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# The importance of identifying underlying process abnormalities in alexithymia: Implications of the three-process model and a single case study illustration

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ABSTRACT

We present an in-depth case study of a rare individual (whom we will refer to as "Jane") who reported an inability to experience emotion. Jane completed a range of assessments measuring alexithymia, emotional awareness, and emotion recognition ability. She, along with 22 control participants, also underwent skin conductance (SC) measurement and facial electromyography (EMG) during exposure to affective images, and self reported the valence/arousal of their responses to those images. Jane scored high on alexithymia and low on emotional awareness; yet she performed well on emotion recognition measures and showed a typical pattern of valence ratings. Her SC responses and subjective arousal ratings were atypically low, and some of her EMG responses were also atypical. Jane's deficit profile highlights the dissociability of self-focused emotional awareness and other-focused emotion recognition ability, as well as the dissociability between the generation and representation of valence and arousal (with both subjective and objective measures).

## 1. Introduction

The deficits in affective processing associated with alexithymia (and low emotional awareness more generally) may be of heterogeneous origin (Van der Velde et al., 2013, 2014). If so, this highlights the importance of providing detailed clinical evaluation and individualized treatment. It further highlights the challenges associated with identifying the relevant factors that contribute to individual cases and that predict effective treatment options. As a first step in this direction, we recently proposed a neuro-cognitive model of affective processing, here termed the "three-process model" (TPM; Fig. 1), which identifies several dissociable processes – including affective response generation (ARG) processes (e.g., generating autonomic and skeletomotor responses to affective stimuli), affective response representation (ARR) processes (e.g., representing bodily feelings and emotion concepts), and cognitive control (CC) processes (e.g., attention to emotion) – that plausibly interact to produce self-reportable emotional experience (Lane, Weihs, Herring, Hishaw, & Smith, 2015; Panksepp, Lane, Solms, & Smith, 2017; Smith and Lane, 2015, 2016; Smith, Thayer, Khalsa, Lane,

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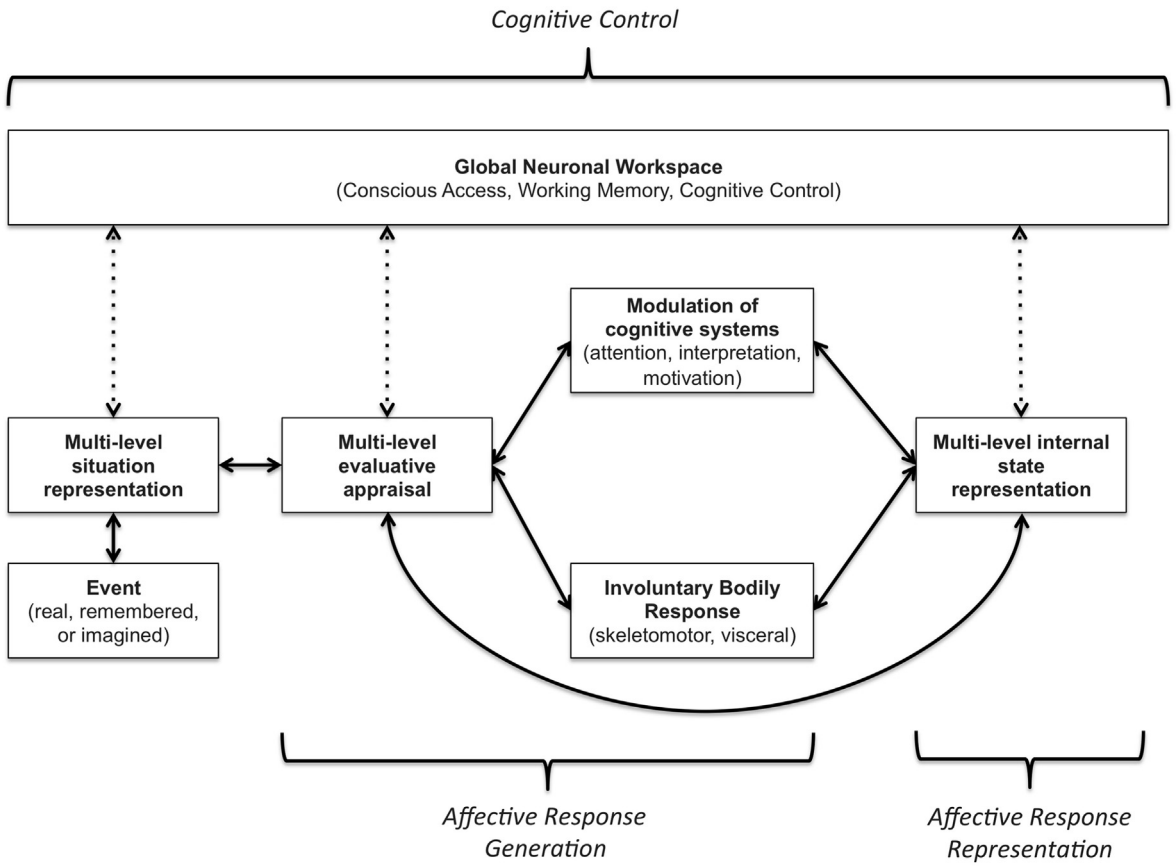


**Fig. 1.** Graphical depiction of the three-process model of affective processing (Smith et al., 2018b).

2017; Smith, Killgore, Lane, 2018); this model also suggests that abnormalities in any of these underlying processes, or abnormalities in their interactions, could theoretically account for clinical phenomena involving the self-reported absence of emotional experience in normatively affective contexts. However, at present it remains an open question regarding which neuro-cognitive process or processes are in fact abnormal in alexithymic individuals, and different process abnormalities could be of primary relevance in different cases. More generally, given the plausible interactions between ARG, ARR, and CC processes, the degree to which alexithymia-related pathology in one process could leave the other processes preserved is poorly understood. Empirically informed answers to these questions would be of great clinical significance, as alexithymia (and low emotional awareness more generally) has been associated with multiple psychiatric conditions (Lane et al., 2015) and with differences in psychotherapeutic treatment outcomes (Beutel et al., 2013).

In this article we present an in-depth case study of a rare individual (who we will refer to as “Jane” for de-identification) with severe alexithymia whose deficit profile may shed light on the separability of, and possible interactions between, the distinct processes contributing to emotional experience highlighted above.

Jane was a 54-year old woman who requested help for a lifelong inability to experience emotion. She had read several research articles on alexithymia (Lane, Ahern, Schwartz, & Kaszniak, 1997), and she believed herself to be suffering from this condition. She characterized herself as “an emotional eunuch” and gave numerous examples of her inability to experience emotions in situations where feelings would be expected (e.g., at a funeral, a child’s graduation, or during sex). She was able to verbally describe what she should be feeling (e.g., sadness, happiness); yet she reported not experiencing these emotions. During a clinical interview for the assessment of alexithymia, her eyes welled with tears when discussing the lack of closeness in her marriage, but, upon questioning, she stated that she felt nothing at the moment (e.g., no awareness of sadness, despite such outward indicators). Therefore, this individual was unusual in that she (1) complained that she did not experience emotions, (2) expressed a strong desire to seek help for her condition, (3) displayed conceptual knowledge about emotions and (as illustrated below) an intact ability to recognize emotions in others, and (4) displayed a striking dissociation between self-reported emotional experience (which was absent) and physiological/behavioral expressions of emotion, some of which were outwardly observable and (as described below) measurable experimentally.

