by the instrument items

Feasibility: Considerations such as the number of items within each instrument, the licencing costs and restrictions, and the availability of the languages of interest

Expert opinion: From the Central & South Genomic Medicine Service Alliance (C&S GMSA) Patient and Public Involvement (PPI) Panel and Research Directors

Method: Critical appraisal

Primary source: Smith et al., (2021). Supplementary sources: Kohler et al., (2017a;b); Hayeems et al., (2021); Duenas et al., (2023).

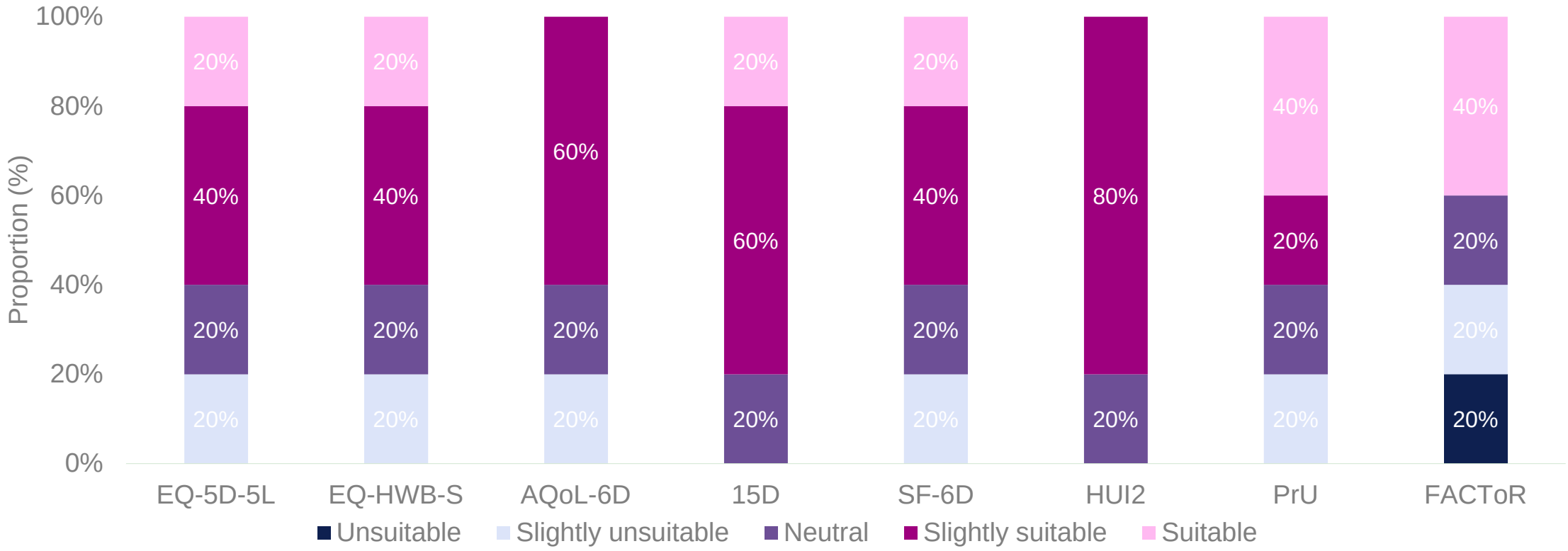
Domain	Sub-domain
Clinical	Diagnostic remarkability (establishment, confirmation, or ruling out a diagnosis)
	Appropriateness of follow-up care
	Informed clinical management
	Monitoring for early disease detection
	Referral to clinical trial
Emotional	Adverse responses (anxious feelings, confusion, depressive symptoms, disappointment, fear, frustration, guilt, sadness, worry)
	Positive responses (empowerment, gratitude, hope, relief, spiritual wellbeing)
Cognitive	Value of knowing information
	Perceived health risk
	Satisfaction of curiosity
	Self-knowledge
Behavioural	Insurance coverage
	Health habits (diet, exercise, smoking, substance use)
	Information seeking
	Future planning (estate, financial, career choices)
	Parenting decisions
	Reproductive decision-making
	Communication with family
Social	Advocacy activities
	Blame
	Access to support services
	Degree of social support
	Discrimination (employment, schooling, insurance)
	Privacy concerns
	Social stigma
	Quality of relationship with care providers
	Helping others

Results: Critical appraisal

	Relevance	Comprehensiveness	Feasibility
Generic health economics	AQoL-8D (71%) EQ-HWB-S (67%) AQoL-6D (50%)	EQ-HWB (15%) AQoL-8D (11%) AQoL-6D (11%)	Costly licence fees Translation limitations Number of items
Genome sequencing specific	GENE-U PD / AS (100%) Parent PrU / PrU (100%) FACToR-P (77%)	GENE-U PD (67%) GENE-U AS (59%) PrU (37%)	Translation availability
Carer-specific quality-of-life	AC-QOL (80%) CES (67%) CarerQoL-7D (63%)	AC-QOL (26%) CarerQoL-7D (15%) CES (11%)	Translation limitations
Generic psychological	Almost all were 100%	CES-D 20 (15%) MHI-38 (11%) MHI-5 (11%)	Number of items Cultural adaptation issues

Results: Critical appraisal

Expert opinion on a sub-set of instruments



Note: 1) Respondents included 2 (66.67%) Central & South Genomic Medicine Service Alliance (C&S GMSA) Research Directors and 3 (42.86%) C&S GMSA Patient and Public Involvement (PPI) Panel members; 2) Two of the C&S GMSA PPI Panel members offered to meet via Zoom to discuss their feedback in more detail, however, neither responded to my emails about this; 3) The GENEtic Utility Pediatric & Adult Diagnostic Scales (GENE-U PD/AD) were not available when these experts were consulted.

Results: Critical appraisal

Instruments selected to be evaluated via a prospective cohort study

Instrument	Acronym/abbreviation
Generic health economics instruments	
European Quality of Life 5 Dimensions 5 Level Version	EQ-5D-5L
European Quality of Life Health and Wellbeing Instrument Short	EQ-HWB-S
Assessment of Quality of Life 6 Dimensions	AQoL-6D
Quality of Life Questionnaire 15 Dimensions	15D
Genome sequencing specific instruments	
GENEtic Utility Pediatric & Adult Diagnostic Scales	GENE-U PD / GENE-U AD
Personal Utility Scale (incl. parent version)	PrU / Parent PrU
Feelings About genomiC Testing Results Questionnaire (incl. parent version)	FACToR / FACToR-P
Carer specific quality of life instrument	
Care-related Quality of Life Instrument	CarerQol-7D
Generic psychological instrument	
Mental Health Inventory-Five Item	MHI-5

Note: Proxy-report versions were also obtained (or developed) and tested.

