

BRIEF COMMUNICATION

Recognition of Positive Vocalizations Is Impaired in Behavioral-Variant Frontotemporal Dementia

Sharpley Hsieh,^{1,2} John R Hodges,^{1,2} AND Olivier Piguet^{1,2}

¹Neuroscience Research Australia, Randwick, New South Wales, Australia

²School of Medical Sciences, University of New South Wales, New South Wales, Australia

(RECEIVED August 3, 2012; FINAL REVISION December 12, 2012; ACCEPTED December 13, 2012; FIRST PUBLISHED ONLINE FEBRUARY 1, 2013)

Abstract

Recognition of negative emotions is impaired in behavioral-variant frontotemporal dementia (bvFTD). Less is known about the identification of positive emotions. One limitation likely arises from the stimulus sets used in previous studies. The widely used Ekman 60 Faces Test, for example, consists of four negative emotions (anger, fear, disgust and sadness) but only one positive emotion (happiness). Here, patients with bvFTD ($n = 9$), AD ($n = 9$), and controls ($n = 15$) recognized a range of experimentally-validated positive and negative non-verbal vocalizations (e.g., cheers for triumph; retching for disgust) that have recently become available. The bvFTD group was impaired in the recognition of both positive and negative vocalizations. In contrast, performance in the AD cohort was comparable to that of controls. Findings in the bvFTD group point to a global emotion recognition deficit in this syndrome. These results are consistent with a growing body of research showing that deficits also extend to positive emotions. (*JINS*, 2013, 19, 483–487)

Keywords: dementia, Alzheimer's disease, Emotion recognition, Faces, Voices, Frontotemporal dementia

INTRODUCTION

Impaired emotion recognition in the behavioral-variant subtype of frontotemporal dementia (bvFTD) is evident particularly for negative emotions, whereas the recognition of positive emotions is generally intact (Kumfor & Piguet, 2012). One limitation, however, is that emotion stimuli sets typically contain more negative than positive emotions. The widely used Ekman 60 Faces Test, for example, consists of four negative emotions (anger, fear, disgust, and sad) and only one prototypical positive (happiness) emotion. Moreover, happiness is visually distinctive (curved lips representing a smile) and is easier to differentiate than the other negative facial emotions in this test. Experimentally validated non-verbal vocalizations have recently become available which communicates several positive (e.g., cheers for triumph) and negative (e.g., retching for disgust) emotions (Sauter & Scott, 2007). Importantly, vocalizations do not contain linguistic information but are able to encompass a wider range of emotions that may not necessarily be expressed in prosody (e.g., groans or bursts of laughter) and are thought to be auditory equivalent to facial

emotions (Belin, Fecteau, & Bedard, 2004). An opportunity exists to investigate whether emotion processing deficits are present also for positive vocalizations in bvFTD. The recognition of positive and negative emotions using vocalizations as well as that of facial emotions (Ekman 60 Faces Test) was investigated in bvFTD in comparison to Alzheimer's disease (AD) group and an age- and education-matched control cohort.

METHODS

Participants

Eighteen patients (9 bvFTD; 9 AD) were recruited from the Frontier Frontotemporal Dementia Research Group where they were diagnosed by a senior neurologist (J.R.H.). Behavioral-variant FTD patients met current consensus criteria for FTD (Rascovsky, et al., 2011) and were characterized by an insidious onset of decline in social behavior (e.g., disinhibition, apathy, loss of empathy), cognition (e.g., executive deficits with relative sparing of other cognitive functions), as well as functional abilities. Imaging results showed predominantly bifrontal atrophy on 3 Tesla magnetic resonance imaging scanner. AD patients met the consensus criteria for probable AD (McKhann et al., 2011) and were characterized by prominent deficits in episodic

