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RESEARCH ARTICLE

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Music appreciation phenotypes in patients with frontotemporal dementia: A pilot study

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Abstract

Objectives: Frontotemporal dementia (FTD) can present with changes in music appreciation. Research has suggested a relationship of altered music appreciation phenotypes with typical socio-emotional changes. We aimed to determine the prevalence and severity of music appreciation phenotypes in FTD and study the relationship with emotion recognition capacities in order to examine whether they could serve as a proxy for changes in socio-emotional functioning.

Methods/Design: Based on reported musical changes in the literature, we developed an informant-based questionnaire to assess musical changes and a music test to assess music emotion recognition. Social cognition was assessed with the Ekman 60 faces test in a subgroup of patients (n = 23). Relationships between measures were assessed with linear regressions.

Results: We included 47 patients (44.7% female, mean age 65.0 ± 8.4, 31 behavioral variant FTD (bvFTD), 10 semantic dementia (SD), and six progressive nonfluent aphasia (PNFA)). Thirty-six caregivers were included in the music emotion recognition test as controls. Altered music appreciation phenotypes were observed in 79% of the FTD patients. Musicophilia was present in a third of bvFTD patients, and only in up to 10% in language FTD variants. Changes in music appreciation were not associated with decreased music emotion recognition or visual emotion recognition. Compared to controls, bvFTD performed worse on the music emotion recognition task ($p < 0.003$), and no differences were found with SD ($p = 0.06$) and PNFA patients ($p = 0.8$).

Conclusions: Music appreciation phenotypes are highly prevalent in FTD patients. Future studies should further investigate the potential diagnostic value of changes in music processing.

KEYWORDS

emotion recognition, frontotemporal dementia, music, musicophilia, social cognition

Key points

- Music appreciation phenotypes are highly prevalent in frontotemporal dementia.
- Music appreciation phenotypes are not related to emotion recognition capacities.
- bvFTD patients perform worse on a music emotion recognition task.

1 | INTRODUCTION

Frontotemporal dementia (FTD) is the second most prevalent cause of early-onset dementia following Alzheimer's disease.[1,2] FTD is a spectrum of diseases that consists of behavioral variant FTD (bvFTD), and two language variants: semantic dementia (SD) and progressive nonfluent aphasia (PNFA). Socio-emotional and behavioral abnormalities are the hallmarks of FTD,[3] ranging from deficits in emotion recognition to failure of higher order moral reasoning.[4,5] However, there is currently no gold standard for measuring social cognition and social cognitive changes in FTD can be difficult to detect with standard clinical tests because of potential interference with other cognitive domains.

Music is argued to be important for forming social bonds and inferring mental states.[6–9] One potential novel diagnostic tool could be measures of music appreciation, which has been reported to be altered in FTD.[10] Patients can present with hedonic changes of music appreciation, ranging from musicophilia (enhanced craving for music),[11] music-aversion,[12] sound-aversion[10,12] and changes in musical taste.[11,13,14] However, studies on music appreciation phenotypes in FTD either consist of case reports or did not study the full spectrum of altered music appreciation phenotypes. Only one study by Fletcher et al. 2015 systematically investigated multiple music appreciation phenotypes and found musicophilia, music aversion or sound aversion in 55% of 56 FTD patients.[12] Studies that investigated individual music appreciation phenotypes found musicophilia in 26%–64%,[11,12,15] sound-aversion in 43% and music-aversion in 37% of FTD patients.[12] Studies have suggested that music appreciation phenotypes might be related to typical socio-emotional dysfunction of FTD.[15,16] For example, patients with musicophilia performed worse on a theory of mind (social inference) task and showed more atrophy in regions associated with social cognition.[11] It remains unknown how other music appreciation phenotypes relate to social cognition, and whether different phenotypes show specific associations with social cognition. Studies found similar brain regions involved in socio-emotional processing and music processing in FTD[12,17,18] and a role for music in the evolution of socio-emotional abilities.[7,8,19] Social cognition is hierarchically defined by multiple components, such as emotion recognition, empathy and social reasoning.[20,21] At present, social cognition in FTD is most commonly assessed with a visual emotion recognition tasks,[22,23] which largely require similar brain regions as music emotion recognition.[24,25]

The aim of this study was to prospectively examine the prevalence and severity of music appreciation phenotypes in FTD. We further aimed to study whether music appreciation phenotypes were related to alterations in social cognition, as measured with Ekman 60 faces test. Finally, we used a music emotion recognition task to study the relationship between music and visual emotion recognition.

