

Participant attitudes toward returning individual results from observational research in Cerebral Autosomal Dominant Arteriopathy with Subcortical Infarcts and Leukoencephalopathy

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Background

- Problem: disagreement about returning individual research results (IRR).
- Aim: explore research participant attitudes toward returning IRR from the United States CADASIL Consortium (USCC), an observational study.

Methods

- 4-item survey: three 5-point Likert items; one open-ended item about IRR.
- Quantitative: Descriptive statistics (means, percentages).
- Quantitative: Independent samples Mann-Whitney U test (SPSS, v. 30.0) to compare results with Clark et al. (2022) “Anticipated reactions to learning Alzheimer’s disease biomarker results”.
- Qualitative: Deductive codebook → pilot coding → calibration → deductive & inductive coding → consensus & theming

Highlights

1. 27.1% response rate (152/560).
2. Most important results: “imaging or MRI scans” and “cognitive testing” (Figure 1).
3. Most important reason for return: “to learn about my disease and its predicted course” (Figure 2).
4. Hypothetical negative outcomes: “a little” to “somewhat” concerning (Figure 3).
5. Comparison with Alzheimer’s disease (AD): USCC participants rated reasons for return higher and negative outcomes lower than AD participants (Table 1).
6. Majority support for qualified disclosure: IRR should be returned to interested participants (Figure 4).
7. Top code: “IRR return will help improve my health and well-being” (Table 2).

Discussion

- Lessons from the literature.
 - Alternate interpretation of strong endorsement: desire for meaningful, packaged risk information.
 - Study limitations: selection bias for most motivated participants.
- Future directions in CADASIL: new IRR guidance for the CADASIL community.
 - Reflect community attitudes.
 - Support successful clinical trials.
 - Meet duty to investigate return.
- Future directions in disclosure science.
 - Comparison with non-AD populations.
 - Assess the personal utility of returning IRR.
 - Measure outcomes like changes in trust or retention secondary to return.

Disclosures

The authors report no conflicts of interest.

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Findings

FIGURE 1. HOW IMPORTANT IS IT TO SHARE THIS TYPE OF INFORMATION? (EXTREMELY/VERY [%])

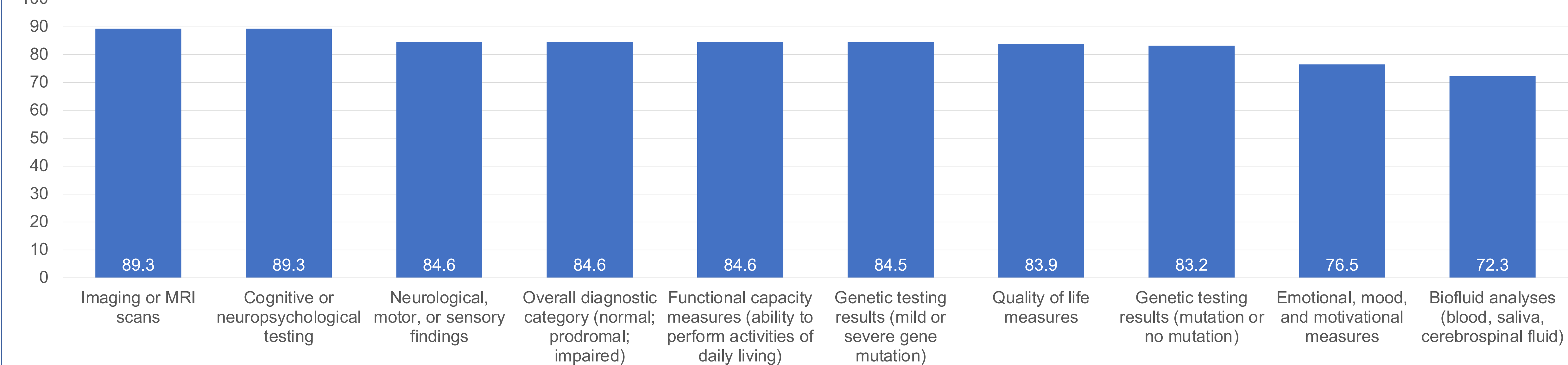


FIGURE 2. HOW IMPORTANT ARE THESE REASONS TO SHARE RESEARCH RESULTS? (EXTREMELY/VERY [%])

