

## **The Fluff test: Improved scoring system to account for different degrees of contralesional and ipsilesional personal neglect in brain damaged patients**

Gianna Cocchini<sup>a</sup> & Nicoletta Beschin<sup>b</sup>

<sup>a</sup>Goldsmiths University of London, London (U.K.)

<sup>b</sup>Neuropsychology Ward, Somma Lombarda Hospital, Gallarate (Italy)

### **ABSTRACT**

The Fluff test is a simple test to assess evidence of personal neglect (PN) in brain damaged patients. While blindfolded, patients are asked to remove targets previously attached on their body and the number of targets detached provides information about possible spatial bias. This test has been widely used for clinical and research purposes. However, the current scoring system presents some limitations, which make difficult to interpret patients' performance in terms of both contralesional and ipsilesional PN when they omit targets on the ipsilesional side. Moreover, it does not consider possible confounding variables, such as non-spatial cognitive deficits or lack of compliance that may affect patients' performance and lead to incorrect diagnosis. The present paper proposes a new scoring method overcoming the limitations mentioned above and it analyses data from a large sample of 243 brain damaged patients. Findings showed that contralesional PN was significantly more severe, but not more frequent, following right (31%) than left (21%) brain damage. We also found evidence of left ipsilesional PN and cases of potential mis-diagnosis that would have passed unnoticed with the original scoring system. The new scoring method allows to identify different degrees of contralesional and ipsilesional PN and potential confounding variable.

**Keywords:** Personal neglect, hemispatial neglect, Fluff test, assessment, brain damage, stroke

## INTRODUCTION

Personal neglect (PN) refers to a form of hemi-inattention where brain damaged patients show a “deficit relative to the side of the body contralesional to the lesion” (Guariglia & Antonucci, 1992; p.1001), showing a reduced tendency to explore or perform movements in the contralesional side of one’s own body. Patients showing PN may ignore their plegic limb, may not dress the contralesional side of their body or they only comb the hair on the ipsilesional side of their head. Unsurprisingly, this has important consequences on patients’ everyday life and on functional rehabilitation. Despite the impact of PN on functional recovery and everyday life, a recent review of the last 123 years of PN literature has identified a rather limited range of tools to assess PN (Caggiano & Jehkonen, 2018). In fact, only 7% of the studies on PN adopted more than two assessment methods, limiting the investigation of PN forms. For example, Committeri et al. (2018) pointed out as performance on some tests (e.g. One item test; Bisiach et al., 1986) may be confounded by directional hypocinesia, measuring patient’s awareness of contralesional hemiplegia rather than PN. Two revision papers (Caggiano & Jehkonen, 2018; Committeri, Piervincenzi & Pizzamiglio, 2018) have both recommended that PN research and clinical assessment consist of a battery of tests to compensate for limitations of individual tasks, provide a more comprehensive investigation of different aspects of PN and assess different body parts. Therefore, it is important to refine available assessment methods to minimise possible limitations of single tests and maximize their use. The Fluff test (Cocchini, Beschin & Jehkonen, 2001) represents one of the four most frequently used tests to assess PN in research studies with brain damaged patients (see Table 3 in Caggiano & Jehkonen, 2018). The test is simple, fast to be administered and it has a high test-retest reliability ( $r= 0.89$ ;  $p< 0.001$ ). However, over the years, the current scoring system has shown some limitations. In detail, i) it does not provide a cut-off to interpret target omissions in the ipsilesional side of the body (ipsilesional PN); ii) the current cut-off for contralesional targets assumes that all

ipsilesional targets are removed. This is clearly not always the case as clinicians and researchers have often observed in everyday practice; iii) it does not allow for discrimination between spatial bias (i.e. PN) and non-spatial deficits that may affect the patient's overall performance regardless of the body side, such as general attentional disorders, lack of compliance, poor comprehension of instructions, executive functions impairment etc. For this reason, we are proposing a new scoring system that allows to account for these three aspects also enabling researchers and clinicians to follow clear guidelines for degree of severity.

## **METHODS AND PROCEDURE**

### **Participants**

We considered a large group of 243 patients showing unilateral right (n=131) or left (n=112) brain damage (RBD and LBD, respectively) on CT or MRI scans. Some of them were considered in previous studies (27 in Cocchini et al., 2001; 197 in Caggiano, Beschin & Cocchini, 2014) but scored according to the original scoring system. All patients were recruited at the Neuropsychology Unit of Somma Lombardo Hospital in Italy. The study was in accordance with the Declaration of Helsinki (BMJ 1991; 302:1194) and approved by the Research Ethical Committee of Gallarate Hospital. All participants provided full consent to enter the original studies. The demographical and clinical details of these patients are shown in Table 1. All patients were then asked to complete at least two tests to assess extrapersonal neglect (i.e., Star cancellation test – Wilson, Cockburn, & Halligan, 1987; Line cancellation task- Albert, 1973). Up to 77% of the RBD patients also completed a verbal reasoning task (Spinnler & Tognoni, 1987) whereas 90% of the LBD patients completed the Brixton test (Burgess & Shallice, 1997), which measures the patients' ability to detect and change rules in sequences of stimuli, and it is suitable for patients showing language impairments. Verbal

