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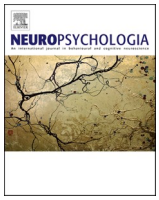

Examining the role of self-reported somatosensory sensations in body (dis) ownership: A scoping review and empirical study of patients with a disturbed sense of limb ownership

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ABSTRACT

Patients with a disturbed sense of limb ownership (DSO) offer a unique window of insight into the multisensory processes contributing to the sense of body ownership. A limited amount of past research has examined the role of sensory deficits in DSO, and even less is known regarding the role of patient self-reported somatosensory sensations in the pathogenesis of DSO. To address this lack of knowledge we first conducted a systematic scoping review following PRISMA-SR guidelines, examining current research into somatosensory deficits and patient self-reported somatosensory sensations in patients with DSO. Eighty studies, including 277 DSO patients, were identified. The assessment of sensory deficits was generally limited in scope and quality, and deficits in tactile sensitivity and proprioception were most frequently found. The reporting of somatosensory sensations was even less frequent, with instances of paraesthesia (pins-and-needles), stiffness/rigidity, numbness and warmth, coldness and heaviness amongst the deficits recorded. In a second part of the study, we sought to directly address the lack of evidence concerning the impact of patient self-reported somatosensory sensations in DSO by measuring DSO and self-reported somatosensory sensations in a large (n = 121) sample of right-hemisphere stroke patients including N = 65 with DSO and N = 56 hemiplegic controls. Results show that feelings of coldness and stiffness modulate DSO symptoms. Sense of heaviness and numbness are more frequent in patients with DSO but do not have a clear impact on disownership symptomology. Although preliminary, these results suggest a role of subjective sensations about the felt body in the sense of limb ownership.

1. Introduction

Contemporary research into self-consciousness that takes an 'embodied' approach seeks to understand how the body, through the processing of sensorimotor information, gives rise to fundamental aspects of the self, such as body ownership (i.e. the sense that "my body belongs to me") (Gallagher, 2000). A large and ever-growing number of psychological and cognitive neuroscience studies with healthy individuals have sought to examine how multisensory integration, amongst other factors, contributes to the sense of body ownership (Kilteni, 2015). In parallel to these highly-informative studies of healthy individuals, neuropsychological studies have provided a unique form of evidence from patients with a disturbed sense of limb ownership (DSO), with specific references to the patient's upper and lower paralysed limbs

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(Baier and Karnath, 2008). DSO is a label mainly used in research setting that clusters symptoms that often co-occur, such as asomatognosia (i.e. a deficit in the feeling or judgment that my body belongs to me and is ever present, Jenkinson et al., 2018) and somatoparaphrenia (i.e. the occurrence of illusional, confabulatory or delusional ideas of disownership or misidentification regarding the affected body parts, such as supernumerary limb, attribution of the body part to another person, personification or objectivation of body parts, Jenkinson et al., 2018). Patients with DSO allow fundamental questions about the nature of body ownership to be addressed in a manner that is impossible via studies that include only health individuals, where the sense of body ownership can be temporarily manipulated but is never completely absent. Thus, patients with DSO allow the mechanisms of body part ownership to be examined and provide additional evidence to that obtained from studies of healthy individuals.

A key area that has long been examined is the role of sensory feedback in limb ownership. Early work investigated the role of individuals sensory channels, such as visual, proprioceptive and somatosensory signals to body ownership (Vallar, 1997; Rossetti and Rode, 2002). Subsequent work has highlighted the importance of multisensory integration of exteroceptive (i.e. external, Zampini et al., 2004; Bolognini et al., 2014; Romano and Maravita, 2019) and interoceptive (internal, Michael et al., 2015; Martinaud et al., 2017; Echalier et al., 2020; Jenkinson et al., 2020) sensory information, as well as higher-order beliefs and representations in body ownership (Apps and Tsakiris, 2014; Fotopoulou, 2015; Samad et al., 2015). For example, Martinaud et al. (2017) suggested that sensations (such as feelings of heaviness, numbness, coldness, and other similar sensations), which are frequently observed in DSO patients in clinical practice, may generate error signals that cannot be integrated into the patient's current body representation. These sensations are not predicted by existing, top-down expectations that arise from outdated representations of the body and cannot be integrated into current body representations. The resulting mismatch between predicted and actual body sensations contributes to feelings of limb disownership (see Apps and Tsakiris, 2014 for further background to these ideas, predictive coding, and self-recognition). Subsequent work by Jenkinson et al. (2020) supported this hypothesis, showing that body ownership was improved when CT-optimal touch (i.e. a specific kind of slow, gentle touch that is associated with subjective feelings of pleasure and has been redefined as an interoceptive modality; see Craig, 2003) was administered to the affected (disowned) limb of DSO patients. The proposed mechanism underlying this improvement was a reduction in feelings of numbness that in these patients arise from a mismatch between seen and felt sensory information (see Longo et al., 2008; Roel Lesur et al., 2020) and prevent the limb from being integrated into the existing, multimodal, body representation. Finally, a few of single case studies suggest that experimental induced multisensory integration and visuo-tactile stimulation ameliorate DSO (Bolognini et al., 2014; D'Imperio et al., 2017). However, further studies that include systematic and comprehensive assessment of somatosensory sensations in patients with and without DSO are needed to substantiate these ideas.

Indeed, despite a growing number of studies, the role of somatosensory deficits and in particular patient-reported somatosensory sensations (i.e. reports of coldness, numbness, pain, pins-and-needles, etc.) remain poorly understood. In fact, although existing work that reports somatosensory deficits in DSO has been previously summarised by two comprehensive (though not systematic) reviews (see Vallar and Ronchi, 2009; and more recently Romano and Maravita, 2019), it is unclear what information is available in the existing literature about patient self-reported somatosensory sensations. With this in mind, we tested the hypothesis that subjective somatosensory sensations play a role in generating DSO. As a first step a scoping review was conducted to summarise the extant research in this area, and to identify any gaps in existing knowledge. We examined what is known from the literature about somatosensory deficits and self-reported somatosensory sensations in brain damaged patients and their impact on DSO symptoms. Based on the findings of this scoping review, an empirical study was devised that specifically aimed to systematically screen self-reported somatosensory sensations in patients with various symptoms of DSO. A new tool for the assessment of DSO was developed along with a questionnaire to capture the range of somatosensory sensations reported by patients following stroke (the Somatosensory Sensations Questionnaire; SSQ). The impact of somatosensory sensations and other clinical variables on DSO symptoms was then analysed to test our hypothesis.

2. PART 1: scoping review

2.1. Methods

Our review was guided by the PRISMA-ScR reporting items (Tricco et al., 2018, see SM - 1 for details) and 5-step framework of (Arksey and O'Malley, 2005), involving the following stages: 1) identifying the research question, 2) identifying relevant studies, 3) study selection, 4) charting the data, and 5) collating, summarising and reporting the results. The protocol for conducting the review was developed by VM and MS in consultation with PMJ and AF, based on the existing work of (Vallar and Ronchi, 2009; Romano and Maravita, 2019), but was not pre-registered. Details are provided below.

2.1.1. Search strategy

The review focused on what is known from the literature about somatosensory deficits and self-reported somatosensory sensations in brain damaged patients with DSO. To identify relevant studies we examined the reference list of two previous reviews that sought to summarise existing literature on DSO (i.e. Romano and Maravita, 2019, which contains studies published up until 2017, and Vallar and Ronchi, 2009, which contains studies published up until July 25, 2008, and is also included in the review of Romano and Maravita, 2019). We also conducted our own search of the literature commencing from 2018 (i.e. the end point of Romano and Maravita, 2019) and including all studies published until 2022. This new search was performed in PubMed, entering the following keywords: "Somatoparaphrenia" OR "Disownership" OR "Misoplegia" OR "Asomatognosia" OR "Main etrangere" OR "Alien hand" AND "Body sensations" OR "Sensory system". Further studies were identified from reading the articles identified from our searches and checking their reference lists.