Correspondence and reprint requests to: Olivier Piguet, Neuroscience Research Australia, PO Box 1165, Randwick, NSW, 2031. E-mail: o.piguet@neura.edu.au

memory as well as mild impairment in other non-memory domains (e.g., visuospatial, language, and executive functions); social behavior was relatively preserved. Control participants were selected from a healthy volunteer panel and were excluded if they had any neurological (e.g., epilepsy, prior significant head injury) or psychiatric (e.g., depression) conditions. All controls scored above the cutoff (88/100) on a cognitive screening measure, the Addenbrooke's Cognitive Examination – Revised (ACE-R) (Mioshi, Dawson, Mitchell, Arnold, & Hodges, 2006). All participants provided informed consent for the study; dual consent was obtained from the carer for some patients. Participants volunteered their time but were reimbursed for travel costs where required. This study was approved by the Southern Sydney and Illawarra Area Health Service and the University of New South Wales ethics committees.

Emotional Stimuli and Procedure

Experimentally validated sets of emotional stimuli were used. Vocalizations included five positive (achievement, amusement, pleasure, relief, and triumph) and four negative (anger, disgust, fear, and sadness) emotions. Ten items were used per category, which resulted in a total set of 90 stimuli (Sauter & Scott, 2007). Each vocalization lasted approximately 1 s. Facial expressions of emotion consisted of the Ekman 60 Faces Test (Young, Perrett, Calder, Sprengelmeyer, & Ekman, 2002), which consisted of two positive (happiness, surprise) and four negative (fear, disgust, sadness, and anger). Each facial emotion was displayed on the computer for 5 s. The two emotion tests were counter-balanced across participants.

On a forced-choice recognition task, participants were required to select one label from a list of choices for each stimulus. The list of response choices was visible throughout testing and consisted of all of the emotion labels available for the test being administered; in other words, the response list contained nine labels for the vocalization test (achievement, amusement, pleasure, relief, triumph, anger, disgust, fear, and sadness) and six labels for the facial emotion test (happiness, surprise, fear, disgust, sadness, and anger). No time limit was enforced for each trial; subsequent trials did not proceed until a response was recorded. Participants were able to listen to the vocalization a second time, if requested. All testing was performed in a quiet room. Practice trials were administered to the participants to ensure that they could see and hear the stimuli being presented.

Statistical Analyses

Data were analyzed using IBM SPSS Statistics (Version 20). Consistent with previous work, scores on the individual vocalizations and facial emotions were averaged to form two composite scores: positive and negative emotions (Hsieh, Hornberger, Piguet, & Hodges, 2012). Because of the sample size, correct responses for the individual emotions and misrecognition errors were not analyzed separately, but this information is provided in the Appendix. First, a Diagnosis (bvFTD, AD, controls) \times Emotion (positive, negative) mixed

analysis of variance (ANOVA) was conducted separately for the set of facial emotions and vocalizations. Next, group comparisons for the recognition of positive and negative emotions were analyzed using one-way ANOVA with *post hoc* comparisons using the Tukey HSD test. The Kruskal-Wallis test was used where data were non-parametric, followed by *post hoc* testing using the Mann-Whitney *U* test. A *p* value of $<.05$ was adopted.

RESULTS

As can be seen in Table 1, groups were matched for age [$H(2) = 0.12$; $p > .05$] and years of education [$F(2,32) = 3.14$; $p > .05$]. Significant group differences on the ACE-R [$F(2,32) = 20.7$; $p < .05$; $\eta^2 = 0.58$] indicated that, as expected, each patient group scored below the control group ($p < .05$ for all pairwise comparisons). Patient groups did not differ on the ACE-R ($p > .05$). Patient groups were also matched on the Clinical Rating Scale-Sum of Boxes score (CDR; Berg et al., 1992) ($U = 10.0$; $Z = -1.22$; $p > .05$). While there were more men than women, sex distribution was not statistically different between groups (Fisher exact test = 0.48; $p > .05$; Cramer's $V = 0.39$; $p > .05$).