In addition to the clinical interview, Jane submitted to a variety of standardized psychometric tests and participated in a study assessing psychophysiological and experiential components of emotional responses to affective images. Below we therefore also describe a range of psychometric and psychophysiological assessments, and a comparison to twenty-two healthy control participants, designed to ascertain which of the underlying neuro-cognitive processes highlighted above were intact in this individual and which

may have been impaired. The results of these analyses suggest that abnormalities in some (but not all) ARG processes may, via their subsequent influence on ARR processes, best account for the lack of self-reported emotional experience in this individual. These results suggest important new hypotheses that should be examined in further alexithymia research. They also highlight the potential importance of this type of more detailed evaluation in routine clinical practice. Sadly, this individual, who was receiving clinical care from a psychiatrist in another community, subsequently committed suicide (about 3 months after this case study was conducted), highlighting the clinical importance of systematically studying severe alexithymia as a potentially life-threatening condition.

## 2. Case report

### 2.1. Patient and control participants

This case study was conducted on a 54 year-old married white female (Jane) who presented to the outpatient psychiatry department at the University of Arizona complaining that she did not feel emotions. Her psychiatrist had diagnosed her with alexithymia, and she wondered if there was a treatment for the condition. She agreed to participate in an informal clinical interview with a psychiatrist (RDL) and a clinical psychologist (JK), as well as in a range of psychometric and psychophysiological assessments (described below), to determine the nature of her difficulties before developing a treatment plan. She signed consent for these procedures.

This study also included twenty-two healthy control participants matched for age and free of depression, neurologic, or other psychiatric disorders (assessed via self-report, using a comprehensive health and demographic questionnaire). These participants were recruited from the local community in order to establish normative values on the previously mentioned psychometric and psychophysiological assessments. The twenty-two participants included 13 Females and 9 males, with a mean age of 57.9 years (SD = 10.84). They signed consent for these procedures.

### 2.2. Psychometric measures

Jane was asked to complete each of the following psychometric measures:

#### 2.2.1. Toronto Alexithymia Scale (TAS-20)

The TAS-20 is a self-report measure that asks individuals to rate 20 items on a 5-point Likert scale (maximum score of 100) (Bagby, Parker, & Taylor, 1994a, 1994b). The 20 items make up three subscales: (a) difficulty identifying feelings; (b) difficulty describing feelings; and (c) externally oriented thinking. Although alexithymia is understood as a dimensional construct, cutoff scores have been established for categorizing respondents as alexithymic (61 + ), intermediate (52–60), and nonalexithymic (51 or below). Jane's data were compared to previously established normative data for this measure (Parker, Taylor, & Bagby, 2003).

#### 2.2.2. Levels of Emotional Awareness Scale (LEAS)

The LEAS is a performance-based measure of emotional awareness that presents 2–4 sentence descriptions of 20 social situations, each involving 2 people. The scenarios are designed to elicit four types of emotion (happiness, anger, sadness, and fear). One scenario is presented per page, followed by two questions: "How would you feel?" and "How would the other person feel?" Participants are instructed to write as much or as little as needed to answer the two questions. They are also told that they must use the word "feel" in their responses.

Each item response is scored separately for self, other, and total score. Self and other item scores are given based on the EA level assigned to the words participants use in their responses: non-feeling words = 0; body sensation words (e.g., "tired") = 1; emotional action or valence words (e.g., "punching," "good") = 2; single emotion terms (e.g., "happy," "angry") = 3; multiple emotion terms (e.g., "sad and afraid") = 4. Total item scores are then given based on the higher of the self- and other-related scores, unless a score of 4 is given for both. In that case, a total score of 5 is given for the item, so long as the self- and other-related responses were differentiable (for more detail, see (Lane, Quinlan, Schwartz, Walker, & Zeitlin, 1990)). The overall LEAS total score is computed by summing the total item scores across all 20 items for a maximum possible score of 100. Jane's data were compared to previously established normative data for this measure (unpublished, based on the dataset reported in (Lane et al., 1996, 1998)).

#### 2.2.3. Perception of Affect Task (PAT)

The PAT (Lane et al., 1996; Rau, 1992; Wright, Riedel, Sechrest, Lane, & Smith, 2017) assesses seven emotional states (happiness, sadness, fear, anger, surprise, disgust, and neutral) via four 35-item subtasks, T1 to T4, varying the verbal versus nonverbal nature of stimulus and response. In subtask T1, participants choose one of the seven emotion-state labels (words) in response to a sentence describing an emotional situation (with an underline indicating the indexed individual(s), e.g., "Several relatives gather at a funeral."). The instruction provided to participants for this subtask asks them to read each sentence and to circle one of the 7 emotion words that seems to best match the emotion of the underlined person or persons in that sentence. In subtask T2, participants choose one of the seven emotion-state labels in response to an Ekman photograph (Ekman & Friesen, 1975) of a facial expression. The instruction provided to participants asks them to view each photograph and to circle the word that seems to be the best match for the emotional expression depicted. In subtask T3, participants choose one of seven T2 photographs in response to the T1 sentences. The instruction provided to participants for this subtask is as follows: "Please read each sentence and circle the letter for the facial expression that seems to be the best match for the underlined person or persons in that sentence." In subtask T4, participants choose

one of seven photographs of scenes without human faces in response to T2 photographs, where each scene is most consistent with one of the seven emotions (with an arrow indicating the indexed individual in each scene, e.g., a person being physically attacked). The instruction provided to participants for this subtask asks them to view each photograph and to circle a letter corresponding to the scene that seems to be the best match for the emotional expression depicted. Thus, T1 (sentences-words) is verbal-verbal, T2 (faces-words) is nonverbal-verbal, T3 (sentences-faces) is verbal-nonverbal, and T4 (faces-scenes) is nonverbal-nonverbal. The score for the overall PAT, or one of its subscales, is the percent of items for which the intended emotion is recognized. Jane's data were compared to previously established normative data for this measure (Wright et al., 2017).

#### 2.2.4. Reading the Mind in the Eyes Test (RMET)

The RMET (Baron-Cohen, Wheelwright, Hill, Raste, & Plumb, 2001) evaluates an individual's ability to infer the mental states of others when viewing pictures of facial expressions that only reveal their eyes. Participants choose from a list of four words, and the options differ depending upon the target mental state depicted in the image (e.g., serious, alarmed, bewildered, ashamed). The score is the total number correct out of 36 items. This was given to Jane as a second measure of emotion recognition ability. Jane's data were compared to previously established normative data for this measure (Baron-Cohen et al., 2001).

#### 2.2.5. The Marlowe-Crowne Scale (MCS) and the Taylor Manifest Anxiety Scale (TMAS)

The MCS (Crowne & Marlowe, 1960) consists of 33 true-false items designed to measure defensiveness. These items include statements that are socially undesirable but likely or socially desirable but unlikely. The Bendig short form of the TMAS (Bendig, 1956) consists of 20 true-false items designed to assess consciously experienced distress/nervousness. A high score on the MCS in combination with a low score on the TMAS constitutes a measure of the repressive coping style. These measures were both given to Jane in order to assess whether or not a repressive coping style might contribute to her self-reported lack of emotional experience.