comprehension was assessed in 101 LBD patients by mean of the Token Test. Scores corrected by age and education were compared to norms (Spinnler & Tognoni, 1987). Overall performance on these tests is shown in Table 1. Finally, patients were asked to perform two tests to assess personal neglect (Comb & Razor test and the Fluff test). On the Comb and Razor/Compact test (Beschin & Robertson, 1997; McIntosh, Brodie, Beschin, & Robertson, 2000) patients are asked to pretend to comb themselves and shave, or apply make-up for 90 seconds. The numbers of strokes applied by the patient are then considered in a formula (McIntosh et al., 2000) to identify possible leftward (negative value) or rightward bias (positive value), which is then compared with norms. The Fluff test consists in applying 15 small targets on the contralesional side of the blindfolded patients' body (6 on the arm, 6 on the leg and 3 on the torso) and 9 small targets on the ipsilesional side (6 on the leg and 3 on the torso). Patients, unaware of the total number of stimuli, are then asked to remove all targets they can find using their ipsilesional hand. The number of targets removed on the contralesional side is compared with norms to diagnose possible contralesional PN. Performance on these two tests is considered in more detail in the Results section.

**Table 1. Demographical and clinical details of RBD and LBD groups**

Groups	N	Age	Education level (yrs)	Gender	Onset from lesion (mths)	Abstract reasoning*	Extraperosnal neglect**	Token Test***
<b>RBD</b>	131	Mean = 64.48 SD = 12.15 Range = 27-85	Mean = 6.89 SD = 3.37 Range = 1-17	49 F	Mean = 4.05 SD = 4.51 Range = .99-28	Mean = 44.63 SD = 7.88 Range = 36-60 N= 0	N= 53 (40.5%)	n/a
<b>LBD</b>	112	Mean = 59.36 SD = 15.82 Range = 19-84	Mean = 8.30 SD = 4.24 Range = 3-23	48 F	Mean = 6.92 SD = 16.35 Range = .33-130.6	Mean = 4.69 SD = 2.65 Range = 1-10 N= 32	N=16 (14.3%)	Mean = 25.8 SD = 10.3 Range = 2-36 N= 35

\* Data related to a subgroup of 202 patients. RBD performed the Verbal abstract reasoning; score range: 0- 60; cut-off<= 33 (Spinnler & Tognoni, 1987); LBD performed the Brixton test; scaled score range: 1-10; cut-off: <=4 (Burgess & Shallice, 1997). N= number of patients showing pathological performance

\*\* Number of RBD and LBD patients showing contralesional extraperosnal neglect on at least one of the neglect tests (i.e. Stars and Line cancellation tests)

\*\*\* Data related to a subgroup of 101 LBD patients; score range: 0-36; cut-off<= 26.50 ( adjusted for age and education; Spinnler & Tognoni, 1987); N= number of patients showing pathological performance

## **Fluff tests- new scoring system**

### *Contralesional and Ipsilesional PN*

The original cut-off for the Fluff test defines patients' performance as normal when at least 13 out of 15 (86.7%) targets are detached in the contralesional side, since no healthy participants omitted more than 2 stickers on this side (Cocchini et al., 2001). We re-analyzed the raw data considered in the 2001 paper and we adopted the same criterion (i.e. lowest score achieved by healthy participants) to define the cut-off for ipsilesional side. No healthy participants omitted more than 1 sticker on the ipsilesional side, thus we considered it as a 'normal' performance for the ipsilesional side if at least 8 out of 9 targets were detached. Therefore, considering both sides, a normal performance is recorded when a patient detached at least 13 targets on the contralesional side and at least 8 on the ipsilesional side. When a patient omits targets only on one side of the body (i.e. the performance on the other side is at ceiling), we can adopt the cut-offs above to determine whether a patient shows contralesional or ipsilesional PN. However, sometimes patients omit targets on both sides and it may not be easy to understand whether their performance should be interpreted as evidence of PN or other non-spatial factors, such as general attentional, comprehension or executive deficits, or even patient's lack of compliance with the task.

To allow for interpretation of patients' performance in these cases, we compared performance from each side to establish whether there is a spatial bias leading to PN. To this aim we identified the crucial pathological difference between side performances (i.e. when performance is worse on one side) and the tolerance value of 13.3 was calculated considering the more conservative cut-off (i.e.  $100 \text{ minus } 86.7 = 13.3$ ). Therefore, a side difference of more than  $\pm 13.3\%$  represents a relevant (i.e., above cut-off) spatial bias towards one side and it should be interpreted as evidence of PN. Since the ipsilesional performance was subtracted from the contralesional one, the negative or positive sign represents the direction of the spatial

bias and determines whether the patient shows contralesional or ipsilesional PN, respectively. Table 2 provided spatial bias values for all possible combinations. For example, if a patient detects 12 out of 15 (i.e., 80%) targets in the contralesional side and all 9 (i.e., 100%) in the ipsilesional side, the side difference will be -20. This spatial bias is higher than  $\pm 13.3$  and should be interpreted as evidence of contralesional PN. However, if a patient detects 12 out of 15 (i.e., 80%) targets in the contralesional side and 8 out of 9 (i.e., 88.9%) in the ipsilesional side, the spatial bias is -8.9%. This value is clearly below the critical difference of tolerance  $\pm 13.3$  and it should not be considered as evidence of contralesional neglect.