2 | METHODS

2.1 | Patients

Patients were recruited prospectively and consecutively from April 2018 to November 2020 through the Amsterdam Dementia Cohort in a specialized outpatient memory clinic of the Alzheimer Center Amsterdam. All patients were diagnosed with a syndrome in the FTD spectrum (i.e. behavioral variant frontotemporal dementia (bvFTD), semantic dementia (SD), or PNFA) by a senior neurologist according to current consensus criteria.[3,26] Patients were excluded if they suffered from hearing loss (e.g. used hearing aids, or hearing loss interfered with the testing procedure), were unable to perform the procedure (e.g. due to difficulties with understanding the procedure, cognitive problems), answers were judged as unreliable by the investigator, or if participants were unable give informed consent. Caregivers were included as a control group for the music emotion recognition task and consisted of spouses, siblings or children that knew the patient well before the disease had developed. Disease severity was assessed with the Frontotemporal Lobar Degeneration Clinical Dementia Rating Scale (FTLD-CDR), Mini-Mental State Examination (MMSE), FTD Rating Scale (FRS) and the Frontal Assessment Battery (FAB). The medical ethics committee of the Amsterdam University Medical Centers approved the study in March 2018. All participants gave informed consent in accordance with the Declaration of Helsinki.

2.2 | Music appreciation phenotypes

To assess music appreciation phenotypes, we performed a semi-structured informant-based interview using the questionnaire by Fletcher et al. 2015, which we extended as follows: we added music appreciation phenotypes (i.e. musical anhedonia, change in musical taste, music agnosia, hallucinations, tone-deafness, singing/dancing) and we included a musical change severity score. Caregivers answered about each music appreciation phenotype whether it was present compared to premorbid behavior (yes/no). For the musical change severity score the 3 most striking hedonic phenotypes (musicophilia, change in musical taste and music anhedonia) were scored on a 5-point rating scale, ranging from 0 (no alteration) to 4 (complet. alteration) (Appendix 1). The scores from these 3

phenotypes were totaled to perform analyses on a musical change severity score. Musicophilia was defined as an increased interest in music compared with premorbid behavior (typically associated with compulsive music listening (>10 h per week) and music associated behaviors)[11,15]; musical anhedonia was defined as decreased interest music. Caregivers were instructed to compare the current situation with before symptoms of the disease were present. If any musical alterations were present, additional details were noted (e.g., the kind of changes and corresponding behavior, summarized in Table S1). Finally, caregivers answered yes or no questions on whether the patient showed sound aversion (a strong negative reaction when hearing sound), music aversion (a strong negative reaction when hearing music), increased singing/dancing, musical hallucinations, musical agnosia (inability to recognize familiar music pieces) and tone-deafness (inability to distinguish differences in pitch). To have an indication on musical knowledge, we noted if the patient and caregiver played a musical instrument (including voice).

2.3 | Music emotion recognition

We tested music emotion recognition as follows: Nine chords were played subsequently on an acoustic guitar, which were either major, minor or dissonant in nature. Three excerpts of each emotional class were played. Each excerpt lasted around 5 s. All chords were played in root position and consisted of four notes. First the full chord was strummed and then the individual four notes of the chord were fingerpicked. After a chord was played, the participants judged a chord as either major, minor or dissonant. However, since not everyone is familiar with these terms, participants could judge the chords as either happy, sad or out of tune/disharmonic. A room without visual or auditory distractions was used. The guitar was tuned prior to each test, and examples were played to become familiar with the exercise and to make sure that the audibility was sufficient. Each couple alternately started with either the patient or caregiver and started with either the first or second set of chords (the chords and an explanation of the procedure can be found in Appendix 2). The patient and caregiver were in the room during both tests. Patients with severe speech impairment were given the option to point their answers on a form displaying multiple choices. At the end of the test the number of correct answers was noted.