#### 2.2.6. CPI socialization scale (CPI-so)

The CPI-so (Gough, 1994) is a common measure of proneness to social deviance. This scale was given to Jane to rule out the presence of socially deviant (psychopathic) tendencies that are sometimes associated with the absence of normative affective responding.

### 2.3. Psychophysiological measures

Jane and the control participants were asked to perform a simple task where they were shown 24 images from the international affective picture system (IAPS) (Lang, Ohman, & Vaitl, 1988) while undergoing (1) facial electromyography (EMG) of the left and right corrugator and zygomatic muscles, and (2) skin conductance responses measured from the middle phalanges of the first and middle fingers of the left hand. The task required that each image be viewed for 6 s (interstimulus interval = 15 s), after which participants were immediately asked to rate the valence and arousal level associated with the image using an electronic version of the 9-point Self-Assessment Manikin (SAM) scales (Lang, Greenwald, & Bradley, 1988). On the basis of normative data (both male/female) provided by Lang et al. (Lang, Bradley, & Cuthbert, 2008), 8 images for each specific valence were selected (unpleasant (U): $M_{valence} = 2.37$, $M_{arousal} = 6.05$; neutral (N): $M_{valence} = 4.92$, $M_{arousal} = 3.54$; pleasant (P): $M_{valence}7.34$, $M_{arousal} = 5.80$) presented in a fixed (pseudo-random) order.

Skin conductance and facial EMG were recorded for 15 s intervals (from 2.5 s pre- to 12.5 s post-stimulus onset) using BIOPAC MP100 EMG and GSR amplifiers sampling at 1000 Hz, with data collected on a PC running AcqKnowledge software (https://www.biopac.com/manual/acqknowledge-5-software-guide/) (see Supplemental methods for more detail). Stimulus presentation (by slide projector) was synchronized with physiologic data acquisition by a computer running MAESTRO, a version of DMASTR software (http://www.u.arizona.edu/~kforster/dmastr/dmastr.htm).

### 2.4. Data analysis

The amplitude of the first skin conductance response (SCR) with onset in the period from 1 to 6 s post-slide onset was measured. All SCRs of 0.03 microSiemens or larger were included for analysis. Mean raw SCRs for each image type were calculated separately for Jane and for the control participants.

Mean EMG responses were recorded for each muscle group for a 2-s pre-stimulus baseline and for a 5-second epoch beginning 1.7 s post slide presentation onset. Change scores from the baseline were computed for each image and then the mean was calculated separately for Jane and for the control participants.

Mean valence and arousal scores on the 9-point SAM scales were calculated for each image type (U, N, P) separately for Jane and for the control participants.

## 3. Results

### 3.1. Clinical case description

Jane requested an expert consultation regarding whether she had alexithymia and whether it could be treated. As she reported, "I do not feel emotions. I've been told that I have alexithymia." She was married (for 21 years) with two adult daughters (21 and

24 years of age, living outside the home), and she was an allied medical professional. She described herself as "manager of the world," and she stated that she finds going to work therapeutic.

Jane had been told during the previous year by a psychiatrist treating her for "depression" that she had alexithymia. She was greatly relieved to know that there was a name for the condition that she had. She did a computer search and came across a paper by one of the authors (RDL) in which alexithymia was described as the emotional equivalent of the neurological condition "Blindsight" (Lane et al., 1997). She stated that this was exactly what she had. She wondered if there was any treatment for the condition.

Throughout her life she had labeled herself as an "emotional eunuch." She stated that on occasions when she should have felt emotions – such as her daughter's graduation from college, or the funeral of a family member – she didn't feel anything. She also stated that she didn't understand why people engaged in sex, as she found nothing pleasurable about it. She had never experienced lust. She and her husband had not had sex for 3–4 years, and they never talked about it. They only talked about sports and day-to-day things. She described the marriage as one involving going through the motions. She also reported that she had never been excited, involved, or passionate about things, that she had no artistic inclinations, and that she liked things "cut and dried." However, she successfully held a professional managerial position that involved interacting with customers and supervising employees.

At age 24 she began taking birth control pills after getting married. She began to have symptoms of crying, depression, and moodiness. She stated that she doesn't think she was sad, blue, or "down in the dumps." She attributed the symptoms to the high estrogen-progesterone ratio of the contraceptive. She had been on a variety of antidepressant medications through the years (at the time of the interview a selective serotonin reuptake inhibitor [SSRI]; Prozac, 40 mg/day, for the last 3 years). She was very clear, however, that SSRIs had not blunted her emotional experience and that it had been this way all her life. When she was not on an SSRI she stated that she "gets nasty to her customers" and could lose her job. However, she has never had a full depressive syndrome; on structured interview (Structured Clinical Interview for DSM-IV; (First, Spitzer, Gibbon, & Williams, 2002)) she did not meet criteria for major depression, either at the time of this case study or further in the past. Based on her self-reported lack of persistent depressed mood, she also would not have met criteria for other depression-related conditions, such as persistent depressive disorder in DSM-5 (300.4; analogous to dysthymia in DSM-IV). She had also never made any suicide attempts before or during the time of this case study.

Four years previously she had been admitted to an inpatient psychiatric unit for a 1-week stay. She was moving out of the house in the context of a marital separation ("we were just co-existing"). She states that she was not upset as she was driving the moving van, but that she developed an upset stomach and sweating. She was seen in the emergency room, and was eventually admitted to the Psychiatry Unit with the diagnosis of "depression." Her therapist told her that she had emotions but didn't recognize them. She later obtained marital counseling after moving out. She had been "living off of her kids" (i.e., relying on them emotionally), and didn't know how to live on her own. The counselor eventually convinced her to move back in with her daughters.

On mental status exam, she was a well-groomed, overweight woman who looked her stated age. She was able to relate her history in a clear and coherent manner. Her affect was flat (i.e., her outward expression of emotion in facial expressions, gestures, and voice tone was relatively subdued and non-animated) and induced some feelings of boredom in the examining clinician, which may have hindered the spontaneous experience of client-clinician connection relative to what is typical in initial interviews. She did not appear overtly depressed. Her thought process appeared logical and goal-directed. There was no evidence of cognitive impairment or psychosis. Her judgment and insight appeared good. At one point during her interview with RDL and JK, when talking about her marriage and the lack of emotional connection with her husband, tears welled up in her eyes. To the external observer she appeared to be on the verge of crying. However, she said that she was not upset, and did not feel sad. She explained that her eyes simply "do this" sometimes.

