

## What matters to people and families affected by cerebral small vessel disease (SVD)? A qualitative grounded theory investigation

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### ABSTRACT

**Background:** Cerebral small vessel disease (SVD) is a common neurological disorder contributing to stroke, dementia, and disability. No treatment options exist although clinical trials are ongoing. We aimed to understand what matters to people and families affected by SVD to inform future research.

**Methods:** We thematically analysed unsolicited correspondences from members of the public addressed to members of the Edinburgh SVD Research Group on a variety of subjects related to SVD. We used inductive thematic codes, categorised under concerns, requests, emotions, and contributions, to form a grounded theory that categorised and ranked concerns raised.

**Results:** 101 correspondents expressed 346 concerns between August 2015 and February 2021, mostly via email. 60 correspondents (59.4 %) disclosed a SVD diagnosis, 39 (38.6 %) disclosed a previous stroke or TIA, and 40 (39.6 %) were family of people living with SVD. Primary concerns related to cognitive problems (number of correspondents (n)=43 (42.6 %)), lack of support or information from healthcare services (n = 41 (40.6 %)), prognosis (n = 37 (36.6 %)), sensory disturbances (n = 27 (26.7 %)), functional problems (n = 24, (23.8 %)), impact on daily life (n = 24 (23.8 %)), and causes of SVD (n = 19 (18.8 %)). 57 correspondents (56.4 %) expressed support for research, 43 (42.6 %) expressed an eagerness to understand SVD, 35 (34.7 %) expressed helplessness, and 19 (18.8 %) expressed frustration.

**Conclusions:** Cognitive decline was the main concern for people and families living with SVD who corresponded with the Edinburgh SVD research group. These findings also indicate a need for more accessible services and better information about SVD for patients and families.

### Introduction

Understanding and involving patients and families' values and experiences is a global priority for healthcare and essential to guide patient centred research [1]. Cerebral small vessel disease (SVD) is a common neurological disorder associated with ageing, stroke, cognitive decline, dementia, and characterised by neuroradiological markers on CT or MRI brain scans [2].

Stroke research has traditionally used hard outcomes of recurrent stroke, death or scales featuring physical disability such as the Rankin Scale [3]. Similarly, dementia research (predominantly into the field of Alzheimer's Disease) focusses on cognitive scores and to a lesser degree

function [4]. Previous research into what matters to patients has focussed on those affected by stroke or dementia – either patient priority setting partnerships in post stroke research priorities [5] or reviews of prioritisation exercises for those living with stroke [6] or cognitive decline [7]. SVD causes multiple other symptoms other than stroke or dementia, including atypical psychiatric symptoms [8]. Although SVD can cause stroke and dementia, patients and families with SVD may consider other outcomes more important and identifying these outcomes is essential to guide SVD research. We did not identify via systematic review any reports detailing what matters to patients with SVD.

We addressed this knowledge gap by analyzing correspondence from the public addressed to the University of Edinburgh SVD Research

Non-standard Abbreviations and Acronyms SVD (Cerebral) Small Vessel Disease Families Small vessel disease stakeholders including family members, friends, and carers who are neither people with small vessel disease, nor small vessel disease healthcare professionals

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Group. Based on these correspondances, we developed a theory of what matters to people living with SVD and their families.

## Methods

The Edinburgh SVD research group regularly receives unsolicited correspondence in a variety of different formats from members of the public. We store all correspondence for future reference. We collated all these correspondences (irrespective of the format e.g. email/written letter/telephone call transcription) that dealt with any aspect of the topic of cerebral small vessel disease between August 2015 and February 2021. We then analysed these correspondences using grounded theory [9] and associated a constructivist paradigm (see Supplement I for further details) to identify common themes and concerns that were expressed by the correspondents. This involved conducting ‘initial coding’ using questions such as “how are correspondents expressing what matters to them?”. Next, we conducted ‘focused coding’ which involved grouping and comparing the most significant initial codes to form main categories: ‘concerns’, ‘requests’, ‘emotions’, and a wish to ‘contribute to research’, and sub-categories (see Supplement II for further details). Correspondents could express multiple points within a sub-category. For example, if a correspondent was concerned about ‘progression to dementia’ and ‘stroke recurrence’, this was counted as two instances of ‘prognostic concerns’. This approach allowed all subject matters to be carefully acknowledged and accounted for to reflect the broad spectrum of SVD manifestations and personal circumstances. Using unsolicited correspondence allowed us to identify pertinent issues that patients actively sought answers or reassurance for, and highlighted areas that may not have been appropriately addressed through other sources of patient information.