2.4 | Social cognition

Social cognition was assessed with a visual emotion recognition task (the Ekman 60 faces test),[27] and was part of standard neuropsychological testing in bvFTD. In this test participants see photographs of actors that express one of six basic emotions (happiness, sadness, fear, disgust, anger and surprise) and judge which emotions best matches the photograph. The maximum score is 60 points for all six emotions and 10 points for each emotion. Most patients in our cohort with in bvFTD had visual emotion recognition assessed during the diagnostic trajectory. In SD and PNFA social cognitive changes contribute less to diagnostics, and as such were not part of standard neuropsychological testing.

2.5 | Statistical analysis

Statistical analyses were performed with R (version 4.1.0) in RStudio (version 1.4.1717). For demographical characteristics we used *T*-tests, Kruskal-Wallis tests and chi-square tests where appropriate. The presence and severity of music appreciation phenotypes were investigated using linear regression analysis corrected for age and sex. The proportions of patients exhibiting music appreciation phenotypes were assessed with Pearson's chi-square tests. We used *t*-tests to investigate the performance of tests in patients with musicophilia and musical anhedonia as they might reflect two sides of a spectrum in music appreciation. The relationship of music emotion recognition and visual emotion recognition was assessed with Pearson's correlation tests. Non-parametric tests were used to compare the music emotion recognition task of patients and caregivers. We investigated the effects of playing a musical instrument on music appreciation phenotypes using chi-square tests, and the association of playing an instrument on both disease severity and emotion recognition capacities using linear regression analysis corrected for age and sex. A threshold of $p < 0.05$ was accepted as a statistically significant difference in all comparisons.

3 | RESULTS

3.1 | Clinical and behavioral characteristics

We included 47 patients (21 female; mean age 65.0 ± 8.4 (SD) years). The music emotion recognition test was performed on 36 caregivers as control group (16 female; mean age 61.9 ± 8.4 (SD) years) (Table 1). Frontotemporal dementia patients and controls showed similar distributions in age, sex, education, and playing a musical instrument. Frontotemporal dementia subtypes showed similar disease durations ($p = 0.61$), and frequency of playing a music instrument ($p = 0.96$). Progressive nonfluent aphasia patients had worse FTLD-CDR scores compared to bvFTD ($p < 0.02$) and SD ($p < 0.003$). The informant-based semi-structured interview was performed for all patients, the music emotion recognition task on 38 patients, and the visual emotion recognition test (Ekman 60 faces test) was performed on 22 bvFTD patients and one SD patient.

3.2 | Music appreciation phenotypes

One or more changes of music appreciation were present in 37 of the 47 included patients (79%). Musicophilia was most often present (12/