2.1.2. Inclusion/exclusion criteria

Selection of relevant studies was determined by inclusion/exclusion criteria, which were developed by the authors and guided by the research question "what sensory deficits, self-reported somatosensory sensations, and clinical variables impact on DSO symptoms?". To be included in the review papers needed to report data on patients suffering from focal brain damage (i.e. stroke, tumour) with subsequent DSO. For the new search, peer-reviewed journal papers were included if they were published between the period of 2018–2022, were written in English, included human participants and reported a clinical or psychometric measure of DSO. Both single case and group studies were included. Papers were excluded if they focused exclusively on syndromes different from DSO (e.g. anosognosia for hemiplegia, personal neglect), or non-focal damages (e.g. traumatic brain injury, dementia, degenerative conditions, peripheral nervous system lesions, amputations).

2.1.3. Screening and selection

After removing duplicates and non-English literature, the eligible records underwent a first selection based on title and abstract scanning. The full text of papers identified as relevant were retrieved and further assessed for relevance. All these steps were conducted independently by two of the authors (VM and VG). In case of a disagreement, a third (MB) author was consulted for the final decision. The search and studies selection followed the PRISMA 2020 guidelines (Page et al., 2021), as illustrated in the flow-chart (Fig. 1).

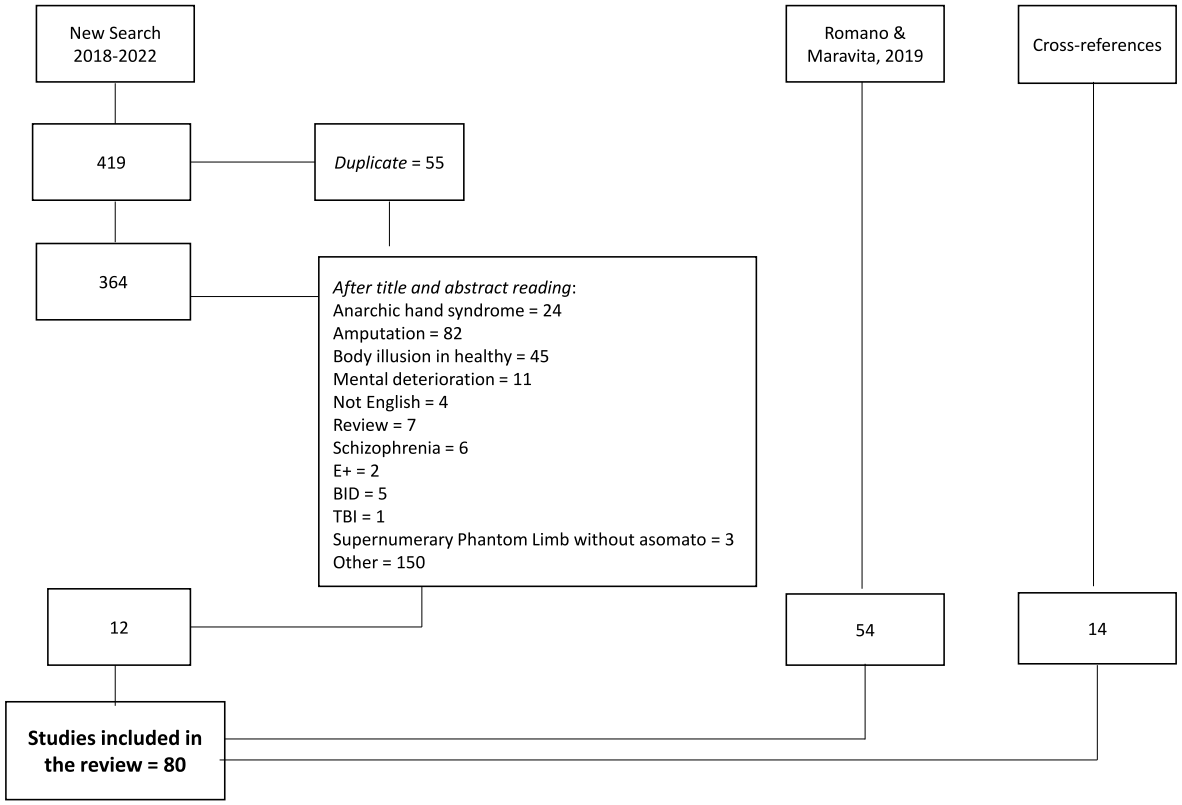

Fig. 1. PRISMA Flow diagram of the steps followed in the literature research from the three sources (new search of papers between 2018 and 2022; papers reported in Romano and Maravita, 2019; and cross-references) and screening of the articles.

2.1.4. Data extraction

Data were extracted and charted using a standardised template which collected data on: authors, year of publication, lesion (i.e. stroke or tumour, side of lesions), DSO patient sample size, somatosensory deficits, and patient self-reported somatosensory sensations. Other reported feelings regarding the paralysed arms were reported as well. At the end of the process, the total number of papers was 80 and the total number of patients taken into consideration were 277 (see Fig. 1).

2.1.5. Data synthesis

We collated data into a table (Table 1), which summarises whether somatosensory deficits (i.e. tactile sensitivity and proprioception) or self-reported sensations (i.e. pain, numbness, heaviness, warmth, cold, needles, stiffness/rigidity) were reported as being present (+), absent (−), or not reported (nr) in each paper and for single patient. Every other feeling described in the original studies was reported as well. We calculated the percentages of patients reporting sensory deficits based on the total number of patients in which it was investigated (i.e. the sum of "+", i.e. the sensation is present, and "-", i.e. the presence of the sensation is investigated and not present).

3. Results and discussion

Our review indicates that DSO is mainly reported after vascular damage, and only rarely in patients suffering from cerebral tumour (2.53%). The symptom is described as localized in the upper limb in 97.12% of cases, in the lower limb in 1.44% and in the whole left side of the body in 1.44% of patients. In general, DSO follows right hemisphere lesions, but 3.61% of patients described were left-hemisphere damaged.

Sensory systems, in particular tactile sensitivity and proprioception are typically investigated in DSO patients as part of routine clinical examinations (in 3.62% and 10.8% data not reported, respectively) and often found to be compromised (93.26% of patients examined for tactile sensitivity and 95.95% for proprioception). By contrast, patient self-reported somatosensory sensations were found to be very poorly investigated, in terms of both reporting frequency and the extent of the symptoms (Table 1).

To sum up, the results of our scoping exercise reveal several gaps and limitations in the current knowledge base. Firstly, the fact that sensory deficits are typically only assessed as part of routine clinical assessments (i.e. relying on brief procedures rather than a comprehensive assessment of sensory functions) is problematic, since such assessments may not be sensitive enough to identify the full range or severity of sensory deficits. A second, related point is that clinical assessments typically focus on specific sensory domains (in particular, proprioception and tactile sensations) but seldom investigate other domains or subjective sensations that might be affecting the patient (e.g. kinaesthetic or thermal sensations).

Crucially for the aim of the current study, patient self-reported somatosensory sensations are particularly neglected, and when patients do report aberrant feelings, these are seldom followed up with a specific, in-depth investigation. A related problem is that a systematic bias in the reporting of somatosensory sensations was present in the existing literature, as only spontaneously self-reported sensations are recorded, without a specific, systematic screening. In this way, only "positive" cases, namely when the symptom is described by the DSO patients, are reported in the literature on the topic, while no data are available on "negative" cases, i.e. DSO patients that when specifically asked report the absence of somatosensory sensations. Only systematic investigations would allow the identification of potential double dissociations between DSO and self-reported sensations, which would be fundamental to understanding the possible impact of these sensations in DSO. With this in mind, we conducted a study to assess self-reported somatosensory sensations in hemiplegic patients with DSO and without DSO.

