Two mental disorders as disease models of language: linguistic (dis)organisation in schizophrenia and Huntington's disease

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Dedication

To the family I chose

Cada nuevo amigo que ganamos en la carrera de la vida

nos perfecciona y enriquece,

más aún que por lo que de él mismo nos da,

por lo que de nosotros mismos nos descubre.

Miguel de Unamuno, *El secreto de la vida* (1906)

In appreciation

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Summary

Language is a core aspect of human cognition and richly interacts with aspects of nonverbal cognition, as evidence from normal development (Perszyk & Waxman, 2018), abnormal development (Schroeder et al., 2020), and aphasia following brain damage (Fonseca et al., 2019; González et al., 2020) has come to suggest. Yet, how cognitive dysfunction relates to language dysfunction in adult-onset mental disorders still remains an open area of research. Profiling language disturbances in this clinical context could be practically significant in helping to discover representative biomarkers of disease progression and early detection, apart from contributing theoretical insights into the relation between language and cognition. In line with this, the main objective of this thesis is to profile deviant linguistic patterns in people with Huntington's disease (HD) and Schizophrenia (SZ), both of which have only rarely been analysed from a linguistic point of view.

Five linguistic studies were conducted. Study 1 (chapter 3) develops the linguistic profile of HD through the analysis of spontaneous speech samples from 20 individuals compared to 20 neurotypical subjects (NTs). Its aims were to replicate and expand a previous study in a different sample (Hinzen et al., 2018), using a total of 56 variables related to grammatical organisation. The second and third studies are two experimental neuropsychological studies, one using a grammaticality judgment task (Study 2, chapter 4), the other a sentence-picture matching task (Study 3, chapter 5). For these two studies, a new sample of 31 NTs and 31 patients with HD classified into pre-symptomatic, early and advanced stages was collected. Motivated by results from the general linguistic profiling of the HD population in the previous study, the focus here was experimental and the purpose was to specifically target the processing of illicit syntactic movement and the processing of *Binding Theory* principles respectively (Chomsky, 1984).

Studies 4 and 5 focus on two unique corpora of speech in SZ. Study 4 (chapter 6) focuses on a rare corpus of spontaneous speech from 38 patients with an unusually high severe form of formal thought disorder (FDT), a core symptom of SZ according to the DSM-5. This corpus was collected and first analysed in Moya (1989). Our study captured patterns of linguistic disintegration across different linguistic strata (referential anomalies, argument structure, lexicon and morphosyntax). Study 5 (chapter 7) is based on a corpus of hallucinated voice talk in patients with a high symptom load of auditory

verbal hallucinations (AVHs). A linguistic profile of hallucinated voice talk based on literal transcriptions was built, through the analysis of transcripts from 19 patients with highly frequent voice talk.

Overall, results from both HD and SZ populations provided evidence for distinctive and specific linguistic effects, which are not easily interpreted as secondary to primary motor dysfunction (in the case of HD) or to intellectual disability or nonverbal neurocognitive impairments (in the case of SZ). Specifically, pre-symptomatic and symptomatic HD patients exhibited distinct but complementary language patterns in certain grammatical domains: in the fluency domain, pre-symptomatic patients manifested patterns marked by prolongations, fill pauses, and repetitions, while symptomatic patients were prone to use more empty pauses, truncations and reformulations. In the domain of sentence connectivity, their speech was generally characterised by poor grammatical connections, since the use of parataxis and coordination was very common. The reduction of subordination could be interpreted as a weakness in building syntactic hierarchy. Following this line of thought, the evidence collected in Study 2 further experimentally confirmed a loss of cognitive control over the structural hierarchy as built through linguistic movement of syntactic constituents. Study 3 expanded this result to for syntactic principles involved in licensing (co-) reference. The latter study specifically demonstrates difficulties in processing syntactic locality constraints in the HD group, as captured by traditional principles of the *Binding Theory* (Chomsky, 1984).

In the case of SZ, Study 4 captured patterns of linguistic disintegration comparatively across hierarchical layers of linguistic organization in patients with FTD. In terms of broad linguistic domains, it turned out that even in FTD at this severe end, the morphosyntax and the lexicon were relatively little affected, and much less so proportionally when comparing it with the total number of referential errors, while argument structure was placed in the middle. In turn, the linguistic analysis of hallucinated voice talk (Study 5) revealed a strong dominance of parataxis (isolated clauses without grammatical connection), use of non-anaphoric noun phrases (without connection with previous units), and the relative absence of the first person, grammatical errors and semantic errors.

Overall, these results show the feasibility and richness of linguistic profiling outside of neurological disorders said to be language-specific (i.e. aphasia). We regard such profiles as necessary new baselines for integration into neurocognitive models of these diseases; and possibly as informing the development of clinical tools for assessing, monitoring, and detecting cognitive changes and related symptoms. They provide a new dimension for neuropsychological profiling as well, where current test batteries may not capture the relevant linguistic phenomena, thereby adding an extra layer of relevant data. In addition, the richer linguistic disorders turn out to be in mental disorders, the more they motivate new theoretical models that rethink the relationship between cognition and language and link them in systematic ways.

Resum

El llenguatge és un aspecte central de la cognició humana i interactua amb aspectes de la cognició no verbal, com evidencien el desenvolupament normal (Perszyk & Waxman, 2018), el desenvolupament anormal (Schroeder et al., 2020), i l'afàsia causada per danys cerebrals (Fonseca et al., 2019; González et al., 2020). No obstant això, la relació entre la disfunció cognitiva i la disfunció lingüística en els trastorns mentals encara està per descobrir. La identificació i la investigació de les pertorbacions lingüístiques en un context clínic pot ajudar a descobrir biomarcadors representatius de la progressió de certes malalties, desenvolupar perfils lingüístics específics per a la detecció precoç i contribuir a l'estudi teòric de la relació entre cognició i llenguatge. Seguint aquesta línia de pensament, l'objectiu principal d'aquesta tesi és detectar patrons lingüístics anormals en pacients amb la malaltia de Huntington (HD) i l'esquizofrènia (SZ). Des d'un punt de vista lingüístic, les anomalies del llenguatge en ambdues malalties han estat analitzades de forma superficial.

Seguint aquest objectiu, es van dur a terme cinc experiments lingüístics. L'Estudi 1 (capítol 3) va desenvolupar el perfil lingüístic de la HD a través de l'anàlisi de mostres de parla espontània de 20 individus. En aquesta investigació vam replicar i expandir un estudi anterior en una mostra diferent (Hinzen et al., 2018), utilitzant un total de 56 variables relacionades amb l'organització gramatical. La segona i la tercera investigació són dos estudis neuropsicològics experimentals on es van dur a terme una tasca de judici gramatical (Estudi 2, capítol 4) i una tasca de concordança d'imatges i oracions (Estudi 3, capítol 5). En aquestes investigacions es van recopilar les dades de 31 subjectes neurotípics (NTs) i 31 pacients classificats en etapes presimptomàtiques, inicials i avançades. Atès que el perfil lingüístic general de la població HD es va extreure en l'estudi anterior, l'objectiu aquí era disseccionar el processament del moviment sintàctic il·lícit i el processament de les restriccions sintàctiques de (co-) referència, capturades pels principis tradicionals de la *Binding Theory* (Chomski, 1984).