TABLE 1 Demographic, clinical, musical and social cognitive characteristics

	bvFTD	SD	PNFA	Total FTD	Controls
Demographic and clinical					
No. (F:M)	31 (13:18)	10 (3:7)	6 (5:1)	47 (21:26)	36 (20:16)
Age in years (±SD)	64.3 ± 9.7	65.3 ± 5.5	68.3 ± 3.6	65.0 ± 8.4	61.9 ± 8.4
Plays an instrument (%)	12 (39%)	4 (40%)	2 (33%)	18 (38%)	6 (16.6%)
Education (±SD)[a]	5.1 ± 1.3	5.3 ± 0.8	4.7 ± 1.2	5.1 ± 1.2	n.a.
MMSE (±SD)	24.3 ± 3.0	25.7 ± 3.0	26 ± n.a.	24.7 ± 3.0	n.a.
FAB (±SD)	14.4 ± 2.7	14.3 ± 2.0	12 ± n.a.	14.3 ± 2.5	n.a.
FTLD-CDR (0–3)	1.1 ± 0.7	0.6 ± 0.2	**2.5 ± 0.7**[b]	1.1 ± 0.8	n.a.
FRS (±SD)	51 ± 22	87 ± n.a.	n.a.	53 ± 23	n.a.
Symptom duration in years (±SD)	5.0 ± 3.0	4.2 ± 2.1	4.0 ± 2.2	4.7 ± 2.7	n.a.
Musical tests					
Percentage of music appreciation phonotypes	25 (81%)	8 (80%)	4 (67%)	37 (79%)	n.a.
Musicophilia	11 (35%)	1 (10%)	0 (0%)	12 (26%)	n.a.
Musical anhedonia	7 (23%)	3 (30%)	1 (17%)	11 (23%)	n.a.
Changes in music taste	8 (26%)	1 (10%)	1 (17%)	10 (21%)	n.a.
Increased singing	7 (23%)	2 (20%)	1 (17%)	10 (21%)	n.a.
Sound aversion	6 (19%)	2 (20%)	1 (17%)	9 (19%)	n.a.
Music aversion	3 (10%)	2 (20%)	1 (17%)	6 (13%)	n.a.
Music agnosia	3 (10%)	1 (10%)	0 (0%)	4 (9%)	n.a.
Increased dancing	2 (6%)	1 (10%)	0 (0%)	3 (6%)	n.a.
Tone-deafness	0 (0%)	0 (0%)	0 (0%)	0 (0%)	n.a.
Musical hallucinations	0 (0%)	0 (0%)	0 (0%)	0 (0%)	n.a.
Percentage music appreciation phenotypes absent	6 (19%)	2 (20%)	2 (33%)	10 (21%)	n.a.
Correct answers music emotion recognition task/9 (±SD)	**3.2**[c,d] **(1.3)**	3.0[d] (1.7)	4.0[d] (1.8)	**3.2**[c] **(1.4)**	4.2 (1.4)
Social cognition					
Total score Ekman 60 faces test/60 (±SD)	31.0 ± 10.2	35 ± n.a.	n.a.	31.2 ± 10.0	n.a.

Note: Significant differences are displayed in bold.

Abbreviations: bvFTD, behavioral variant frontotemporal dementia; FAB, Frontal Assessment Battery; FTD, Frontotemporal dementia; FTLD-CDR, Frontotemporal Degeneration Clinical Dementia Rating Scale; FRS, Frontotemporal Dementia Rating Scale; MMSE, Mini-Mental State Examination; n.a., not available; PNFA, progressive nonfluent aphasia; SD, semantic dementia.

[a]Level of education was classified using the Verhage system ranging from 1 (no or little education) to 7 (highest academic degree).[35]

[b]the FTLD-CDR score of was higher in PNFA than bvFTD and SD ($p < 0.05$).

[c]the FTD patients combined ($p < 0.003$) and subgroup bvFTD ($p < 0.003$), performed worse on the music emotion recognition task compared to controls.

[d]The music emotion recognition task was completed in 25 out of 31 bvFTD patients, in nine out of 10 SD patients, in four out of six PNFA patients.

47 patients, 26%), followed by musical anhedonia (11/47 patients, 23%), change of musical taste (10/47 patients 21%), increased singing (10/47 patients, 21%), sound-aversion (9/47 patients, 19%), music-aversion (6/47 patients, 13%), music agnosia (4/47 patients, 9%) and increased dancing (3/47 patients, 5%) (Table 1; Figure 1; Table S1). When comparing FTD subtypes, we observed that musicophilia was more frequently present in bvFTD on a trend level (11/31 cases, 35%) compared to SD (1/10 cases, 10%) and PNFA (0% of cases), but this did not reach significance (X^2 (2, $N = 47$) = 4.94, $p = 0.08$). There were no differences in the severity score of music appreciation phenotypes between the subgroups ($p = 0.21$, Figure S1). Other musical changes were evenly distributed across the FTD subtypes. Of the 10 patients who showed increased singing, two patients were able to sing despite having aphasia. Out of 9 patients with sound aversion, 6 had music aversion. No musical hallucinations or tone-deafness were reported. We found no relationship between the presence of music appreciation phenotypes and disease severity as measured with the MMSE (F (1,25) = 0.16, $p = 0.69$), FRS (F (1,13) = 1.77, $p = 0.21$), FTLD-CDR (F (1,25) = 0.50, $p = 0.48$) and FAB (F (1,23) = 0.21, $p = 0.65$). Finally, when investigating these