Table 1
Sensory deficits and somatosensory sensations associated with disturbed sense of limb ownership.

	Clinical data				Sensory		Sensations							others
	n. DSO	etiology	lesion side	limb	T	P	pain	numbness	heaviness	warmth	cold	needles	stiffness (rigid)	
Moro et al. (2023)	23	stroke	RBD	UL	nr	nr	nr	nr	nr	nr	nr	nr	nr	nr
Errante et al. (2022)	4	stroke	RBD	UL	3+	4+	nr	nr	nr	nr	nr	nr	nr	E+
Maggio et al. (2021)	1	tumor	LBD	LL	-	+	nr	nr	nr	nr	nr	nr	nr	misoplegia, capricious, “alien and separate from me”, “an uncooperative enemy"
Saetta et al. (2021a)	1	stroke	RBD	UL	+	nr	–	nr	nr	nr	nr	nr	nr	nr
Saetta, et al. (2021b)	1	tumor	RBD	UL	+	–	nr	+	nr	nr	nr	nr	+	nr
Yamada et al. (2021)	1	stroke	RBD	UL	+	+	+	nr	nr	nr	nr	nr	nr	nr
Ronchi et al. (2020)	8	stroke	RBD	UL	6+	nr	nr	nr	nr	nr	nr	nr	nr	nr
Spitoni et al., 2016	10	stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	nr
Randev et al. (2019)	1	stroke	RBD	UL	–	–	nr	nr	nr	nr	nr	nr	nr	nr
Sakamoto et al. (2019)	1	stroke	RBD	UL	–	+	nr	nr	nr	nr	nr	nr	nr	nr
Romano and Maravita (2019)	3	stroke	RBD	UL										
pt.1		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
pt.2		stroke	RBD	UL	–	+	nr	nr	nr	nr	nr	nr	nr	nr
pt.3		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Salvato et al. (2018)	1	stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	nr
D’Imperio et al. (2017)	1	stroke	LBD	UL	+	+	+	nr	nr	nr	nr	nr	nr	nr
Martinaud et al. (2017)	7	stroke	RBD	UL	nr	+	nr	nr	nr	nr	nr	nr	nr	nr
Salvato et al. (2016)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Spitoni et al. (2016)	1	stroke	RBD	UL	–	–	+	+	+	nr	nr	nr	nr	nr
Moro et al. (2016)	17	stroke	RBD	UL	nr	nr	nr	nr	nr	nr	nr	nr	nr	nr
Perren et al., 2014	1	stroke	RBD	LL	+	+	nr	nr	nr	nr	nr	nr	nr	a piece of wood, not feel it, misoplegia
Romano et al. (2014)	5	stroke	RBD	UL	+	+4	nr	nr	nr	nr	nr	nr	nr	nr
Bolognini et al. (2014)	2													
pt 1		stroke	RBD	UL	+	+	–	nr	nr	nr	nr	nr	nr	nr
pt 2		stroke	RBD	UL	+	+	–	nr	nr	nr	nr	nr	nr	nr
Invernizzi et al. (2013)	5													
pt 1		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
pt 2		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
pt 3		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
pt 4		stroke	RBD	UL	+	+	nr	nr	+	nr	nr	nr	nr	nr
pt 5		stroke	RBD	UL	–	+	nr	nr	nr	nr	nr	nr	nr	fat, puffy and disobedient
Ronchi et al. (2013)	1	stroke	LBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Gandola et al. (2012)	11													
pt 1		stroke	RBD	UL	+	nr	nr	nr	nr	nr	+	nr	nr	sweaty, plump
pt.2		stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	
pt3		stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	unconfortable, artificial
pt 4		stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	too short
pt 5		stroke	RBD	UL	+	nr	nr	nr	+	warm	nr	nr	nr	nr
pt 6		stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	on my stomach

(*continued on next page*)

Table 1 (*continued*)

	Clinical data				Sensory		Sensations							others
	n. DSO	etiology	lesion side	limb	T	P	pain	numbness	heaviness	warmth	cold	needles	stiffness (rigid)	
pt 7		stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	a puppet
pt 8		stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	nr
pt 9		stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	does not work
pt 10		stroke	RBD	UL	+	nr	nr	nr	+	nr	nr	nr	nr	on my stomach
pt 11		stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	too small, on the bed
Zeller et al. (2011)														
	12	stroke	RBD	UL	–	nr	nr	nr	nr	nr	nr	nr	nr	nr
	6	stroke	LBD	UL	–	nr	nr	nr	nr	nr	nr	nr	nr	nr
Fotopoulou et al. (2011)	2	stroke	RBD	UL										
pt 1 (also reported in Jenkinson et al., 2013)		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
pt 2		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Pugnaghi et al. (2012)	1	stroke	RBD	UL	nr	nr	nr	nr	nr	nr	nr	nr	+	nr
Van Stralen et al. (2011)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	misoplegia
Feinberg et al. (2010)	13	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Beato et al. (2010)	1	tumor	LBD	UL	+	–	nr	nr	nr	nr	nr	nr	nr	strangness
Baier and Karnath (2008)	11	stroke	RBD	UL	nr	nr	nr	nr	nr	nr	nr	nr	nr	nr
Srivastava et al. (2008)	1	stroke	RBD	UL	–	+	nr	nr	nr	nr	nr	nr	nr	more painful supernumerary limbs
Loetscher et al. (2006)	1	tumor	RBD	LL	–	–	nr	nr	nr	nr	+	nr	nr	misoplegia, anosognosia, personification
Moro et al. (2004)	2													
pt 1		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
pt 2		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Paysant et al. (2004)	14	stroke	RBD	UL	12+	+11	nr	nr	nr	nr	nr	nr	nr	nr
Cereda et al. (2002) (case 4)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Bottini et al. (2002)	1	stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	nr
Paulig et al. (2000)	1	stroke	RBD	UL + LL	+	nr	nr	nr	nr	nr	+	nr	nr	nr
Feinberg et al. (2000)	5	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	illusory limb movements (5)
Daprati et al. (2000)	1	tumor	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Schiff and Pulver (1999)	1	stroke	LBD	UL	nr	nr	nr	nr	nr	nr	nr	nr	nr	nr
Aglioti et al. (1996)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Halligan et al. (1995)	1	stroke	RBD	UL	+	+	nr	nr	+ (foot)	nr	nr	+	nr	detachment, alienation, cow's foot, willies, foreign
Halligan et al. (1993)	1	stroke	RBD	UL	+	+	nr	nr	nr	+ (the third arm)	+ (the third arm)	nr	nr	iti's like a sack of coal, detached, dead, artificial
Rode et al. (1992)	1	stroke	RBD	UL	+	+	nr	nr	+	nr	nr	nr	nr	kind
Levine et al. (1991)	2													
pt 4		stroke	RBD	UL	+	+	nr	nr	+	nr	nr	nr	nr	husband's hand
pt 6		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	objectivation

(*continued on next page*)

Table 1 (*continued*)