#### *Degree of severity for PN*

We also attempted to provide some guidelines to determine the degree of severity of PN considering the magnitude of the side difference and establishing two further limits to differentiate between mild-moderate and between moderate-severe degrees. Based on the previous cut-off for spatial bias (i.e.,  $\pm 13.3$ ), side differences between  $\pm 13.4$  and  $\pm 100$  indicate evidence of PN. We then divided the 'pathological window' (i.e.  $100 - 13.3 = 86.7$ ) in three equal sections ( $86.7/3 = 28.9$ ) to consider two limits equally distant as moderate (i.e.  $13.3 + 28.9 = 42.3$ ) and severe (i.e.  $42.3 + 28.9 = 71.2$ ). Therefore, a spatial bias between  $\pm 13.4$  and  $\pm 42.2$  represents a mild PN, between  $\pm 42.3$  and  $\pm 71.1$  represents a moderate PN and between  $\pm 71.2$  and  $\pm 100$  a severe form of PN (negative and positive signs indicate contralesional and ipsilesional PN, respectively). The different degrees are represented in the Table 2 with different shaded areas (darker for more severe).

### *Non-spatial underperformance (NSU)*

While a side difference above the 13.3% cut-off suggests a spatial disorder (PN); a side difference below this cut-off may not be interpreted as evidence of PN even if the overall performance is clearly pathological. In this case, patients' performance can be poor but similar on both sides, therefore there is not a clear spatial bias. These values are shown in Table 2 as white cells. There may be several different reasons for this performance such as, general attentional disorder, lack of comprehension, poor compliance, motor difficulties to bend over and reach lower parts of both legs, or planning disorders. We labelled this type of performance as 'non-spatial underperformance' (NSU). The vague definition is intentional as the Fluff test and its new scoring system are not intended to guide any interpretation of this type of performance and, on the bases of these results, examiners should only exclude evidence of hemispacial deficit, such as PN. In these cases, the side difference value is not indicative of severity as it only indicates that there is no spatial bias. As a general guideline, the cells on the top left represent poorer performance on both sides. Considering that missing more than half of the targets on each side would be more than three times the maximum number of omissions allowed in a normal performance (i.e., 13.3%), clinicians may want to consider as evidence of severe NSU when patients detached less than half of the targets on each side (i.e. less than 7 contralesional and less than 4 ipsilesional targets) and showed no spatial bias.

### *Scoring table*

Following the calculations mentioned above, we developed a scoring table to allow easy interpretation of individual performance on the Fluff Test. Table 2 shows the side difference in percentages for every combination of targets detached from each side. This scoring table is very easy to use as the examiner only needs to enter the number of targets detached on



contralesional and ipsilesional sides and check the value in the corresponding cell. The sign represents the direction of the spatial bias. If the cell values are higher than 13.3%, positive values indicate ipsilesional PN and negative values indicate contralesional PN (grey cells). Cells representing pathological performance without evidence of specific spatial bias should be considered as non-spatial underperformance (NSU; white cells in Table 2). The combination of the cell values with the cut-offs for normal performance and degree of severity for pathological performance allows examiners to interpret patients' performance.

**Table 2. Scoring table to assess contralesional and ipsilesional PN**

		CONTRALESIONAL TARGET DETACHED															IPSILESIONAL PN		
		0	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15		
IPSILESIONAL TARGET DETACHED	0	0	6.7	13.3	20.0	26.7	33.3	40.0	46.7	53.3	60.0	66.7	73.3	80.0	86.7	93.3	100.0		
	1	-11.1	-4.4	2.2	8.9	15.6	22.2	28.9	35.6	42.2	48.9	55.6	62.2	68.9	75.6	82.2	88.9	SEVERE	
	2	-22.2	-15.6	-8.9	-2.2	4.4	11.1	17.8	24.4	31.1	37.8	44.4	51.1	57.8	64.4	71.1	77.8		
	3	-33.3	-26.7	-20.0	-13.3	-6.7	0.0	6.7	13.3	20.0	26.7	33.3	40.0	46.7	53.3	60.0	66.7		
	4	-44.4	-37.8	-31.1	-24.4	-17.8	-11.1	-4.4	2.2	8.9	15.6	22.2	28.9	35.6	42.2	48.9	55.6	MODERATE	
	5	-55.6	-48.9	-42.2	-35.6	-28.9	-22.2	-15.6	-8.9	-2.2	4.4	11.1	17.8	24.4	31.1	37.8	44.4		
	6	-66.7	-60.0	-53.3	-46.7	-40.0	-33.3	-26.7	-20.0	-13.3	-6.7	0.0	6.7	13.3	20.0	26.7	33.3		
	7	-77.8	-71.1	-64.4	-57.8	-51.1	-44.4	-37.8	-31.1	-24.4	-17.8	-11.1	-4.4	2.2	8.9	15.6	22.2	MILD	
	8	-88.9	-82.2	-75.6	-68.9	-62.2	-55.6	-48.9	-42.2	-35.6	-28.9	-22.2	-15.6	-8.9	-2.2	4.4	11.1		
	9	-100.0	-93.3	-86.7	-80.0	-73.3	-66.7	-60.0	-53.3	-46.7	-40.0	-33.3	-26.7	-20.0	-13.3	-6.7	0.0		
		SEVERE			MODERATE			MILD			NORMAL								
CONTRALESIONAL PN																			

The examiner should identify the cell corresponding to the number of targets detached from the contralesional and ipsilesional sides. Values in the cells indicate spatial bias between sides. Cells in the rectangle at the bottom right indicate normal performance. Cells in grey shade indicate a spatial bias toward one side and evidence of PN. Severity of PN is represented by darker shades. White cells outside the rectangular area suggest poor performance without clear spatial bias (i.e. Non-spatial underperformance – NSU) and should NOT be interpreted as evidence of PN. Particular caution should be given to performances falling in the cells in the top left corner of the table, in these cases, performance may have been affected by various reasons, including lack of comprehension of test instructions, patient’s poor compliance or severe general attentional disorders.