She described her father as a civil/mechanical engineer who was tall, thin, dark-haired, intelligent, even-tempered, well-read, a provider; he was middle class, there every night, and kept everything fixed up. She described her mother as shorter, rounder, and a manager of the household. She was practical, kept the kids fed and clothed, a bookkeeper, sold real estate, and a great cook. She stated that there was no discussion of emotions when she was growing up. Her mother was also described as overreacting to things (catastrophizing). For example, when her brother was 13 years old his breasts were enlarged; her mother bought a cemetery plot because it meant breast cancer and that he would die. Another example: her sister was in college and had to move because of a fire; her mother wouldn't allow her brother to help, and said he would hurt his back and that she would have to push him around the rest of his life. Her mother was at times quite cruel to her, telling her that she was so ugly she would never find a man to marry. She reported that her mother and father had never seen a psychiatrist, and she was unaware of any medical or psychiatric problems experienced by her brother or sisters (1 sister 4 years older; 1 sister 3 years younger; and 1 brother 11 years younger). Within this context, Jane also stated that as an adolescent she couldn't go on a date and eat anything; she would vomit because she was too nervous.

Her other medical history included bilateral carpal tunnel syndrome and painful bone spurs in her feet (she said she couldn't stand exercising because it was boring). She took Naproxyn for pain in ankles, feet, hips, bunions, and bone spurs (as well as 2 calcium tablets per day) at the time of the interview. She was also perimenopausal and took Estradiol 1 mg and Provera 2.5 mg for hot flashes.

Example transcript from interview:

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Doctor: OK. Now, let me ask you a question. Is it that you're having feelings and you have trouble putting them into words? Or is it that you have no feelings? That you draw a blank? Nothing.

Patient: I draw a blank.

Doctor: There's nothing there when you try to access feelings. You do not experience anything consciously. It's just zero.

Patient: Zip.

| | |
|---|---|
| Doctor: | OK. |
| Doctor: | Is your father still alive? |
| Patient: | No. Died before my mother so (inaudible). |
| Doctor: | OK. Did you discuss with your father important and personal issues in your life? |
| Patient: | No. |
| Doctor: | Can you recall and describe a single important issue that you shared with your father? |
| Patient: | No. |
| Doctor: | OK. |
| Doctor: | Did your father discuss with you important and personal issues in his life? |
| Patient: | No. |
| Doctor: | There wasn't much discussion of emotions in your family? Was there any discussion of emotions growing up? |
| Patient: | No. Not that I can remember from my childhood; not that I filed. |
| Doctor: | OK. So how would you describe your relationship with your husband? |
| Patient: | Right now we are sort of at an impasse. Because once I found out what this is…alexithymia… …I don't…(sigh) we just, well, for years sort of co-existed brother and sister apparently (inaudible). We laugh and joke and be together and discuss things but it's not emotional. |
| Doctor: | Was it ever? |
| Patient: | Ahhh Here we go with the (inaudible) Years back … I kind of decided—I used to just fake everything. You know that's how I got through this life. You know. I would try to do what I thought I was supposed to do so I was happy; I was glad, concerned, subservient, I did all the things I thought were right at the time but I didn't know why. So….it was, you know. I'd say we were pretty happy most of the time. I mean, you know it was a type of boarding house effect. You know. The four of us, the two girls, we all just sort of lived there. I did the cooking, they did the whatever and that's the way it's been. |
| Doctor: | (Jane appears on verge of tears) Is it difficult for you to share this information with us today? |
| Patient: | I can feel this is going. (patient motions at face; laughing nervously). My jaw jiggles. No, this does not bother me. |
| Doctor: | OK. |
| Patient: | I'm interested that someone else thinks this exists. |
| Doctor: | Do you feel upset now? |
| Patient: | No. |
| Doctor: | You don't feel sad? Or… |
| Patient: | No. |
| Doctor: | OK. |
| Doctor: | Your eyes look red. |
| Patient: | I know. I know. I don't have any control over that. |
| Doctor: | OK. So you don't know why your eyes are red. Because it would seem to an external observer that you would feel sad and that you were crying. |
| Patient: | uh huh. |
| Doctor: | But you don't feel anything in that regard. |
| Patient: | No. (pause) I think this is one of those sort of blindsight things. Where the brain is triggering the tears because of the something but it's not telling the other part that this is because you're sad or you are upset. I don't know. |

### 3.2. Psychometric and psychophysiological results

Jane's psychometric scores on standardized assessments are reported here as z-scores (relative to previously established normative data; for raw scores, see Table 1). Her LEAS total score was quite low ($z = -2.19$). Her especially low score for the LEAS self subscale is noteworthy. On the TAS-20, her total score ($z = 2.94$) and factor scores (difficulty identifying feelings: $z = 1.63$; difficulty describing feelings: $z = 2.15$; and externally oriented thinking: $z = 3.19$) were quite high, particularly for externally oriented thinking. On the PAT, her total score was high (95% correct; $z = 0.80$), indicating intact emotion recognition ability. This was further confirmed by her performance on the RMET, where her score was 34/36 ($z = 2.38$). On the MCS and TMAS, her scores (22/33 and 3/20, respectively) placed her in the repressor category by the most stringent standards in the literature (Lane, Sechrest, Riedel, Shapiro, & Kaszniak, 2000). Jane showed no evidence of deviant/psychopathic tendencies, as indicated by a score of 29/54 on the CPI-so.

With respect to the affective image-rating task, control participants showed the expected pattern of differentiation among image groups on all experiential measures, with arousal and valence ratings consistent with established norms for the chosen IAPS stimuli.

**Table 1**
The patient's scores on all psychometric measures compared to established norms.

| | Jane's score | Female test norms |
|---|---|---|
| **LEAS Total** | 42 (z = -2.19) | 64.3 ( ± 10.2) |
| *Self* | 22 | NA |
| *Other* | 36 | NA |
| **TAS-20 Total** | 77 (z = 2.94) | 44.15 ( ± 11.19) |
| *Difficulty identifying feelings* | 23 (z = 1.63) | 14.51 ( ± 5.22) |
| *Difficulty describing feelings* | 21 (z = 2.15) | 11.96 ( ± 4.21) |
| *Externally oriented thinking* | 33 (z = 3.19) | 17.93 ( ± 4.72) |
| **MCS** | 22 | NA |
| **TMAS** | 3 | NA |
| **PAT Total** | 95% correct (z = 0.80) | 87% correct ( ± 10) |
| **RMET** | 34 (z = 2.38) | 26.4 ( ± 3.2) |
| **CPI-so** | 29 | NA |

NA indicates that data are not available.

