

The controversy could be partly due to the material in the test; it is a prerequisite that difficulty level of lexical-semantic aspects is even among sentences used in the tests. Thus, we conducted the present study to evaluate the deficits of metaphor and sarcasm in AD using a questionnaire that consists of the same type of sentences with similar difficulty levels and whose efficacy was validated for differential diagnosis of developmental disorders in children.²² For a better understanding of characteristics of AD, error patterns were analyzed. We hypothesized that comprehension might be deteriorated at the early stages of disease and sarcasm comprehension might be deteriorated earlier than metaphor comprehension.

Methods

Participant

The participants were 31 young normal controls (YNC), 104 aged normal controls (ANC), 42 patients with amnesic mild cognitive impairment (aMCI), and 30 patients with mild AD in Clinical Dementia Rating scale (CDR) 1. The YNC were university students and ANC were recruited from community dwellers, who underwent clinical interviews by a clinician who specialized in evaluation of dementia. Patients were recruited from the outpatient clinics. The exclusion criteria were psychiatric diseases and delirium. Verbal incomprehension was also an exclusion criterion. The participants were required to read out the questions and those who lacked fluency were excluded. Concerning language ability, the participants received the Mini-Mental State Examination (MMSE) and were confirmed to have the capacity to name simple objects, repeat phrases, follow written commands, and write a sentence with a noun and a verb. The participants were diagnosed based on the criteria for AD by National Institute of Neurological and Communicative Disorders and Stroke-Alzheimer's Disease and Related Disorders Association²³ and on the criteria for aMCI by the report of the International Working Group on Mild Cognitive Impairment.²⁴ Patients with aMCI were limited to those free from objective symptoms of other types of dementia such as dementia with Lewy bodies or frontotemporal dementia. The Ethics Board of the Gunma University School of Health Sciences approved all procedures (No. 21-26), and written informed consent was obtained from all the participants.

Task

Metaphor and sarcasm comprehension was evaluated by the Metaphoric and Sarcastic Scenario Test (MSST), which was developed for discrimination of high functioning pervasive developmental disorders from attention deficit/hyperactivity disorders in young children.²² This test consists of 5 metaphoric and 5 sarcastic sentences; metaphoric sentences are odd numbered and sarcastic sentences even. The words and sentences in MSST were selected from standard textbooks of Japanese language (Mitsumura Press) for 1st, 2nd, and 3rd grades in elementary school. Therefore, the lexical-semantic components were not above the levels for those who completed

6 years of elementary school education. The test employed a multiple-choice style, that is, 1 choice was correct and 4 were incorrect. The wrong choices included a literal interpretation, an answer associated with part of the sentence, misunderstanding of the sentence, and not knowing. The number of correct answers represented the metaphor score and sarcasm score, respectively. Each pattern of incorrect answers was totaled. Cognitive performance was assessed using MMSE.

Analysis

Group comparison of scores and the 4 error scores were conducted using the repeated measures analysis of variance (metaphor/sarcasm vs 4 participant groups).

Among aged groups, we conducted the repeated measures analysis of covariance (metaphor/sarcasm vs 3 participant groups) with covariates of age, sex, education, and MMSE scores. A post hoc test was conducted with multiple comparisons with Bonferroni correction. All analyses were conducted using the Japanese version of SPSS for Windows version 19.0 (IBM Corporation, New York). Significance was set as $P < .05$.

Results

Demographic scores are shown in Table 1. The results of the MSST are shown in Table 2 and Figure 1. The main effect indicated that sarcasm was more difficult to comprehend than metaphor ($F_{1,203} = 54.634, P < .001$), and interaction with participant groups was also significant ($F_{3,203} = 3.354, P = .020$). According to within-subject post hoc analysis, no significant difference was observed between metaphor and sarcasm scores in YNC ($P = .442$), whereas in ANC, aMCI, and mild AD, scores of sarcasm was significantly lower than that of metaphor ($P < .001$ in all the groups). According to between-subject post hoc analysis, metaphor scores were not different between YNC and ANC, whereas metaphor scores were significantly better in ANC than in aMCI ($P = .011$) and in aMCI than mild AD ($P < .001$). Sarcasm scores were significantly better in YNC than in ANC ($P = .040$), in ANC than in aMCI ($P = .005$), and in aMCI than in mild AD ($P = .002$).

Concerning the error patterns, group differences were observed only in literal interpretation and there were no group differences in the other 3 error patterns (an answer associated with a part of the sentence, misunderstanding of the sentence, and not knowing; Table 3, Figure 2). The main effect was significant ($F_{1,203} = 34.283, P < .001$) and interaction was also significant ($F_{3,203} = 6.887, P < .001$). According to the between-subject post hoc analysis, errors of literal interpretation of metaphor and sarcasm comprehension were not different between YNC and ANC ($P = 1.000$ in both), and ANC and aMCI ($P = .115, P = .349$, respectively), whereas a significant difference was observed between aMCI and mild AD ($P < .001$ in both). According to within-subject post hoc analysis, the errors of literal interpretation were more in sarcasm than in metaphor in aMCI ($P = .038$) and in mild AD ($P < .001$), whereas there was no significant difference in YNC ($P = .187$) and in ANC ($P = .072$).