	Clinical data				Sensory		Sensations							others
	n. DSO	etiology	lesion side	limb	T	P	pain	numbness	heaviness	warmth	cold	needles	stiffness (rigid)	
Bisiach et al. (1991)	1	stroke	RBD	UL	+	+	nr	nr	nr	warm	nr	nr	nr	her mother's hand, strong
Bisiach and Geminiani (1991)	1	stroke	RBD	UL	+	+								
Bisiach et al., 1990	1	stroke	RBD	UL	nr	nr	nr	nr	nr	nr	nr	nr	nr	nr
Feinberg et al., 1990	12	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	personification, objectivation
Starkstein et al. (1990)	2													
Case 1		stroke	RBD	UL	–	–	nr	nr	nr	nr	nr	nr	nr	disjointed, and separated, motor conflict
Case 2		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Assal (1983)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Healton et al., 1982	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Nightingale (1982)	1	tumor	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	nr
Verret and Lapresle (1978)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	etrangere
Cutting, 1978 (11RBD + 3 LBD)														
RBD	18	stroke	RBD	UL	+	+	nr	nr	nr	nr	3+	nr	nr	"It disobeyed me" ""I've got no feeling" ""My fingers shrank to short fat fingers" lifeless and clammy"), misoplegia personification, kinaesthetic illusions
LBD	9	stroke	LBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	misoplegia
Frederiks (1963)	3													
pt 1		stroke	RBD	UL + LL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
pt 2		stroke	RBD	UL + LL	–	–	nr	nr	nr	nr	nr	nr	nr	supran legs are heavy and tired
pt 3		stroke	RBD	UL + LL	+	–	nr	nr	nr	nr	nr	nr	nr	supran- legs are fleshy, real and painful
Hécaen et al., 1954	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	personification
Weinstein et al. (1954) (pt. 1)	1	stroke	RBD	UL	–	+	nr	nr	+	+	nr	nr	nr	Thicker, fatter, darker, bigger
Weinstein and Kahn, 1950 (pt 7)	1	tumor	RBD	UL	–	+	nr	nr	nr	nr	nr	nr	nr	nr
Roth (1949)	2													
pt 1		stroke	RBD	UL	–	+	nr	nr	nr	nr	+	nr	+	paraesthesia
pt 2		stroke	RBD	UL	–	+		nr	+	nr	nr	nr	nr	pressing on his body +
Sandifer (1946)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	facial expression of pain but verbal deny
Halloran (1946)	1	stroke	LBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Gerstmann, 1942 (pt 2)	1	stroke	RBD	UL	+	+	+	nr	+	+	nr	nr	nr	paraesthesia
Wortis and Dattner (1942)	1	stroke	RBD	UL	+	+	nr	nr	+	nr	nr	nr	nr	smaller
Rubinstein (1941) (pt 4)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	The head of a reptile, queer, strange, dead
Nielsen (1938) (pt 8)	1	stroke	LBD	UL	+	+	nr	+	+	nr	nr	nr	nr	water logged

(*continued on next page*)

Table 1 (*continued*)

	Clinical data				Sensory		Sensations							others
	n. DSO	etiology	lesion side	limb	T	P	pain	numbness	heaviness	warmth	cold	needles	stiffness (rigid)	
Von Hagen and Ives (1937)	3													
pt 1		stroke	RBD	UL	+	+	+	nr	nr	nr	nr	nr	nr	that's an old man, a spirit
pt 2		stroke	LBD	UL	+	+	nr	+	+	nr	nr	nr	nr	arm not attached, waterlogged
pt 5		stroke	RBD	UL	+	+	+	+	+	nr	nr	nr	nr	
Olsen, 1937 (quoted by Nielsen, 1938)	1	stroke	RBD	UL	nr	nr	nr	nr	nr	nr	nr	nr	nr	"But my eyes and my feel¬ ings don't agree, and I must believe my feelings. I know they look like mine, but I can feel they are not, and I can't believe my eyes
Lhermitte and Tcherazi (1937)	1	stroke	RBD	UL	nr	nr	nr	nr	nr	nr	nr	nr	nr	nr
Ives and Nielsen (1937)	2													
pt 1		stroke	RBD	UL	+	nr	nr	nr	nr	nr	nr	nr	nr	nr
pt 2		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	the arm does not seem as his own, stuck on
Potzl, 1935 (quoted by Lhermitte, 1952)	2													
pt 1		stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	long, dead, as a snake
pt 2		stroke	RBD	UL	nr	nr	nr	nr	nr	nr	nr	nr	nr	nr
Schilder (1935) (case b)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	fingers fat and swallen
Barkman (1925)	1	stroke	RBD	LL	+	+	nr	nr	nr	nr	+	nr	nr	nr
Ehrenwald, 1930 (quoted in Assal, 1983)	1	stroke	RBD	UL	nr	nr	nr	nr	nr	nr	nr	nr	nr	a monster, a stump, a prosthetic
Barré et al., 1923	1	stroke	RBD	UL	–	–	nr	nr	nr	nr	nr	nr	nr	nr
Kramer, 1915 (quoted in Barkman, 1925, obs V)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Zingerle (1913) (quoted in Benke et al., 2004)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Biancone, 1909 (quoted in Lhermitte, 1952)	1	stroke	RBD	UL	+	+	nr	nr	nr	nr	nr	nr	nr	nr
Anton (1893) (quoted in Benke et al., 2004)	1	stroke	RBD	UL	+	+	+	nr	nr	nr	nr	nr	nr	nr

Various different measures of Asomatognosia/DSO (e.g. clinical assessment, neuropsychological scales, VAS) were used in the studies (reported in the table from the most recent to the oldest). Symptoms are here reported as present (+) or absent (−) regardless the methods used in the assessment. T = tactile sensitivity; P = proprioception; nr = not reported; UL = upper limb; LL = lower limb; n.+ = in group studies, the number of patients with the symptom/sensation; other = other feelings reported by the patients. Despite widespread research, some of the oldest studies were not available to us but these were considered when the quotations were detailed enough in secondary sources.

4. Part 2: assessing self-reported somatosensory sensations in DSO

4.1. Methods

4.1.1. Participants

121 right hemisphere stroke patients (F = 57; age M = 63.72, SD = 33.3, range = 36–88; education M = 10.95, SD = 4.08, range = 5–17) were recruited at two stroke units located in Italy (Rehabilitation Unit, IRCSS Sacro Cuore Hospital, Negrar, Verona) and in the United Kingdom (Acute Stroke Rehabilitation Unit, St. Thomas Hospital, London). A subset of patients had previously taken part in three studies (Pacella et al., 2019; Moro et al., 2021, 2023). All patients met the following inclusion criteria: (i) unilateral right hemisphere damage, secondary to a first ever stroke, as confirmed by clinical neuroimaging (MRI or CT) and (ii) paralysis of the contralesional upper limb (Medical Research Council, 1976 – MRC Scale = 0). Exclusion criteria were: (i) previous history of neurological or psychiatric illness; (ii) medication with severe cognitive or mood side-effects; and (iii) severe language, general cognitive impairment, or mood disturbance that precluded completion of the assessment. In 24 patients the damage was haemorrhagic, while in the others there was an ischemic lesion, and the examination was carried out at different intervals from the lesion onset (mean = 38.97 days, SD = 43.27, range = 1–222). All patients gave written, informed consent and the research was conducted in accordance with the guidelines of the Declaration of Helsinki (World Medical Association, 2013) and approved by the Local Ethical Committees of each location.

4.1.2. Neuropsychological assessment

A neuropsychological battery was administered to assess general cognitive functions (MMSE, Folstein et al., 1975), executive functions (FAB, Dubois et al., 2000), short-term memory (verbal span forward, Baddeley et al., 1975), extrapersonal and personal neglect (Line Crossing, subtest of the Behavioural Inattention test, Wilson et al., 1987 and Comb test, subtest of the Comb and Razor test McIntosh et al., 2000), and anosognosia for hemiplegia (MUNA, Moro et al., 2021). Proprioception was assessed by asking the patients to state whether or not they felt a passive movement administered to an upper limb joint (i.e. index finger, wrist, and elbow; Vocat et al., 2010). See Table 2.