We created and tested a standardized data extraction form before applying the coding framework to all correspondences (see Supplement III for further details). We defined a ‘correspondence chain’ as multiple correspondences from the same correspondent regarding the same subject. The last stage of coding involved ‘theoretical coding’ by analysing relationships between codes and forming a theory about what matters to people with SVD and their families.

Secondarily, we determined the main concern of each correspondence chain and compared responses based on the disclosed disease status: confirmed SVD diagnosis, previous stroke or TIA, both, or unspecified.

### Trustworthiness

The lead author kept a reflective diary guided by Lipp’s framework [10] which involved reflecting on the personal impact on the investigation and personal biases. The thematic codes were first generated by the lead author (SW), a medical student with no previous experience or expectations relating to SVD. These codes were verified by a stroke physician with training in and experience of qualitative techniques (FD) and a neuroradiology specialist (JW) with plentiful experience managing patients with cerebral small vessel disease. This ensured that codes accurately reflected the meanings within correspondences from multiple investigator standpoints. Using correspondences meant that the investigators had minimal influence on the data generation process, unlike with focus groups or interviews.

All personal data were kept secure and strictly confidential throughout the entire research process and all presented data were completely anonymised in line with local regulations and within the current research governance framework in the University of Edinburgh. Only the project investigators had access to correspondences which were only accessed if necessary for data extraction. Following assessment by the UKRI MRC HRA decision making tool [11], this project did not require NHS REC review for ethics.

## Results

We received 107 correspondence chains from 101 correspondents between August 2015 and February 2021. 94 (87.9 %) correspondences were emails, 6 (5.6 %) were letters, 5 (4.7 %) were records of a phone call, and 2 (1.9 %) were records of a direct interaction. We collected and included all correspondences received on the topic of SVD from the lay public, none were excluded. Of the 58 correspondents who disclosed their location, 42 were from the UK, 6 from mainland Europe and 10 from elsewhere. We recorded how the correspondent found out about the SVD research group – for the 50 correspondents where this was stated, 26 found our details online (8 through the British Heart Foundation website), 14 from a published scientific article, 5 from a printed newspaper or magazine, and 10 from a variety of sources (radio, podcast, Stroke Association conference, discussion with healthcare professional). Where stated, 61 correspondents contacted us regarding themselves, whilst 34 correspondents contacted us regarding a friend or relative with SVD. Among the 55 correspondents who stated their age, the median age of people with, or suspected to have, SVD was 56 years old. 60 correspondents disclosed that the individual had a confirmed SVD diagnosis via brain scan and 39 disclosed a previous stroke or TIA.

### Coding framework

Thematic codes were categorised under concerns, requests, (offers of) contributions, and emotions. We identified 526 themes during initial coding, from which we developed 71 focused codes. These were arranged into a hierarchy – 44 codes were grouped into 10 concern categories, 9 into the 6 requests categories, 13 into 3 contribution categories, and 5 emotion categories.

### Concerns

We identified 346 concerns with a median of two different concerns per correspondent (IQR 4, min 0- max 18). Table 1 one details the frequency of concerns expressed.

Cognitive problems were the most common concern (70/346 concerns from 43 (42.6 %) correspondents). People with SVD regarded memory essential for “being themselves” while families considered memory important for independence. Families were also concerned about unusual behaviours in relation to loss of self-control potentiating self-harm while people with SVD emphasised disinterest in hobbies.

“She cannot remember how to get to some shops and other places that she used to frequent” – Daughter of a patient with a confirmed diagnosis of vascular dementia.

Many correspondents were concerned by suboptimal support or information from healthcare services (62/346,  $n = 41$  (40.6 %)). Those with a confirmed SVD diagnosis felt dismissed after being told that SVD

**Table 1**

Table showing the most common ranked SVD topics of concerns arising from thematic analysis among 101 correspondents. Correspondents often raised more than one concern in one correspondence (median two concerns).