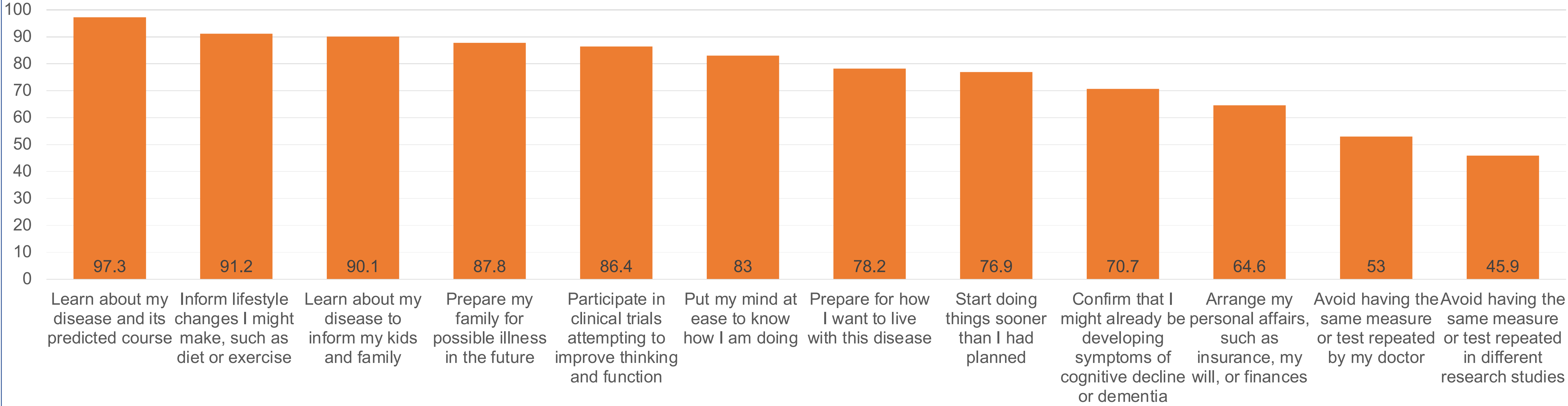


FIGURE 3. HOW CONCERNED WOULD YOU FEEL ABOUT THE SHARING OF RESULTS? (EXTREMELY/VERY [%])