Table 1. Demographic Data^a

	n	Age	Gender	Education	MMSE
		Mean \pm SD	Male, Female	Mean \pm SD	Mean \pm SD
YNC	31	19.3 \pm 1.4	M10, F21	13.3 \pm 0.6	
ANC	104	72.1 \pm 4.2	M25, F79	12.0 \pm 2.3	28.4 \pm 1.4
aMCI	42	74.0 \pm 5.4	M18, F24	11.1 \pm 3.0	25.8 \pm 1.7
AD	30	78.0 \pm 7.2	M6, F24	9.3 \pm 2.3	21.4 \pm 4.0

Abbreviations: YNC, young normal controls; ANC, aged normal controls; aMCI, amnesic mild cognitive impairment; AD, patients with mild Alzheimer's disease in clinical dementia rating 1; MMSE, Mini-Mental State Examination; SD, standard deviation.

^a The rate of gender difference was not different among the groups ($P = .088$, chi-squared statistic). Concerning age, there was no difference between ANC and aMCI, but patients with mild AD were significantly older than ANC and aMCI ($P < .001$, $P = .004$, respectively). Concerning years of education, there was no difference between ANC and aMCI, but patients with mild AD received significantly shorter education than the patients with ANC and aMCI did ($P < .001$, $P = .006$, respectively). Scores of MMSE was significantly different among groups ($P < .001$, among all the groups).

Table 2. Correct Answers

	Metaphor		Sarcasm		P
	Mean \pm SD	P Value ^a	Mean \pm SD	P Value ^a	
YNC	5.0 \pm 0.2		4.8 \pm 0.4		.442
YNC vs ANC		1.000		.040*	
ANC	4.8 \pm 0.7		4.1 \pm 1.2		<.001**
ANC vs aMCI		.011*		.005*	
aMCI	4.3 \pm 1.2		3.4 \pm 1.3		<.001**
aMCI vs AD		<.001**		.002*	
AD	3.3 \pm 1.2		2.3 \pm 1.6		<.001**

Abbreviations: YNC, young normal controls; ANC, aged normal controls; aMCI, amnesic mild cognitive impairment; AD, patients with mild Alzheimer's disease in clinical dementia rating 1; SD, standard deviation.

^a The difference among groups analyzed by between-subject post hoc analysis of 2×4 analysis of variance (metaphor and sarcasm; 4 groups).

^b The difference between metaphor and sarcasm analyzed by within-subject post hoc analysis of 2×4 analysis of variance (metaphor and sarcasm; 4 groups).

* $P < .05$.

** $P < .001$.

There was weak correlation between MMSE scores and metaphor ($r = .362$, $P < .001$) and sarcasm scores ($r = .337$, $P < .001$).

The difference among the aged groups of ANC, aMCI, and mild AD remained by the repeated measures analysis of covariance with covariates of age, sex, education, and MMSE scores. According to within-subject post hoc analysis, in ANC, aMCI, and mild AD, scores of sarcasm was significantly lower than that of metaphor ($P < .001$, $P < .001$, $P = .004$, respectively). According to between-subject post hoc analysis, metaphor scores were significantly better in ANC than in aMCI ($P = .040$) and in aMCI than mild AD ($P = .002$). Sarcasm comprehension was significantly better in ANC than in aMCI ($P = .021$) and in aMCI than in mild AD ($P = .023$).

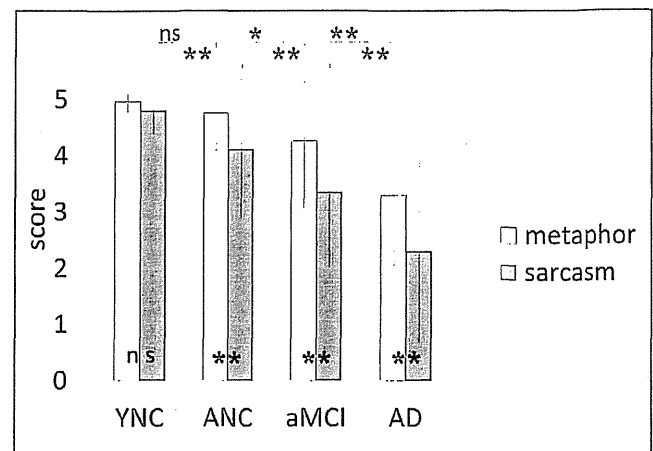


Figure 1. Scores of correct answers. Sarcasm scores were significantly lower in ANC than YNC, whereas metaphor scores were not different between the 2 groups. Metaphor scores were deteriorated from MCI. Post hoc analysis of 2×4 analysis of variance (metaphor and sarcasm; 4 groups) was conducted; * in upper row indicates statistical significance of between subject analysis of metaphor, * in middle row indicates that of sarcasm, and * in the bottom row indicates statistical significance calculated by intrasubject analysis. * $P < .05$, $P < .001$. YNC indicates young normal controls; ANC, aged normal controls; aMCI, amnesic mild cognitive impairment; AD, patients with mild Alzheimer's disease in clinical dementia rating 1.

Table 3. Errors of Literal Answers

	Metaphor		Sarcasm		
	Mean \pm SD	P Value ^a	Mean \pm SD	P Value ^a	P Value ^b
YNC	0.00 \pm 0.00		0.19 \pm 0.40		.187
YNC vs ANC		1.000		1.000	
ANC	0.05 \pm 0.21		0.19 \pm 0.44		.072
ANC vs aMCI		.115		.349	
aMCI	0.21 \pm 0.47		0.48 \pm 0.77		.038*
aMCI vs AD		<.001**		<.001**	
AD	0.87 \pm 0.82		1.77 \pm 1.72		<.001**

Abbreviations: YNC, young normal controls; ANC, aged normal controls; aMCI, amnesic mild cognitive impairment; AD, patients with mild Alzheimer's disease in clinical dementia rating 1; SD, standard deviation.

^a The difference among groups analyzed by between-subject post hoc analysis of 2×4 analysis of variance (metaphor and sarcasm; 4 groups).