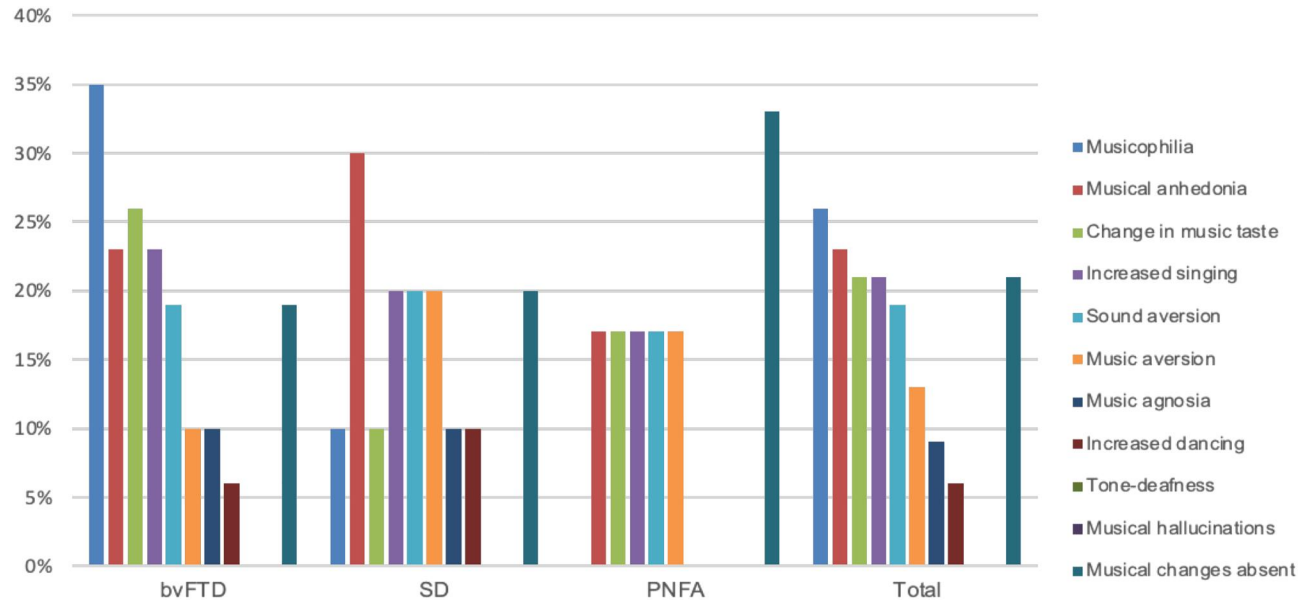

FIGURE 1 Music appreciation phenotypes

measures in musicophilia in comparison to musical anhedonia we found no significant differences between these groups.