Pel que fa al perfil lingüístic de la població amb la SZ, els estudis 4 i 5 es van centrar en dos corpus únics de dades. l'Estudi 4 (capítol 6) es va centrar en un corpus de discurs espontani de 38 pacients amb una forma inusualment alta de trastorn del pensament formal (FTD), un símptoma central de la SZ segons el DSM-5. Aquest corpus va ser recollit i analitzat per primera vegada a Moya (1989). L'estudi 4 capturava patrons de desintegració lingüística, fent ús d'un conjunt de variables relacionades amb

l'organització lingüística (anomalies referencials, estructura d'arguments, lèxic i morfosintaxi). A l'estudi 5 (capítol 7) es va confeccionar un corpus de parla de veu al·lucinada en pacients amb al·lucinacions verbals auditives (AVHs). Es va construir un perfil lingüístic per a l'AVHs mitjançant l'anàlisi de transcripcions de 19 pacients.

Els resultats mostren de forma inequívoca l'existència d'efectes lingüístics distintius i específics en les dues malalties, que no es poden explicar per una disfunció motora primària (en el cas de HD) o per una discapacitat intel·lectual prèvia o deficiències neurocognitives no verbals (en el cas de SZ). En la població amb HD, els pacients pre-simptomàtics i simptomàtics exhibien diferents patrons lingüístics, però complementaris en certs dominis gramaticals: en el domini de fluència, els pacients pre-simptomàtics manifestaven patrons marcats per les prolongacions, pauses plenes i repeticions, mentre que els pacients simptomàtics eren propensos a utilitzar més pauses buides, truncacions i reformulacions. En el domini de la connectivitat oracional, el seu discurs es caracteritzava generalment per connexions gramaticals pobres, ja que l'ús de la parataxis i la coordinació era molt comú. La reducció de l'ús de la subordinació es podria interpretar com una debilitació de la jerarquia sintàctica. Seguint aquesta línia de pensament, a l'Estudi 2 es va descobrir una pèrdua de control cognitiu sobre la jerarquia estructural, construïda a través del moviment lingüístic dels constituents sintàctics. L'Estudi 3 va confirmar aquesta tendència: es va descobrir que els pacients amb HD presentaven una reducció de la sensibilitat als principis sintàctics de construcció d'estructures. Els resultats van mostrar dificultats en el processament de les restriccions sintàctiques de la localitat en el grup HD, seguint els principis establerts en la *Binding Theory* (Chomski, 1984).

En el cas de SZ, l'Estudi 4 va capturar patrons de desintegració lingüística a través de les capes jeràrquiques d'organització lingüística en pacients amb FTD. El resultats mostren que, fins i tot en el cas extrem del FTD, la morfosintaxi i el lèxic es trobaven relativament preservats en comparació al nombre total d'errors referencials. D'altra banda, l'estudi 5 va confeccionar el perfil lingüístic de les AVHs, caracteritzat pel domini de la parataxis (clàusules aïllades sense connexió gramatical), l'ús de frases nominals no anafòriques (sense connexió amb unitats anteriors) i l'absència relativa de la primera persona, d'errors gramaticals i d'errors semàntics.

En general, aquests resultats mostren la viabilitat i la riquesa de l'elaboració de perfils lingüístics fora dels trastorns neurològics que tradicionalment s'han investigat des d'un punt de vista lingüístic, com és el cas de l'afàsia. Considerem que aquests perfils són

rellevants per a l'anàlisi tant de la SZ com de la HD i que s'haurien d'integrar dins dels models neurocognitius d'ambdues malalties. Permetrien desenvolupar, així, eines clíniques més precises capaces de detectar, avaluar i rastrejar canvis cognitius i símptomes relacionats. També proporcionarien una nova dimensió per a l'elaboració de perfils neuropsicològics més complets, ja que les bateries clíniques actuals no poden capturar fenòmens lingüístics tan específics. A més, l'aparició constant de pertorbacions lingüístiques dins dels trastorns mentals motiva de forma directa la creació de nous models teòrics que qûestionin la relació entre la cognició i el llenguatge i que els vinculin de manera sistemàtica.

Resumen

El lenguaje es un aspecto central de la cognición humana e interactúa con aspectos de la cognición no verbal, como evidencian el desarrollo normal (Perszyk &Waxman, 2018), el desarrollo anormal (Schroeder et al., 2020), y la afasia causada por daños cerebrales (Fonseca et al., 2019; González et al., 2020). Sin embargo, la relación entre la disfunción cognitiva y la disfunción lingüística en los trastornos mentales todavía está para descubrir. La identificación y la investigación de las perturbaciones lingüísticas en un contexto clínico puede ayudar a descubrir biomarcadores representativos de la progresión de ciertas enfermedades, desarrollar perfiles lingüísticos específicos para la detección temprana y contribuir a la investigación de la relación entre cognición y lenguaje. Siguiendo esta línea de pensamiento, el objetivo principal de esta tesis es detectar patrones lingüísticos anormales en pacientes con la enfermedad de Huntington (HD) y la esquizofrenia (SZ). Desde un punto de vista lingüístico, las anomalías del lenguaje en ambas enfermedades han sido analizadas de forma superficial.

Siguiendo este objetivo, se llevaron a cabo cinco experimentos lingüísticos. El Estudio 1 (capítulo 3) desarrolló el perfil lingüístico de la HD a través del análisis de muestras de conversación espontánea de 20 individuos. En esta investigación replicamos y expandimos un estudio anterior en una muestra poblacional distinta (Hinzen et al., 2018), utilizando un total de 56 variables relacionadas con la organización gramatical. La segunda y la tercera investigación son dos estudios neuropsicológicos experimentales donde se llevaron a cabo una tarea de juicio gramatical (Estudio 2, capítulo 4) y una tarea de concordancia de imágenes y oraciones (Estudio 3, capítulo 5). En estas investigaciones se recopilaron los datos de 31 sujetos neurotípicos (NTs) y 31 pacientes clasificados en etapas presintomáticas, iniciales y avances. Dado que el perfil lingüístico general de la población HD se extrajo en el estudio anterior, el objetivo aquí era diseccionar el procesamiento del movimiento sintáctico ilícito y el procesamiento de las restricciones sintácticas de (co-) referencia, capturadas por los principios tradicionales de la *Binding Theory* (Chomski, 1984).

En cuanto al perfil lingüístico de la población con la SZ, los estudios 4 y 5 se centraron en dos corpus únicos de datos. El Estudio 4 (capítulo 6) se centró en un corpus de discurso espontáneo de 38 pacientes con una forma inusualmente alta de trastorno del pensamiento formal (FDT), un síntoma central en la SZ según el DSM-5. Este corpus fue recogido y analizado por primera vez en Moya (1989). Este estudio buscaba capturar

patrones de desintegración lingüística empleando un conjunto de variables relacionadas con la organización lingüística (anomalías referenciales, estructura de argumentos, léxico y morfosintaxis). En el Estudio 5 (capítulo 7), por otro lado, se confeccionó un corpus de habla de voz alucinada en pacientes con alucinaciones verbales auditivas (AVHs). Se construyó un perfil lingüístico para el AVHs tras el análisis de las transcripciones de 19 pacientes.