^b The difference between metaphor and sarcasm analyzed by within-subject post hoc analysis of 2×4 analysis of variance (metaphor and sarcasm; 4 groups).

* $P < .05$.

** $P < .001$.

Discussion

Scores for both metaphor and sarcasm were not significantly different from each other in YNC, which confirmed that the difficulty level of metaphor and sarcasm comprehension tested by MSST was not different, at least among young participants.

The result suggested that deterioration of sarcasm comprehension was an age-related change. Sarcasm scores were

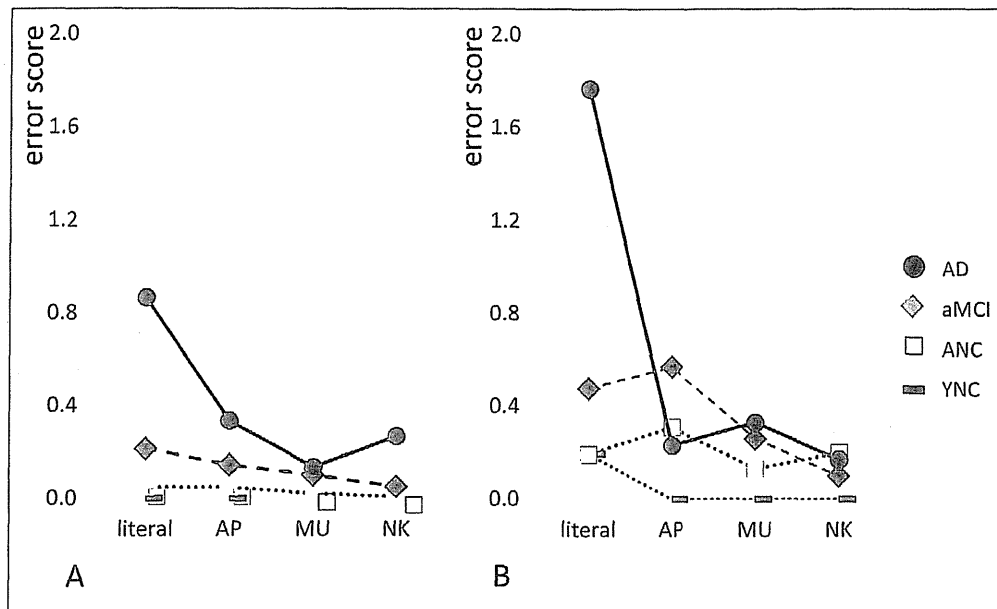


Figure 2. Error patterns. Error patterns of metaphor (A) and sarcasm (B). Significant differences among groups were observed in literal errors in both metaphor and sarcasm and the other 4 patterns of error were not significantly different among groups. AD indicates patients with mild Alzheimer's disease in clinical dementia rating 1; aMCI, amnesic mild cognitive impairment; ANC, aged normal controls; YNC, young normal controls; literal, literal interpretation; AP, answers associated with part of the sentence; MU, misunderstanding of the sentence; NK, not knowing.

significantly lower in ANC than in YNC, whereas no difference was observed in metaphor comprehension. Empirical developmental studies of normal children have found that metaphors are comprehended at an earlier age than ironies.⁴ One factor critical for understanding verbal irony (sarcasm) is an individual's ability to attribute appropriate second-order ToM.⁴ The success of the second-order ToM task emerges at around age 5 or 6²⁵ and it has been revealed that age-related decline occurred directly in the second-order ToM and indirectly in the first-order ToM.²⁶ The influence of difference in difficulty level could not be ruled out. Colston and Gibbs have shown that it takes healthy adults longer to read ironic than metaphoric statements, which suggests that irony (sarcasm) processing requires more cognitive load than metaphor processing.⁵

Age-related decline in metaphor comprehension was not shown in the present study. The deterioration was reported in the early stage of AD by a study that did not include the participants with MCI,¹⁶⁻¹⁸ and the present study showed that comprehension begins to decline even during aMCI, the prodromal stage of AD.

Another issue was with the comprehension of conventional metaphor. In the present study, conventional metaphor comprehension was deteriorated as well as nonconventional novel expressions, as shown in previous studies.^{16,17} However, Amanzio et al reported the deficits in nonconventional novel metaphors, while no impairment was observed in conventional metaphors.¹⁸ The study assumed that conventional metaphors might be interpreted automatically through frequent usage, whereas novel metaphors recruited ToM

processes. However, the patients might tend to avoid complicated pragmatic wording and without usage in everyday speech, conventional metaphors could recruit ToM processes as novel metaphors.

Deficits of AD were characterized by literal interpretation; concerning error patterns, group differences were observed only in the pattern of literal interpretation. Decline of inhibition could be related to choosing literal interpretation. Metaphor and sarcasm comprehension requires contextual coherence judgment, as literal interpretation can be taken out of context. It has been proposed that both the literal and the nonliteral meaning are activated concurrently and the inappropriate meaning is inhibited by the context.²⁷⁻³¹ However, patients with AD had difficulty suppressing inappropriate literal interpretation, which is concurrently activated.^{32,33} Literal interpretation of metaphor causes misunderstanding and that of sarcasm could be more problematic. In sarcastic expression, the speakers say the opposite of what they mean¹⁵ and thus the patients with AD may interpret the utterance as admiration, which would be opposite to the speakers' intention. Such misinterpretation would result in social miscommunication.

Miscommunication between patients and caregivers could lead to behavioral and psychological symptoms of dementia (BPSD) in patients and distress in caregivers.³⁴⁻³⁷ Therefore, caregivers' understanding of decreased communication abilities in patients may reduce BPSD and caregiver distress.^{38,39}

As a limitation, the groups of the present study were not matched for age and education. Based on the results of the present study, further study is required with a larger group of participants for consideration of clinical relevance.