## RESULTS

Fluff test performance of the 243 brain damaged patients was scored following the original and the new adapted system. Individual final interpretations were then compared for RBD and LBD patients.

### *RBD group*

According to the new scoring system, we identified different degrees of severity of contralesional PN in 41 patients (31.30%). Nineteen (46.3%) were deemed as mild, 13 (31.7%) as moderate and 9 (22.0%) as severe PN. Of the 41 patients showing contralesional PN, 31 (75.6%) also showed extrapersonal neglect.

Comparing both scoring systems, performance of 129 out of 131 (98.5%) RBD patients was deemed either as normal (88; 67.2%) or showing contralesional PN (41; 31.3%) by means of both scoring systems. Two patients (1.5%) were diagnosed as showing contralesional PN by means of the original score whereas following the new scoring system both patients were classified as NSU (see Table 3). The first patient (n. 49) detected 12 targets on the contralesional side and 7 on the ipsilesional side (spatial bias 6.7); the second patient (n. 110) detached 11 targets on the contralesional side and 6 on the ipsilesional side (spatial bias -4.4). No patients showed ipsilesional PN according to either scoring method.

### *LBD group*

According to the new scoring system, we identified different degrees of severity of contralesional PN in 23 patients (21.43%). Fourteen (60.9%) were deemed as mild, 7 (30.4%) as moderate and 2 (8.7%) as severe contralesional PN. Furthermore, 7 patients (6.25%) showed

mild (6; 85.7%) or moderate (1; 14.3%) ipsilesional PN. Of the 30 patients showing PN, 9 also showed extrapersonal neglect.

Comparing the two scoring systems, performance of 96 out of 112 (85.7%) LBD was deemed either as normal (73; 65.2%) or showing contralesional PN (23; 20.5%) by means of both scoring systems. However, 16 patients out of 112 (14.3%) received a different diagnosis depending on the scoring system used (see Table 3). In detail, 5 of the 16 patients, who were diagnosed as normal by means of the original scoring system, showed ipsilesional neglect when the new scoring system was adopted (i.e.  $5/112 = 4.5\%$ ); 2 out of 16 patients, who were diagnosed as showing contralesional PN by means of the original scoring system, showed ipsilesional PN when the new scoring system was used (i.e.,  $2/112 = 1.8\%$ ); 9 of the 16 patients, who were diagnosed as showing contralesional PN by means of the original scoring system, showed a non-spatial pathological performance (i.e., NSU;  $9/112 = 8\%$ ) using the new scoring system. A 'NSU performance' does not lead to a specific interpretation. However, it may be interesting to note that 6 of these 9 LBD patients whose performance was scored as 'NSU' also showed a pathological performance on the Token test. We cannot exclude that their poor performance was, at least in part, due to lack of comprehension of the instructions. On the other hand, of the 35 patients showing comprehension deficits on the Token test (See Table 1), 16 (46%) of them performed within norms on the Fluff test and 13 showed evidence of PN. Therefore, it seems that the new scoring system allows to identify potential ambiguous (non-spatial) underperformances and aphasic patients should not be automatically excluded from PN assessment.

**Table 3. Performance details of patients obtaining a different diagnosis with the original and new scoring system**

PT code	Target detached (%)		Fluff Test			Comb and Razor/Compact test	
	Contralesional	Ipsilesional	Original scoring Interpretation (CONTRALESIONAL only) *	Spatial Bias (negative value indicates contralesional PN)	Adapted scoring Interpretation #	Spatial bias (negative value indicates right PN)	Adapted scoring Interpretation~
RBD 49	80	77.8	PN_contra	2.2	NSU	27.9	PN_contra
RBD 110	73.3	66.7	PN_contra	6.7	NSU	72.2	PN_contra
LBD 174	73.3	77.8	PN_contra	-4.4	NSU	-1.7	Normal
LBD 180	0	0	PN_contra/?	0	NSU	-4.8	Normal
LBD 203	0	11.1	PN_contra	-11.1	NSU	2.8	Normal
LBD 204	66.7	55.5	PN_contra	11.1	NSU	0.9	Normal
LBD 207	6.7	0	PN_contra/?	6.7	NSU	-2.2	Normal
LBD 227	80	66.7	PN_contra	13.3	NSU	-2.4	Normal
LBD 228	86.7	77.8	Normal	8.9	NSU	5.4	Normal
LBD 199	20	33.3	PN_contra	-13.3	NSU	-86	PN_contra
LBD 206	0	0	PN_contra/?	0	NSU	-49.2	PN_contra
LBD 181	40	22.2	PN_contra	17.8	PN_ipsi mild	6.9	Normal
LBD 202	60	33.3	PN_contra	26.7	PN_ipsi mild	-1.6	Normal
LBD 217	93.3	55.5	Normal	37.8	PN_ipsi mild	48.1	PN_ipsi
LBD 177	100	66.7	Normal	33.3	PN_ipsi mild	-63.6	PN_contra
LBD 179	100	55.5	Normal	44.4	PN_ipsi moderate	7.7	Normal
LBD 209	86.7	66.6	Normal	20.0	PN_ipsi mild	-35.6	PN_contra
LBD 212	86.7	66.6	Normal	20.0	PN_ipsi mild	-10	Normal

\* The interpretation refers to the contralesional side only. Cut-off for contralesional PN: Targets detached < 86.7%.