4.1.3. Disturbed sense of limb ownership

To assess DSO, 18 questions were selected to capture various different aspects of aberrant limb ownership reported in the existing literature (Table 3 and SM - 2 for details on the sources consulted to develop the questionnaire). This provides two types of scores: 1) a preliminary Limb Disownership Score, which mirrors the assessment of DSO as carried out during clinical routine (Feinberg et al., 2010), and 2) a DSO Score, which provides a comprehensive characterisation of various elements that make up disturbed sensations of body part ownership (Baier and Karnath, 2008). Specifically, after the first three questions (Q1-Q3), 5 questions (Q4-Q8) refer to the sense of upper limb presence and position (also controlling for the actual position of the arm in space, Q8); 5 questions investigate phenomena of personification and objectification (Q9-Q14); 2 questions assess the presence of supernumerary phantom limb (Q15-Q16). 2 other questions (Q17-Q18) refer to the ipsilesional arm and investigate sensations of increased strength in the healthy arm (Marcel et al., 2004; Cocchini et al., 2022). These were not considered in the final scoring since they refer to functioning of the ipsilesional arm rather than disturbances relating to the contralesional arm (Table 3).

The score for each item ranges from 0 to 2, with 0 meaning that: i) the symptom is absent, and the patient recognizes the arm as belonging to him/her (items Q1-Q3); ii) the specific feeling is not reported (Q4-Q15); iii) the patient correctly reports having two arms (Q16). The score 1 is attributed to uncertain answers (i.e. symptom slightly present) indicating doubts about ownership and sensations, while the score 2 indicates the presence of clear signs of disownership, the attribution of the arm to somebody else, symptoms of personification or objectivation, supernumerary arm.

The Preliminary Limb Disownership Score calculated using the standard questions asked during initial clinical investigations (Feinberg et al., 2010, i.e. the responses to items Q1-Q3) allowed us to divide the patients into different groups (No disownership, Mild, Moderate and Severe disownership – SM – 3 for details). The remaining 16 items were then used to calculate the full 'DSO Score'. This score was computed following a previously established item weighting method used to develop other neuropsychological measures (Bizzozero et al., 2000), as the ratio between the frequency of each symptom in the Severe disownership group and the frequency of the no Disownership group (see SM – 4 for details). Then, the rank of these ratios was used as weighting scores, obtaining the highest weights for symptoms more frequent in the severe group and less frequent in the no Disownership group (See SM – 4 for details). Each raw score was multiplied by its weight to calculate the weighted scores and the final DSO score was computed as the sum of the weighted scores. Finally, a ROC analysis was performed to identify the DSO Scores and the cut-offs able to distinguish between the disownership severity groups (SM – 4). In order to control for the consistency of the two steps of the assessment (the first three questions on Disownership and the other questions of the DSO questionnaire (i.e. the three questions used to distinguish the DSO groups were also included in the

Table 2
Demographic and clinical data of the patients recruited for the study. The mean and (standard deviations) of the four groups emerging from the preliminary analysis on the scores at the DSO questionnaire are reported (see the text for details) along with the ranges of scores. NO Disownership = absence of the symptoms in the Preliminary Disownership Score; MILD, MODERATE and SEVERE refer to the different severity of disownership symptoms; Interval (d) = days between the lesion onset and neuropsychological assessment; Vocat = proprioception assessment; MMSE = Mini-mental state examination; FAB = Frontal Assessment Battery; Line Canc = line cancellation; Comb = the comb subtest of the Comb and Razor test; MUNA = Motor Unawareness Assesment. Statistical tests (t-tests or χ^2) showed no significant differences among groups.

	NO Disownership		MILD		MODERATE		SEVERE	
	M (SD)	range	M (SD)	range	M (SD)	range	M (SD)	range
age	66.51 (12.6)	34–88	59.7 (14.48)	36–89	67.83 (13.63)	44–97	63.61 (15.18)	41–86
education	10.06 (3.94)	5–17	11.56 (4.5)	5–20	11.26 (3.96)	5–18	12.15 (4)	5–18
interval	48.98 (48.55)	1–222	30.48 (38.73)	1–160	30.64 (33.98)	1–120	35 (44.03)	1–171
Lesion Size	105695.70 (103330.44)		110342.52 (116550.51)		130745.73 (141222.67)		126831.07 (125536.63)	
Vocat	4.86 (3.62)	0–9	5.15 (2.76)	2–9	7.05 (1.85)	4–9	5.44 (2)	2–9
MMSE	23.1 (4.70)	10–30	24.27 (4.88)	12–30	22.64 (4.62)	12–30	23.12 (5.48)	16.4–29
FAB	9.5 (3.86)	3–15.9	9.1 (4.57)	0–16	9.59 (3.64)	2–14	10.39 (3.49)	2–14
Digit span	6.83 (2.78)	1–13	6.48 (2.06)	4–12	6.43 (2.04)	4–12	5.5 (1.65)	4–12
Line Crossing	24.54 (12.11)	1–36	26 (10.67)	6–36	23.52 (11.74)	2–36	16.55 (12.9)	2–36
Comb	−0.44 (1.05)	(-6) - (0.84)	−0.35 (0.3)	(-0.7) - (0.14)	−0.31 (0.25)	−0.75 - (0.21)	−0.29 (0.32)	−0.75 -(0.21)
MUNA	13.92 (8.71)	0–33	10.89 (10.19)	0–27	9.4 (6.61)	1.5–25	11.46 (10.82)	1.5–25

Table 3
The Disturbed Sense of Ownership questionnaire. Each item is scored with 0 = when the symptom is absent and the patient recognizes the arm as belonging to him/her (items Q1-Q3); the specific feeling is not reported (Q4-Q15); or the patient correctly reports having two arms (Q16); 1 = uncertain answers (i.e. symptom slightly present) indicating doubts about ownership and sensations; 2 = the symptom is clearly present. Qualitative notes on the patient's specific reports are also taken. For the final score, the scores (0,1, 2) from each item (Q 1-Q16) are multiplicated for the weight of the item (which is reported in the column "rank"). Finally, the sum of the scores of Q1-Q16 items is calculated for the final total score.

Disturbed Sense of Limb Ownership Questionnaire			score 0-2	Rank
Preliminary Limb disownership score	Q1	Is this your hand?		6
	Q2	Does it ever feel like it does not belong to you?		15
	Q3	Does it ever feel like it belongs to someone else? If yes, ask: "Anyone in particular?"		16
Presence and spatial position	Q4	Does it ever feel like your arm is not attached to your shoulder?		11.5
	Q5	Does it ever feel like your arm in elsewhere in the room/in space?"		14
	Q6	Do you ever feel that your arm is missing?		8
	Q7	Do you ever feel that your arm has disappeared?		11.5
Place the patients left hand in their right visual field, on the table,	Q8	What is this?"; "Is that your hand?"		3
Personification and objectivation	Q9	Do you ever call it names?		4
	Q10	Does it ever feel your arm is a separate person?		2
	Q11	Does it ever feel like your arm is a pet or a child?		9.5
	Q12	Is your arm ever naughty?		1
	Q13	Does it ever feel like your arm is an object?		7
	Q14	Does it ever feel like your arm is an alien, or foreign arm?		13
Supernumerary limb	Q15	Does it ever feel a third arm is lying beside your left arms?		5
	Q16	How many arms do you have?		9.5
Overestimation of the ipsilesional limb (not scored)	Q17	Does your right arm feel particularly strong?		
	Q18	Do your senses in your right side of your body feel heightened?		
		Total score		/272

whole battery), the same ROC analysis was performed on the DSO questionnaire with the exclusion of the first three questions (Q1-Q3, SM – 4). However, as the battery has been devised as a comprehensive screening tool, it would be used as a whole questionnaire and thus considering the global scores.