Phase 3: Cohort study

Method: Cohort study

Study design

- Informed by **40+ professional stakeholder consultations**
- Obtained **£50,000 funding** to support:
  - Translation into Urdu and Punjabi – **totalled £20,000**
  - Interpreters for Urdu and Punjabi
  - Paper and accessible digital (i.e., Qualtrics) versions
  - NHS Genomic Medicine Service (GMS) staff identifying and referring potential participants

Method: Cohort study

Participant identification and referral

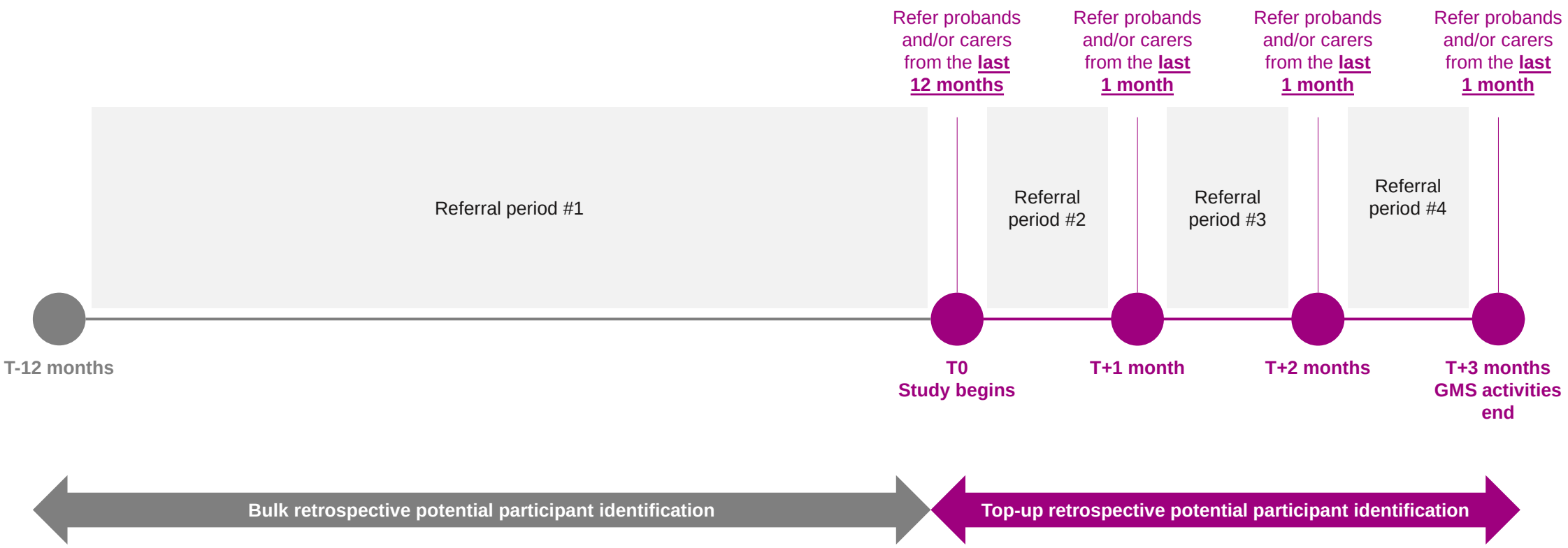
Facilitated by Genomic Practitioners and Research Nurses within the NHS Genomic Medicine Service (GMS) in Oxford, Birmingham, and Southampton (i.e., the Central & South Region)

Target populations

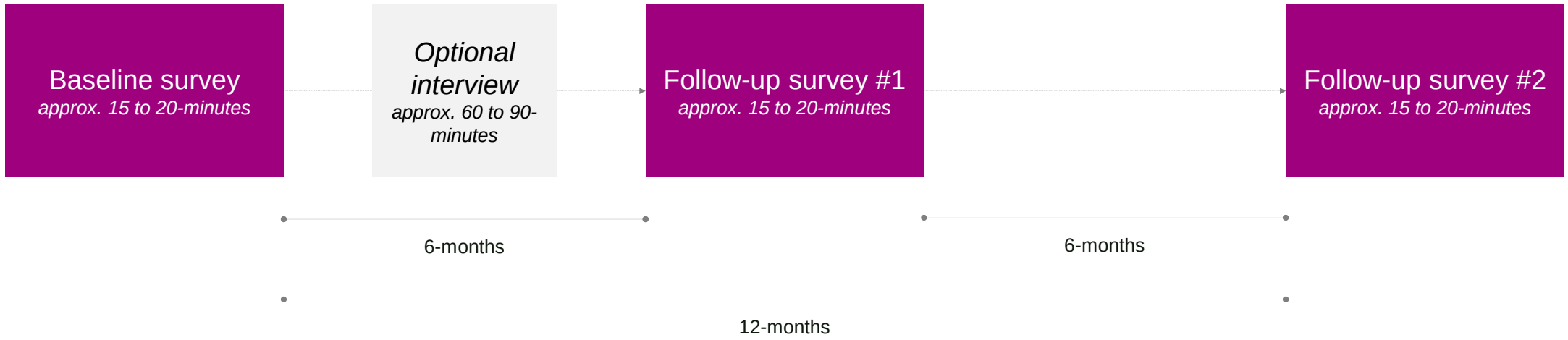
1. Adults undergoing GS for rare disease diagnosis (self-report)
2. Unpaid carers of adults undergoing GS for rare disease diagnosis (proxy or self-report)
3. Unpaid carers of children undergoing GS for rare disease diagnosis (self-report)

Note: Carers include parents, guardians, spouses/partners, adult children, and other adults who have an informal (i.e., unpaid) caring relationship with the person with a rare disease.

Method: Cohort study



Method: Cohort study



Key:

- Core study activity
- Optional study activity

Note: Participants who express interest in participating in an interview when completing the baseline survey will only be invited to complete one interview. This interview may take place either in-between the baseline and first follow-up survey, or in-between the first and second follow-up surveys.

Method: Cohort study

Survey start	Instruments	Survey end
Welcome	4 x generic health economics	General feedback Qs
Participant information	3 x genome sequencing specific	Optional random prize draw Q
Screening and eligibility Qs	1 x generic psychological	Thank you and next steps
Informed consent Qs	1 x carer specific quality of life	
Demographic Qs	Instrument relevance Qs	
Exposure status Qs		
Health status Qs		

Note: 1) The participant information, screening and eligibility questions, and informed consent questions are only included in the baseline survey; 2) The health status questions are only included in the follow-up surveys; 3) The instrument order is randomised in the online survey platform; 4) The instrument relevance questions are asked after each instrument, 5) Carer specific quality of life instruments are only displayed to carers; 6) Genome sequencing specific instruments are only displayed to participants who have received their results; 7) The optional random prize draw question is only included in the second follow-up survey.