# The interpretation considers both sides cut-offs and cut-off for spatial bias (i.e. PN; Spatial bias > ± 13.3%)

~ Spatial bias cut-off > ± 11 (McIntosh et al., 2000).

PN\_contra and PN\_ipsi indicate contralateral and ipsilateral PN, respectively. NSU = non-spatial underperformance

? - indicates a particularly poor performance and some degree of uncertainty about the diagnosis

### *RBD and LBD comparison*

Both groups showed a spatial bias towards the ipsilesional side, in other words a neglect-trend for the contralesional side. We ran two separate chi-square analyses to explore whether the new scoring system identified different frequencies of contralesional and ipsilesional PN between groups. When we analyzed the frequency of contralesional PN, we considered it as ‘non-contralesional PN’ all diagnosis of ipsilesional PN, NSU and normal performance. Similarly, when we considered the frequencies of ipsilesional PN, we classified them as ‘non-ipsilesional PN’ all diagnosis of contralesional PN, NSU and normal performance.

Contralesional PN was not significantly more frequent following RBD (41/131; 31.30%) than LBD (23/112; 20.53%; chi-square = 3.071;  $p = .06$ ); however RBD patients showed a significantly stronger spatial bias (mean = -16.83; SD = 26.87) than LBD patients (mean = -7.00; SD = 22.75;  $F [1,241] = 9.283$ ;  $p = .003$ ).

Fisher’s exact test was used to compare the frequency of ipsilesional PN between groups and ipsilesional PN was significantly ( $p = .004$ ) more frequent in LBD (6.25%) than RBD (0%).

### *Comb and Razor/Compact test*

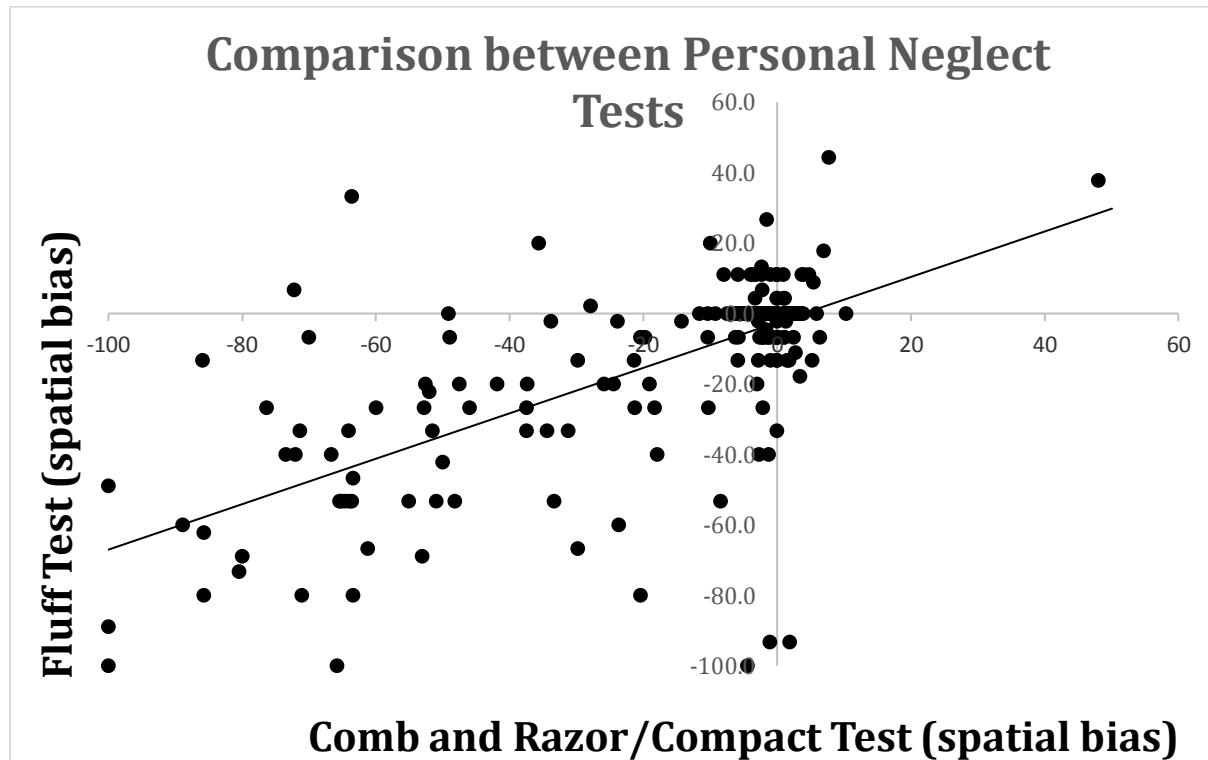
Up to 238 out of 243 patients managed to complete the Comb and Razor/Compact test. In this task, 45/128 RBD (35.2%) and 23/110 LBD (21%) patients showed PN for the contralesional side whereas 2 LBD (2%) patients showed PN for the ipsilesional side. To facilitate comparison between groups and with the Fluff test, we changed the sign value (i.e., positive/negative) of the Comb and Razor/Compact bias for the RBD patients. This was necessary as the sign of the spatial bias for the Comb and Razor/Compact test indicates which side is neglected (i.e. left

PN and right PN, respectively), whereas the sign of the spatial bias for the Fluff test indicates when the PN is contralesional (negative) or ipsilesional (positive).