Los resultados muestran de forma inequívoca la existencia de efectos lingüísticos distintivos y específicos en las dos enfermedades, que no se pueden explicar por una disfunción motora primaria (en el caso de HD) o por una discapacidad intelectual previa o deficiencias neurocognitivas no verbales (en el caso de SZ). En la población con HD, los pacientes pre-sintomáticos y sintomáticos exhibían diferentes patrones lingüísticos, pero complementarios en ciertos dominios gramaticales: en el dominio de fluencia, los pacientes pre-sintomáticos manifestaban patrones marcados por las prolongaciones, pausas llenas y repeticiones, mientras que los pacientes sintomáticos eran propensos a utilizar más pausas vacías, truncaciones y reformulaciones. En el dominio de la conectividad oracional, su discurso se caracterizaba generalmente por conexiones gramaticales pobres, puesto que el uso de la parataxis y la coordinación era muy común. La reducción del uso de la subordinación se podría interpretar como una debilitación de la jerarquía sintáctica. Siguiendo esta línea de pensamiento, en el Estudio 2 se descubrió una pérdida de control cognitivo sobre la jerarquía estructural, construida a través del movimiento lingüístico de los constituyentes sintácticos. El Estudio 3 confirmó esta tendencia: se descubrió que los pacientes con HD presentaban una reducción de la sensibilidad a los principios sintácticos de construcción de estructuras. Los resultados mostraron dificultades en el procesamiento de las restricciones sintácticas de la localidad en el grupo de HD, siguiendo los principios establecidos en la *Binding Theory* (Chomski, 1984).

En el caso de la SZ, el estudio 4 capturó patrones de desintegración lingüística a través de las capas jerárquicas de organización lingüística en pacientes con FTD. Los resultados mostraron que, incluso en el caso extremo del FTD severo, la morfosintaxis y el léxico se encontraban relativamente preservados en comparación al número total de errores referenciales. Por otro lado, el estudio 5 confeccionó el perfil lingüístico de las AVHs, caracterizado principalmente por el dominio de la parataxis (cláusulas aisladas sin conexión gramatical), el uso de frases nominales no anafóricas (sin conexión con

unidades anteriores) y la ausencia relativa de la primera persona, de errores gramaticales y de errores semánticos.

En general, estos resultados muestran la viabilidad y la riqueza de la elaboración de perfiles lingüísticos fuera de los trastornos neurológicos que tradicionalmente se han investigado desde un punto de vista lingüístico, como es el caso de la afasia. Consideramos que estos perfiles son relevantes para el análisis tanto de la SZ como de la HD y que se tendrían que integrar dentro de los modelos neurocognitivos de ambas enfermedades. Permitirían desarrollar, así, herramientas clínicas más precisas capaces de detectar, evaluar y rastrear cambios cognitivos y síntomas relacionados. También proporcionarían una nueva dimensión para la elaboración de perfiles neuropsicológicos más completos, puesto que las baterías clínicas actuales no permiten capturar fenómenos lingüísticos tan específicos. Además, la aparición constante de perturbaciones lingüísticas dentro de los trastornos mentales motiva de forma directa la creación de nuevos modelos teóricos que cuestionen la relación entre la cognición y el lenguaje y que los vinculen de manera sistemática.

Table of Contents

Key acronyms

AVH Auditory verbal hallucination

BDAE Boston Diagnostic Aphasia Examination test

CL Criterion location (CL),

DCL Diagnostic Confidence Level

fMRI Functional MRI

FTD (+/-) Formal thought disorder

HD Huntington's Disease

HTT Huntingtin gene

ICC Intraclass Correlation Coefficient

INT Interviewer

IQ Intelligence Quotient

MMSE Mini-mental State Examination

NP Noun phrase

NT Neurotypical population

P Person

PAT Patient

PSYRATS Psychotic Symptom Rating Scales

SZ Schizophrenia

TAP Word Accentuation Test, Spanish version

TFC Total Functional Capacity scale

TMT Trail Making Test

ToM Theory of mind

UHDRS Unified HD Rating Scale

UPF Universitat Pompeu Fabra

VP Verb phrase

WAYS Wechsler Adult Intelligence Scale

List of original publications

Chapter 3

Tovar, A., Soler, A. G., Ruiz-Idiago, J., Viladrich, C. M., Pomarol-Clotet, E., Rosselló, J., & Hinzen, W. (2020). Language disintegration in spontaneous speech in Huntington's disease: a more fine-grained analysis. *Journal of Communication Disorders*, *83*, 105970.

http://dx.doi.org/10.1016/j.jcomdis.2019.105970

Chapter 4

Tovar, A., Perry, S., Ruiz-Idiago, J., Viladrich, C. M., Muñoz, J. E., Painous, C., Santacruz, P., & Hinzen, W. Detection of illicit phrasal movement in Huntington's disease (under review)

Chapter 5

Tovar, A., Perry, S., Ruiz-Idiago, J., Viladrich, C. M., Muñoz, J. E., Painous, C., Santacruz, P., & Hinzen, W. Understanding of referential dependencies in Huntington's disease (to be submitted in September 2022)

Chapter 6

Torres, A. T., Nieto, W. S. S., Soler, A. G., Matamalas, C. M., & Hinzen, W. (2019). Language disintegration under conditions of severe formal thought disorder. *Glossa: A Journal of General Linguistics*, *4*(1).

http://dx.doi.org/10.5334/gjgl.720

Chapter 7

Tovar, A., Fuentes-Claramonte, P., Soler-Vidal, J., Ramiro-Sousa, N., Rodriguez-Martinez, A., Sarri-Closa, C., & Hinzen, W. (2019). The linguistic signature of hallucinated voice talk in schizophrenia. *Schizophrenia research*, *206*, 111-117.

http://dx.doi.org/10.1016/j.schres.2018.12.004

1. INTRODUCTION

1.1 Personal motivations

As a Hispanic Philology student, my take on language used to focus on how combinatorial rules constrain and manipulate linguistic units, while observing how speakers perceive these supposedly innate rules. From the beginning, I have been interested in the human component of language capacity, and discovering the GracLab group research (UPF) made me realise that it was possible to study the relationship between language and cognition from a more naturalistic point of view: by analysing language deterioration in individuals suffering from certain pathologies. This involves a close working relationship with the clinical team, patients and families, and allows us to observe the real impact of deterioration affecting language on people's daily lives. At a theoretical level, I was drawn to the idea of how the progression of certain neuropsychological disorders makes it possible to trace cognitive changes through language impairment patterns and thus clarify the relationship between cognition and language.

It is in this particular context that my thesis has developed. At the beginning of my master's course, I joined an ongoing Huntington's disease (HD) project. Following this research and previous work done in the GracLab (Hinzen et al., 2018), I continued with the linguistic analysis of this population, which was made feasible through our collaborators at Hospital Mare de Déu de la Mercè and Hospital Clínic de Barcelona. Without the collaboration of the formidable clinical staff at these institutions, it would have been impossible to carry out such a study. In addition, being able to have access to and include the vast amount of neuropsychological data collected in the ENROLL-HD study (CHDI Foundation, Inc.), has been a crucial reason for continuing my research in this direction.

Along the way, an opportunity arose for me to work on two independent projects on schizophrenia (SZ), in collaboration with the Universitat de Barcelona and the Fidmag neuroimaging unit at Benito Menni Hospital. At this point, a decision had to be made and I chose to continue with those investigations. Why? Because linguistic and thought disturbances have a high relevance in the clinical picture of SZ, and it can shed some light on the relationship between language and thought.