Method: Cohort study

Instrument relevance questions

- **Adults with a rare disease:** To what extent are the questions from the previous screen relevant to the impact that genetic testing for rare disease diagnosis has had on you?
- **Proxies for adults with a rare disease:** To what extent would the Study Person think that the questions from the previous screen are relevant to the impact that genetic testing for rare disease diagnosis has had on them?
- **Carers of adults with a rare disease:** To what extent are the questions from the previous screen relevant to the impact that the Study Person's genetic testing for rare disease diagnosis has had on you?
- **Carers of children with a rare disease:** To what extent are the questions from the previous screen relevant to the impact that the Study Child's genetic testing for rare disease diagnosis has had on you?

Note: Responses are collected via a 5-point Likert scale including “Very irrelevant”, “Somewhat irrelevant”, “Neither relevant nor irrelevant”, “Somewhat relevant”, and “Very relevant”

Method: Cohort study

Domain	Psychometric property	Sub-component	Qualitative analysis	Quantitative analysis
Validity	Content validity	Relevance	X	X
		Comprehensiveness	X	
		Comprehensibility	X	
	Construct validity	Known-groups validity		X
		Convergent validity		X
		Divergent validity		X
		Structural validity		X
Responsiveness	Responsiveness	-	X	X
Interpretability	-	-		X
Acceptability and feasibility	-	-	X	X

Method: Cohort study

Changes made following the pilot study

1. Changed the eligibility criteria to allow people who have received their results to participate in the baseline survey

2. Refined the wording of the instrument relevance questions to ensure it is clear they are asking about relevance with respect to the impact of genome sequencing and not participants' condition

3. Refined the wording of the question asking respondents if they had a rare disease or care for someone who has a rare disease

4. Simplified the question type for collecting the date of sample collection and return of results (i.e., from DD/MM/YYYY to MM/YYYY)

Method: Cohort study

Other key changes made during the study design process

1. Narrowing the scope by excluding children and proxies for children from the target population
2. Changing the study design from “pre-test post-test” to “baseline and follow-up” to allow for the retrospective recruitment of patients who had consented to genome sequencing in the past to increase the likelihood of achieving the target sample size within the target timeframe
3. Reducing the prospective component (from 6-months to 3-months) and increasing the retrospective component (from 6-months to 9-months and then to 12-months) to increase the likelihood of achieving the target sample size within the target timeframe
4. Organising for a 3rd NHS GMS site to join the study to increase the likelihood of achieving the target sample size within the target timeframe
5. Broadening the scope of eligible participants to include those who have received their genome sequencing results at the point of completing the baseline survey to allow conclusions regarding the content validity of the instruments to be made as soon as possible

Results: Cohort study

Recruitment by site

Site	Invitations sent (n)		Responses received (n)			Response rate (%)		
	Letters	Reminder texts	Eligible	Ineligible	Unfinished	Eligible	Ineligible	Unfinished
Birmingham	806	669	89	29	36	11.04%	3.60%	4.47%
Southampton	692	-	63	26	41	9.10%	3.76%	5.92%
Oxford	769	-	78	23	29	10.14%	2.99%	3.77%
Total	**2,267**	**669**	**230**	**78**	**106**	**10.15%**	**3.44%**	**4.68%**

Note: 1) Eligible and ineligible figures, and finished and unfinished figures, are not additive. This is because some participants accidentally respond incorrectly to an eligibility question and then try again. Additionally, some participants may open and close the survey multiple times before completing and submitting their response. 2) Birmingham is the only site with the ability to send text message reminders to potential participants.

Results: Cohort study

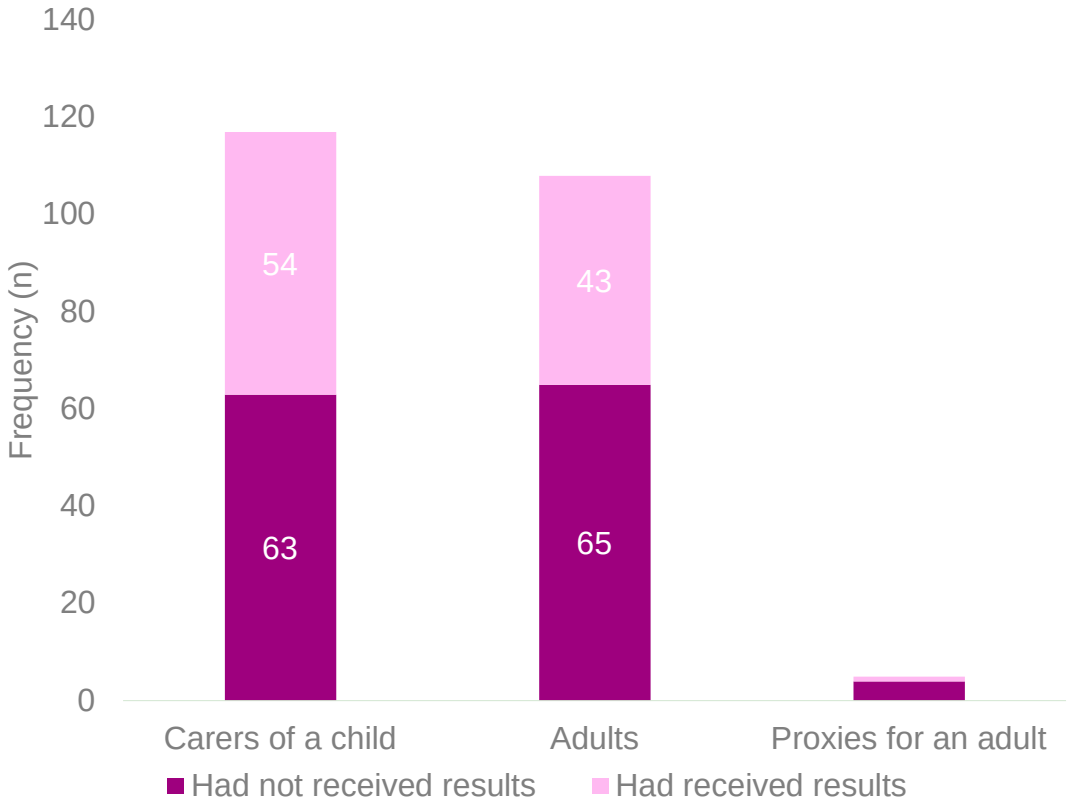
Recruitment by participant category

Participant category	Target		Actual	
	Original	Revised	Original	Revised
Adults	156	156	108 (69.23%)	113 (72.44%)
Proxies for adults	156	-	5 (3.21%)	-
Carers of an adult	156	-	0 (0.00%)	-
Carers of a child	156	156	117 (75.00%)	117 (75.00%)
Total	**624**	**312**	**230 (36.86%)**	**230 (73.72%)**