Concern	Total number of concerns raised	Number of correspondents
Cognitive problems	70	43
Healthcare services	62	41
Prognosis	55	37
Sensory disturbances	38	27
Functional problems	32	24
Impact on daily life	31	24
Causes of SVD	22	19
Pain	14	13
Impact on family	8	8
Fatigue	5	5

is “mild”, “just part of ageing”, or “not a major stroke”. Some felt that “people don’t seem to understand [their] difficulties”. Some of those without a confirmed SVD diagnosis were worried about having SVD brain scan markers which were recorded in their notes, but which no-one had discussed with them. Three correspondents expressed the burden of having multiple brain scans with little resultant explanation.

“No-one will talk to me about it... they just keep saying he hasn’t had a stroke.” – Partner of a patient with negative MRI findings for stroke and suspected diagnosis of SVD.

Concerns about long-term prognosis were common (55/346,  $n = 37$  (36.6 %)). Many referred to dementia as their “greatest fear”. Only six of 101 correspondents mentioned mortality, most of whom disclosed a family history of vascular dementia. Seven correspondents were concerned about future stroke events, of whom four had previously experienced a stroke.

“I have been told this condition is not life threatening [but] I am worried about my future.” – Patient with a confirmed diagnosis of SVD on MRI brain scan.

Sensory disturbances (38/346,  $n = 27$  (26.7 %)), such as vision problems and tingling, caused concern as they were thought to indicate worsening disease, although few considered sensory changes as their main concern. Functional problems (32/346,  $n = 24$ , (23.8 %)), such as poor mobility and reduced hand function, caused concern as they impinged on independence. Anxieties after receiving a SVD diagnosis were barriers to fulfilling working and social roles (31/346,  $n = 24$  (23.8 %)). In some cases, families stated that they felt more anxious than their relative with SVD. Ascribing cause to SVD was considered important for understanding the diagnosis (22/346,  $n = 19$  (18.8 %)), especially for young correspondents who considered themselves to be healthy. Some correspondents were concerned about pain and migraines (14/346,  $n = 13$  (12.9 %)) but acknowledged that these were reasonably managed. Some people with SVD expressed concern about the impact on family (8/346,  $n = 8$  (7.9 %)) such as how they would cope, and fatigue (5/346,  $n = 5$  (5.0 %)), which was considered to hinder healthy living.

“I don’t appear to be a person you would typically associate with stroke... I keep trying [to] figure out where I went wrong.” – Patient with a confirmed diagnosis of lacunar infarct on MRI brain scan.

“I know that I should do some exercise & probably eat better & I know that I should overcome the fatigue to ensure that I do both.” – Patient with a past medical history of stroke and epilepsy and a suspected diagnosis of SVD.

We report in Table 2 the concerns compared between different groups by disclosed disease status. In general, there was minimal variation depending upon whether the patient had SVD or a previous stroke with cognition, healthcare services, and prognosis featuring strongly.

## Requests

We identified 96 direct requests, the most common being for general

SVD information or advice (44/96,  $n = 44$  (43.6 %)). Correspondents also requested medication reviews (11/96,  $n = 10$  (9.9 %)), local expert referrals (10/96,  $n = 10$  (9.9 %)), private consultations (10/96,  $n = 10$  (9.9 %)), or brain scan reviews (7/96,  $n = 7$  (6.9 %)).

## Emotions

More than half of correspondents expressed gratitude towards healthcare staff and research ( $n = 57$  (56.4 %)) as it “brings hope and dignity [to patients]”. Many expressed eagerness to understand SVD ( $n = 43$  (42.6 %)), helplessness as to what to do next ( $n = 35$ , (34.7 %)), confusion as to what SVD means ( $n = 26$ , (25.7 %)) and frustration ( $n = 19$  (18.8 %)), often accompanied by concerns about the lack of patient-friendly information.

## Contributions

54 (53.5 %) correspondents wished to contribute towards SVD awareness and understanding, by participating in research ( $n = 45$  (44.6 %)), offering theories as to SVD pathophysiology and treatments ( $n = 10$  (9.9 %)), or offering anecdotes ( $n = 6$  (5.9 %)).