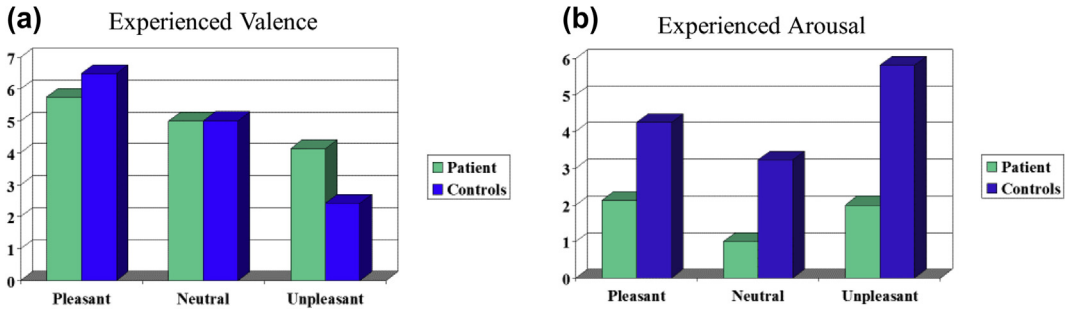


**Fig. 2.** Self-reported valence (A) and arousal (B) ratings for the patient (Jane) and control participants.

Jane's pattern of valence ratings appeared broadly comparable to that of control participants (see Fig. 2a); however, she showed a somewhat restricted range of experience on the 9-point scale (where 9 = extremely positive, 5 = neutral, and 1 = extremely negative) relative to controls with ratings for unpleasant images (M = 4.125; z = 1.74) and pleasant images (M = 5.75; z = −0.749) deviating only slightly from the neutral point on the scale. Her ratings for neutral images were comparable to those of control participants (M = 5, z = −0.009). Relative to control participants, Jane's arousal ratings on the 9-point scale (where 1 = calm and 9 = excited) were markedly attenuated for all image categories, with this attenuation being especially pronounced for unpleasant images (see Fig. 2b): pleasant (M = 2.125, z = −1.213), neutral (M = 1, z = −1.732), and unpleasant (M = 2, z = −2.038). All of these findings indicate that the standardized psychometric assessments corroborated her self-report as well as our clinical observations.

Descriptive results comparing Jane to control participants on these valence and arousal measures, as well as on the psychophysiological measures described below, are detailed in Table 2. Unfortunately, after calculating the z scores and descriptive results reported here, the raw data for control participants were lost. Due to this issue, we were not able to include information about some standard deviations in control participants, nor could we calculate additional z-scores. However, for all available data the control participants reported valence and arousal ratings consistent with IAPS valence/arousal norms. Control participants also showed the expected pattern of differentiation among image groups (i.e., the same patterns observed in previous studies; (Larsen, Norris, & Cacioppo, 2003)) on all psychophysiological measures, with increased corrugator activity to unpleasant images, decreased corrugator activity to pleasant images, increased zygomatic activity to pleasant images, and increased SCRs to pleasant and unpleasant images.

Relative to control participants, Jane's skin conductance responses to all types of IAPS images were markedly smaller and less differentiated (see Fig. 3A): pleasant (M = 0.016, z = −0.59), neutral (M = 0.0038 , z = −0.44), and unpleasant (M = 0.005 , z = −0.53). Jane's pattern of EMG responses was also abnormal and noisy. The magnitudes of Jane's *right corrugator* responses to all IAPS images were markedly blunted compared to control participants, showing little differentiation according to valence (Fig. 3B): pleasant M = 0.00747 μV, neutral M = 0.06657 μV, unpleasant M = 0.10376 μV (z = −0.676 for unpleasant; i.e., notably blunted compared to control participants). Jane's right zygomatic, left corrugator, and left zygomatic responses to IAPS images instead

**Table 2**
Descriptive results comparing the patient to control participants on valence ratings, arousal ratings, and psychophysiological measures.

| | Pleasant | Neutral | Unpleasant |
|---|---|---|---|
| **Valence ratings** | | | |
| Patient | 5.75 | 5 | 4.125 |
| Control | 6.48 ( ± 0.98) | 5.01 ( ± 0.63) | 2.42 ( ± 0.98) |
| **Arousal ratings** | | | |
| Patient | 2.125 | 1 | 2 |
| Control | 4.25 ( ± 1.75) | 3.22 ( ± 1.28) | 5.81 ( ± 1.87) |
| **SCR (microSiemens)** | | | |
| Patient | 0.016 | 0.0038 | 0.005 |
| Control | 0.1696 ( ± 0.26) | 0.0941 ( ± 0.21) | 0.1701 ( ± 0.31) |
| **Left corrugator EMG (microVolts)** | | | |
| Patient | 0.07991 | −0.314 | 0.4509 |
| Control | −0.3295 (NA) | −0.0042 (NA) | 0.33041 (NA) |
| **Right corrugator EMG (microVolts)** | | | |
| Patient | 0.00747 | 0.06657 | 0.10376 |
| Control | −0.2592 (NA) | −0.0051 (NA) | 0.32848 ( ± 0.33) |
| **Left zygomatic EMG (microVolts)** | | | |
| Patient | 0.19884 | 0.25715 | −0.3199 |
| Control | 0.24231 (NA) | −0.0805 (NA) | −0.1523 (NA) |
| **Right zygomatic EMG (microVolts)** | | | |
| Patient | 0.08776 | 0.52489 | −0.4258 |
| Control | 0.24778 ( ± 0.34) | −0.106 (NA) | −0.143 (NA) |

NA indicates that data are not available.

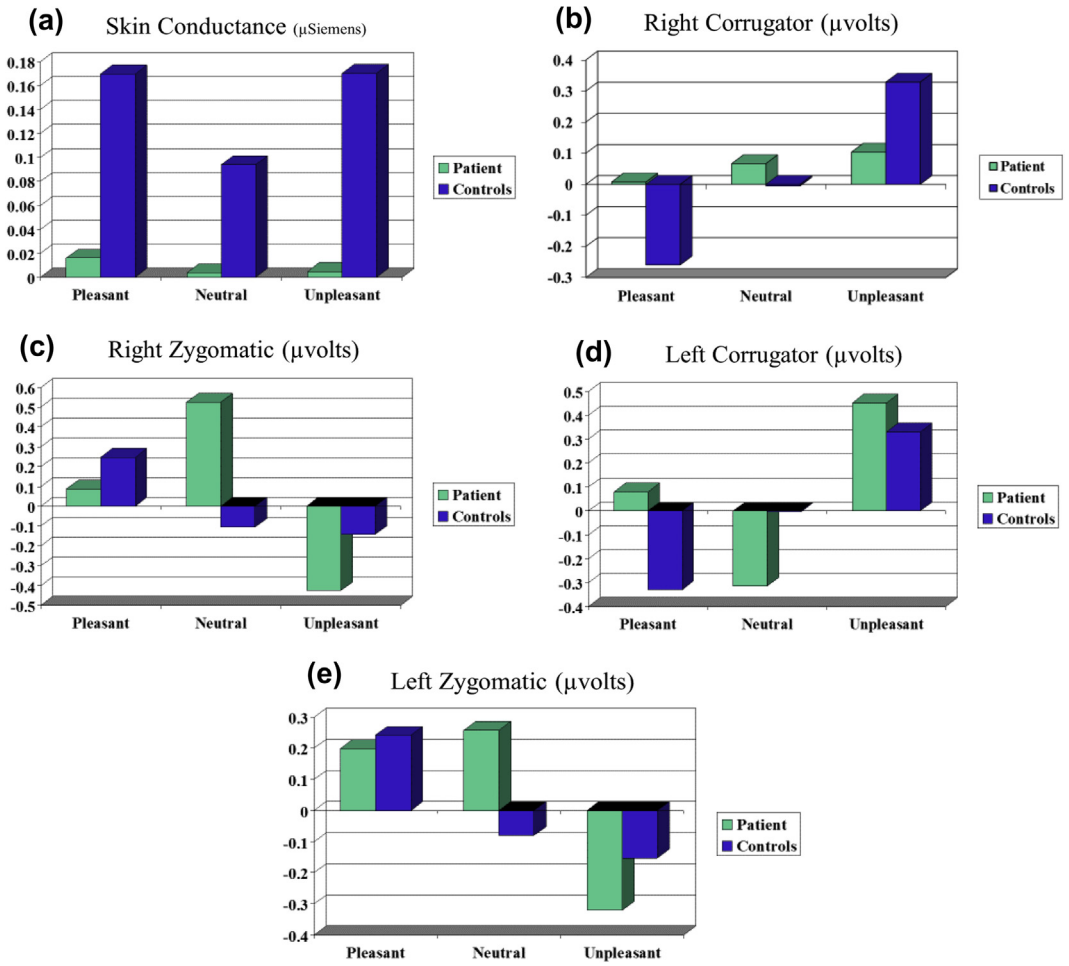


**Fig. 3.** Skin conductance (A) and facial electromyography (B-E) ratings for the patient (Jane) and control participants.