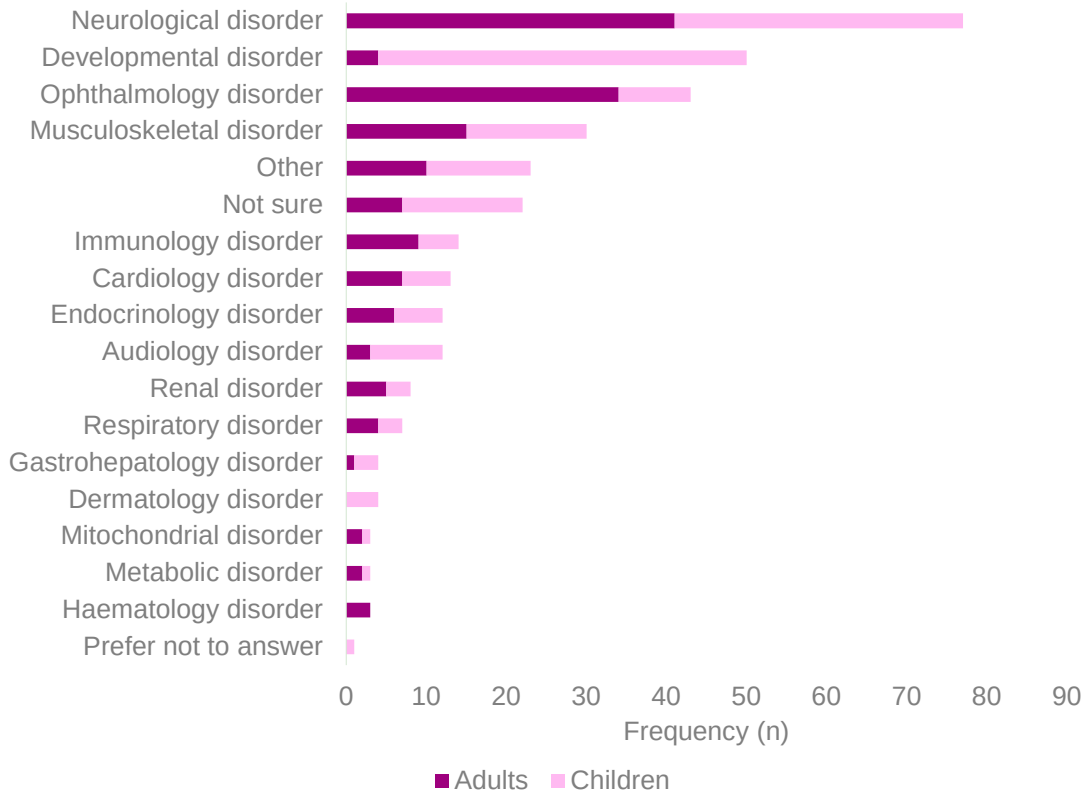
Note: Participants who meet the criteria for both “proxies for an adult” and “carers of an adult” are preferentially directed to complete the proxy pathway until the sample size for this sub-group is achieved.