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Declaration of Conflicting Interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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Original Research Article

Yamaguchi Facial Expression-Making Task in Alzheimer's Disease: A Novel and Enjoyable Make-a-Face Game

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Key Words

Dementia · Facial expression · Dementia care · Caregivers · Emotional reactions to dementia · Cognitive tests · Behavioral/psychiatric symptoms of dementia

Abstract

Background: To assess the ability to make emotional facial expressions, we newly developed the Yamaguchi facial expression-making task (Y-FEMT). **Method:** We recruited 20 normal controls and 61 outpatients: 10 with amnesic mild cognitive impairment (aMCI), 34 with mild Alzheimer's disease (AD), and 17 with moderate AD. In the Y-FEMT, smile and anger expressions were made by arranging face parts. We examined the relationship between each Y-FEMT score and the Mini-Mental State Examination (MMSE) score or overlapping figure identification test (Fig-test). **Results:** The Total score (0–20) was nearly achieved in controls (18.9 ± 1.4) and declined with AD progression (aMCI 17.2 ± 2.4 , mild AD 15.7 ± 2.6 , moderate AD 12.3 ± 2.7). The Anger score (0–10) was significantly lower than the Smile score (0–10) in mild and moderate AD ($p = 0.007$ and $p = 0.006$, respectively). The Structure score (0–6 each) correlated well with both the MMSE score ($r = 0.44$, $p < 0.001$) and Fig-test ($r = 0.45$, $p < 0.001$), whereas the Expression score (0–4 each) correlated only with the MMSE score ($r = 0.33$, $p = 0.01$). The Subjective scores (0–4), evaluated by 10 therapists, highly correlated with the Total score. Additionally, the Y-FEMT promoted laughter and a convivial atmosphere. **Conclusion:** The Y-FEMT pleasantly assessed the ability to make emotional facial expressions without special equipment. Furthermore, the Y-FEMT may provide helpful clues for caregivers to achieve good communication with AD patients for better care.

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Introduction

In the care of Alzheimer's disease (AD) patients, deficits in the recognition of emotional facial expressions may cause problems between AD patients and their caregivers because emotion perception deficits, rather than general cognition, have been linked to interpersonal difficulties [1, 2]. Furthermore, problems in emotional perception, but not cognitive function or mood, predict quality of life [3]. Patients with AD have deficits in recognizing facial emotion, which may be independent of their impairment in recognizing non-emotional features of faces [4]. Discriminating facial identities has been shown to be impaired in AD patients [5]. Recognition of emotional facial expressions can be impaired in mild cognitive impairment (MCI) prior to the diagnosis of AD [4, 6]. In addition, the ability to recognize emotional facial expressions is impaired with the progression of AD [7], but recognition and reaction to emotional facial stimuli are partially preserved in the advanced stages of dementia, in which the most sensitively identified emotion is happiness [8].

As described above, there are many reports on disturbed recognition of emotional facial expressions in MCI and AD. However, no study has investigated the ability of AD patients to make facial expressions. Furthermore, it is not easy to detect the disturbed recognition of emotional facial expression in a clinical setting because special techniques or instruments, such as a computer with morphing technology, are needed for detection.

We thus developed a new cognitive task, the Yamaguchi facial expression-making task (Y-FEMT), to assess the ability of patients to make emotional facial expressions by arranging pieces of face parts. The origin of the Y-FEMT is the traditional Japanese make-a-face game 'Fuku-warai', meaning 'Lucky Laugh'. The aim of this game is to put face parts (e.g. eyebrows, eyes, nose and mouth) on a face outline while the player's eyes are covered with a hand towel. Then, all participants enjoy laughing at the funniness of these facial expressions. Fuku-warai is traditionally played during the New Year's holidays. The Y-FEMT consisted of two tasks to make up 'Smile' and 'Anger' faces without a blindfold. We describe the development of the Y-FEMT, how we used to investigate patients' ability to make emotional facial expression, and its association with other cognitive tests. After defining the objective standard of the Y-FEMT, we added a subjective scoring method of the Y-FEMT for easy use in clinical practice.

Methods

Participants

Normal controls (NC; $n = 20$), aged 61 to 80 years, were recruited from community dwellers who participated in the 'Prevention of mental decline' project in Takasaki City, Gunma, Japan. These participants were judged normal based on the results of cognitive tests and medical interviews by clinicians specializing in dementia. We recruited 61 participants who were diagnosed as having amnesic MCI (aMCI) or AD at the outpatient clinic of the Geriatrics Research Institute and Hospital in Maebashi, Gunma, Japan.

The AD patients were diagnosed based on the criteria of the National Institute of Neurological and Communicative Disorders and Stroke and Alzheimer's Disorders and Related Disorders Association (NINCDS-ADRDA) [9]. Similarly, the aMCI patients were diagnosed based on a previous study [10]. In the present study, 61 participants were classified according to the Clinical Dementia Rating (CDR) by an experienced neurologist. The discriminant criteria of mild and moderate AD were CDR 1 ($n = 34$) and CDR 2 ($n = 17$), respectively.