Due to the arrival of Covid and various sick leaves, the original thesis plan had to be modified. However, the UPF allowed the option of a thesis by compendium of papers, and the result is a thesis much more heterogeneous than initially planned.

1.2 Organisation of this thesis

The remainder of this thesis is structured as follows. After stating the aims and rationale of the thesis in section 1.3, the relevant bibliography is reviewed as research contextualisation in Chapter 2 (Background literature). In section 2.1, the relevance of clinical linguistics as a discipline is described, and the significant role of aphasia so far. Section 2.2 focuses on HD, more specifically on the clinical symptoms associated with this disorder (section 2.2.1) and the linguistic profile in HD (section 2.2.2) across different linguistic domains. Section 2.3 pursues the same aims for SZ.

Chapters 3 to 7 include the 5 papers that constitute this thesis by compendium of publications. The original format and layout of the papers have been respected.

In Chapter 8 (General discussion and conclusion) I close the thesis with a general summary of the presented studies (section 8.1), an overall discussion of the results (section 8.2) and the main implications of this research (section 8.3). This chapter closes with section 8.4, in which several future lines of investigation are described.

1.3 Thesis objectives

The relation between language and cognition has been a fundamental cornerstone of linguistic theory, though linguistics today still remains focused on language at the expense of cognition, reflecting a crucial dichotomy retained in current thinking and academic practice. The starting point of this thesis was the idea that so-called ‘cognitive’ or ‘mental’ (as opposed to ‘linguistic’) disorders also mask linguistic diversity, which plays a crucial role both for illuminating the clinical picture of these disorders and for discussions in theoretical linguistics. Based on this, the basic and overarching objective of this thesis is to illustrate this role for one neurological and one psychiatric disorder, where linguistic research is still in its infancy: HD and SZ, respectively. My goal was to enhance current knowledge both on the nature and the specificity of the forms of linguistic diversity seen in these disorders. Next, we present the specific objectives which have guided each of the five studies that comprise this project.

Objective of Study nº 1: Creating a specific linguistic profile for spontaneous conversation in HD.

In the case of this disease, Hinzen et al. (2018) had sought to construct a first comprehensive profile based on the production of narrative discourse (story re-telling) within a given group of individuals. The aim here is to replicate this study, but in a larger sample, and in a more natural linguistic environment, intending to simulate the normal social use of language: spontaneous conversation.

Objective of Study nº 2: Analysing illicit syntactic movements processing in the HD population.

Previous studies showed that HD population exhibited difficulties in processing non-canonical sentences, such as passive constructions (Teichmann et al., 2015; Szalisznyo et al., 2017), and a reduction of the syntactic complexity (Hinzen et al., 2017; Tovar et al., 2020). These findings together motivated Study 2. Through a grammaticality judgment task, the sensitivity to locality constraints on syntactic movements in the HD population is tested.

Objective of Study nº 3: Analysing referential dependencies understanding in the HD population.

As observed in previous investigations, the subcortical damage in HD has a direct impact on reference (Hinzen et al. 2018, Tovar et al. 2020). Specifically, difficulties were detected in applying the syntactic rules governing co-referentiality (Sambin et al., 2012). Through a sentence-picture matching task, the purpose of this experimental study is to target the sensitivity to locality constraints in HD, testing the processing of the Binding Theory (BT) principles.

Objective of Study nº 4: Capturing referential abnormalities in patients with SZ and FTD.

Recent studies led to consider several grammatical deficits as part of language impairment in FTD, a core symptom of SZ according to the DSM-5. This study aims to captured patterns of linguistic disintegration across different linguistic strata (referential anomalies, argument structure, lexicon and morphosyntax). Following the work of Sevilla et al. (2018) and Cokal et al. (2018, 2022), the exploration of the referential anomalies is based on an exhaustive analysis of the NPs.

Objective of Study nº 5: Creating a specific linguistic profile for AVHs in SZ.

Very few studies have investigated the formal linguistic aspects of AVHs, one of the central criterion symptoms of SZ. The objective of this research is to determine if there is a distinctive linguistic profile of the internal (hallucinated) language.

2. LITERATURE REVIEW

2.1 Starting points: language and cognition in the clinical context

Language plays a fundamental role in our daily lives: it characterises people culturally as a society and serves as a communication tool. It is also now considered as a cornerstone in the regular cognitive development of any human being, since a bidirectional connection between language and cognition is established from very early stages (Perszyk & Waxman, 2018). More broadly, language has been conceived as a 'mirror of the mind': it allows to structure and share our inner world and enter the minds of other individuals (Chomsky, 1984). It would be unsurprising, then, if diseases of the mind/brain would be reflected in language, giving rise to a rich pattern of clinical linguistic diversity. Studying this diversity may allow to:

(i) Discover biomarkers of disease progression;
(ii) Define characteristic linguistic patterns helping in the early detection of the disease;
(iii) Detect cognitive change concomitant to language impairment;
(iv) Contribute to the analysis and/or refutation of theoretical linguistic concepts.

One of the first attempts to apply language theory in the clinical context was made by Roman Jakobson in 1941. Using a structuralist approach, he analysed the linguistic profile of adults with acquired aphasia. Aphasia is a disorder mainly characterised by language impairment, originating from either cardiovascular accidents (acquired aphasia) or neurodegenerative diseases (progressive primary aphasia). Specifically, acquired aphasia results from damage to left-hemisphere temporal, frontal, and parietal brain regions that are critical for the language system. Non-linguistic cognitive abilities can be affected in this disorder too, but there is an important variability between the different types of aphasia (Marinelli et al., 2017, Fonseca et al., 2018, Gonzalez et al., 2020). Deficits in attention (Murray, 2012), executive control (Meier et al. 2022), complex reasoning (Baldo et al., 2015) and memory (Ghoreyshi et al. 2021) have been consistently documented. These evidences directly challenge the language-cognition dichotomy that has been perpetuated for years in the field of clinical linguistics (even for aphasia). I will return to this issue later.

Although one of the main research topics within clinical linguistics has been linguistic impairment in aphasia, there are many mental disorders of a different nature that have a direct impact on language. Linguistic richness in the clinical context does not end with aphasia. On a daily basis, clinicians are faced a wide variety of neurodevelopmental disorders, and linguistic analysis can help them to identify and assess changes in the patient's communicative behaviour (Priyadarshi & Mahesh, 2022). Given the multifaceted nature of these disorders, clinicians themselves have highlighted the lack of tools to effectively assess, describe and manage such populations. In this thesis, I focused on two mental disorders that have only rarely been analysed from a linguistic point of view: HD and SZ. In the following sections, their main characteristics are presented.

2.2 Huntington's disease

In 1872, the American medical practitioner George Huntington described, for the first time, the effects of what would later become known as Huntington's disease (HD). Already in these first investigations, choreic movements, dementia and hereditary transmission were indicated as defining features of this disorder:

> 'The name 'chorea' is given to the disease on account of the dancing propensities of those who are affected by it, and it is a very appropriate designation. The disease, as it is commonly seen, is by no means a dangerous or serious affection, however distressing it may be to the one suffering from it, or to his friends. Its most marked and characteristic feature is a clonic spasm affecting the voluntary muscles [...] As the disease progresses the mind becomes more or less impaired, in many amounting to insanity, while in others mind and body gradually fail until death relieves them of their suffering. When either or both the parents have shown manifestations of the disease, one or more of the offsprings invariably suffers from the condition [...]' (Huntington, 1872)

HD is an autosomal dominant genetic neurodegenerative disease with a primary aetiology of striatal pathology (Bano et al., 2011). The Huntingtin gene (HTT) has a unique DNA trinucleotide repeat feature: in the neurotypical population (NT), the repeat length ranges from 10 to 35. A repeat between 36 and 39 causes HD with reduced penetrance (some subjects will develop the disease, others will not). Exceeding 40 repeats causes HD with full penetrance (everyone with this length or more will develop the

disease). The number of repeats in HTT is inversely related to the appearance of HD; that is to say, the higher the number is, the sooner symptoms emerge (Lee et al., 2012).