displayed a more complex pattern of abnormality relative to control participants. Jane's *right zygomatic* muscle showed blunted responses to pleasant images (0.08776 Microvolts, $z = -0.475$; i.e., weaker activation compared to control participants), but also increased activation to neutral ($M = 0.52489\ \mu V$) and deactivation to unpleasant images ($M = -0.42578\ \mu V$) (Fig. 3C). Jane's *left corrugator* showed increased activation, similar to controls, for unpleasant images ($M = 0.4509\ \mu V$), but no deactivation to pleasant images ($M = 0.07991\ \mu V$) and large response magnitudes to neutral images ($M = -0.314\ \mu V$) (Fig. 3D). Finally, Jane's *left zygomatic* muscle showed increased activation similar to controls for pleasant images ($m = 0.19884\ \mu V$), but also unusually strong activation to neutral ($m = 0.25715\ \mu V$) and somewhat pronounced deactivation to unpleasant images ($m = -0.3199\ \mu V$) (Fig. 3E). Thus, almost all right facial muscle response patterns were notably different in Jane. However, while Jane's overall left facial muscle response patterns were also distinct from the patterns seen in healthy participants, with unusually strong activation (zygomatic) and deactivation (corrugator) to neutral images, they did reveal evidence of some partially normative response patterns as well. Specifically, Jane showed a pattern of relatively greater left zygomatic responses to pleasant than unpleasant images, and relatively greater left corrugator responses to unpleasant than pleasant images, similar to that observed in healthy populations.

## 4. Discussion/Conclusions

In this case study we provide a detailed characterization of the affective functioning of an individual (Jane) with severe alexithymia, as confirmed by both her very high TAS-20 score and her very low LEAS score. The pattern of normal and abnormal responses in Jane provides important insights into the dissociability of distinct affective processes, and into how these processes may contribute to deficits in experiencing/recognizing one's own emotions. As reviewed in the introduction, however, alexithymia is likely quite heterogeneous; so these results are most plausibly viewed as one empirically confirmed combination of process abnormalities, out of many possible combinations that may contribute to different cases of alexithymia. In what follows, we review the possible implications of the particular pattern of preserved and abnormal processes/functions observed in Jane. Given the qualitative/descriptive nature of our results, however, the discussion below should be viewed as suggestive and preliminary, with the

potential to guide future research on the dissociability and heterogeneity of symptoms within alexithymic populations.

### 4.1. Preserved function

First, Jane's PAT and RMET scores provide evidence that she can recognize emotions in others at normal levels, and her own statements in her interview further suggest that she has acquired some conceptual knowledge about discrete emotions (e.g., knowing that she "should" feel certain emotions in specific contexts, like a graduation ceremony or a funeral); it was also interesting in this context that her LEAS OTHER subscale score was somewhat higher than her LEAS SELF subscale score – further suggesting relatively greater awareness for the emotions of others. Previous studies suggest that alexithymia is typically associated with impaired emotion recognition for others, and impaired semantic representation of emotion concepts, suggesting that Jane is an unusual case in this regard (for a review, see (Grynberg et al., 2012)).

Her interview statements, and clinical observations during the interview, also suggest that Jane is able to generate some aspects of affective responses in a normative manner (e.g., crying when discussing a sad topic, laughing at jokes, vomiting due to nervousness before a date, becoming irritable or "nasty" in response to customers at work, getting a stomach ache while separating from her husband, etc.). As also reported in previous studies of alexithymic individuals (e.g., (Peasley-Miklus, Panayiotou, & Vrana, 2016; Roedema & Simons, 1999)), Jane's self-reported valence ratings for all affective images were also broadly consistent with those of control participants (although they appeared somewhat restricted in range). Some of Jane's facial EMG responses to some affective image categories were also normal (i.e., her left corrugator responses to unpleasant images and her left zygomaticus responses to pleasant images appeared similar to those in control participants). These findings suggest that Jane's brain was capable, at least in part, of representing both simple affective discriminations as well as more complex emotion concepts (i.e., intact ARR processes), and that some of her response generation (i.e., ARG) processes were also normal. These areas of preserved function are notable because, in certain contexts, this could lead someone like Jane to use emotion words appropriately. For most clinicians, who are not specialists in alexithymia, the occasionally appropriate use of emotion words creates confusion as to whether alexithymia (technically meaning "lacking words for emotions") is present.

### 4.2. Atypical function

#### 4.2.1. Arousal responses

In contrast, relative to control participants, Jane displayed markedly blunted skin conductance responses; her self-reported arousal ratings for all affective images were similarly much lower than control participants. These results suggest that Jane did not generate, or subsequently represent/experience, typical autonomic arousal responses (i.e., possibly an example of a deficient affective arousal generation process leading to a failure in normative arousal representation). They are also consistent with previous studies that have found autonomic hypoarousal in alexithymia (Neumann, Sollers, Thayer, & Waldstein, 2004; Peasley-Miklus et al., 2016; Pollatos, Schub, Herbert, Matthias, & Schandry, 2008, Pollatos et al., 2011; Roedema & Simons, 1999; Wehmer, Brejnak, Lumley, & Stettner, 1995) (however, other studies instead find evidence of hyperarousal; see (Cecchetto, Rumiati, & Aiello, 2017; Friedlander, Lumley, Farchione, & Doyal, 1997; Luminet, Rimé, Bagby, & Taylor, 2004)). Given the nature of the affective stimuli (i.e., IAPS images depicting complex scenes), however, it remains unclear whether this failure to generate arousal reflects a (possibly congenital) lack of unconditioned arousal responses, or whether it instead represents a failure to learn normatively appropriate conditioned arousal responses during development. Although a somewhat similar lack of arousal responses has previously been associated with psychopathy (Lorber, 2004), Jane's score on the CPI-so showed no indication of deviant/psychopathic tendencies.

#### 4.2.2. Valence responses

Relative to control participants, Jane's right corrugator responses did not differentiate between pleasant and unpleasant affective images, while her left corrugator appeared to display more typical responses to unpleasant images, but less typical patterns for neutral and pleasant images. Normative studies have shown that corrugator responses reliably index both pleasantly and unpleasantly valenced responses (i.e., with decreased and increased activation from baseline, respectively; (Larsen et al., 2003)), although more recent work suggests this effect may only occur when valence is task-relevant (i.e., as it was in the presently used rating task; (Weinreich & Funcke, 2014)). Thus, while these results suggest that Jane's left corrugator generated normative responses to unpleasant stimuli, all other left and right corrugator responses to the different valence categories were either absent or atypical. It remains unclear, however whether this reflects a basic deficit in ARG processes, or whether it might also reflect a deficiency in appropriately treating valence as task-relevant (and thus failing to simulate/represent valence by using the corrugator muscles, as suggested by (Weinreich & Funcke, 2014)). Given that Jane showed somewhat intact valence discrimination abilities in her self-report ratings (suggesting normal valence representation ability), we suggest an ARG process deficit appears more plausible.