None of the participants demonstrated other psychiatric disorders or had problems with alcoholism, motor deficits such as paralysis, major heart disease or neurological or psychi-

Table 1. Demographics and clinical characteristics

	All (n = 81)	NC (n = 20)	aMCI (n = 10)	Mild AD (n = 34)	Moderate AD (n = 17)
Male/female	27/54	7/13	6/4	12/22	2/15
Age, years	77.5 ± 6.8	72.3 ± 4.9	74.3 ± 7.0	79.6 ± 6.6	81.3 ± 4.4
Education, years	10.3 ± 2.7	12.5 ± 2.0	10.6 ± 4.0	9.5 ± 2.1	9.2 ± 2.4
MMSE score	21.6 ± 6.5	28.8 ± 1.4	26.5 ± 2.6	20.2 ± 3.2	12.9 ± 4.2
Y-FEMT score (objective)					
Total	16.0 ± 3.2	18.9 ± 1.4	17.2 ± 2.4	15.7 ± 2.6	12.3 ± 2.7
Smile	8.4 ± 1.6	9.6 ± 0.9	9.0 ± 1.2	8.4 ± 1.6	6.9 ± 1.2
Anger	7.5 ± 2.1 ***	9.3 ± 1.1 n.s.	8.2 ± 1.6 n.s.	7.4 ± 1.7 **	5.4 ± 2.0 **
Structure	10.4 ± 1.8	11.6 ± 0.8	11.0 ± 1.1	10.6 ± 1.3	8.5 ± 2.3
Expression	5.5 ± 2.0	7.3 ± 0.9	6.2 ± 1.9	5.1 ± 1.8	3.8 ± 1.3
Subjective score [#]	2.8 ± 1.1	3.5 ± 0.6	3.3 ± 0.8	2.8 ± 1.0	1.8 ± 1.1

Data are presented as mean ± standard deviation. n.s. = Not significant.

** p < 0.01, *** p < 0.001: comparison between the Smile and Anger scores by a paired t test. # Subjective score was assessed by 10 evaluators.

atric disorders other than the primary diagnosis of AD or aMCI. Demographic data and clinical characteristics are shown in table 1. All participants reported normal or corrected-to-normal vision, and they were naive with regard to the purpose of the experiment. The Ethics Board of Gunma University School of Health Sciences approved all procedures (No. 21–47), and signed informed consent was obtained.

All participants underwent the Mini-Mental State Examination (MMSE) [11] and overlapping figure identification test (Fig-test), which consists of line-drawn figures of six objects: hat, eyeglasses, vase, apple, tulip and hammer. The participants were asked to identify each figure, and the score range was 0–6.

Procedure

The Y-FEMT contains two tasks. Participants were asked to arrange parts of a face puzzle (fig. 1a) to make two kinds of emotional facial expressions, 'Smile' and 'Anger' (fig. 1b, c). The face puzzle used in this study was made of thick, stiff paper and consisted of one outline plate of a face with hair (25 cm long and 19 cm width) and six facial parts: two eyebrows (or mustache; 5.5 cm long and 2 cm width), two eyes (6.5 cm long and 3.5 cm width), one nose (4.5 cm long and 4 cm width) and one mouth (10 cm long and 3.5 cm width) (fig. 1a).

Protocol

The protocol was as follows: (1) The participant sits at their desk. The face outline plate (fig. 1a) is placed in the center of the desk. (2) The Y-FEMT starts with the Smile task (fig. 1b) followed by the Anger task (fig. 1c). (3) For the Smile task, the examiner hands six randomly-oriented parts of a human face (fig. 1a) to the participant, and gives the simple instruction: 'You must use all of these parts and arrange the pieces to make a smiling face'. The instruction can be repeated, if required. (4) Then, the participant arranges the face parts on the face outline plate to make a smiling face. (5) The examiner records the completed task by taking a photograph (fig. 1d–f). If the participant cannot complete the task within 2 min, the uncompleted task is scored. (6) Avoid giving advice or pointing out any error until the end of the next task. (7) For the 'Anger' task, the examiner hands the participants six face