Please see Fig. 1 for an infographic depicting results.

## Discussion

In this study of unsolicited correspondences to the Edinburgh SVD research group from lay members of the public who have been affected by SVD we have demonstrated several emergent themes— major concerns about cognition, access to healthcare services, and a lack of information about prognosis. This is consistent with previous research priority setting partnerships in stroke as a whole that demonstrated cognition post stroke was a very highly ranked topic [5]. This also however varies from a systematic review of priority setting exercises in stroke that found rehabilitation to be the most common concern [6] (as would perhaps be expected in stroke survivors) illustrating that although there is overlap, patients affected by SVD are not the same as patients affected by stroke. However, whilst quality of life (QoL) in stroke survivors is multifactorial [12], most of our correspondents specifically attributed their difficulties to cognitive impairment, a prominent feature of SVD [2]. Interestingly, the concerns reported in our study differ slightly from international dementia research priorities where more attention is given to the needs of the care giver, support, and education about the disease [7]. In our study we did not have the granularity to delve into this question; nonetheless, this reflects that although overlap exists, patients affected by SVD may have different concerns from those affected by dementia.

Stroke recurrence and mortality were a less common concern in this group. A post-hoc analysis of the Secondary Prevention of Small Subcortical Strokes (SPS3) trial highlighted that patients who experienced a lacunar stroke suffered continuously worsening disability over time even without stroke recurrence as other patient-centred factors influence quality of life measures [13]. This may have been due to the

**Table 2**

Table showing different ranking of main concerns raised divided by disease status disclosed by the correspondent.

Code	Overall rank	Confirmed SVD diagnosis	Previous stroke/TIA	Previous stroke	Confirmed SVD diagnosis and previous stroke
Cognitive problems	1	1	1	1	1
Healthcare services	2	2	2	2	3
Prognosis	3	3	3	3	2
Sensory disturbances	4	5	5	7	6
Functional problems	5	4	4	4	4
Impact on daily life	6	6	6	6	7
Causes of SVD	7	7	7	8	5
Pain	8	8	10	9	9
Impact on family	9	9	9	10	10
Fatigue	10	10	8	8	8