Relative to control participants, Jane's right zygomatic responses also showed several abnormalities. Normative studies have shown that zygomatic responses reliably index strongly pleasant stimuli, and that these responses tend to remain near baseline for unpleasant and neutral stimuli (Larsen et al., 2003); a largely similar pattern was also observed in our control participants. In examining Jane's zygomatic responses, while her left zygomatic muscle responded in a typical manner to pleasant images, all her other left and right zygomatic responses showed a complex pattern of amplified, blunted, and sometimes reversed responses to differently valenced images relative to healthy controls (i.e., with greater activation to neutral stimuli and deactivation to unpleasant stimuli). These findings further suggest that some valence-related ARG processes in Jane are abnormal. It is notable that previous work has failed to find EMG abnormalities in alexithymic individuals (e.g., (Peasley-Miklus et al., 2016; Roedema & Simons, 1999)),

suggesting that Jane may be unusual in this regard.

#### 4.2.3. Additional observations

In reviewing Jane's self-reported statements, it also appears plausible to suggest that her learned concept of "emotions" may be somewhat restrictive. For example, despite claiming that she does not feel emotions, and that this has been true her whole life, she nonetheless stated that (1) she had felt "nervous" before going on dates, (2) that she felt "relieved" to learn that her condition had a name, and (3) that she was "bored" by exercise. She also indicated a general dissatisfaction with the lack of closeness in her marriage. So it appears that these experiences did not count as "feeling emotions" in her mind. It is therefore possible that, for Jane, failing to experience heightened arousal and fully valence-appropriate facial changes may have played a role in leading her to infer that she was not really feeling emotions, even if other aspects of her experience (e.g., tears and stomach pain in normatively affective contexts) were consistent with the emotion knowledge she had acquired. For example, without sufficient arousal, perhaps her boredom and relief simply felt too "cold" to count as real emotions in her mind (although it is less clear how to reconcile this lack of arousal with her self-reported feeling of nervousness in adolescence).

It is also worth highlighting that Jane was capable of representing positive and negative affect. For example, aside from her valence ratings being relatively typical, she was also motivated to seek help because she did not like her alexithymic condition (i.e., she communicated a general sense of emptiness/hopelessness about a life without emotions – suggesting that her experience was very different from simply feeling emotionally "neutral"); she further reported many unpleasant/painful bodily sensations. Her dissatisfaction with her life without emotions (especially without positive, motivating, high-arousal emotions – perhaps reflecting anhedonia), and her failure to find treatment for her condition, also appear to have contributed to her eventual decision to commit suicide (although, as there was no suicide note, the motivations for making this decision cannot be known with certainty). Moreover, her preserved ability to recognize emotion in others may have contributed to her awareness of her own deficits by comparison and a sense that she was missing out on a vitally important aspect of life. Thus, despite her severe deficit in experiencing/recognizing discrete emotions in herself, she does appear to have experienced a general sense of negative affect (some of which was caused by her dissatisfaction over a life that felt empty without emotions or meaningful social relationships). This, when combined with a lack of hope that things could improve (e.g., a belief that there were no effective treatment options), may have been sufficient for her to choose to end her life. Feelings of social isolation, such as Jane's sense of detachment in her relationships (not feeling connected to her husband, not experiencing feelings at funerals, graduations, during sex, etc.), have also been shown to directly increase suicide risk generally (Trout, 1980). These considerations highlight how dissatisfying and life threatening severe alexithymia can be, even in the absence of an individual meeting criteria for any other mental disorder.

### 4.3. Implications for treatment

Treatment is a critical issue in this case history because it was the reason for her seeking medical attention. The lack of accepted treatments for alexithymia that could be offered quite plausibly contributed to her giving up hope and committing suicide (i.e., consistent with previous studies suggesting that alexithymia is a risk factor for suicide; (Carano et al., 2012; De Berardis et al., 2017; Hintikka et al., 2004; Kušević, Ćusa, Babić, & Marčinko, 2015)). Moreover, the conceptual advances since her death, including the description of "affective agnosia" and the three-process model, have greatly informed the possibilities for treatment.

At the time that Jane sought consultation, her primary question was whether her condition could be treated. She was told that a recommendation regarding treatment could be made after a detailed evaluation. At that time, it was well-known that alexithymia was difficult to treat (Krystal, 1979; Ogrodniczuk, Piper, & Joyce, 2011). A consultation was sought from another leading expert in alexithymia, who, consistent with prevailing wisdom, recommended that group therapy be offered (Apfel-Savitz, Silverman, & Bennett, 1977), but added that it would likely not help (although more recent positive findings have been reported from multimodal treatments including a combination of individual, group, and body-oriented treatments; (Subic-Wrana, Beutel, Garfield, & Lane, 2011)). In addition to the absence of a coherent treatment that we could offer, further treatment logistics that delayed intervention included that (1) long-term treatment would likely be needed; (2) she lived over 100 miles away, such that traveling to frequent appointments over an extended period of time would have been difficult; (3) in the absence of a specific plan or course of action a therapist can be challenged to find the motivation to make a long-term commitment; (4) she specifically denied having suicidal feelings or any history of suicide attempts; (5) she was in treatment with a psychiatrist in her hometown, whom we had also informed that we would be making treatment recommendations by a certain date following this consultation. Attempts to contact Jane were delayed for 2–3 weeks in the hope of finding something positive to offer in the interim. When the call was made to discuss treatment possibilities, her husband reported that she had committed suicide.

An important conceptual advance with treatment implications was the description of "affective agnosia" as a severe form of alexithymia (Lane et al., 2015). Rather than lacking words for emotions, the concept of affective agnosia holds that the primary deficit is in the capacity to acquire and/or appropriately activate concept-level representations of emotional states (for recent empirical studies of emotion conceptualization ability, see (Kashdan, Barrett, & McKnight, 2015; Smith, Alkozei, et al., 2017; Smith, Bajaj, et al., 2018; Smith, Lane, et al., 2017; Smith, Lane, Alkozei, et al., 2018; Smith, Lane, Sanova, et al., 2018; Smith, Sanova, Alkozei, Lane, & Killgore, 2018; Wilson-Mendenhall, Barrett, Simmons, & Barsalou, 2011)). Instead of lacking words for emotions as the primary deficit, such patients lack the ability to know/understand what they are feeling; difficulty in describing feelings in words follows from the deficit in being aware of and understanding their affective responses. This corresponds to affective response representation or ARR deficits in the three-process model, and it has important clinical implications because techniques to promote mentalization of affective states are now well established (Fonagy & Luyten, 2009; Lumley et al., 2017; Luyten, Houdenhove, Lemma,

Target, & Fonagy, 2012). Had this conceptual advance been made at the time Jane sought consultation, our response to her request for treatment could have been more positive and specific.