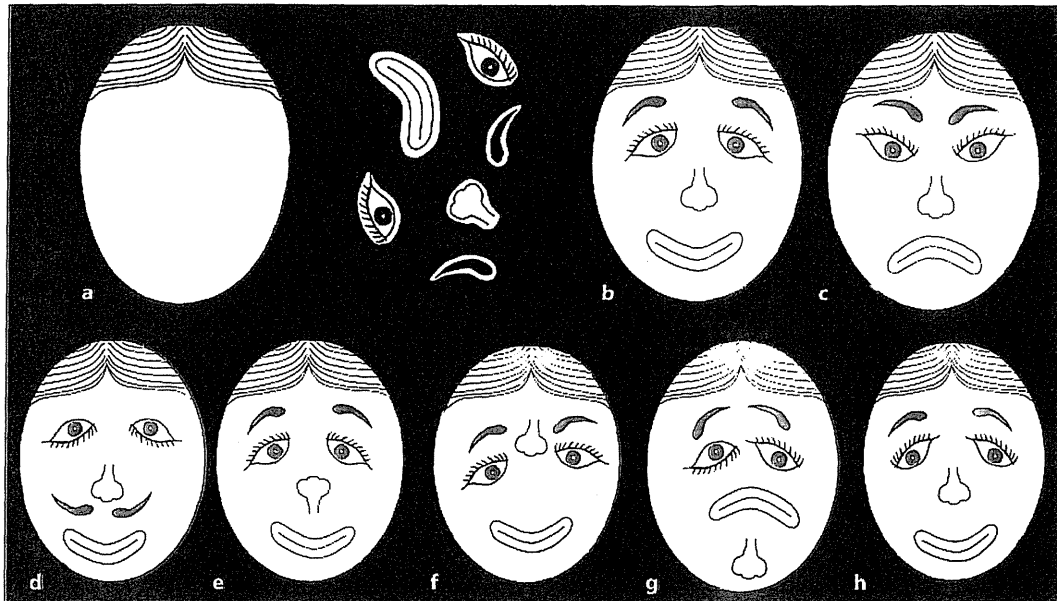


Fig. 1. Parts of the Y-FEMT, exemplary results and error patterns. **a** The Y-FEMT consists of one outline plate and six simple parts of the human face. Exemplary results of the Smile task (**b**; Smile score was 10/10) and Anger task (**c**; Anger score was 10/10) in healthy elderly people. **d–f** Examples of the Smile task in AD patients. The score was decreased by 1 point for incorrect orientation of the eyes, but adopting the eyebrows as a moustache was not considered a mistake (**d**; Smile score was 9/10). Orientation of the nose is upside-down (**e**; Smile score was 9/10). Placement of the nose and symmetric property are different and expression score was 3 (**f**; Smile score was 7/10). **g, h** Examples of the Anger task in mild-to-moderate AD patients. Only the position of the eyebrows and eyes are correct, each gaining 1 point (**g**; Anger score was 2/10), and the fundamental structure of the face was good, except that the orientation of the eyes is left-right reversed, but the facial expression is different (**h**; Anger score was 5/10).

parts and gives a similar instruction: ‘You must use all of these parts and arrange the pieces to look like an angry face’. (8) The participant arranges the six parts on the face outline to make an angry face. (9) The examiner records the completed task by taking a photograph (fig. 1g, h).

There is the option to look at the photos and laugh together after finishing both tasks.

Guideline for Objective Scoring (table 2)

Structure Score

Assessment of the fundamental structure of the face (0–12 points; 0–6 points each for Smile and Anger structures):

- Eyebrows are judged for placement in the uppermost part of the face and above the eyes or not (1 point). If participant perceived the eyebrows as a moustache, judge whether they are placed between the mouth and nose, and placed on the lower face (1 point). Point allocation is 0–1.
- Eyes are judged for placement and orientation. The criteria for placement of eyes is whether the pupil of the left eye is in the upper left quarter of the face and the pupil of the right eye is in the upper right quarter of the face (1 point). Orientation of the eyes is assessed for left-right reversal or upside-down (1 point). Therefore, point allocation for the eyes is 0–2.

Table 2. Instructions for scoring

	Sum	Smile task	Anger task
<i>Objective scoring</i>			
Structure score for each Smile and Anger task	0–12		
Eyebrows or mustache (uppermost part of face or lie between nose and mouth)		0–1	0–1
Placement of eyes (pupil is in the upper left or right quarter of face)		0–1	0–1
Orientation of eyes (no left-right reversal or upside-down placement)		0–1	0–1
Placement and orientation of nose (around the center and under eyes)		0–1	0–1
Placement and orientation of mouth (bottommost part of face)		0–1	0–1
Symmetric property (bilaterally symmetric placement)		0–1	0–1
Expression score for Smile task/Anger task	0–8		
Outer corners of eyes slant downward/upward (each eye 0–1)		0–2	0–2
Outer corners of mouth slant upward/downward		0 or 2	0 or 2
Total score (sum of Smile score and Anger score)	0–20		
Smile score and Anger score are the sum of each structure score and expression score		0–10	0–10
<i>Subjective scoring</i>			
Subjective score for each Smile and Anger task	0–4		
Subjective evaluations: quite good = 2, approximate = 1, different = 0		0–2	0–2

- Appropriate placement of the nose is within one-half of the distance from the center of the face to the outline of the face and located between eyes and mouth. When both placement and orientation are appropriate, the score for the nose is 1 point.
- Appropriate placement of the mouth is as the bottommost of the face parts. Appropriate orientation of the mouth is with the length in the horizontal direction and the slant of the mouth within 45 degrees, regardless of whether it is upside-down. When both placement and orientation are appropriate, the score for the mouth is 1 point.
- Symmetric property (1 point) is given for bilaterally symmetric placement of the face parts.
- The 'Structure' score (0–12 points) is the sum of both the Smile and Anger structures (0–6 points each).

Expression Score

Assessment of facial expression (0–8 points; 0–4 points each for Smile and Anger expressions):

- For the Smile task, the outer corners of each eye must slant downward (each eye 1 point) and the outer corners of the mouth must slant upward (2 points).
- For the Anger task, the outer corners of each eye must slant upward (each eye 1 point) and the outer corners of the mouth must slant downward (2 points). These directions are the opposite of those in the Smile task. Eyebrows (or mustache) and nose are exempt from directional judgment. However, if there is an error in the placement of the eyes or mouth, it is not pointed out.

Smile Score and Anger Score

The Smile and Anger scores are the summations of the Structure score and the Expression score for each task. Each point allocation is 0–10.

Total Score

The Total score is the sum of the Smile and Anger scores, which are equal to the sums of the Structure scores and the Expression scores. Point allocation is 0–20.

Guideline for Subjective Scoring (Point Allocation 0–2 for Each Task)

If an evaluator thinks each facial expression is quite appropriate, the score is 2, and if it is approximately appropriate or totally different, the score is 1 or 0, respectively. In this study, the facial expressions in each task were assessed by 10 evaluators, who were experienced occupational therapists.

Data Analyses

We scored the pictures created by all participants. To investigate group differences of objective Y-FEMT scores, we carried out analyses of covariance (ANCOVA) with covariates of age and educational years. Post hoc analysis for each group was conducted with Bonferroni correction in all 81 participants. We examined the relationship between each objective Y-FEMT score and MMSE or Fig-test scores by Pearson's product-moment correlation coefficient in the 61 patients. The influence of the order of tasks (c.f. protocol) was analyzed in the 20 NC participants. Test-retest reliability (test-retest interval 127.4 ± 51.4 days) was examined in 22 mild-to-moderate AD patients (age 80.0 ± 7.4 years, education 9.5 ± 1.9 years, MMSE score 17.1 ± 5.4; 13 mild AD and 9 moderate AD patients). Furthermore, we examined the relationship between the objective Y-FEMT score and the Subjective score assessed by 10 experienced therapists, and investigated inter-rater reliability by intraclass correlation coefficients (ICC) and correlation with the objective Y-FEMT score for clinical utility. All statistical analyses were performed with the Japanese version of SPSS 19.0 for Windows (IBM Com.). Results are reported at a significance level of $p < 0.05$.