Results: Cohort study

Participant categories and result status



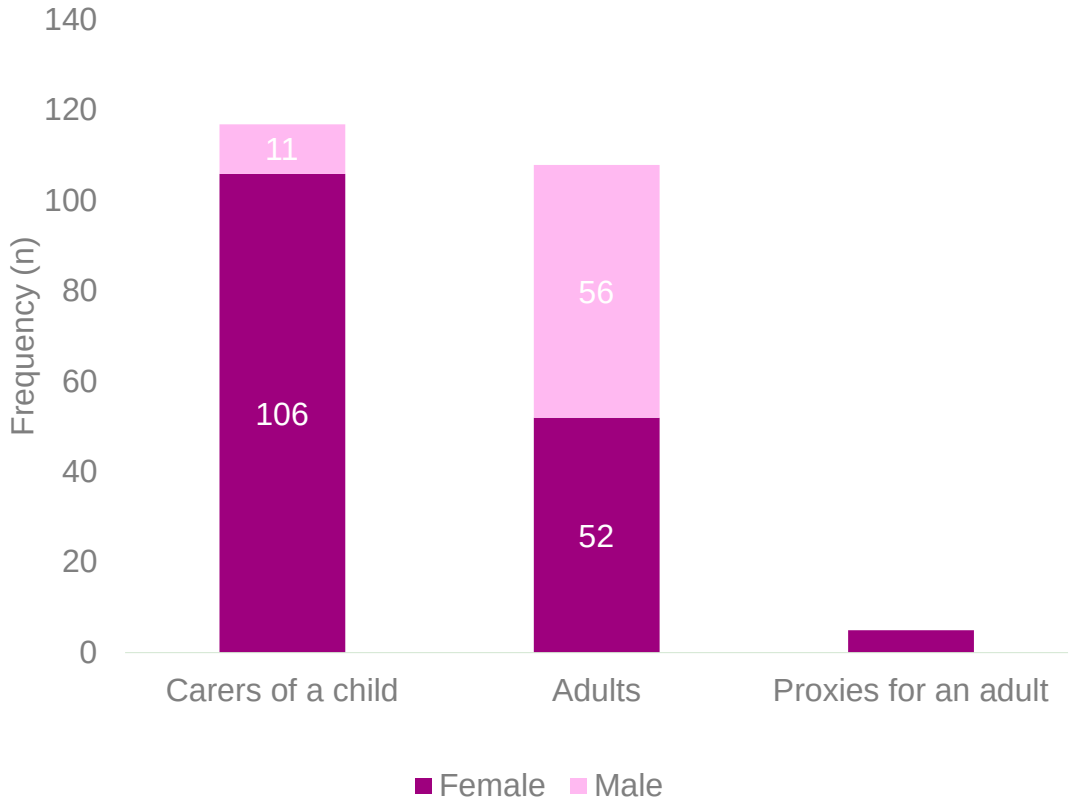
Disease areas



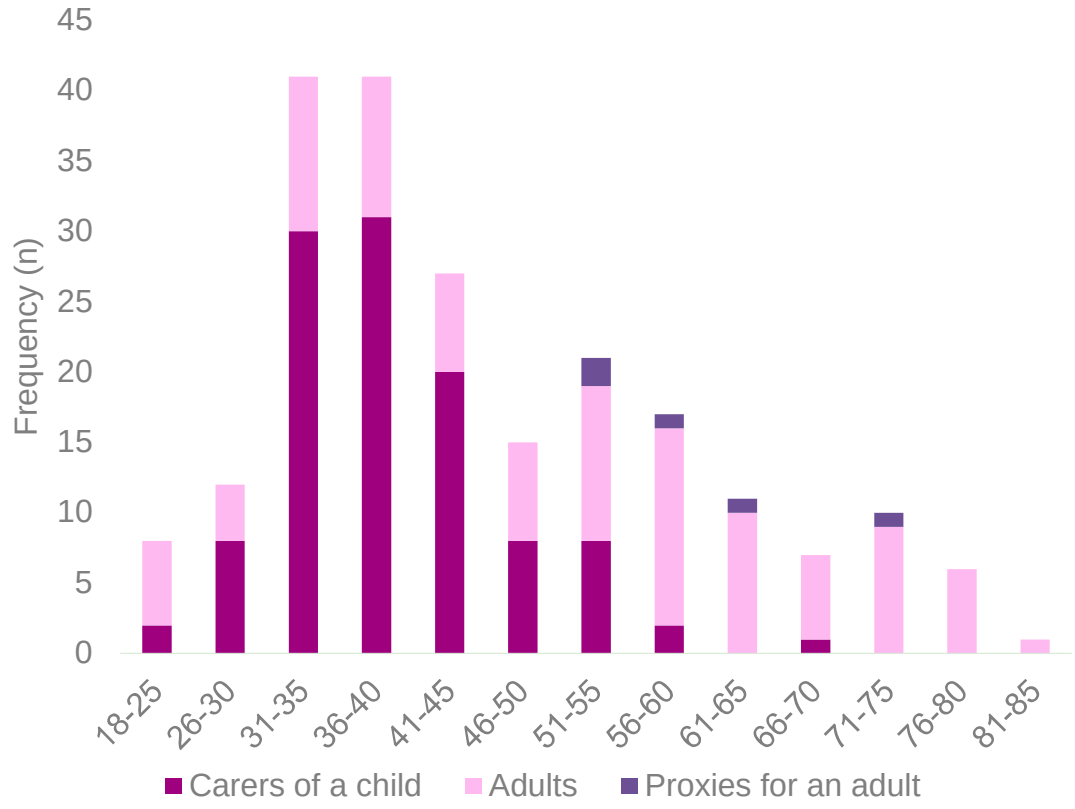
Note: Total exceeds number of participants as rare diseases can affect multiple systems.

Results: Cohort study

Sex at birth (person completing the survey)



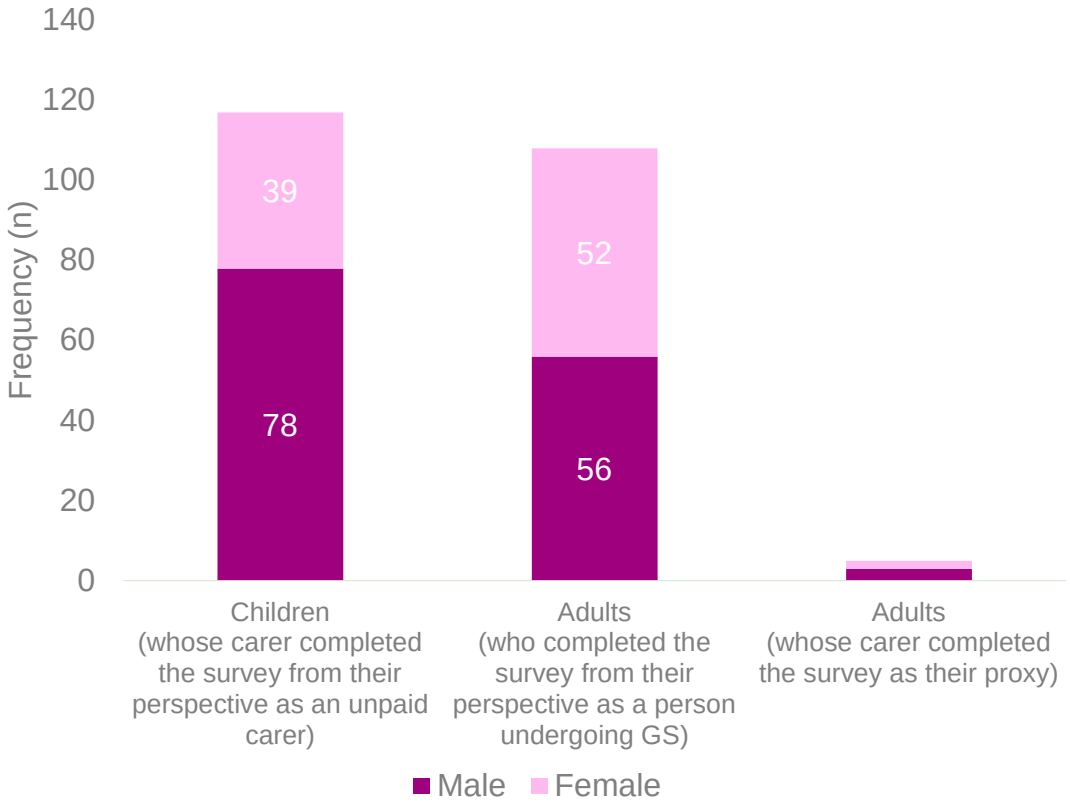
Age (person completing the survey)



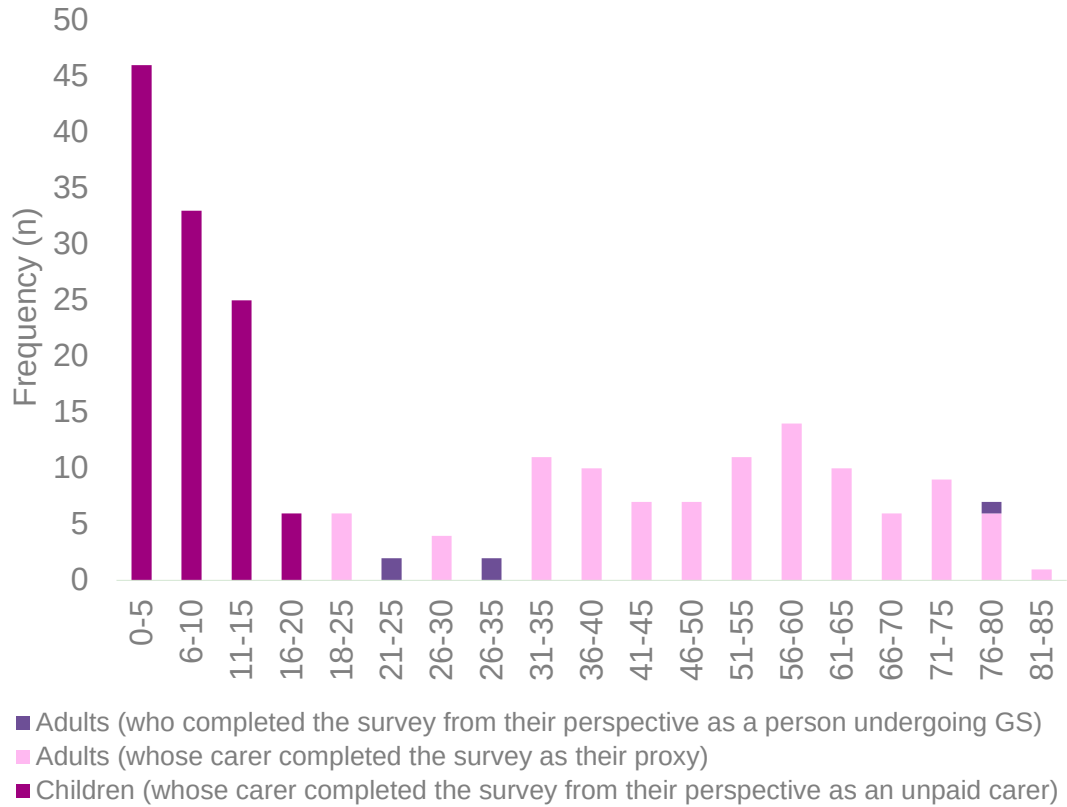
Note: Participants who meet the criteria for both “proxies for an adult” and “carers of an adult” are preferentially directed to complete the proxy pathway until the sample size for this sub-group is achieved.