The overall 3×2 ANOVA on the composite negative and positive vocalizations across groups revealed a significant Diagnosis \times Emotion interaction [$F(2,30) = 4.41$; $p < .05$; $\eta^2 = 0.23$] indicating that the recognition of positive and negative vocal emotions differed across the diagnostic groups as a function of the stimuli valence. The main effect of Diagnosis was significant [$F(2,30) = 10.3$; $p < .05$; $\eta^2 = 0.41$] with recognition accuracy significantly worse in the bvFTD than AD and control groups ($p < .05$ for both comparisons). The main effect of Emotion was not significant ($p > .05$). To clarify the interaction further, separate analyses were conducted on each composite score. These analyses showed a significant group effect for both positive [$F(2,32) = 14.6$; $p < .05$; $\eta^2 = 0.49$]

Table 1. Demographic data and recognition accuracy of positive and negative vocalisations and facial emotions in the patient and control groups

	bvFTD	AD	Controls
<i>n</i>	9	9	15
Male/Female	9/0	7/2	9/6
Age	62.5 (8.7)	64.2 (7.1)	64.8 (6.2)
Education (years)	10.9 (2.2)	14.1 (3.9)	13.5 (2.7)
CDR-SB	4.86 (2.9)	3.20 (2.1)	N/A
ACE-R (/100)	78.3 (10.8) ^a	83.2 (4.4) ^a	95.0 (3.6)
Vocalizations			
Negative	55.3 (20.4) ^a	70.0 (13.5)	74.0 (11.1)
Positive	49.1 (20.9) ^a	68.2 (13.1)	81.1 (8.6)
Facial emotions			
Negative	50.8 (11.2) ^a	68.3 (15.1) ^a	81.8 (8.3)
Positive	88.9 (15.8)	93.9 (6.5)	92.7 (6.8)

Standard deviation is presented in brackets. ^a $p < .05$ vs. controls
CDR-SB = Clinical Dementia Rating-Sum of Boxes score; ACE-R = Addenbrooke's Cognitive Examination-Revised

and negative [$F(2,32) = 4.67$; $p < .05$; $\eta^2 = 0.24$] emotions. *Post hoc* comparisons revealed that the bvFTD group was significantly worse in comparison to controls for both positive and negative vocalizations ($p < .05$ for all comparisons); the AD group, in contrast, did not differ significantly from controls ($p > .05$ for both comparisons). In addition, the bvFTD group was also significantly worse than the AD group at the recognition of positive vocalizations ($p < .05$).

The overall 3×2 ANOVA on the composite negative and positive facial emotion scores across groups revealed a significant Diagnosis \times Emotion interaction [$F(2,30) = 15.3$; $p < .05$; $\eta^2 = .51$] indicating that the recognition of positive and negative facial emotions also differed across the diagnostic groups as a function of the stimuli valence. The main effects of Diagnosis [$F(2,30) = 10.9$; $p < .05$; $\eta^2 = 0.42$] and Emotion [$F(1,30) = 136.5$; $p < .05$; $\eta^2 = 0.82$] were also significant. Again, the bvFTD group was significantly worse at recognizing facial emotions when compared to AD and control groups. Further analyses were conducted on each of the composite scores for positive and negative facial emotions separately to clarify the previous interaction. These analyses showed a significant group effect for negative emotions only [$F(2,32) = 21.5$; $p < .05$; $\eta^2 = 0.59$]. *Post hoc* comparisons revealed that both patient groups differed significantly from controls ($p < .05$ for both comparisons). The bvFTD cohort was also significantly worse than the AD group at recognizing negative facial emotions ($p < .05$). As expected, patient groups did not differ in the recognition of positive facial emotions [$F(2,32) = 0.64$; $p > .05$].

DISCUSSION

This study is the first to show that the recognition of a range of positive vocalizations is impaired in bvFTD. Findings are consistent with recent work showing impaired identification of positive emotions in non-facial modalities, such as music, and also in the comprehension of emotion words in bvFTD (Hsieh et al., 2012; Omar et al., 2011). This study highlights, therefore, that the disproportionate bias against the recognition of negative emotions reported in the literature may be partially attributed to the stimulus sets used in previous studies.