Neuropathologically, degeneration predominates in the neostriatum (caudate nucleus, putamen, and accumben nucleus), including grey and white matter. This divergent pattern of neurodegeneration explains the diversity of functional alterations in the HD population (Petrasch-Parwez et al., 2022). In Vonsattel et al. (2008) a post-mortem analysis of 1000 HD brains is developed, which has led to discover the stages of striatal atrophy throughout the progression of the disease. According to these authors, the neuropathologic hallmarks of HD are the gradual atrophy of the caudate nucleus, putamen, and external segment of the globus pallidus. With the disease progression, the atrophy gradually involves the cerebral white matter, thalamus, cerebral cortex, and cerebellum. At final stages, HD brains are smaller, and most of the degenerative changes affect the striatum.

HD is a rare disease with a prevalence of approximately 10 to 12 individuals of European descent per 100.000 (Evans et al., 2013). The onset of the disease is considered to occur with the manifestation of significant motor or neurological symptoms and emerge on average around 40 years of age. Nevertheless, cases have also been collected of patients with childhood/juvenile-onset HD, who present diverse and more severe symptoms compared to subjects who develop the disease during adulthood. Apart from the atypical clinical picture, the disease progression is faster and survival is shorter (Bakels et al., 2022). In addition to this, the morphological changes of the brain structure are more severe and are characterised by a reduction in intracranial volume. This pattern of brain modifications could explain the occurrence of hypokinetic motor symptoms in JHD, which are not seen in adult-onset HD (Tereshchenko et al. 2019). At present, there is no cure for HD, and only the symptoms can be treated. Its progression, therefore, is inexorable and usually leads to death within 15 to 20 years.

The discovery of the gene in 1993 led to the possibility of performing genetic tests to determine if a subject is a carrier of HTT. Due to the autosomal dominance of the mutation, a child of a parent with HD has a 50% chance of inheriting the disease. The diagnosis of HD relies on clinical outcomes: it is based on a neurological evaluation with the manifestation of an unequivocal extrapyramidal movement disorder together with a positive genetic test for CAG expansion of HD, or a confirmed family history of HD.

Very few people at risk make the decision to take part in this genetic test, approximately between 5% and 10% (Nopoulos, 2022). The diagnosis is usually confirmed by identifying an increased CAG repeat length in the HTT. Although it is frequently and relatively simple, there might be some cases in which it is difficult to conclude when someone has gone from being an asymptomatic carrier to an early stage of the disease (Stoker, 2022).

2.2.1 Clinical symptomatology

The clinical symptoms of HD are mainly classified into three domains: motor, cognitive, and psychiatric, which are consistent with the pathophysiology of fronto-striatal circuit malfunction.

The movement disorder of HD includes both involuntary movements, such as chorea and dystonia, and disturbances of voluntary movement, characterised by clumsiness, dysarthria, swallowing difficulties, falls, bradykinesia, and rigidity (Rosenblatt, 2022). Within all the psychiatric manifestations of HD, apathy, irritability, and perseveration are the most common ones (De Paepe et al. 2019). In addition to this, subjects with this disorder tend to become disinhibited, impulsive and obsessive (Worrall, 2011).

Regarding the cognitive profile, HD presents a complex spectrum of deficits, including abnormalities in executive functions, working and episodic memory, attention, information processing speed and social cognition (Ho et al., 2003; Papoutsi et al., 2014; Foroud et al., 1995). The profile of deficits differs significantly between manifest and prodromal stages of the disease, not only in quantitative terms but also from a qualitative point of view (Cavallo, 2022). In connection with this, dementia in HD is subcortical in nature. Unlike the classic cortical dementia that appears in other diseases such as Alzheimer's disease, memory loss does not manifest itself in early stages of HD. On the other hand, disorders that characterise cortical processes, such as aphasia and apraxia, are not common in this disease (Nopoulos, 2022).

Following this clinical symptomatology, four stages have been described in HD: the pre-symptomatic stage is characterised by the absence of motor or neurological abnormalities, although cognitive deficits may precede motor symptoms by many years (Stout et al. 2007). The initial stage involves the appearance of involuntary movements,

difficulty in solving simple tasks, irritability, depression, apathy and impulsiveness. In the middle stage of HD, choreic movements are exacerbated, speech and swallowing begin to be affected, and cognition is gradually diminished. There are also signs such as disorientation and short-term memory loss. The late stage presents severe choreic movements and periods of muscular rigidity, swallowing problems are accentuated, weight loss appears and the ability to walk and speak deteriorates considerably, which causes the patient to become more dependent (Camargo-Mendoza et al., 2017, Ho et al., 2011).

2.2.2 Language in patients with HD

Although the motor and cognitive consequences of changes in brain morphology have been widely documented, only a limited but growing literature has examined the integrity of language skills in this population. As previously explained, HD involves primary neuronal death in the striatum that progressively spreads to other cortical areas. The striatum is part of the cortico-subcortical language network, which makes HD become a model to determine how language ability deteriorates under the effect of striatal neurodegeneration. In this section a review of the most relevant studies on linguistic changes in HD will be developed (*see* Gagnon et al., 2018).

2.2.2.1 Language production and comprehension

Comprehensive language assessment batteries, such as the Boston Diagnostic Aphasia Examination or the Aachen Aphasia Test, are one of the most widely used methods when providing a general characterization of language disturbance in individuals with HD in the clinical setting environment. Some of these diagnostic tests have been included in our experimental protocols in order to determine their accuracy within this concrete population.

Oral expression in HD is characterised by sentence construction deficits (Chenery et al, 2002), sentence repetition (Azambuja et al, 2012), decreased sentence length (Gordon & Illes, 1987; Illes, 1989; Murray, 2000; Murray & Lenz, 2001) and syntactic complexity reduction in narrative discourse (Hinzen et al., 2018). Jensen et al. (2006) conducted a picture description experiment and agreed that HD patients produce significantly more grammatical errors and fewer action verbs compared to subjects with nonthalamic subcortical lesions following stroke. On the other hand, HD leads to poorer

narrative writing (Azambuja et al., 2012) and dysgraphic errors are observed, mainly characterised by letter omission (Podoll et al., 1988).

HD also affects individuals' language comprehension regarding words, sentences, commands and general pragmatic information (Podoll et al., 1988; Wallesch & Fehrenbach, 1988; Azambuja et al., 2012; Chenery et al., 2002). In particular, it has been reported that HD subjects are impaired in language processing of sentences with subordinate clauses (García et al., 2018) and non-canonical sentences such as passive constructions (Teichmann et al., 2005).