Results: Cohort study

Sex at birth (person being sequenced)

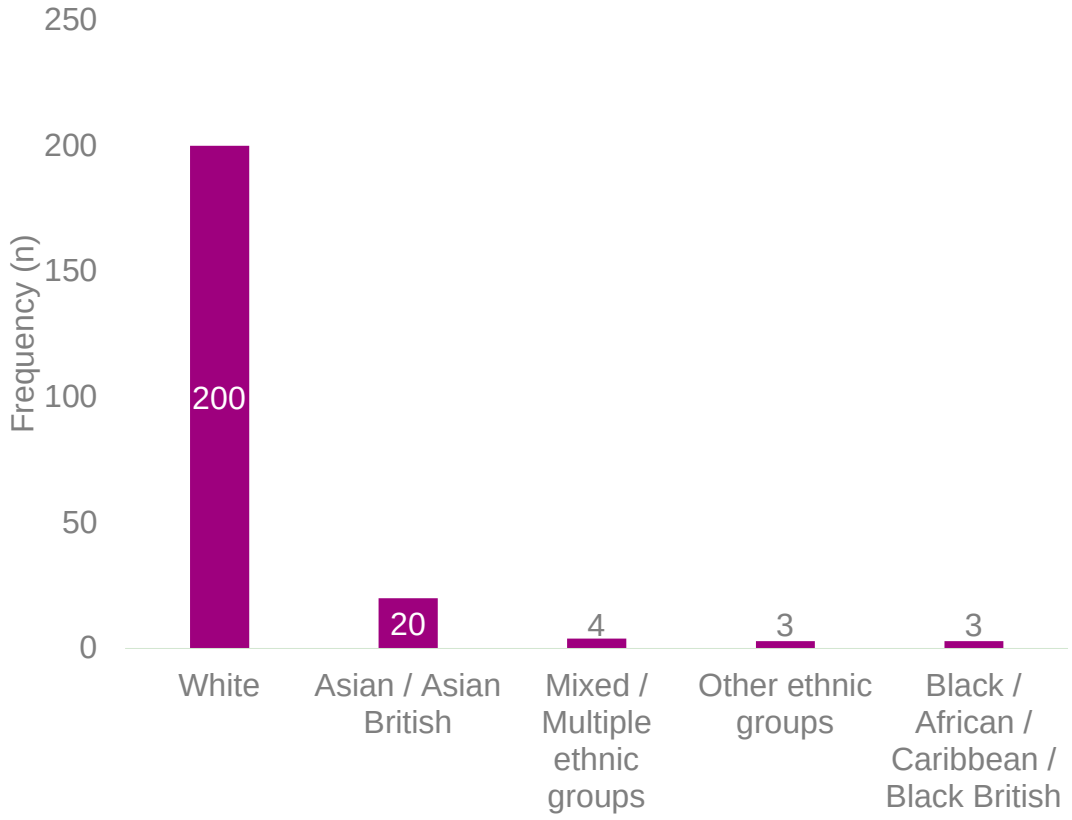


Age (person being sequenced)

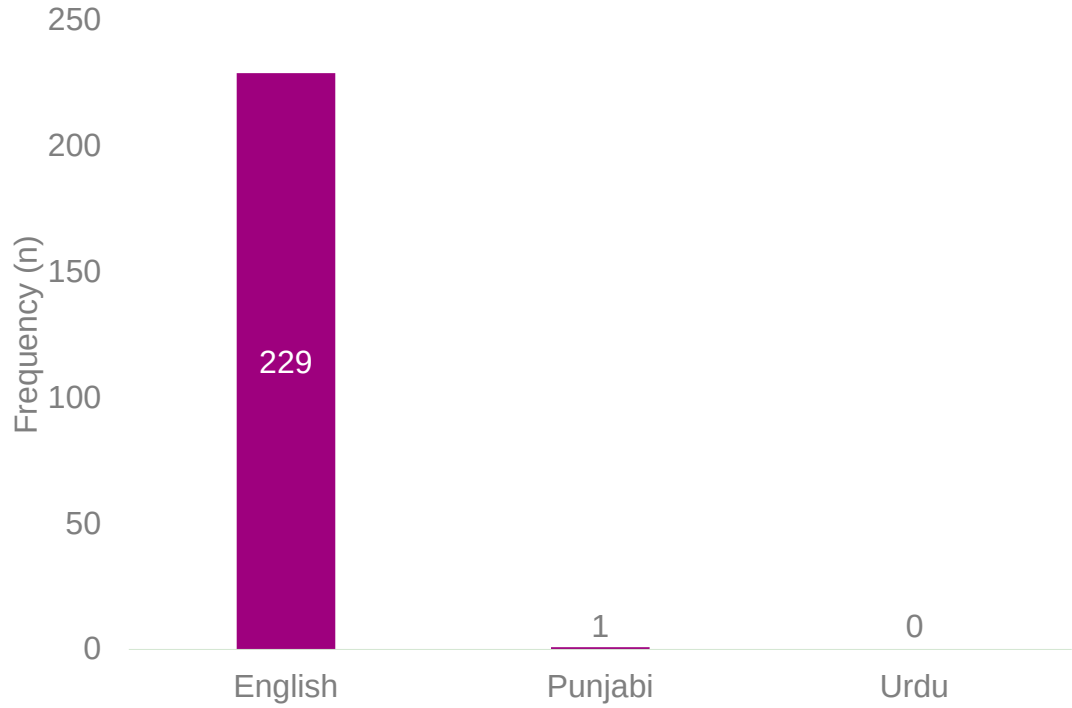


Results: Cohort study

Ethnicity (person completing the survey)