Comparing this test data with the Fluff test results, we found that the spatial bias of the two tests correlated (Pearson correlation) significantly (overall group  $r = .626$ ;  $p < .001$ ; RBD  $r = .696$ ;  $p < .001$ ; LBD  $r = .501$ ;  $p < .001$ ) and Figure 1 illustrates the dispersion graph.

Finally, we considered performance on the Comb and Razor/Compact task for the 18 patients (2 RBD and 16 LBD) whose performance on the Fluff test was interpreted differently by the original and the adapted scoring system (see Table 3). Seven patients (n. 174, 180, 203, 204, 207, 227, 228) did not show a relevant spatial bias on both tasks and one patient (n. 217) showed ipsilesional PN on both tasks. However, other 10 patients performed differently on the two tasks. Four patients (n. 49, 110, 199, 206) did not show spatial bias on the Fluff test but they showed contralesional PN on the Comb and Razor/Compact test; whereas other 6 patients (n. 181, 202, 177, 179, 209, 212) showed ipsilesional PN on the Fluff, only.

**Figure 1. Individual performance (spatial bias) on the Fluff test and the Comb and Razor/Compact Test**



Positive/negative bias scores of the Comb and Razor/Compact Test have been reversed for LBD patients to allow comparison with Fluff test scores (see text).

After the above transformation, negative values for both tests indicate contralesional spatial bias, positive values indicate ipsilesional spatial bias.

## **DISCUSSION**

The new scoring system capitalizes on the original normative data and it simply proposes a more detailed analysis of the scores to address, as mentioned in the Introduction, three main limitations of the original scoring system. Firstly, i) the original scoring only provides a cut-off for contralesional PN and does not offer any interpretation for flawed performance on the ipsilesional side. We have now identified a tolerance score (i.e. no more than 1 omission) to interpret performance on the ipsilesional side. The new scoring system allowed us to



systematically investigate evidence of ipsilesional PN. Interestingly, while none of the RBD patients showed evidence of ipsilesional PN, we found 7 LBD patients (6.25%) showing ipsilesional PN who would have passed unnoticed with the previous scoring system.

Secondly, ii) the original scoring system provides interpretation of contralesional performance assuming a flawless performance on the ipsilesional side. This is not always the case, remaining very unclear on how to interpret patients' performance when they also omit targets on the ipsilesional side. The proposed scoring system takes into account performance on both sides and it considers the difference between side performance rather than the absolute score from one side only. For example, let us consider a case where a patient detects 12 out of 15 targets (80%) on the contralesional side and 8 out of 9 (88.9%) on the ipsilesional side. According to the original scoring system, this patient would be diagnosed with contralesional PN as detecting 12 targets is below the tolerance score of 13.3% of missed targets. However, considering the ipsilesional performance, which is not flawless, the difference between sides is only 8.9%, and this is within the tolerance score. This does not necessarily indicate that this patient's performance is 'normal'; it simply indicates that there is no evidence for clear spatial asymmetry, which is crucial in the diagnosis of PN and neglect in general. On a different case, missing ipsilesional targets can be part of a severe contralesional PN where a patient's attention is dramatically shifted to the most extreme part of the ipsilesional side. In this case, patients tend to omit all contralesional targets and the ipsilesional targets closer to the body midline. While the original scoring system will not differ between this patient and another patient 'only' omitting all contralesional targets, the new system can provide an interpretation of severity and differ between these two patients' performance.

Thirdly, iii) lack of compliance or other cognitive impairments, such as general attention disorders or executive deficits, may worsen patients' performance on various tests, including

the Fluff test. It is therefore useful to identify those pathological performance (‘non-spatial underperformance’; NSU) that may be due to deficits other than PN. The new scoring system proposes to diagnose PN only when patient’s performance is pathological and shows a clear spatial asymmetry between sides, which is crucial for a diagnosis of neglect. Pathological performance, not showing this spatial asymmetry, should be categorized as NSU. Adopting the new scoring system, we identified 11 patients (4.5%) whose performance was originally mis-diagnosed as either normal or evidence of contralesional PN (See Table 3). These apparent dissociations are resulting from the improved new scoring system and should not lead to any theoretical interpretation. It would be inappropriate to make any conclusion about the nature of the non-spatial underperformances, but it seems appropriate to suggest that more missing targets represent poorer performance. Interestingly, 16 of the 35 patients showing comprehension problems performed within the norm on the Fluff test. These findings suggest that patients with comprehension difficulties can reliably complete the Fluff test and aphasic patients should not be excluded a priori from PN assessment for clinical purposes. However, despite the new scoring can identify non-spatial underperformances, examiners should always be cautious in interpreting pathological performance associated with aphasia.

Assessing PN on a large sample of right and left brain damaged patients led to some interesting findings. Frequency of PN in the literature is rather variable (see Table 2 in Caggiano & Jehkonen, 2019; Committeri, Piervincenzi, & Pizzamiglio, 2018) depending on the test used and the body parts assessed. In line with a recent study using the Fluff test (Baas et al., 2011), we found that 31% of our RBD patients showed contralesional PN. A similar percentage (35%) was also found in our clinical sample when we considered the Comb and Razor/Compact performance. However, unlike our previous study in 2001 (Cocchini et al., 2001) we now found a significant correlation between the Fluff test and the Comb and

Razor/Compact test. This may be due to the fact that the clinical sample is now considerably larger and the lack of correlation was probably due to a lack of power of the 2001 study. We exclude that the new scoring system *per se* has had a significant role on this as a correlation between the two tests was also observed when the original scoring system was adopted in a large sample (Caggiano et al., 2014).