4.1.4. Self-reported somatosensory sensations

Self-reported somatosensory sensations were assessed by asking patients whether they experienced a range of sensations identified by our scoping review (Table 1; items S3, S5–S10 in Table 4), with the addition of a question (S4 "Does your arm feel burning?", Table 4) that we often observed to be spontaneously reported after the question about sensations of heat. One more general question (S1 "Does your arm feel different than it felt before?", Table 4) was asked to cover potential other sensations not explicitly requested. However, this question was not considered in the final score because of difficulties categorizing responses that did not provide sensory-specific information, which were

Table 4
The Somatosensory sensations questionnaire. Yes = the symptom is present (score 1), No = the symptom is absent (score 0).

	Self-Reported Somatosensory Sensations		Yes/ No
S1	(not scored)	Does your arm feel different than it felt before?	
S2		Does it feel painful?	
S3		Does it feel numb?	
S4		Does it feel heavy?	
S5		Does it feel hot?	
S6		Does it feel burning?	
S7		Does it feel like pins and needles?	
S8		Does it feel stiff?	
S9		Does it feel cold?	
S10		Does your left arm ever feel strange to you beyond the sensations I mentioned above?	

the focus of the current study. Kinaesthetic sensations were not included in this assessment, as these represent feelings associated with motor awareness (Moro et al., 2021; Pacella and Moro, 2022; Beccherle et al., 2023) rather than with body ownership.

Responses to all questions were recorded as Yes/No; however, only questions S2–S10 (which refer to specific sensory sensations) were scored as 1 (Yes/Present) or 0 (No/Absent), giving a total frequency of somatosensory sensations score ranging from 0 to 9 (higher scores indicating a greater frequency of somatosensory sensations).

4.1.5. Procedure

Patients were tested in a quiet room or at the patient's bedside, with only the patient and two examiners present and standing on the patient's right side (to account for the possible effects of any hemi-neglect). Questions to assess DSO and self-reported somatosensory sensations were asked one at a time without any time limit or pressure for the answer. For each question, the patients' responses were accurately transcribed and later independently scored by 2 expert clinicians. In case of disagreement, the patients' responses were discussed with a third examiner until a common decision was taken. Assessment of DSO took between 15 and 30 min depending on the patient's answers. Self-reported somatosensory sensations took 5–10 min to assess. The two assessments were carried out in the same or two different sessions, depending on the patient's degree of fatigue, while the neuropsychological assessment was carried out in a separate session.

4.1.6. Data analysis

Scores from the DSO, self-reported somatosensory sensations and standard neuropsychological assessments were calculated and used in a series of analyses that aimed to explore which somatosensory and neuropsychological factors explain DSO. First, we determined the frequency of DSO and its severity in our sample using the Preliminary Limb Ownership severity scores derived from the first three questions of the DSO questionnaire (Feinberg et al., 2010). This allowed us to classify patients as No disownership, Mild disownership, Moderate disownership and Severe disownership. Second, in order to confirm this patient classification in groups of severity, we conducted ROC analyses to identify the full DSO questionnaire score cut-offs able to distinguish among the 4 groups of disownership severity (see SM - 4 for details). Third, to examine whether somatosensory sensations varied depending on the disownership severity we analysed the frequencies of self-reported somatosensory sensations in each of the four severity groups (see SM – 5) and then correlated the total number of sensations reported with the full DSO score (Pearson's rho).

Finally, we explored which demographic, clinical, neuropsychological and somatosensory factors modulate DSO. For this, we ran separate linear models using the full DSO questionnaire score as the dependent variable in all cases (for details see SM – 6) and as independent variables: Model 1) the demographic and clinical data (i.e. age, sex, education,

lesion onset/test interval, lesion size); Model 2), the general cognitive state (i.e. MMSE, digit span and FAB); symptoms often associated with DSO, in particular Model 3) personal neglect (Comb test) and extrapersonal neglect (Line crossing) – and Model 4) anosognosia for hemiplegia (MUNA). In all of these models, continuous independent variables were converted to z-scores to put all measures on the same scale, while the categorial independent variables, if any, were entered as factors. In a fifth and final linear model we assessed the role of somatosensory sensations in DSO, using the presence/absence of each somatosensation as categorical independent variable and the full DSO score again as dependent variable. To further exclude a potential impact of lesion size, an additional ANCOVA was carried out, using as factors the presence/absence of somatosensory sensations, the lesion size and the Line Crossing score. In all analyses missing data were imputed by means of the mice R package (Van Buuren & Groothuis-Oudshoorn, 2011- see SM - 6 for details). Examples of qualitative statements offered by the participants spontaneously in the administration of the two questionnaires have also been provided where possible (see SM - 8), to enhance the phenomenological understanding and subjective experience of these feelings of non-belonging and somatosensory sensation.

5. Results

Severity and frequency of limb disownership (based on the clinical routine assessment Feinberg et al., 2010). The frequency of each score (i.e. 0, 1, 2) attributed to the participants' responses to the first three questions (Q1 "Is this your hand?"; Q2 "Does it feel as if it does not belong to you?"; Q3 "Does it belongs to someone else?") and their sum are reported in SM - 3. According with a clinical criterion described in detail in SM - 3, 46.28% of our participants fell in the No disownership group (scores 0 or 1), 20.66% suffered from Mild disownership (score 2), 21.49% from Moderate disownership (scores 3 or 4) and 11.57% from Severe disownership (scores 5 or 6).

Assessment of DSO symptoms. The division in the four groups of disownership severity was confirmed by the Receiving Operating Characteristics (ROC) analyses on the full DSO questionnaire (full details in SM - 4 that show that the Areas Under the Curves (AUC) are greater than 0.7 in all the comparisons. The values of the cut-offs with confidence intervals, AUC values and values of specificity and sensitivity are reported in Table 5 and SM – 4. The ROC analysis with the exclusion of the three preliminary questions (to control for the risk of circularity) confirms the capacity of the tool to discriminate patients with and without disownership (AUC = 0.78). In this way the four groups identified for disownership severity remain the same also for the full score of DSO. However, for clarity, in the text below, when the analyses compare the four groups these will continue to be called with the label 'Disownership' (No Disownership, Mild, Moderate, Severe). Some examples of patients' responses are reported in SM – 8.

Self-reported somatosensory sensations in DSO (as measured in the full DSO questionnaire). A significant difference in the overall presence versus absence of somatosensory sensations (i.e. irrespective of sensation type; 1 = presence of any somatosensory sensation, 0 = no somatosensory sensation) was found between the four Disownership severity groups ($\chi^2(3) = 9.443$, $p = 0.024$, No Disownership group = 86%, Mild = 88%, Moderate = 100%, Severe = 100%). In general, the majority of patients in all the groups reported that their upper limb felt different than before their stroke (i.e. gave an affirmative response to item S1). However, this item ("Does your arm feel different than it felt before?") was not taken into consideration in this analysis as it represents too vague a claim, not specifically connected to somatic sensations.

Post-hoc binomial tests showed that the No Disownership and Mild reported lower frequency of somatosensory sensations than the Severe and Moderate groups (Bonferroni corrected $p < 0.001$ in all the four comparisons), while no other comparison showed differences in the frequencies. The sensations reported more frequently are the feeling of heaviness, the sense of stiffness and numbness (Table 6). Furthermore, the total number of sensations reported by the patients correlates with DSO scores (Pearson's rho = 0.25 ($t(119) = 2.884$, $p = 0.005$), with more sensations being associated with greater DSO severity in the DSO questionnaire.