3.3 | Music appreciation phenotypes and emotion recognition

Next, we investigated whether music appreciation changes were related to alterations in visual emotion recognition as measured by the Ekman 60 faces test, and music emotion recognition as measured with a novel music emotion recognition task. Within a subgroup of FTD patients (n = 23) with the Ekman 60 faces test scores available, we did not observe an association between the presence ($\beta \pm SE = -4.4 \pm 5.5$, $p = 0.43$) or severity score ($\beta \pm SE = -0.9 \pm 1.2$, $p = 0.47$) of music appreciation phenotypes and visual emotion recognition scores. When investigating the association of visual emotion recognition with the severity scores of musicophilia, musical anhedonia and change in musical taste separately, we found no significant association (all $p > 0.05$). Moreover, we found no associations between the presence ($\beta \pm SE = 0.15 \pm 0.6$, $p = 0.79$) or severity score ($\beta \pm SE = -0.16 \pm 0.17$, $p = 0.34$) of musical changes and music emotion recognition. Frontotemporal dementia patients performed worse on the music emotion recognition task compared to controls ($p < 0.003$; Table 1; Figure 2). Post-hoc analyses showed that compared to controls, bvFTD performed worse on the music emotion recognition task ($p < 0.003$), SD patients had lower scores, but this reached no significance ($p = 0.06$) and no difference was found in PNFA patients ($p = 0.82$) (Figure 2). No differences were observed on music emotion recognition performance between the FTD subtypes. Finally, when comparing both emotion recognition tasks, we found no association between the music emotion recognition task and the visual emotion recognition task ($t(19) = 0.96$, $p = 0.35$; Figure 3).

3.4 | Playing an instrument

Patients that played a musical instrument tended to have music appreciation phenotypes more frequently compared to patients that did not play a musical instrument, but this did not reach significance (X^2 (1, $N = 45$) = 2.92, $p = 0.08$). There was no difference in disease severity between participants that played a music instrument and participants that did not as measured with MMSE ($p = 0.94$), FRS ($p = 0.87$), FTLD-CDR ($p = 0.19$), and FAB ($p = 0.23$). Moreover, we found no difference in performance on the music emotion recognition task ($p = 0.37$) and visual emotion recognition task ($p = 0.51$) in participants that played a musical instrument.

4 | DISCUSSION

In this study, we investigated the prevalence of music appreciation phenotypes in a prospective cohort study in 47 consecutive FTD patients, and whether these musical changes are related to socio-emotional changes. We observed musical changes in 79% of FTD patients, of which musicophilia was the most prevalent. The occurrence of music appreciation phenotypes was not related to performance on emotion recognition tasks, suggesting that changes in music appreciation are not related to altered emotion recognition in FTD patients.

The prevalence of music appreciation changes was 79% in our cohort, which was higher than described the previous study by Fletcher et al. 2015.[12] A potential explanation is that Fletcher et al., only looked at the presence of musicophilia, sound aversion and musical anhedonia, and we extended our questionnaire with other music appreciation phenotypes (e.g. changes in taste, music agnosia, singing). Of note is that some of the phenotypes (such as tone