The detailed case history above, however, indicates that the deficit in self-reported emotional experience/recognition is more complex. The minimal skin conductance responses indicate some degree of impairment in affective response generation or ARG processes, which likely corresponds to others perceiving her as boring. Although genetic factors could play a role, another possibility is that sub-optimal early interactions with parents during development could also promote abnormalities in affective responding. It is possible, given Jane's history, that interactions with her mother were quite deficient in this regard. In a psychotherapeutic context, it might be important to create an interpersonal relationship where affective responses could be activated and attended to. It would then be important to pick up on any instances of such responses, such as when Jane's eyes welled up with tears, and to link those responses to the eliciting context and possible verbal labels that would give them emotional meaning. By so doing, the goal would be to induce feelings in the here and now that could then be reflected upon and described, encouraging those feelings to be conceptualized and recognized as emotions. Based on what is known about how emotional awareness typically develops within early social relationships (Gergely & Watson, 1996), if a greater sense of client-clinician connectedness could have been fostered in Jane's case, perhaps she would have benefited from this treatment approach.

The psychometric data suggest that the third process regarding cognitive control in the three-process model might be relevant as well. Specifically, Jane's scores on the Marlowe Crowne and Bendig Short Form of the Taylor Manifest Anxiety Scale placed her in the repressor or repressive coping style category. This designation is potentially confusing, given that the co-occurrence of alexithymia and repression could be seen as inconsistent; alexithymia is considered a deficit, not a defense. The alexithymia concept arose specifically because clinical attempts to overcome defenses in these patients did not yield the expected expression of emotion, and because usual therapeutic techniques did not work (Nemiah & Sifneos, 1970). Nevertheless, in the context of deficits, defenses may still be present. Consistent with her description of her attempts to be pleasant at work, she did show signs of actively striving to behave in a socially desirable manner (corresponding to items endorsed on the Marlowe Crowne scale). Drawing upon psychometric evidence that repressive coping style and alexithymia both involve deficits in emotion recognition (Lane et al., 2000), but that those deficits are more severe in alexithymia, one of us (RDL) proposed that alexithymia may constitute a more severe form of affect recognition impairment. From a clinical standpoint, when people with affective agnosia or severe alexithymia have affective responses, they are often dysregulated (Taylor, 2010). Jane's nervousness to the point of difficulty eating before dates could be such an example. Defensiveness or avoidance strategies might well be ways to avoid such emotional upheavals.

Thus, the therapeutic task from the perspective of the three-process model is quite complex. One may need to overcome defensive cognitive habits (e.g., thought suppression, avoidant attention, etc.) in a way that would alter affective response generation and representation. Affective responses may need to be attended to and put into words to help increase understanding of their source and conceptual meaning. Considerable effort could be needed to learn a vocabulary of emotion words and to link those words to particular bodily feelings in particular contexts. Another important step would be to model and practice ways of handling or regulating emotions so that they are not overwhelming. The patient could then learn to make use of his or her own affective responses as a guide to decision making in the social world. Indeed, the three-process model would serve as a useful guide in clinical treatment and – if it had been available at the time of Jane's consultation – could have led to a more satisfying response and averted tragedy. In fact, with a psychotherapeutic treatment plan formulated in this way, it might have been possible to serve as guidance to a psychotherapist in her local community.

### *4.4. Limitations and conclusions*

It is important to be cautious in drawing conclusions from single cases with the type of qualitative/descriptive results that we have described. However, the case of Jane does appear to illustrate the dissociable nature of different aspects of an affective response. Her case demonstrates, for example, that the processes that generate a crying response may remain intact even if the generation of skin conductance responses is severely compromised. Further, it illustrates how some facial EMG responses to affective stimuli (which reliably distinguish valence in healthy individuals) can be atypical/absent while others can apparently remain normal. Jane's case also suggests that these selective abnormalities in ARG processes may lead an individual to believe that they are not experiencing emotions, even when (1) some aspects of emotional experience are intact, (2) they have acquired sufficient knowledge about specific emotions, and (3) they can recognize emotions accurately in others (i.e., ARR processes are intact). Finally, her case illustrates how dissatisfaction over having a perceived inability to feel emotions can be sufficient to lead to suicide.

We do not have sufficient information to determine the origin of Jane's deficits. It is possible that some or all of the ARG deficits are congenital. For example, her mother appeared to seek resolution to emotional concerns in a very concrete, non-negotiable manner (e.g., buying a cemetery plot), which could suggest alexithymic traits in her as well. It is also possible that interactions with parents during development (e.g., failing to discuss emotions during childhood, repeatedly observing unhealthy catastrophizing responses in her mother, being treated in a harshly critical manner by her mother, a possible lack of responsiveness and warmth in early mother-infant interactions) could have contributed to the ARG abnormalities we observed. Whatever their origin, we suggest that Jane did not self-report experiencing discrete emotions, at least in part, because only some ARG processes were functioning normally and not others. In the absence of feeling a more complete and coherent affective response, it is understandable why recognizing such responses *as emotions* would not occur. This would be analogous to an individual failing to visually recognize that they see a particular object in an image (e.g., a table) because the image is noisy (e.g., low resolution) or because the image only actually shows part of that object (e.g., only one corner of the table) (for a more detailed discussion of structural similarities between visual and self-directed emotion recognition abilities, see (Lane et al., 2015; Smith and Lane, 2015)). Further, if Jane only experienced these relatively noisy/

incomplete affective reactions during development, it could have also plausibly hindered her ability to fully learn about and understand the somatovisceral aspects of emotion (i.e., impoverished emotion concept learning).

In conclusion, this case study highlights how psychometric and psychophysiological response abnormalities may account for a case of severe alexithymia; it also demonstrates how some ARG processes may be abnormal while others are preserved, as well as how such abnormalities may hinder emotion concept learning and lead an individual to infer that they do not feel emotions. Future research should evaluate whether similar patterns are observed in other individuals with alexithymia, or whether alexithymia may instead represent a more heterogeneous condition in which different process abnormalities account for different cases. Answering such questions will be an important first step in designing and testing individualized treatments that may be effective in reducing the subjective and objective manifestations of alexithymia, and in potentially preventing the life threatening consequences that were unfortunately observed in this case.

## 5. Statement of ethics

The authors have no ethical conflicts to disclose.

## 6. Disclosure statement

The authors have no conflicts of interest to declare.

## 7. Disclaimer

The content is solely the responsibility of the authors and does not represent the official views of the NIH or federal government.

## Author contributions

Ryan Smith took the lead in writing the manuscript and participated in data analysis. Alfred W. Kaszniak participated in the psychometric and psychophysiological testing of controls subjects performed in his laboratory. Joanna Katsanis participated in assessing the patient in this case report and also aided in writing the manuscript. Richard D. Lane participated in assessing the patient in this case report and also aided in writing the manuscript. Lisbeth Nielsen participated in acquiring and analyzing the psychometric and psychophysiological data and in the writing of the manuscript.

## Appendix A. Supplementary material

Supplementary data to this article can be found online at https://doi.org/10.1016/j.concog.2018.12.004.

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