With regard to the significant differences in these specific somatosensory sensations, an effect of Group was found in the sense of heaviness ($\chi^2(3) = 9.489$, $p = 0.023$) but post-hoc analyses did not show any significant difference among groups. A group effect was found also in the sense of numbness ($\chi^2(3) = 11.709$, $p = 0.008$), with post-hoc analysis showing the only statistically significant difference being between the No Disownership and Moderate groups ($p = 0.018$, Bonferroni corrected). Finally, a significant effect of Group ($\chi^2(3) = 21.945$, $p < 0.001$) was found for stiffness. Post-hoc analysis showed significant differences between the No Disownership and Moderate groups ($p < 0.001$, Bonferroni corrected) and between Mild and Moderate groups ($p = 0.036$, Bonferroni corrected). Some examples of patients' responses are reported in Table 7.

The impact of neuropsychological variables and somatosensory sensations on DSO score

An original contribution of this study consists in the possibility of investigating not only the presence of clinical and neuropsychological variables and the differences in the frequencies of symptoms among the groups, but their impact on DSO symptoms as measured by the DSO questionnaire. The linear models show that only extrapersonal neglect as measured by Line Crossing modulates DSO symptoms ($F(1, 110) = 5.889$, $p = 0.017$, $\eta_p^2 = 0.05$), while no effects were found for any other clinical and neuropsychological variables. With regards to somatosensory sensations, results indicate that DSO symptoms is modulated by the presence of self-reported coldness ($F(1, 110) = 8.200$, $p = 0.005$, $\eta_p^2 = 0.07$), stiffness ($F(1, 110) = 15.150$, $p < 0.001$, $\eta_p^2 = 0.12$), and a trend for numbness sensations ($F(1, 110) = 3.494$, $p = 0.064$, $\eta_p^2 = 0.03$). Adding as a control variable the lesion size to the latter analysis, results from the ANCOVA (SM7) show that not only the statistically significant somatosensory sensations do not change, but that Lesion Size is not statistically significant with a negligible effect size (pη2 = 0.003).

6. Discussion

The study aimed to examine the role of subjectively experienced somatosensory sensations in the sense of limb (dis)ownership. We conducted a systematic scoping review focused on what is currently known about the role of somatosensory sensations in body part (dis)ownership, via existing studies in patients with DSO. Our scoping review indicated that despite existing reports of somatosensory functions in DSO

Table 5
The results from the ROC analyses, with the cut-off value to distinguish DSO versus No DSO patients and the various degrees of severity in DSO. Cut-off: the cut-off computed by means of bootstrap (1000 iterations) maximizing the Specificity – Sensitivity product; AUC = Area Under the Curve; Specificity = true negative rate; Sensitivity = true positive rate; 95% CI = 95% confidence interval; Disown = Disownership.

Groups	DSO Cut-off	95% CI	AUC	95% CI	Specificity	95% CI	Sensitivity	95% CI
No Disown v. disownership	39.01	25.94 - 45.06	0.91	0.84-0.96	0.85	0.79 - 0.93	0.88	0.75 - 0.95
No Disown. v. Mild	18	18–41	0.85	0.76-0.92	0.66	0.56 - 0.91	1	0.7–1
Mild v. Moderate	67	47–71	0.76	0.6 - 0.89	0.8	0.46 - 0.95	0.65	0.55–1
Moderate v. Severe	118	80–173	0.84	0.7 - 0.95	0.85	0.56–1	0.71	0.54–1

Table 6
Patients' responses to the Somatosensory Sensations Questionnaire. The number of positive responses (Yes) and frequencies of the presence of the specific sensations asked in the questionnaire are reported for each group of patients. * = difference in the frequency of sensation with respect to No Disownership; ^ = difference in the frequency of sensation with respect to MILD.

	Does your arm feel …	NO disownership (n. 56)		TOT Disownership (n. 65)		MILD (n.25)		MODERATE (n.26)		SEVERE (n.14)	
		n.	%	n.	%	n.	%	n.	%	n.	%
S1	Different than it felt before	37	66.07	46	70.63	13	52	23	88.46	10	71.43
S2	Painful	21	37.50	23	36.49	8	32	9	34.62	6	42.86
S3	Burning	9	16.07	12	17.79	4	16	6	23.08	2	14.29
S4	Numb	22	39.29	44	67.07	15	60	20	76.92*	9	64.29
S5	Pins and needles	14	25.00	23	36.44	7	28	10	38.46	6	42.86
S6	Heavy	39	69.64	54	82.24	18	72	25	96.15	11	78.57
S7	Stiff	19	33.93	43	66.68	11	44	22	84.62*^	10	71.43
S8	Hot	25	44.64	26	40.29	7	28	13	50.00	6	42.86
S9	Cold	16	28.57	12	16.74	5	20	6	23.08	1	7.14
S10	Something else	9	16.07	24	37.62	5	20	13	50.00	6	42.86

Table 7
Examples of spontaneous patient responses to questions about somatosensory sensations.

	Somatosensory sensations questionnaire	Spontaneous qualitative responses
S1	Does your arm feel different than it felt before?	"Yes, it feels so heavy, all the time." "It feels different, heavy, it feels so strange." "It does not feel like it is mine. It feels heavy and it feels burning hot." "It feels like it is not mine, it felt like a stone yesterday."
S2	Does it feel painful?	"It often feels painful. A tingling feeling actually."
S3	Does it feel numb?	"Yes, it feels numb, like it is not mine."
S4	Does it feel heavy?	"It feels like a weight." "Yes, it feels heavy, like a stone." "It feels like a brick. It weighs me down."
S5	Does it feel hot?	"It burns. Yesterday it was burning"
S6	Does it feel burning?	"Yes, burning hot."
S7	Does it feel like pins and needles?	"It tingles." "Yes, it often feels like pins and needles, a tingling sensation."
S8	Does it feel stiff?	"It feels sharp sometimes."
S9	Does it feel cold?	"Yes, it feels Icey cold." "Sometimes I think it is frozen. It is frozen still."
S10	Does your left arm ever feel strange to you beyond the sensations I mentioned above?	"It feels dead."

(especially proprioceptive and tactile deficits), very little is currently known about the role of self-reported somatosensory sensations in body (dis)ownership, and that empirical research focused on this question is needed. We subsequently conducted an empirical study to specifically examine self-reported somatosensory sensations in patients with DSO. We assessed self-reported somatosensory sensations in one of the largest samples of DSO patients to date, finding that these somatosensory sensations are more common in patients with moderate and severe disownership (and DSO symptoms) compared to those with mild or no Disownership, and such sensations are associated with greater DSO features. Findings from the combined scoping review and empirical study suggest that self-reported somatosensory sensations may contribute to limb ownership but are a severely neglected area of research in need of further work.

Indeed, the scoping review showed that existing research has not paid sufficient attention to the role of somatosensory deficits or self-reported somatosensory sensations in DSO. Although clinical examination of tactile sensitivity and proprioception are commonly reported and found to be affected in DSO patients, other somatosensory abilities (e.g. thermoception) and patient self-reported somatosensory sensations are seldom investigated. As already discussed (see section Part 1: Discussion), only a narrow selection of somatosensory abilities is usually assessed as part of routine clinical assessments, but such assessments may not identify the full range and severity of sensory deficits. These studies are also subject to a reporting bias, as the lack of a systematic assessment of self-reported aberrant sensations results in the reporting of the 'positive' cases (i.e. when the patients spontaneously report the sensations) and the omission of 'negative cases'(i.e. patients with DSO who are specifically interviewed and do not report any sensations). These systematic biases limit our understanding of how somatosensations might impact body ownership and lead to DSO.