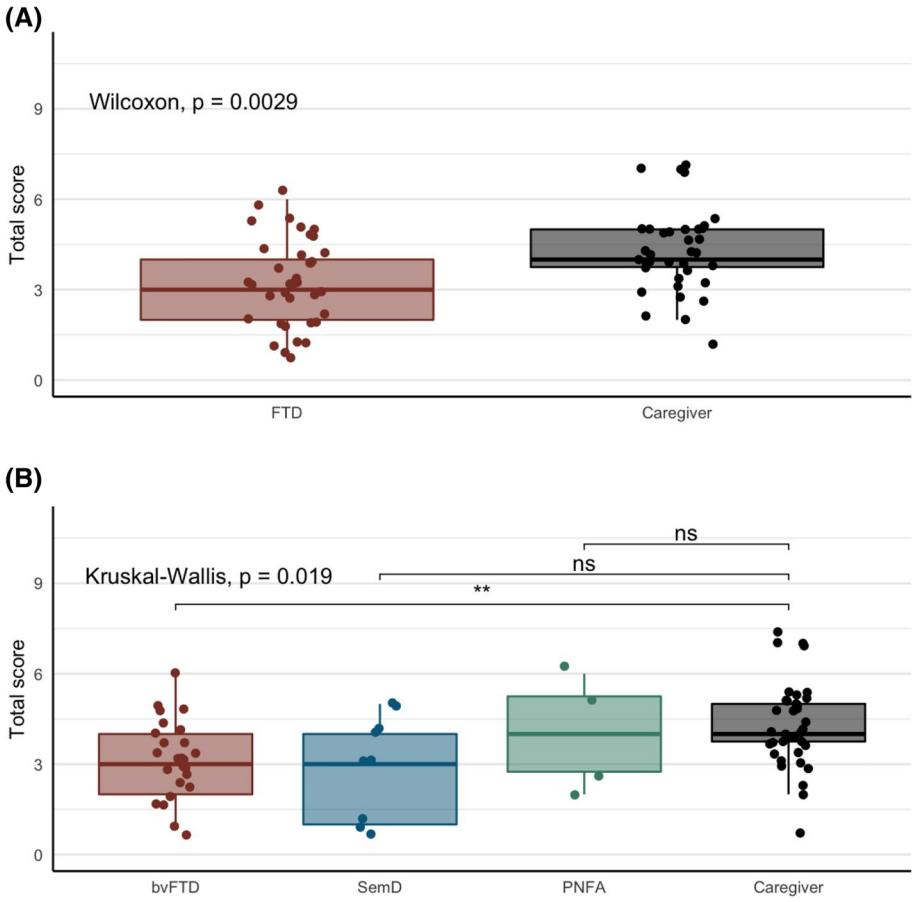

FIGURE 2 Performance on the music emotion recognition task. (A) Frontotemporal dementia (FTD) versus controls, and (B) FTD subtypes versus controls

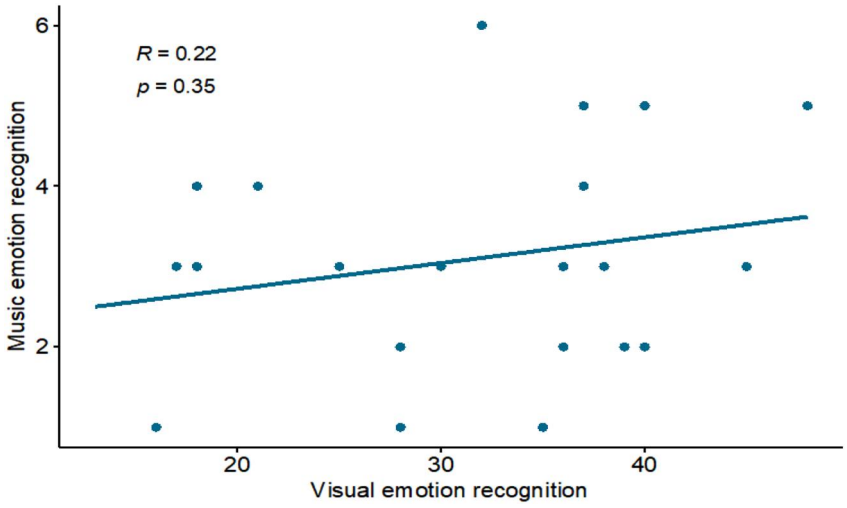

FIGURE 3 The association of music emotion recognition and visual emotion recognition

deafness and musical hallucinations) may be difficult to detect for caregivers, and potential underreporting is also possible. In line with Fletcher et al., we observed that musicophilia was most often present in bvFTD. However, in SD we observed musicophilia was present in only 10% of patients, compared to 64% in the report of Fletcher et al.[10–12] This difference might be explained by our smaller sample size. On the other hand, it might be related to our definition of musicophilia since we did not consider patients as musicophilic if they reported increased dancing or singing without an increased interest in music. Although singing and dancing are important musical activities, some patients in our cohort displayed increased singing or dancing without an increased interest in music. None of our PNFA