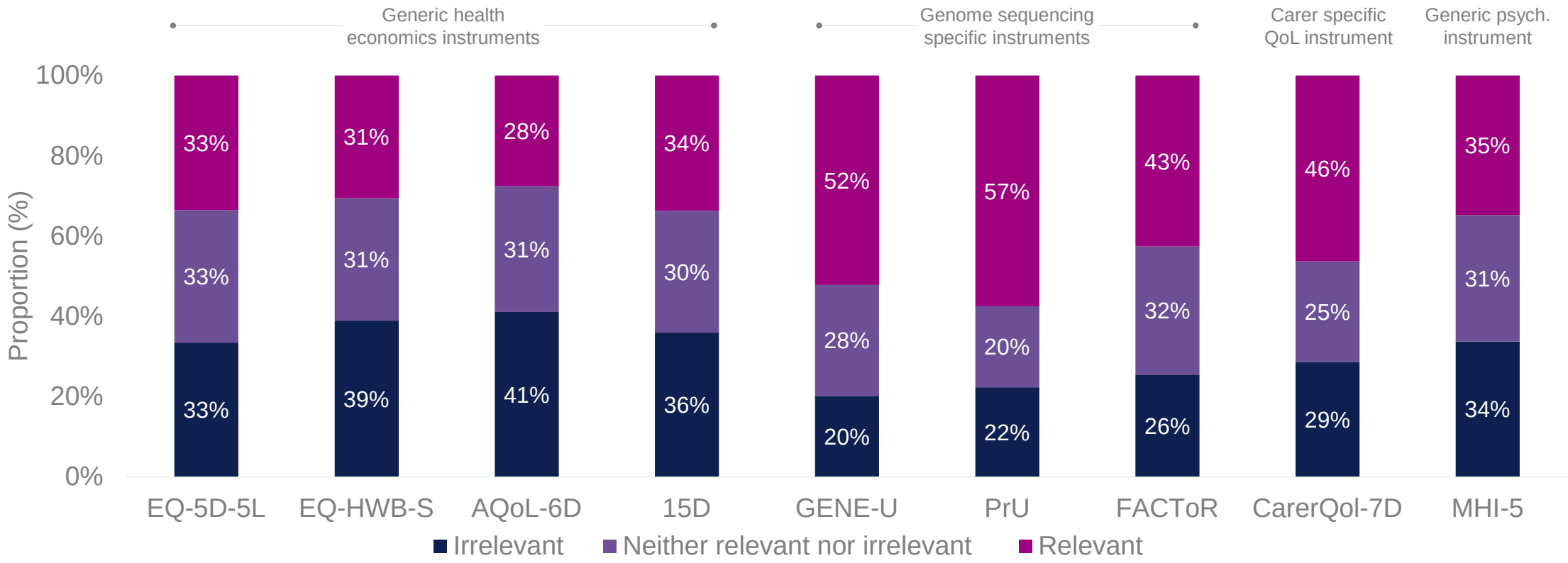


Survey language



Results: Cohort study

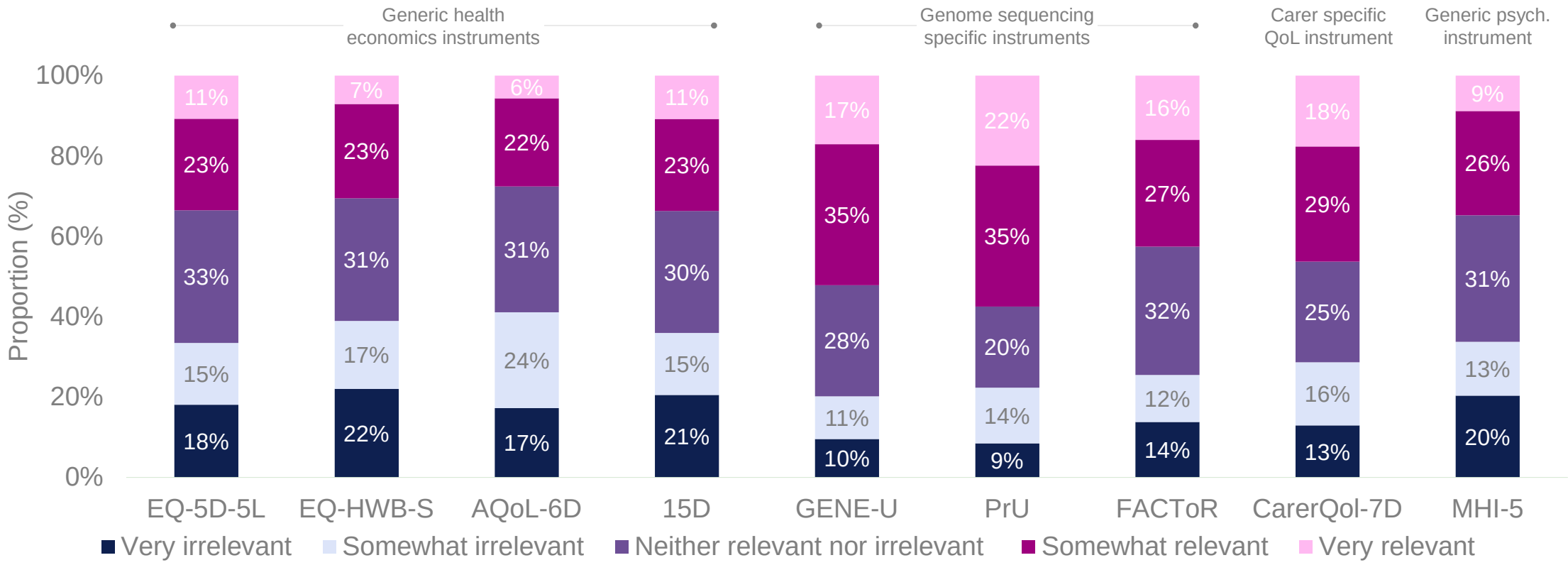
Instrument relevance (summarised Likert categories)



Note: 1) Carer specific quality of life instruments are only displayed to carers, 2) Genome sequencing specific instruments are only displayed to participants who have received their results.