The current study shows that self-reported somatosensory sensations are more common in patients with more severe forms of Disownership (i.e. greater frequency of patients with aberrant sensations in Moderate and Severe Disownership relative to Mild or No Disownership groups), and a positive correlation between the number of sensations reported and the symptoms of DSO. Furthermore, heaviness, stiffness and numbness are significantly more frequent in patients with greater severity of Disownership. Moreover, we found that feelings of coldness, stiffness and numbness impact on DSO features.

The finding that self-reported somatosensory sensations are more common in DSO is consistent with the previous suggestion that limb disownership is the result of an error signal that cannot be integrated into the patient's body representation (Martinaud et al., 2017). Somatosensory sensations, which are not predicted by existing, top-down expectations arising from outdated representations of the body, and cannot be integrated into current self-body representations, may lead to feelings of limb disownership. The feeling of coldness might be associated with the perception of the internal state of the body (interoception) in particular with stroke-induced alterations in tissue trophism, blood circulation and thermoregulation. Coldness, numbness and parasthesia (i.e. pins-and-needles) sensations are cardinal features of Raynaud's phenomenon, which is caused by abnormal blood flow in affected areas of the body (Herrick, 2012). No data exists in the literature regarding the assessments of these various physiological aspects in DSO, and unfortunately these data were not collected in the current study. However, a previous study by Romano et al. (2014) found a reduced anticipatory skin conductance response to stimuli directed to the ipsilesional hand in patients with somatoparaphrenia, and a drop in skin temperature has been related to feelings of disownership in studies with healthy individuals and other clinical samples characterized by feeling limb disownership (Crivelli et al., 2021, 2023; Salvato et al., 2022). Interoception might also be associated with the sense of numbness and stiffness. Previous studies of our group (Jenkinson et al., 2020) show that the administration of pleasant touch (i.e. a slow, light touch able to activate the unmyelinated C-fibres that project to insula cortex and are

part of the interoceptive system, Craig, 2003) can increase the sense of limb ownership in patients with DSO. This may be via the enhancement of pleasant interoceptive sensations and reduced feelings of paraesthesia ('pins-and-needles') or deafference that are caused by multisensory mismatches (Jenkinson et al., 2020; Longo et al., 2008; Roel Lesur et al., 2020). Stiffness and numbness might be further linked to motor deficits and spasticity. However, although all the patients in our sample suffered from hemiplegia, only a subset reported these sensations, and stiffness was also reported by patients without spasticity. A specific assessment of spasticity was not carried out and further investigations are needed to understand the role of spasticity in somatosensory sensations.

Although we identified group differences in somatosensory sensations, with certain sensations being more frequent or a significant regressor of DSO, there were no somatosensory sensations that were unique to patients suffering from limb disownership. Indeed, patients with no Disownership also experience the same somatosensory sensations – and so the presence of such sensations cannot be sufficient for the occurrence of Disownership and DSO. However, the presence of at least one somatosensory sensation may be a necessary condition for the occurrence of limb disownership. Indeed, we found seven patients without Disownership who did not report any somatosensory sensations, while all of our Disownership patients reported at least one sensation, and those in the moderate or severe groups reported at least three sensations. Further research is needed to explore whether (unexpected) somatosensory sensations are necessary for the emergence of Limb disownership and DSO, and if they therefore play a key role in the sense of body ownership. Crucially, as shown by the responses to the first question of the Self-Reported Somatosensory Sensations questionnaire (where percentages of responses are sometimes less than in specific questions), specific questions are needed during DSO assessment in order to identify somatosensory sensations, as sometimes patients who do not report symptoms to a general question (i.e. Does your arm feel different than it felt before?) respond positively to more specific questions.

In terms of other clinical and neuropsychological factors that might impact on DSO, consistent with existing literature (Romano and Maravita, 2019), we found extrapersonal neglect to be the only significant regressor of DSO as measured by the DSO questionnaire. General cognitive functioning, executive functions, anosognosia and personal neglect were not associated with DSO. The lack of an association between DSO, anosognosia and personal neglect is consistent with recent evidence that these are distinct neuropsychological conditions caused by different lesion patterns (Pacella et al., 2019; Monai et al., 2020; Bertagnoli et al., 2022; Moro et al., 2023). In the current study, only one patient in the Severe DSO group showed a pathological score on the MUNA battery for anosognosia for hemiplegia. Although a diagnosis of personal neglect should include more than one task (Bertagnoli et al., 2022), we found that in all the groups the percentage of patients suffering from personal neglect was high, although Severe and Moderate DSO suffer from personal neglect more frequently (80% and 81% pathological scores, respectively) than Mild DSO (79%) and No DSO (69.09%).

In addition to being a novel exploration of self-reported somatosensory sensations in DSO, the current study provides a more comprehensive assessment of DSO, which includes the sense of limb disownership and several other features of DSO described in the extant literature. The new measure scoring is validated using ROC analysis with a balance of specificity and sensitivity. This statistical procedure allows the identification of various different degrees of severity of DSO, which may be particularly useful in clinical practice, in both the initial diagnosis and in recording changes in symptoms over time. Furthermore, this may help in experimental studies, where differences in severity of symptoms (when not controlled) may affect results. Crucially, after this first screening, a more in-depth analysis of the qualitative characteristics of the patient's symptoms may be carried out through specific interviews (e.g. Saetta et al., 2021) An advantage of this questionnaire is the inclusion of cut-offs that may be useful for the first clinical diagnosis and for tracking changes in symptoms over time. Likewise, our assessment of self-reported somatosensory sensations is the first of its kind, aiming to capture the subjective experiences of DSO patients in a systematic manner. It was developed based on an extensive scoping review of the existing literature and provides a new measure of somatosensory sensations, Replications of our findings in other samples are needed, which include also patients suffering from left hemisphere damage that were not considered in this study as DSO is typically reported after right-hemisphere lesions.

A possible limitation of the study is represented by the presence of 'leading' questions in the DSO questionnaire, that might suggest otherwise undetected symptoms to the patient. The fact that the patient can be suggestible in his responses must be held in high regard during questionnaire administration. Our data indicate that patients can discriminate among symptoms and also describe carefully the presence of aberrant sensations. On the other hand, only questions directly focused on specific symptoms allow to identify patients with and without those symptoms (and reduce the risk to consider only the 'positive' cases).

In summary, the paper reports the first in-depth analysis of self-reported somatosensory sensations in DSO patients, thus offering a first contribution to fill a gap in the literature and responding to the clinical and experimental question regarding the role of these subjective sensations in the sense of body ownership. Furthermore, it offers a quantitative measure for the clinical screening of limb disownership and DSO symptoms.

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CRediT authorship contribution statement

Valentina Moro: Writing - review & editing, Writing - original draft, Visualization, Supervision, Resources, Project administration, Methodology, Investigation, Funding acquisition, Data curation, Conceptualization. **Michele Scandola:** Writing - review & editing, Formal analysis, Data curation. **Valeria Gobbetto:** Writing - review & editing, Investigation. **Sara Bertagnoli:** Writing - review & editing, Investigation, Conceptualization, Writing - review & editing, Investigation. **Maddalena Beccherle:** Writing - review & editing, Investigation. **Sonia Ponzo:** Writing - review & editing, Investigation. **Aikaterini Fotopoulou:** Writing - review & editing, Supervision, Resources, Funding acquisition, Conceptualization. **Paul M. Jenkinson:** Writing - review & editing, Writing - original draft, Supervision, Resources, Project administration, Methodology, Funding acquisition, Conceptualization.

Declaration of competing interest

The authors report there are no competing interests to be declared.

Data availability

URL: https://osf.io/2f8p9/

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Appendix A. Supplementary data

Supplementary data to this article can be found online at.https://doi.org/10.1016/j.neuropsychologia.2023.108776.

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