Another interesting finding is observed in a relatively small group of patients who showed different performance on the Fluff test and the Comb and Razor/Compact test. It should be noted that despite these two tests are investigating unilateral attentional disorder in the personal domain, they also show crucial differences: i) they target different body parts and some recent studies have shown as some brain areas (e.g. Extrastriate Body area) can selectively represent different body parts (e.g., Orlow, Makin & Zohary, 2010). Committeri et al. (2018) suggested that tests involving the face (e.g. Comb and Razor/Compact test) may rely on anterior regions within the inferior parietal cortex whereas tasks involving the rest of the body (e.g. Fluff test) may rely on more posterior portions of the parietal cortex.

Therefore, we cannot exclude that different networks may underlie the performance of tasks involving different body areas. ii) These two tests also differ on the type of tasks, repetitive and automatic for the Comb and Razor/Compact test, but goal-oriented for the Fluff test.

Again this may justify different performances. Finally, iii) some methodological aspects of the two tests place different emphasis on visual input and mental representation processes as the Comb and Razor/Compact test is performed with eyes open whereas the Fluff test is performed with eyes closed. Similarly, previous studies have observed dissociations between perceptual and representational extrapersonal neglect (e.g., Guariglia, Padovani, Pantano & Pizzamiglio, 1993; Beschin, Cocchini, Della Sala & Logie, 1997; see also Bartolomeo, 2002 for a revision). A detailed interpretation of this dissociation, based on current data, may lead to speculative conclusions; however, it is important to note that this observation reinforces

the importance to assess PN by means of a battery of tests to capture the complexity of this attentional disorder rather than rely on a single task.

Our findings also suggest that contralesional PN is significantly more severe, but not necessarily more frequent, after right rather than left brain lesion, confirming a crucial role of right hemisphere areas on spatial attention. However, the lack of a clear hemispheric asymmetry of frequency, which is commonly observed in cases of extrapersonal neglect (e.g., Stone et al., 1992), suggests that visuo-attention processes for the personal and extrapersonal domains may rely on different mechanisms. Some studies reported a selective difficulty of PN patients to perform mental representation tasks when these concern body images (Di Vita et al., 2017) and objects that are used with body parts (e.g. with hands; Bass et al., 2011), confirming that PN is a disorder specifically related to body representation (Di Vita et al., 2017). Committeri et al. (2007) identified different neuroanatomical substrates for these subtypes of neglect. According to these authors, awareness for extrapersonal space relies more on integrity of ventral circuits involving frontal and superior temporal areas; whereas personal neglect is linked to lesions of the inferior parietal areas causing functional disconnection between somatosensory information and abstract representations of the body. More recent studies confirmed that different lesion patterns are responsible for neglect in the personal and extrapersonal domains (e.g., Baas et al., 2011). A recent revision of the literature suggests that PN may result from a disconnection between parietal areas processing proprioceptive and somatosensory information with areas involved in the coding of abstract spatial information related to egocentric frames (Committeri et al., 2018). In particular, Di Vita and colleagues (2019) observed that the disconnection of the frontomarginal tract was significantly predicting PN.

Furthermore, ipsilesional neglect in the extrapersonal domain is rarely reported but it is not unusual (Robertson et al., 1994), especially following lesions in the left hemisphere (e.g. Stone, Halligan, & Greenwood, 1993) or when verbal tasks/stimuli are involved (e.g., Katz & Sevush, 1989; Cubelli, Nichelli, Bonito, De Tanti, & Inzaghi, 1991; Haywood & Coltheart, 2001). To our knowledge, the current study represents the first attempt to systematically investigate ipsilesional neglect in the personal domain. Interestingly, none of the right brain damaged patients in our sample showed evidence of ipsilesional PN; however, 6% of the LBD sample showed this type of PN. It is difficult to reconcile ipsilesional neglect with current attentional theories for neglect (see Danckert & Ferber, 2006 for a critical revision); however, our findings seem to imply that the left representational space is more susceptible to attentional distortion regardless the site of lesion. On the other hand, Robertson et al. (1994) suggested that ipsilesional neglect may represent a form of overcompensation over time, implying that originally these patients showed contralesional neglect. A possible overcompensation mechanism may explain some fluctuation in performance and possible different diagnosis for these patients when tested by two different tests. However, we would expect to observe ipsilesional neglect in more chronic phases. On the contrary, 5 out of 7 ipsilesional PN cases suffered from a brain lesion in the last 20-30 days, which is well below the LBD group average (i.e., 6.92 months). Moreover, it remains unclear why only LBD patients would show this effect of compensation in the personal domain. As mentioned above, PN seems to differ once more from extrapersonal neglect, suggesting different underlying mechanisms that need to be investigated in future studies.

To conclude, the new scoring system and interpretation of the Fluff test performance are based on original normative data and tolerance cut-off. Without inflating the number of PN cases, the proposed scoring system overcomes some limitations allowing to systematically

assess contralesional and ipsilesional PN, to evaluate the degree of PN severity and to exclude confounding variables. Findings on a sample of 243 brain damaged patients suggest that PN presents a more complex picture than previously thought, especially for ipsilesional neglect in LBD patients.

### **Disclosure statement**

No potential conflict of interest was reported by the authors.