The pattern of performance between the dementia groups differed according to modality and valence: the bvFTD group was impaired in the identification of emotions in faces and vocalizations, with the exception of positive emotion conveyed by faces. In contrast, AD cohort was impaired only in the recognition of negative facial emotions. These findings emphasize the primary emotion processing deficit characteristic of bvFTD. In addition to the orbitofrontal-insular and anterior temporal cortices, atrophy in bvFTD also involves the amygdala, a structure central to the processing of emotional stimuli. We would like to suggest that it is, therefore, likely that amygdalar atrophy underlies this deficit in bvFTD. The role of the amygdala in the processing of facial expressions of emotion is well recognized in both lesion studies as well as neuroimaging work (Adolphs, Tranel, Damasio, &

Damasio, 1994; Phelps & LeDoux, 2005). Involvement of the amygdala in the processing of negative as well as positive emotional vocalizations has been demonstrated in recent fMRI work (Fecteau, Belin, Joanette, & Armony, 2007). In AD, however, impaired performance across the two emotion modalities may be attributable, at least in part, to concurrent cognitive deficits (such as visuospatial deficits) that are found in the disease. Loss of gray matter in posterior parietal and occipital cortices is more severe in AD than in FTD whereas lesions to the limbic system in FTD outweigh those found in AD (Rabinovici et al., 2008). The pattern of performance in the AD group highlights the need to examine the integrity of auditory and visual systems and determine their contribution to the network of structures involved for emotion processing (Rohrer, Sauter, Scott, Rossor, & Warren, 2012).

We were not able to examine the individual emotions separately. Such an approach could help characterize the positive emotion deficits in bvFTD. This issue is relevant particularly for the emotion surprise; whether this emotion is regarded as positive, negative, or exists separately as a category of its own (Toivonen et al., 2012), remain contentious. Furthermore, analyses of misrecognition errors for individual emotion categories in future work may also reveal response biases that differentiates between bvFTD and AD cohorts. It is possible that bvFTD patients use some labels disproportionately (e.g., confusing amusement with disgust or fear; see Appendix) which are not present in either the AD or control groups. A useful statistical method which takes into account the disproportionate use of a particular category on forced-choice paradigms is that of the "Hu" score, which calculates an unbiased hit rate based upon the participant's use of the different response alternatives (Wagner, 1993).

Finally, a useful adjunct to pursue in the future is to further examine the neural correlates of individual emotion categories; certain brain regions are thought to be biased for specific emotions (e.g., insula for disgust; Chapman & Anderson, 2012) but those for positive emotions remain elusive. The neural basis of non-verbal auditory emotions has been examined in the dementias using total recognition scores (Omar et al., 2011; Rohrer et al., 2012; Hsieh, Hornberger, et al., 2012). The lack of differentiation, particularly for positive emotions, in the literature so far is likely to have resulted from the loss of statistical power from ceiling effects for the recognition of happiness using stimuli that were previously available. While sample sizes in the patient groups in this study are in keeping with the cohorts that have been studied (e.g., $n = 7$; Drapeau, Gosselin, Gagnon, Peretz, & Lorrain, 2009), larger cohorts will be needed to examine the neural correlates of vocal emotion recognition.

ACKNOWLEDGMENTS

This research was supported by a National Health and Medical Research Council (NHMRC) of Australia Project Grant (510106) and the ARC Centre of Excellence in Cognition and its Disorders (CE110001021). J.R.H. is supported by an Australian Research

Council Federation Fellowship (FF0776229). O.P. is supported by an NHMRC Clinical Career Development Fellowship (APP1022684). We thank participants who support the Frontier Frontotemporal Dementia Research Group. There are no conflicts of interest.