2.2.2.2 Lexical-semantic deficits

Semantic processing of objects and actions was found to be impaired within the HD population (Smith et al, 1988; Frank et al., 1996; Kargieman et al., 2014). However, it should be noted that research by Frank et al. (1996) and Smith et al. (1988) include participants with mild and moderate dementia, which could explain the presence of semantic impairment. Nonetheless, according to Kargieman et al. (2014), HD participants without dementia also show semantic impairment in both object and action concepts.

Access to this population's lexicon was explored through word generation tasks. Lepron et al. (2009) reported difficulties in HD subjects without dementia in terms of reaction time and error rate, with no significant differences between nouns and verbs. In advanced stages of the disease, naming and lexical retrieval difficulties have also been documented (Gordon & Illes, 1987; Illes, 1989; Caine, et al., 1986). However, other studies reported that HD patients process idioms such as *It is raining cats and dogs* without difficulty (Teichmann et al., 2008). Since idioms are stored in the lexicon as a set, semantic processing is much easier. As suggested by Jacquemot and Bachoud-Levi (2021), the semantic deficit in HD is related to executive control requirements: the striatum has an impact on higher lexical-semantic operations requiring higher control demands.

2.2.2.3 Application of language rules

Another grammar aspect that was investigated within the HD population is the application of language rules. In the field of phonology, a reduction in phonemic discrimination ability was detected (Teichmann et al., 2009). In relation to the

morphological domain, patients have difficulties in applying infrequent linguistic rules, producing errors in verb conjugation (Nemeth et al, 2012; Teichmann et al., 2005; 2008b; Ullman et al, 1997) and in the acceptability judgement of conjugated verbs (Teichmann et al, 2006). As proposed in Nemeth et al. (2012), the striatal neurodegeneration present in the HD population causes a disinhibition of frequent rules, leading to the production of over-regularisation errors. In the syntactic field, problems were detected with regards to the application of syntactic rules governing co-referentiality (Sambin et al., 2012) and the construction of non-canonical sentences in passive voice (Teichmann et al., 2005, 2008a, 2008b).

Finally, Diego-Balaguer et al. (2008) assessed the ability to extract lexical rules in pre-symptomatic and symptomatic participants by means of a simplified artificial grammar. They attributed significant impairment in learning new linguistic rules to the lack of semantic information. Moreover, this impairment correlates significantly with working memory and attentional control, suggesting the involvement of both systems in language acquisition.

2.2.2.4 Language deficits at a prodromal stage

Recent studies showed that cognitive impairment in HD subjects may occur long before it is possible to perform a clinical diagnosis of the disease based on neurological or motor criteria (Camargo-Mendoza et al. 2017). Prodromal changes were also noted in the field of language: production of over-regularisation errors in irregular nouns (Nemeth et al., 2012), deficits in syntactic processing of complex tasks (García et al. 2017) and reduced narrative syntactic complexity and referential disfluencies (Hinzen et al. 2018). Changes were also detected in certain acoustic aspects of speech, such as vocal phonation, speech rate and alterations in acoustic firmness in syllable repetition tasks (Vogel et al., 2012; Rusz et al., 2014; Skodda, et al. 2016).

Overall, the decline of language skills begins at pre-symptomatic stages, before neuropsychological or motor tests are able to detect it. Moreover, according to findings, some authors suggested that, in early HD, language is affected beyond the motor aspects of articulation. This also challenges the traditional conception that there is no primary language deficit in this disease: in other words, this language decline occurs only concomitantly with other neurobiological and neuropsychological changes (Podoll et al., 1988, Murray & Lenz, 2001; Gagnon, et al., 2018). Including pre-symptomatic patients

in research may therefore shed some light on this debate, with the intention of reinforcing the idea that there is a primary language impairment in HD.

2.2.2.5 The role of the striatum in language

HD involves primary neuronal death in the striatum, which progressively spreads to generalised cortical areas. Due to its neurodegenerative pattern, HD has been essential when researching on the striatum role in language (Jacquemot and Bachoud-Levi, 2021). This brain region is part of the cortico-subcortical language network, although its role and degree of specificity in language production and comprehension are still unresolved questions at present.

The striatum may influence language processing in several ways, as it is the main input nucleus to the basal ganglia. Several neuroimaging studies suggest that the striatum plays a key role in linguistic articulation (Wildgruber et al., 2001; Wise et al., 1999), syntactic processing (Moro et al. 2001), lexical processing (Friederici & Kotz, 2003; Kotz et al., 2002), lexical retrieval (Rosen et al., 2000) and handwriting (Siebner et al., 2001). However, the debate focuses on determining how specific the impact of the striatum is within the linguistic system. Generically, it contributes to relevant executive functions in language processing, such as attention, planning and working memory (Dominey & Inui, 2009, Dominey et al., 2009). Following this line of research, Jacquemot and Bachoud-Levi (2021) proposed an anatomo-functional model in which the striatum is a central node of the executive control network and it regulates limited cognitive resources such as verbal working memory and verbal attention. Thus, they combine traditional levels of language processing and relevant language executive functions. According to the authors:

> 'In this model, the striatum is part of a verbal executive network that improves the efficiency and fluidity of language, enabling online processing of language at each level (phonetics, phonology, morphology, syntax, and lexico-semantics) by regulating, monitoring, and controlling the allocations of limited cognitive resources (verbal working memory and verbal attention) for processing linguistic units and rules.'

On the one hand, the novelty of this proposal lies in considering that executive control, working memory and attention are part of the language system through its verbal component, and language processing would lose efficiency without these resources. On

the other hand, unlike cortical models of language, they proposed that the striatum plays a crucial role in the network connecting executive functions with language levels.

In this context, investigating the characteristics of language impairment in patients with striatal neurodegeneration may help clarify the striatum's role in language processing. At the beginning of this section, it was noted that early studies characterised HD as a (mainly) motor disorder accompanied by disturbances in the motor aspects of speech. Given that the striatum regulates several motor functions, HD patients suffer from chorea, psychomotor slowing and articulation problems (Snowden, 2017). In terms of language, dysarthria is often the first reported symptom (Podoll et al., 1988), and some authors do not interpret it as a language deficit *per se*, but as another manifestation of motor dysfunction (Ludlow et al., 1987). However, over the years, a primary linguistic and cognitive impairment has been demonstrated prior to the onset of motor symptomatology (section 2.3.3.5) and affecting various phases of language processing, such as language production and comprehension (section 2.3.2.1), lexical-semantic domain (section 2.3.2.2) or the application of linguistic rules (section 2.3.2.3). Therefore, the striatum appears to be linked to more than one level of language.

2.2.4 Summary: the relevance of HD as a disease model of language

HD is the perfect model of language impairment under the influence of striatal neurodegeneration. Striatal damage has an impact on the application and learning of grammatical rules (Sambin et al., 2012; Teichmann et al, 2005, 2008a, 2008b, De Diego-Balaguer et al., 2008), naming, lexical retrieval, and lexical selection (Gordon & Illes, 1987; Illes, 1989; Caine, et al., 1986, Giavazzi et al., 2018) and syntactic complexity, by decreasing sentence length (Gordon & Illes, 1987; Illes, 1989; Murray, 2000; Murray & Lenz, 2001) and reducing structural complexity (Hinzen et al. 2018).

2.3 Schizophrenia

Paul Eugen Bleuler (1857-1939) first used the term 'schizophrenia' in a lecture in 1908, in order to separate this disorder from other types of dementia. According to this author, the general cognitive manifestations of SZ reflect the disturbance and splitting of the associative processes of the mind, and this ''splitting' of the different psychic functions' is a characteristic feature of SZ (Bleuler, 1911: 8).