Results

Y-FEMT in Normal Controls

Demographic data and the results of the Y-FEMT in NC ($n = 20$) are shown in table 1. In NC, the total Y-FEMT score was nearly achieved, although some participants showed a decreased Anger score. To analyze the influence of the order of the tasks, NC were randomly divided into two groups: 'Smile-Anger' group ($n = 8$, age 72.8 ± 5.7 years, education 12.1 ± 2.2 years, MMSE score 28.5 ± 1.2) and 'Anger-Smile' group ($n = 12$, age 72.0 ± 4.5 years, education 12.8 ± 1.9 years, MMSE score 29.0 ± 1.5). The task order did not influence the scores ($p = 0.578$, Student's *t* test; online suppl. table 1; for all online suppl. material, see www.karger.com/doi/10.1159/000339425).

Making Emotional Facial Expressions and Relationship with Disease Progression

All 81 participants accomplished the two tasks within the 2-min time limit. Results and representative faces made during the tasks are presented in table 1 and figure 1, respectively. In all participants, the Total score decreased with the progression of AD, as demonstrated by ANCOVA with covariates of age and educational years ($F(5,75) = 17.08$, $p < 0.001$). The results of the post hoc analysis with Bonferroni correction indicated a significant difference only between mild and moderate AD ($p < 0.001$). Some strange and funny faces were created by AD patients, as shown in figure 1d–h. Smile, Anger and Structure scores were significantly different between mild and moderate AD patients ($p < 0.01$, $p < 0.01$ and $p < 0.001$, respectively), but not between NC and aMCI patients, or between aMCI and mild AD patients. The Expression score was significantly different between NC and

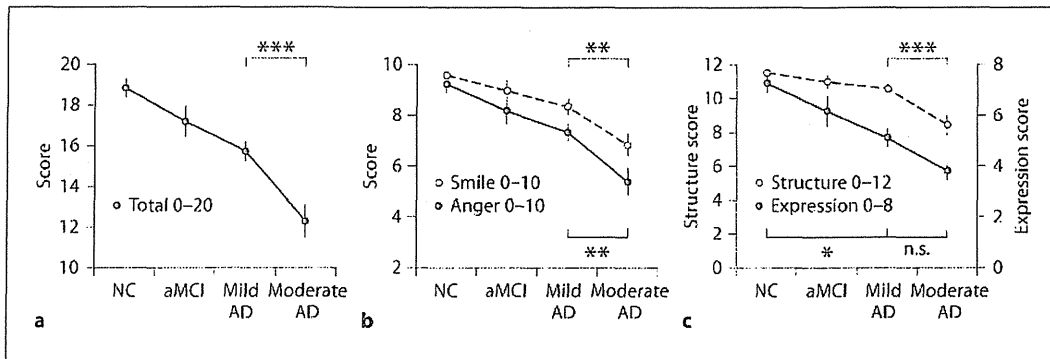


Fig. 2. Y-FEMT scores in four groups. Numbers on the graph indicate mean \pm SE. Point allocation for the Total score is 0–20 (a), for the Smile score and Anger score it is 0–10 (b), for the Structure score 0–12 and for the Expression score it is 0–8 (c). All scores decreased with the progression of AD. There was a significant difference between mild and moderate AD except for the Expression score. * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$; n.s. = Not significant. ANCOVA with covariates of age and educational years.

aMCI patients ($p = 0.01$), but not between mild AD and moderate AD patients ($p = 0.069$). Thus, objective Y-FEMT scores decreased with the progression of AD especially in moderate AD patients.

Smile Score versus Anger score, and Structure Score versus Expression Score

The comparison between the Smile score and Anger score for all participants demonstrated that the Smile score was significantly higher than the Anger score ($p < 0.001$, paired t test; table 1). In the analysis of each group, the Smile score and Anger score were not significantly different in the NC or aMCI groups ($p = 0.286$, $n = 20$ and $p = 0.137$, $n = 10$, respectively), while the Smile score was significantly higher than the Anger score in both the mild AD and moderate AD groups by paired t test ($p = 0.007$, $n = 34$ and $p = 0.006$, $n = 17$, respectively; table 1). Thus, the AD participants achieved a higher score for the Smile task than for the Anger task (fig. 2b).

The Structure score and Expression score also declined with the progression of AD (table 1; fig. 2c). In the four groups, both Expression and Structure scores showed significant differences ($F(5,75) = 11.08$, $p < 0.001$ and $F(5,75) = 10.27$, $p < 0.001$, respectively). Post hoc analysis with Bonferroni correction indicated that the Expression score significantly decreased between NC and mild AD ($p = 0.01$), and that the Structure score significantly decreased between mild AD and moderate AD. Thus, the Expression score declined earlier than the Structure score.