patients showed musicophilia, which is in line with previous studies.[12,28] We further found that sound aversion was not always associated with music aversion and vice versa. One patient presented with an interesting combination of sound aversion and musicophilia, portraying the difference between music processing and general sound processing. This is in line with studies that found that music has distinct reward responses.[29–31] Furthermore, the presence of musical changes was not related to performance on the music emotion recognition task. This suggests that changes of music appreciation are not associated with altered capability of extracting emotions from music. An alternative explanation could be that musical changes might result from altered emotion attribution and meaning rather than emotion recognition, due to damage in brain regions involved in reward and behavior,[32,33] as previous studies on music processing in FTD have also suggested.[12,18] We further found that bvFTD patients performed worse than controls on a music emotion recognition task, which is in line with previous research,[15,17] and suggests that changes in musical processing could particularly be of assistance in the diagnostic work-up of bvFTD. Future studies should assess how musical changes might aid to distinguish bvFTD from other neurodegenerative diseases and psychiatric diseases with larger comparison groups.

When further studying associations between music appreciation phenotypes and emotion recognition, we found no relationships with either music emotion recognition or visual emotion recognition. Of note is that the visual emotion recognition task was performed predominantly on bvFTD participants. Furthermore, we found no association of the music appreciation severity scores with visual emotion recognition capacities. These results suggest that the severity of music appreciation phenotypes is not associated with socio-emotional dysfunction. Of note however is that not all the music appreciation phenotypes were included in the music appreciation phenotype severity score, and future studies should score each music appreciation changes to investigate how they correlate with socio-emotional and behavioral changes (e.g. musical anhedonia could be an expression of apathy). Our results are in line with Fletcher et al., who found no worse performance on an emotion recognition task in musicophilic patients.[11] We added to the literature by showing that music appreciation phenotypes are not associated with altered emotion recognition. However, Fletcher et al. showed worse performance on a theory of mind (social inference) task in 12 musicophilic patients. Of note is that the social inference task relies on auditory social cues, whereas we used a visual emotion recognition test. It might be argued that changes of music appreciation are related to socio-emotional dysfunctions on auditory cues and are not generalizable to visual cues. This is not likely however, as our music emotion recognition task also relies on auditory cues and was not related to the music appreciation phenotypes. Another explanation might be that the two tasks measure different components of social cognition. Whereas the Ekman 60 faces test assesses emotion recognition, the social inference task requires theory of mind, and thus uses different components of social cognition. The fact that musicophilic patients performed worse on this task and not emotion recognition suggests that music appreciation phenotypes relate to theory of mind, and not emotion recognition. This is also supported by research that has suggested a role for musical capacities to interpret mental states of others.[6,7,16] Future studies should further assess which components of social cognition relate to music appreciation phenotypes.

A potential limitation of this study is that group sizes for the FTD subtypes were small, particularly in the PNFA and SD subgroups, which may have limited the statistical power of the subgroup comparisons, and these should be interpreted with caution. Furthermore, there was no control group to compare the changes in music appreciation. Disease severity scores (FRS, FTLD-CDR, MMSE, FAB) and Ekman 60 faces tests were not available for all patients because they were not part of standard neuropsychological testing in all patients. Furthermore, we only had one test for social cognitive functioning (i.e., Ekman 60 faces test), which only tests one of the multicomponential processes of social cognition. We acknowledge that while new diagnostic tools for social cognition are needed, and no official gold standard exists, the Ekman 60 faces test is currently widely used for assessing social cognitive functioning.[34] Furthermore, due to loss of insight in FTD, we depended on informant-based questionaries to assess music appreciation phenotypes. Finally, we used a novel music emotion recognition test, which is not validated, and needs future replication to also take into account the validity and variability of responses in patients with FTD and to compare them with other music emotion recognition tests. However, the music emotion recognition test was also a strong aspect of the study since we used a live music instrument to ensure a more naturalistic stimulus. Another strength was that we included a wide range of musical changes in our questionnaire, which resulted in a better understanding of the complete spectrum of musical changes in our FTD patient population.

5 | CONCLUSION

Changes in music appreciation are highly prevalent in FTD patients, and impaired music emotion recognition was observed in bvFTD. These results are indicative that atrophy patterns in FTD result in altered music behavior and musicality. Future studies should further investigate diagnostic value of changes in music appreciation and study their relationship multiple components of social cognition.

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CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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