Results: Cohort study

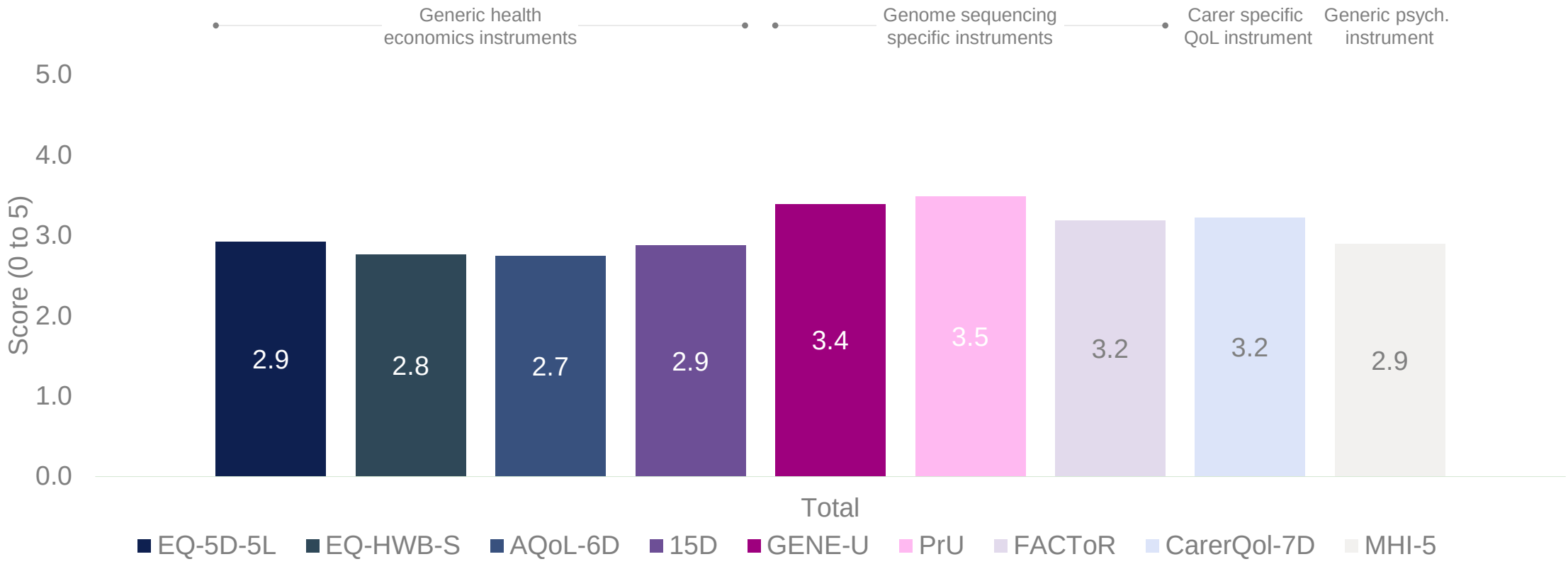
Instrument relevance (detailed Likert categories)



Note: 1) Carer specific quality of life instruments are only displayed to carers, 2) Genome sequencing specific instruments are only displayed to participants who have received their results.

Results: Cohort study

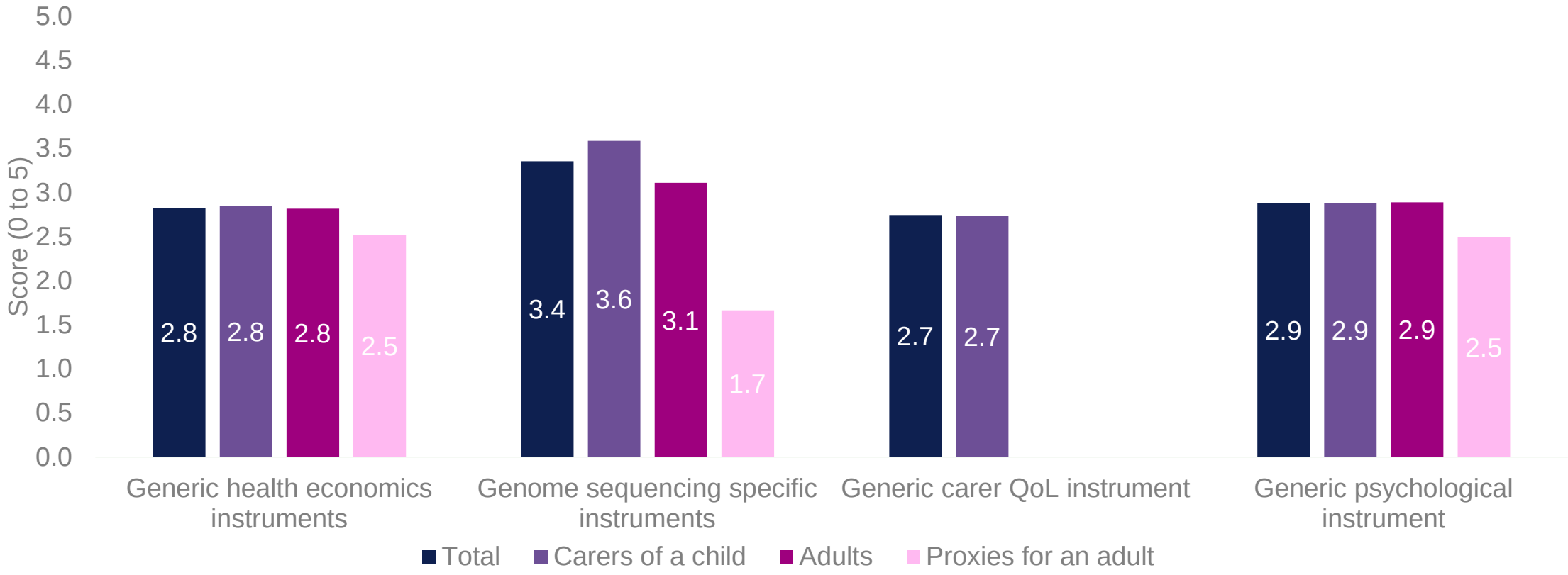
Instrument relevance (mean score)



Scoring: Very relevant = 5, Somewhat relevant = 4, Neither relevant nor irrelevant = 3, Somewhat irrelevant = 2, Very irrelevant = 1
Note: 1) Carer specific quality of life instruments are only displayed to carers, 2) Genome sequencing specific instruments are only displayed to participants who have received their results.

Results: Cohort study

Instrument relevance (mean score) by instrument type and participant category



Scoring: Very relevant = 5, Somewhat relevant = 4, Neither relevant nor irrelevant = 3, Somewhat irrelevant = 2, Very irrelevant = 1

Note: 1) Carer specific quality of life instruments are only displayed to carers, 2) Genome sequencing specific instruments are only displayed to participants who have received their results.

Impact: What will this work enable?

- Improved **instrument selection** decisions in this context
- Improved **understanding** of where generic health economics instruments may fall short in this context
- Improved **interpretation** of evidence generated using generic health economics instruments in this context

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Thank you


sally.sansom@dph.ox.ac.uk


Sally Sansom


@sallysansom.bsky.social


@SallySansom