Relationship between Expression-Making Tasks and Cognitive Tests

Next, we examined the relationship between Y-FEMT scores and two cognitive tests, the MMSE and Fig-test, in participants other than NC ($n = 61$) by Pearson's product-moment correlation coefficient. On analysis of the correlation between Y-FEMT and MMSE scores, each Y-FEMT score was mild-to-moderately and significantly correlated with the MMSE score (Total $r = 0.463$, $p < 0.001$; Smile $r = 0.346$, $p = 0.006$; Anger $r = 0.434$, $p < 0.001$; Structure $r = 0.440$, $p < 0.001$, and Expression $r = 0.328$, $p = 0.01$; online suppl. table 2). On analysis of the correlation between Y-FEMT and Fig-test scores, Y-FEMT scores other than the Expression score were mild-to-moderately correlated with the Fig-test score (Total $r = 0.387$, $p = 0.002$; Smile $r = 0.357$, $p = 0.005$; Anger $r = 0.308$, $p = 0.016$, and Structure $r =$

0.452, $p < 0.001$). Note that the Expression score was not significantly correlated with the Fig-test score ($r = 0.189$, $p = 0.145$), but was significantly correlated with the MMSE score. In contrast, the Structure score showed significant correlations with both the MMSE and Fig-test scores.

Test-Retest Reliability of the Y-FEMT

The test-retest reliability was based on the ratings of 22 mild-to-moderate AD patients. The intra-rater reliability of the Total score was sufficiently high (ICC(1,1) = 0.927, $p < 0.001$). Similarly to the analysis of correlation between the first and second tries, the Total score showed a high correlation ($r = 0.936$, $p < 0.001$, Pearson's correlation).

Subjective Score of the Y-FEMT for Clinical Practice

The inter-rater reliability of the Subjective scores ($n = 81$) by 10 evaluators was good (ICC(2,1) = 0.760, $p < 0.001$). Furthermore, the Subjective scores showed a high correlation with both the Total and Expression scores ($r = 0.789$, $p < 0.001$ and $r = 0.770$, $p < 0.001$, respectively).

Discussion

Comparison with Previous Studies

This study investigated how the ability to make emotional facial expressions changes with the progression of AD. We developed the Y-FEMT for this purpose.

In the objective scoring, the Total score of the Y-FEMT decreased with the progression of AD. This finding about the ability to make emotional facial expressions is in agreement with results from previous studies on the recognition of emotional facial expressions, which start to become impaired in MCI [6, 12] and then become more severely impaired with the progression of AD [7, 13, 14].

Comparing the Structure score and Expression score, the Structure score decreased between mild and moderate AD, whereas the Expression score started to decrease between NC and mild AD. This finding suggests that the ability to make the fundamental structure of the face is maintained in the early stages of AD. Furthermore, the Structure score was significantly and moderately correlated with the Fig-test score, whereas the Expression score did not correlate with the Fig-test score. This suggests that the ability to make the fundamental structure of the face is closely related to visual function, which is impaired in the later stages of AD.

Comparing the Smile score and Anger score, the Anger score was significantly lower than the Smile score in AD patients. This findings agree with those from previous studies on the recognition of facial expressions, in which a happy face was most easily recognized cross-culturally (both Japanese and Caucasian) [15] among both healthy people and elderly subjects with AD [7, 12, 16, 17], as well as people with severe dementia [8]. In addition, the coefficient of correlation between the MMSE and Anger scores ($r = 0.434$) tended to be higher than that between the MMSE and Smile scores ($r = 0.346$). Thus, the findings of this study suggest that the ability to make an angry face is impaired in parallel with general cognitive function, whereas the ability to make a smiling face is maintained for longer.

Utility of the Y-FEMT

Most previous studies on emotional facial expression examined the ability to recognize computer-created faces. However, the Y-FEMT assesses the ability to make the fundamental structures of the face as well as emotional facial expressions. Most participants responded

when completing the Y-FEMT. They said 'I'm not great with games like this' but smiled happily while performing the tasks. In addition, the Y-FEMT induced laughter and a convivial atmosphere, especially when we praised the participants' results after finishing the two tasks. This is a primary characteristic of the traditional Japanese game 'Fuku-warai'. In 'Fuku-warai', the resultant facial expressions are comical and funny, because the game is played while blindfolded. When dementia patients performed the Y-FEMT without a blindfold, they also created comical faces and everyone including themselves could laugh (fig. 1d–h). However, their family caregivers showed various responses; some were shocked and disappointed on seeing the results or reprimanded the patients' mistakes, while other caregivers had fun together with a warm smile even if the patients scored poorly on the tasks. We can estimate some common treatment or communication problems between caregivers and patients based on the Y-FEMT, whether caregivers accept or become angry with the patients' mistakes. Thus, the Y-FEMT is a useful tool for family education on appropriate care because emotional communication and support are very important in dementia care [8]. Problems with emotion perception or emotion comprehension, rather than deterioration of cognitive function and mood, influence the quality of life and problems of interpersonal behaviors in AD patients [1, 3]. Thus, it is better for caregivers to understand the level of social cognition of AD patients. The Y-FEMT provides helpful clues for caregivers to understand the ability of the AD patients to make emotional facial expressions.

In addition, the Y-FEMT is a convenient task for assessing the ability to make faces showing expression and fundamental structure, and is less stressful than conventional cognitive tests. It is easy to do, simple to understand the rules, takes a short time, has non-committal answers and protects patients from a feeling of 'failure'. Furthermore, the Y-FEMT does not require special instruments such as a computer or monitor, unlike other recently published methods.

In this study, the Subjective score correlated highly with the objective Y-FEMT scores, Total score and Expression score. Moreover, the inter-rater reliability of the Subjective score was sufficiently high. Thus, objective scoring can be replaced by the Subjective score in clinical practice. We hope that the Y-FEMT will provide helpful clues for caregivers to improve care and communication with AD patients, and that the task performance will be enjoyable for both the patients and their caregivers.

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Disclosure Statement

The authors have no conflict of interest to declare.

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