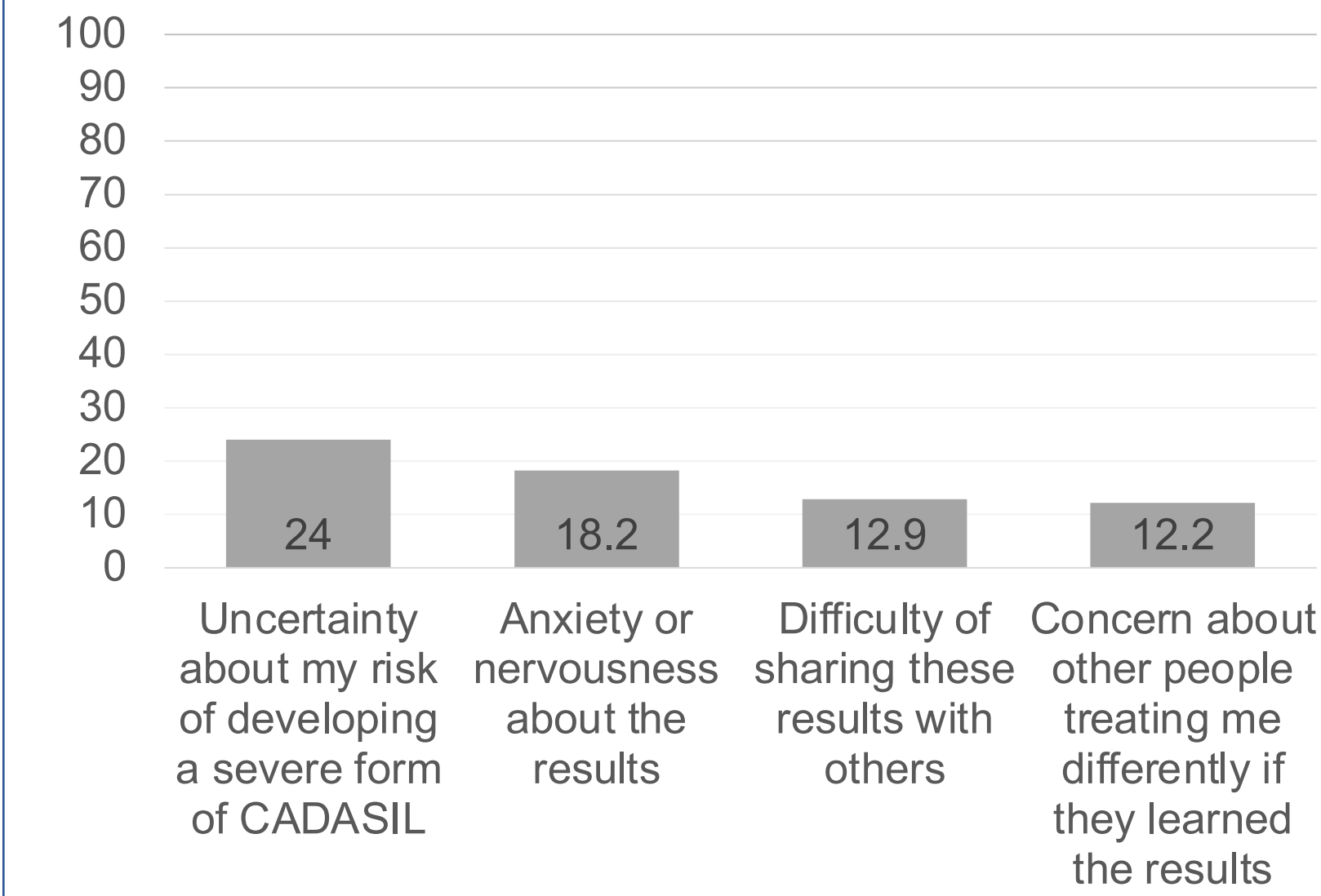


FIGURE 4. GENERAL ATTITUDES TOWARDS IRR RETURN

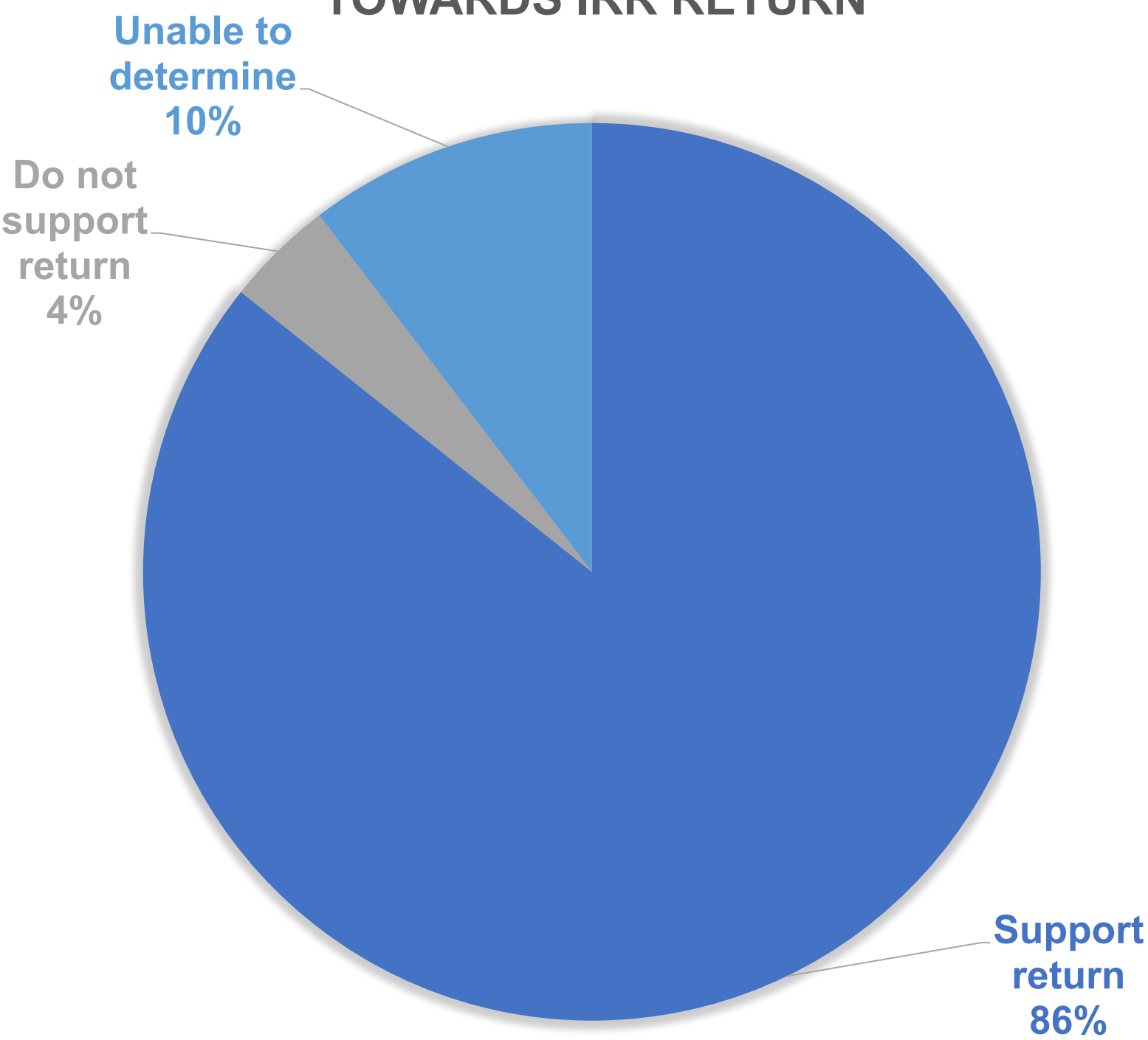


Table 1. Comparison of CADASIL & Alzheimer’s participant responses

Table 1. Comparison of CADASIL & Alzheimer’s participant responses	USCC rating (mean) (SD) (N)	AD rating (mean) (SD) (N)	p-value (* = significant)
How important it is to receive IRR to...			
...learn about my disease and its predicted course	4.68 (0.64) (146)	3.93 (1.20) (331)	<0.001*
...inform lifestyle changes I might make, such as diet or exercise that might help prevent declines in thinking and functioning	4.56 (0.83) (147)	4.26 (1.00) (334)	<0.001*
...put my mind at ease to know how I am doing	4.33 (0.99) (147)	4.13 (1.00) (334)	0.015*
...participate in clinical trials attempting to improve thinking and function	4.35 (0.88) (147)	3.91 (1.00) (332)	<0.001*
...arrange my personal affairs, such as insurance, my will, or finances	3.85 (1.20) (147)	3.73 (1.31) (334)	0.392
...confirm that I might already be developing symptoms of cognitive decline or dementia	3.98 (1.24) (147)	3.77 (1.20) (333)	0.031*
...prepare my family for possible illness in the future	4.32 (1.02) (147)	3.99 (1.10) (334)	<0.001*
...start doing things sooner than I had planned	4.15 (1.11) (147)	3.77 (1.20) (334)	<0.001*
If told my prognosis, I would feel...			