SZ is a severe psychotic disorder that affects 1% of the adult population worldwide. Overall, it causes emotional, behavioural, sensory, psychomotor and cognitive disturbances (Tandon et al. 2010). The course of the disease is chronic, with relapses in psychotic episodes. Antipsychotic treatments are the main intervention method for the stabilisation of acute psychotic episodes and prevention of their symptoms (Spina & Zoccali, 2008). From a neural perspective, SZ is a chronic, progressive disease caused by structural brain changes in both white and grey matter, temporal lobe volume reduction and, particularly, abnormalities in the superior temporal gyrus and in the white matter connections of the temporal and frontal lobes, arcuate, uncinate and fornix (Shenton et al., 2001; Kubicki et al., 2005, for a full review of the literature, Reichenberg, 2022).

The complexity of SZ, as in most neuropsychiatric disorders, lies in its heterogeneous symptomatological profile. The primary aetiology of SZ is still unknown and it is characterised as a complex or multifactorial disorder arising from the combination of genetic, environmental, social and psychological factors. The diagnosis of SZ is therefore based on the presence or absence of a set of clinical signs and symptoms, commonly categorised as positive and negative symptoms, as they describe the excess or lack of features of standard functioning in healthy individuals. Under the label of positive symptomatology, hallucinations, delusions and 'positive thought disorder' (disorganised language production) are included. Negative symptoms include lack of motivation, apathy and 'negative thought disorder' (poor language production) *(American Psychiatric Association*; APA, 2013).

The neuropsychological profile of SZ is characterised by specific deficits in memory, learning, executive functions, attention and processing speed, against the background of a generalised cognitive impairment (Reichenberg, 2022). Linguistic dysfunctions have also been observed (DeLisi, 2001; Kuperberg, 2010), affecting expressive abilities (Tan et al., 2014a) and daily functioning and social interactions of the patients (Holshausen et al., 2014; Whitford et al., 2018; Rojas et al., 2021). In general terms, SZ symptomatology, as in most psychotic disorders, indicates a generalised disturbance of the higher cognitive functions of the mind, resulting in abnormal states of consciousness where perceptions and beliefs do not match reality.

On the other hand, premorbid impairments have been detected prior to the onset of psychotic episodes (Olsen & Rosenbaum, 2006; NCCMH, 2014). At this prodromal stage, subjects show poor social development, lower intelligence and academic performance, and poor speech and motor development (Reichenberg et al., 2010; Keefe, 2014; Davidson, 2022). A major goal within the field of SZ research is identifying which symptoms appear at this prodromal stage and how they develop with the progression of the disease, intending to predict the onset of the first psychotic episode and apply early protective treatments (ARMS). As explained in the next section, disturbed language plays a key role in the search for possible biomarkers to improve clinical diagnosis. For instance, it has already been shown that communication and thought impairments are a feature of the premorbid stage of SZ and have enough predictive power to detect psychosis in adolescents identified as 'high risk' (Bearden et al., 2011). More recent research also indicates that reduced syntactic complexity, in terms of total word count, seems to be a relevant feature in the prodromal population (Haas et al. 2020). Other studies suggest that, with current MRI technology, it is possible to detect cortical atrophic changes and language processing abnormalities at an early stage. This way, it is possible to predict later SZ development and anticipate progressive cortical brain change with early pharmacological treatments (DeLisi et al., 2022).

In summary, SZ is a disorder with a heterogeneous symptomatology that alters a wide variety of vital aspects in cognitive and social human development, such as thought, perception and language.

2.3.1 Language disturbance in SZ

As mentioned in the previous section, SZ is a multifaceted disorder. Not all patients show the same symptoms, and such symptoms may vary throughout the different phases of the disorder (Kuperberg, 2010). However, the combination of disorganisation and language impoverishment are crucial aspects of SZ diagnosis even in prodromal stages (Palaniyappan, 2021). As this disease has a direct impact on patients' daily functioning and quality of life, there is an extensive search for potential biomarkers to track the progression of SZ, including markers related to impaired language ability (McKenna & Oh 2005, Kuperberg, 2010). In general terms, language decline in SZ is characterised by:

(i) Poor content (little information, alogia), loss of topic (deviation from the topic), chaining of topics (glossomania) and other types of discursive density decrease (Rezaii et al., 2019).

(ii) Aprosodia, anomalous and monotonous tone. Fluency characterised by frequent and prolonged pauses, with incorrect timing and correlated with other negative symptoms (Barra & Herrera, 2018).

(iii) Decreased verbal production (poor speech). Considering the total word number, it is possible to distinguish between NT, SZ patients on antipsychotic treatment and unmedicated patients. The latter group shows the highest decrease in the number of linguistic units used (De Boer et al., 2020).

(iv) Preserved morphology and syntax. The 'word salad' (extremely incoherent speech) even stands on correct syntactic structures. However, a deterioration in morpho-syntactic comprehension and a reduction in structural complexity was observed (Tavano et al., 2008).

In psychiatry readings, many of the linguistic anomalies of SZ are categorised as part of a generalised 'formal thought disorder' (FTD). This term is currently used descriptively and refers to a variety of linguistic phenomena that generate communicative anomalies. Although not exclusive to SZ, FTD is one of the criterion symptoms of this disease in the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5-TR; APA, 2022). It is detected at the level of language form and characterised by abnormal and disorganised linguistic patterns. Such patterns worsen during episodes of acute psychosis (Harrow et al., 1986).

Although FTD is naturally associated with a language impairment, linguistic abnormalities in SZ have traditionally been conceptualised as a reflection of an underlying disturbance of thought or a general intellectual decline (Oh et al., 2002). However, as suggested by more recent studies (Hinzen & Roselló, 2015), language disturbance could explain thought abnormalities in this disease, on the assumption that thought structures are at least partially dependent on language. Moreover, the three main positive SZ symptoms originate when dealing with language-mediated thought units: disordered linguistic perception (auditory verbal hallucinations, AVHs), disorganised linguistic production (FTD) and production of abnormal language content (delusions).

Following this line, Hinzen & Rosselló (2015) proposed a different treatment for positive symptomatology, as shown in *Figure 1*. According to these authors, human language articulates perception, production and content in a co-dependent triangle. Depending on the affected domain (that is, the corner of the triangle involved), one symptom or another may occur: FTD is primarily a disorder at the level of speech production, AVHs are linked to speech perception, and delusions produce a disorder at a content level.

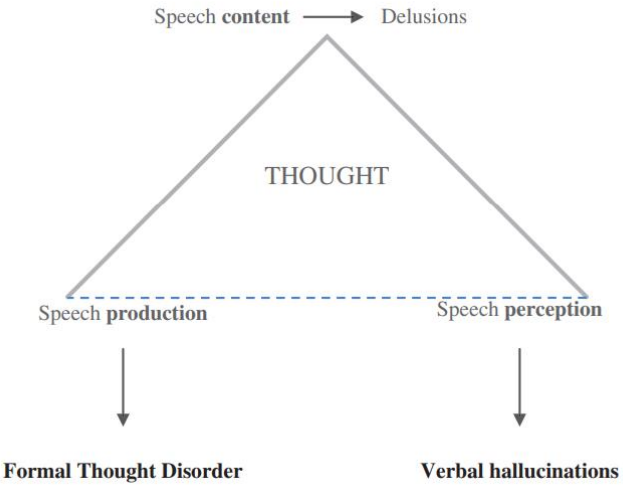


Figure 1. The three positive symptoms organised through linguistic thought (Hinzen & Rosselló, 2015)

Therefore, taking into account the relevance of language within the symptomatology of SZ, and especially in FTD, the following sections present a comprehensive review of the most influential research on language and SZ. Traditionally, literature divides the symptoms of disorganised speech according to the classical grammatical domains: the syntactic level (structure anomalies) and the semantic level (content anomalies).