### **Acknowledgements.**

We would like to thank Andrea Defilippi for proof-reading our manuscript.

## References

1. Albert, M. L. (1973). A simple test of visual neglect. *Neurology*, 23, 658–664.
2. Baas, U., Haan, B., Grässli, T., Karnath, H. O., Mueri, R., Perrig, W.J., Wurtz, P., Gutbrod, K. (2011). Personal neglect—a disorder of body representation? *Neuropsychologia*, 49(5), 898-905.
3. Bartolomeo, P. (2002). The relationship between visual perception and visual mental imagery: A reappraisal of the neuropsychological evidence. *Cortex*, 38, 357-378.
4. Beschin, N., Cocchini, G., Della Sala, S., Logie, R. (1997). What the eyes perceive, the brain ignores: A case of pure unilateral representational neglect. *Cortex*, 33, 3-26.
5. Beschin, N., & Robertson, I.H. (1997). Personal versus extrapersonal neglect: A group study of their dissociation using a reliable clinical test. *Cortex*, 33, 379-384.
6. Bisiach, E., Perani, D., Vallar, G., Berti, A. (1986). Unilateral neglect: Personal and extrapersonal. *Neuropsychologia*, 24, 759-767.
7. Burgess, P.W., & Shallice, T. (1997). *The Hayling and Brixton Tests*. London: Thames Valley Test Company.
8. Caggiano, P., & Jehkonen, M. (2018) The ‘Neglected’ Personal Neglect. *Neuropsychological Review*, 28(4): 417-435.
9. Caggiano, P., Beschin, N., Cocchini, G. (2014). Personal neglect following unilateral right and left brain damage. *Procedia-Social and Behavioral Sciences Journal*, 140, 164-16.
10. Cocchini, G., Beschin, N., Jehkonen, M. (2001). The Fluff Test: A simple task to assess body representation neglect. *Neuropsychological Rehabilitation*, 11 (1), 17-31.
11. Committeri, G., Pitzalis, S., Galati, G., Patria, F., Pelle, G., Sabatini, U., Castriota-Scanderbeg, A., Piccardi, L., Guariglia, C., Pizzamiglio, L. (2007). Neural bases of personal and extrapersonal neglect in humans. *Brain*, 130, 431–441.
12. Committeri, G., Piervincenzi, C., Pizzamiglio, L. (2018). Personal neglect: A comprehensive theoretical and anatomo–clinical review. *Neuropsychology*, 32(3), 269–279.
13. Cubelli, R., Nichelli, P., Bonito, V., De Tanti, A., Inzaghi, G. (1991). Different patterns of dissociation in unilateral spatial neglect. *Brain and Cognition*, 15: 139–59.
14. Danckert, J. & Ferber, S. (2006). Revisiting unilateral neglect. *Neuropsychologia*, 44, 987-1006.
15. Di Vita, A., Palermo, L., Boccia, M., Guariglia C. (2019) Topological map of the body in post-stroke patients: Lesional and Hodological aspects. *Neuropsychology*, 33(4), 499-507.

16. Di Vita, A., Palermo, L., Piccardi, L., Di Tella J., Propato, F., Guariglia, C. (2017) Body representation alterations in Personal but not in extrapersonal neglect patients. *Applied Neuropsychology: Adult*, 24, 308-317.
17. Guariglia, C., & Antonucci, G. (1992). Personal and extrapersonal neglect: A case of neglect dissociation. *Neuropsychologia*, 30, 1001-1009.
18. Guariglia, C., Padovani, A., Pantano, P., Pizzamiglio, L. (1993). Unilateral neglect restricted to visual imagery. *Nature*, 364, 235-237.
19. Haywood, M., & Coltheart, M. (2001). Neglect dyslexia with a stimulus-centred deficit and without visuospatial neglect. *Cognitive Neuropsychology*, 18: 577–615.
20. Katz, R.B., & Sevush, S. (1989). Positional dyslexia. *Brain and Language*, 37, 266–289.
21. McIntosh, R.D., Brodie, E.E., Beschin, N., Robertson, I.H. (2000). Improving the clinical diagnosis of personal neglect: A reformulated comb and razor test. *Cortex*, 36(2), 289–292.
22. Orlov, T., Makin T.R., Zohary, E. (2010). Topographic Representation of the Human Body in the Occipitotemporal Cortex. *Neuron*, 68, 586-600.
23. Robertson, I.H., Halligan, P.W, Bergego, C., Hömberg, V., Pizzamiglio, L., Weber, E., et al. (1994). Right neglect following right hemisphere damage? *Cortex*, 30: 199–213.
24. Spinnler, H., & Tognoni, G. (1987). Gruppo italiano per lo studio neuropsicologico dell'invecchiamento. Standardizzazione e taratura italiana di test neuropsicologici. *The Italian Journal of Neurological Sciences*, 6: 20–119.
25. Stone, S.P., Halligan, P.W, Greenwood, R.J. (1993). The incidence of neglect phenomena and related disorders in patients with an acute right or left hemisphere stroke. *Age and Ageing*, 22: 46–53.
26. Stone SP, Patel P, Greenwood RJ, Halligan PW. (1992). Measuring visual neglect in acute stroke and predicting its recovery: the visual neglect recovery index. *J Neurol Neurosurg Psychiatry*, 55: 431–436.
27. Wilson, B. A., Cockburn, J., Halligan, P. (1987). *Behavioural inattention test*. Flempton: Thames Valley Test Company.