REFERENCES

- Adolphs, R., Tranel, D., Damasio, H., & Damasio, A. (1994). Impaired recognition of emotion in facial expressions following bilateral damage to the human amygdala. *Nature*, *372*(6507), 669–672.
- Belin, P., Fecteau, S., & Bedard, C. (2004). Thinking the voice: Neural correlates of voice perception. *Trends in Cognitive Sciences*, *8*(3), 129–135.
- Berg, L., Miller, J.P., Baty, J., Rubin, E.H., Morris, J.C., & Figiel, G. (1992). Mild senile dementia of the Alzheimer type. 4. Evaluation of intervention. *Annals of Neurology*, *31*(3), 242–249.
- Chapman, H.A., & Anderson, A.K. (2012). Understanding disgust. *Annals of the New York Academy of Sciences*, *1251*, 62–76.
- Drapeau, J., Gosselin, N., Gagnon, L., Peretz, I., & Lorrain, D. (2009). Emotional recognition from face, voice, and music in dementia of the Alzheimer type. *Annals of the New York Academy of Sciences*, *1169*, 342–345.
- Fecteau, S., Belin, P., Joanette, Y., & Armony, J.L. (2007). Amygdala responses to nonlinguistic emotional vocalizations. *Neuroimage*, *36*(2), 480–487.
- Hsieh, S., Foxe, D., Leslie, F., Savage, S., Piguet, O., & Hodges, J.R. (2012). Grief and joy: Emotion word comprehension in the dementias. *Neuropsychology*, *26*(5), 624–630.
- Hsieh, S., Hornberger, M., Piguet, O., & Hodges, J.R. (2012). Brain correlates of musical and facial emotion recognition: Evidence from the dementias. *Neuropsychologia*, *50*(8), 1814–1822.
- Kumfor, F., & Piguet, O. (2012). Disturbance of emotion processing in frontotemporal dementia: A synthesis of cognitive and neuroimaging findings. *Neuropsychology Review*, *11*, 11.
- McKhann, G.M., Knopman, D.S., Chertkoff, H., Hyman, B.T., Jack, C.R. Jr., Kawas, C.H., ... Phelps, C.H. (2011). The diagnosis of dementia due to Alzheimer's disease: Recommendations from the National Institute on Aging-Alzheimer's Association workgroups on diagnostic guidelines for Alzheimer's disease. *Alzheimers & Dementia*, *7*(3), 263–269.
- Mioshi, E., Dawson, K., Mitchell, J., Arnold, R., & Hodges, J.R. (2006). The Addenbrooke's Cognitive Examination Revised (ACE-R): A brief cognitive test battery for dementia screening. *International Journal of Geriatric Psychiatry*, *21*(11), 1078–1085.
- Omar, R., Henley, S.M., Bartlett, J.W., Hailstone, J.C., Gordon, E., Sauter, D.A., ... Warren, J.D. (2011). The structural neuroanatomy of music emotion recognition: Evidence from frontotemporal lobar degeneration. *Neuroimage*, *56*(3), 1814–1821.
- Phelps, E.A., & LeDoux, J.E. (2005). Contributions of the amygdala to emotion processing: From animal models to human behavior. *Neuron*, *48*(2), 175–187.
- Rabinovici, G.D., Seeley, W.W., Kim, E.J., Gorno-Tempini, M.L., Rascovsky, K., Pagliaro, T.A., ... Rosen, H.J. (2008). Distinct MRI atrophy patterns in autopsy-proven Alzheimer's disease and frontotemporal lobar degeneration. *American Journal of Alzheimer's Disease and Other Dementias*, *22*(6), 474–488.
- Rascovsky, K., Hodges, J.R., Knopman, D., Mendez, M.F., Kramer, J.H., Neuhaus, J., ... Miller, B.L. (2011). Sensitivity of revised diagnostic criteria for the behavioural variant of frontotemporal dementia. *Brain*, *134*(9), 2456–2477.
- Rohrer, J.D., Sauter, D., Scott, S., Rossor, M.N., & Warren, J.D. (2012). Receptive prosody in nonfluent primary progressive aphasia. *Cortex*, *48*(3), 308–316.
- Sauter, D.A., & Scott, S.K. (2007). More than one kind of happiness: Can we recognize vocal expressions of different positive states? *Motivation and Emotion*, *31*(3), 192–199.
- Toivonen, R., Kivela, M., Saramaki, J., Viinikainen, M., Vanhatalo, M., & Sams, M. (2012). Networks of emotion concepts. *PLoS One*, *7*(1), e28883.
- Wagner, H. (1993). On measuring performance in category judgment studies of nonverbal behaviour. *Journal of Nonverbal Behavior*, *17*(1), 3–28.
- Young, A.W., Perrett, D.I., Calder, A.J., Sprengelmeyer, R., & Ekman, P. (2002). *Facial expressions of emotions: Stimuli and test (FEEST)*. Thurstone, UK: Thames Valley Test Company.