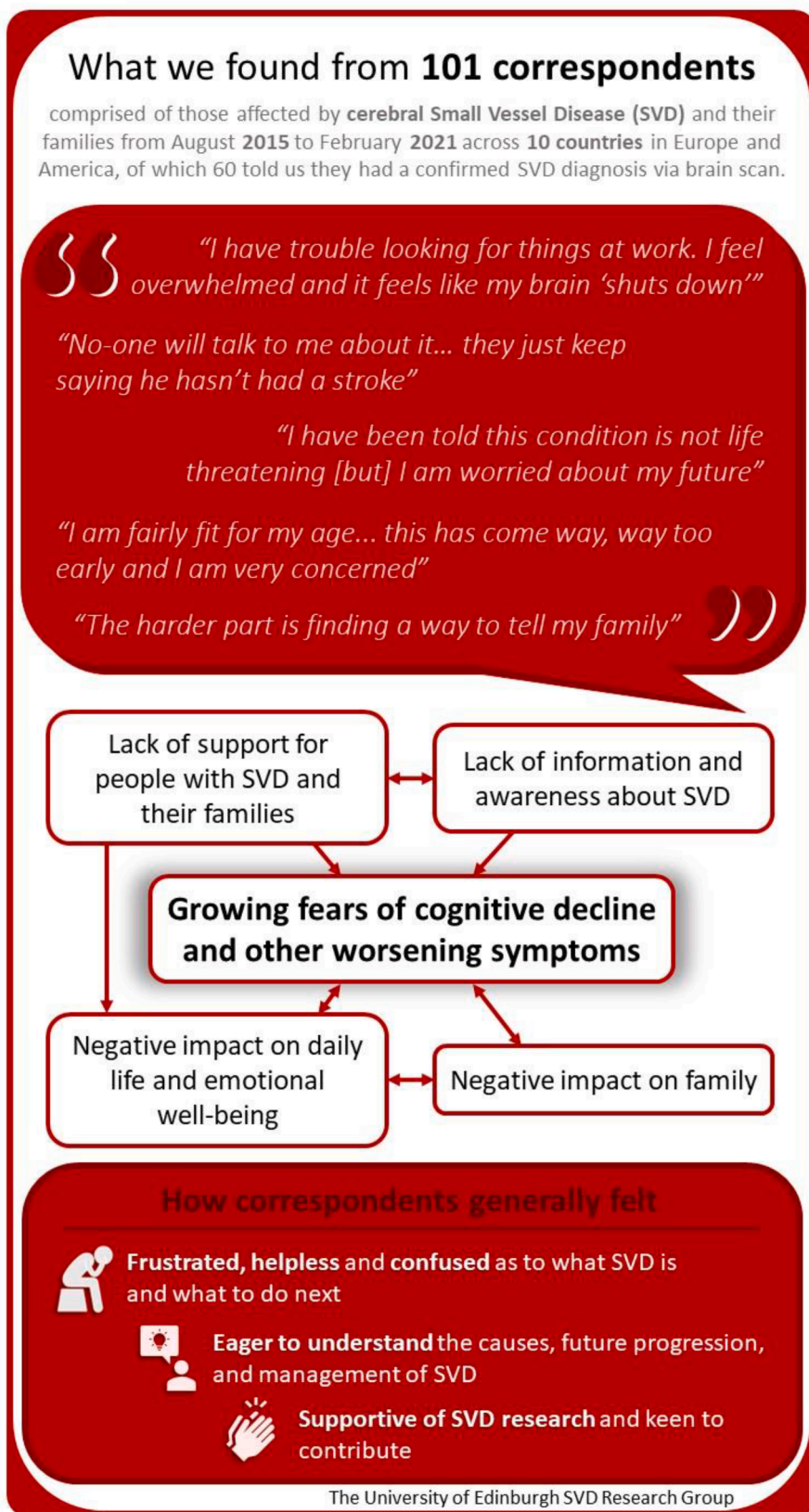


Fig. 1. Infographic summary of the findings of the study illustrating some of the quotes from the source material and the key messages.



progressive nature and low mortality of SVD compared to other stroke subtypes [2].

Concerns regarding a lack of information and support from healthcare services were prominent. This may stem from limited understanding about patient experience and SVD management among health professionals. Some family members exhibited higher levels of anxiety than their relative with SVD, perhaps because apathy is a key neuropsychiatric SVD symptom [14]. Stroke research highlights that receiving appropriate support is essential for effective rehabilitation and self-acceptance [5].

This study has certain strengths – the correspondences were numerous allowing for thematic analysis and were also unsolicited, therefore without the introduction of bias from the researchers as can exist in focus groups or semi-structured interviews. Persons affected by SVD are a relatively understudied population. The analysis was without preconceptions, therefore the themes that arose were introduced by the correspondents. We had a wide geographical spread and, where disclosed, a range of correspondents with different phenotypes.

The nature of this study however also inevitably leads to limitations, the main one being selection bias in that only those motivated enough or inclined or able to write will have corresponded. Since persons with SVD may suffer from apathy [14], this may lead to underrepresentation of those with more severe SVD. Moreover, we had no control over our sampling. Since the correspondences were unsolicited, we did not have a standard proforma for correspondents to complete, therefore there are inevitable episodes of incomplete data. This will limit the generalisability of these results.

These findings have implications for future research in that it should focus on patients concerns and will guide outcome assessment for future trials of promising therapies. Given the that lack of support and information from healthcare services followed cognition as a concern this indicates a need to focus on improving both clinical services and information sharing.

#### Grounded theory

By analysing correspondence addressed to the University of Edinburgh SVD Research Group over six years, we theorise that people affected by SVD and their families are most concerned about cognitive decline, lack of support and information from healthcare services and prognosis.

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#### Disclosures

None.

#### Declaration of competing interest

I declare that in accordance with the ICMJE guidance none of the above mentioned authors have any declarations of interest that could inappropriately bias the results of this paper. Please see individual ICMJE form submissions for further details.

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#### Supplementary materials

Supplementary material associated with this article can be found, in the online version, at [doi:10.1016/j.cccb.2024.100202](https://doi.org/10.1016/j.cccb.2024.100202).

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