...anxious or nervous about the results	2.66 (1.07) (148)	3.20 (1.10) (333)	<0.001*
...uncertain about my risk of developing severe disease	2.82 (1.13) (146)	3.02 (1.00) (332)	0.029*
...the difficulty of sharing these results with others	2.07 (1.15) (147)	2.17 (1.20) (333)	0.489
...concerned about other people treating me differently if they learned the results	2.03 (1.19) (148)	2.64 (1.30) (334)	<0.001*

Table 2. Why should or should not study participants receive results? (n=77)

Table 2. Why should or should not study participants receive results? (n=77)	Occurrences
Deductive or inductive codes	
IRR return will improve my health or well-being	23
IRR return will help me plan for the future	21
IRR return will provide me valuable information	17
IRR return will improve my knowledge of my condition	14
IRR return will satisfy my rights to my information	14
IRR return will help me cope with my situation	11
IRR return will demonstrate justice, fairness, or reciprocity	8
No further information provided	8
IRR return will alleviate research participant burden	5
IRR return will promote respect for persons and the research community	4
IRR return will facilitate family communication and social support	3
IRR return will raise concerns of stigma or discrimination	3
IRR return will make me feel good for helping others	2
IRR return will improve my spiritual well-being	1
IRR return will raise concerns of psychological harms	1