APPENDIX

Confusion matrices in the patient groups and controls for the nine non-verbal vocalizations

	Response								
	Angry	Disgust	Fear	Sad	Surprise	Triumph	Amusement	Pleasure	Relief
bvFTD (n = 9)									
Angry	50.0	20.0	10.1	4.4	4.4	5.6	3.3	1.1	1.1
Disgust	4.4	76.7	7.8	1.1	1.1	1.1	0.0	3.3	4.4
Fear	7.8	2.2	45.6	7.8	14.4	3.3	10.0	2.2	6.7
Sad	3.3	8.9	4.4	48.9	1.1	2.2	3.3	14.4	13.3
Surprise	2.2	13.3	11.1	4.4	41.1	3.3	10.0	7.8	6.7
Triumph	1.1	3.3	1.1	1.1	12.2	26.7	11.1	7.8	12.2
Amusement	0.0	2.2	1.1	1.1	12.2	1.1	67.8	7.8	6.7
Pleasure	1.1	2.2	2.2	3.3	7.8	8.9	3.3	46.7	24.4
Relief	0.0	2.2	0.0	1.1	5.6	10.0	6.7	11.1	63.3

(Continued)

Continued

	Response								
	Angry	Disgust	Fear	Sad	Surprise	Triumph	Amusement	Pleasure	Relief
AD (<i>n</i> = 9)									
Angry	56.7	24.4	6.7	3.3	3.3	2.2	0.0	0.0	3.3
Disgust	0.0	93.3	1.1	0.0	0.0	0.0	0.0	1.1	4.4
Fear	2.2	7.8	61.1	3.3	18.9	2.2	3.3	0.0	1.1
Sad	1.1	3.3	4.4	68.9	2.2	3.3	1.1	5.6	10.0
Surprise	0.0	15.6	3.3	1.1	65.6	4.4	0.0	2.2	7.8
Triumph	3.3	0.0	4.4	0.0	18.9	55.6	5.6	10.0	2.2
Amusement	0.0	0.0	0.0	5.6	2.2	2.2	77.8	12.2	0.0
Pleasure	3.3	2.2	2.2	5.6	3.3	0.0	0.0	60.0	23.3
Relief	1.1	2.2	2.2	4.4	0.0	3.3	0.0	4.4	82.2
Controls (<i>n</i> = 15)									
Angry	59.3	15.3	8.0	2.7	2.0	4.0	0.0	2.7	6.0
Disgust	0.0	96.7	0.0	0.0	1.3	1.3	0.0	0.7	0.0
Fear	0.7	3.3	72.7	3.3	16.0	1.3	0.7	0.0	2.0
Sad	2.0	2.7	5.3	67.3	2.7	0.0	0.0	12.0	8.0
Surprise	2.0	7.3	4.0	1.3	76.0	2.7	0.7	0.0	6.0
Triumph	2.7	0.0	2.7	0.7	20.0	62.7	0.7	3.3	7.3
Amusement	0.0	0.0	0.0	0.0	0.0	0.0	99.3	0.7	0.0
Pleasure	0.0	0.7	0.0	3.3	2.7	2.0	0.0	76.7	14.7
Relief	0.0	1.3	0.7	2.0	0.0	2.0	0.0	3.3	90.7

All rows add to 100%. Correct responses are marked in bold.