2.3.1.1 The morpho-syntactic domain

At first glance, the formal structure of schizophrenic speech does not present any particularly evident shortcomings. However, recent studies noted certain linguistic features shaping a specific morphological profile for SZ patients. Language in this population is characterised by being less complex in morphological terms, more associative and more self-focused (Ziv et al., 2022).

In terms of syntax, we must remark that 'word salad' is one of the most abnormal manifestations of linguistic disorganisation in SZ. In general terms, the SZ discourse is characterised by a reduction in structural complexity and an increase in syntactic errors

were documented as part of the linguistic profile of SZ (Tavano et al., 2008). Compared to neurotypical subjects (NTs), patients with chronic SZ produce fewer coordinated and subordinate clauses and show reduced verbal production, as reported by DeLisi (2001). As for the absolute proportions of syntax errors, some studies have detected no differences between SZ patients and the NTs group (Sevilla et al., 2018), while others do (Cokal et al., 2018). Moreover, according to Cokal et al. (2018), syntax anomalies can characterise the linguistic profile in FTD.

SZ is also accompanied by an impairment of syntactic processing. Recent studies show a sentence comprehension decline in traditional clinical tests such as the Test for Reception of Grammar (TROG; Bishop, 1983; in Little et al., 2019). Di San Pietro et al. (2022) found that the SZ group -compared to the NT group- was impaired on comprehension of syntactically complex (but not simple) sentences. Difficulties were also found in processing grammatical violations in syntactically manipulated sentences. In Moro et al. (2015), a binary acceptability judgement task was constructed by using a set of sentences in Italian with different types of structural and semantic violations. Results showed a decreased ability to detect syntactic anomalies in SZ patients compared to NTs. In contrast, no significant differences were found in the detection of semantic violations, suggesting an impairment of syntactic processing that does not affect the ability to derive meaning from different grammatical components and the syntactic structure.

.2.3.1.2 The semantic domain

Whilst the form of schizophrenic speech has been extensively researched, the semantic content raised even greater interest in research on language and thought in SZ. Rochester & Martin (1979) already highlighted semantic impairment as a defining feature of schizophrenic language, characterising it as confused and difficult to understand. Subsequently, other studies defined semantic impairment as a prominent linguistic feature within SZ (Tan et al., 2014b; Brown & Kuperberg, 2015). In particular, an increase in semantic priming was detected, i.e., the use of words with increased semantic similarity that are not contextually appropriate (Almeida & Radanovic, 2021), thus contributing to redundant discourse with reduced information content (Alonso-Sánchez et al., 2022a,b). It was suggested that this anomalous semantic processing does not occur because of anomalies in lexical knowledge, but because SZ has an impact on the organisation and/or access to lexical material (Kuperberg et al., 2009). According to recent studies, moreover,

this unsuccessful lexical activation mechanism especially characterises the FTD population (Pomarol-Clotet et al., 2008).

Generally, semantic deficits have been described as part of the symptoms of discursive disorganisation, incoherence and intelligibility (so-called 'word salad'). Following this speech characterisation in SZ, it is logical to focus on patterns of semantic cohesion in discourse. This tradition began with Rochester & Martin (1979), who focused their research on the linguistic mechanisms that establish discourse cohesion by means of applying the theoretical framework of Halliday & Hassan (1976). This type of cohesion consists of the logical organisation of meaning and reference by using interrelated linguistic structures. The cohesion markers they explored include anaphoric pronominal reference, substitution, ellipsis, conjunction and lexical cohesion. After comparing the discourse of patients with and without FTD they found there is no significant difference in the total number of cohesion discourse markers. However, the discourse of FTD subjects is riddled with anaphoric pronouns and vague demonstratives, thus reducing cohesion at the referential level.

More recent studies applied this line of research to automated language analysis in SZ. Results suggested that these current tools can be used to predict the first psychotic episode in high-risk patients, both in written narrative (Gupta et al., 2018) and oral samples of spontaneous speech (Figueroa-Barra et al. 2022).

In the area of semantic comprehension, deficits have been detected in the understanding of figurative language, especially idioms and metaphors (Titone et al., 2002). As reported by Kuperberg et al. (2009), SZ impacts on the processing of the figurative meaning of words. This may suggest that failures to inhibit literal meaning block access to alternative meanings, sometimes leading to the aforementioned semantic issues.

In short, SZ has an impact on both the form (reduced syntactic structure) and content (semantic anomalies) of language, and can be detected at the level of language production and comprehension. The following section will take a closer look at the analysis of reference in SZ. Such linguistic function does not belong to either of these two traditional grammatical domains, but it is halfway between semantics, syntax and pragmatics.

2.3.1.3 The referential function

The referential function is a particularly important linguistic mechanism of discourse cohesion. It establishes the co-reference relations between two (or more) linguistic units. Referential cohesion allows references to be introduced and taken up again later in a different grammatical form. For instance, one same person can be referred to as a given name *(Laura*), a definite description *(the waitress*) or a pronoun *(she*).

Current language models in SZ noted the problems of referential definition present in this population (Ditman & Kuperberg, 2010; Docherty et al., 2013; Hinzen & Rosselló, 2015; Hinzen, 2017), particularly reflected in ambiguous and vague use of pronouns. Considering that pronouns are 'the most grammaticalized form of reference that exists in language' (Hinzen & Roselló, 2015, p.10), these linguistic elements may indicate the progression of linguistic disturbance in SZ. In traditional papers, referential alterations, vagueness and lack of definiteness are often found (Rochester & Martin, 1979; Docherty et al., 2003, McKenna and Oh, 2005). Recent studies show that, compared to NTs, SZ patients produce proportionally more personal pronouns in written narratives (Buck & Penn., 2015), and more third-person plural than first-person singular pronouns (Fineberg et al., 2015). In her doctoral thesis, de Freitas (2022) investigated the pronominal use of SZ patients speaking colloquial Portuguese from Brazil. Results showed that the SZ group used significantly (and anomalously) more third person null pronouns. The author interpreted these results in conjunction with the decrease in syntactic complexity attested in SZ *(section 2.4.1.1*). She concluded that the increase of third person null pronouns in a partially pro-drop language, where the use of this type of pronominal forms is more restricted, suggests that SZ causes syntax deficits in the functional layers at nominal and sentence level, resulting in impoverished structures, both syntactically (reduced structural complexity) and referentially (use of null pronouns with less referential load).

In the specific case of the FTD population, a recent study on Spanish language (Sevilla et al., 2018) investigated the proportion of anomalous nominal phrases (NPs) in Spanish-speaking patients. The results showed significant differences between NTs, patients with FTD and patients without FTD, in terms of the incorrect use of pronouns and definite NPs. The study by Cokal et al. (2018) on Turkish language obtained similar results: participants with FTD exhibited more referential anomalies than NTs, and produced a lower number of defined NPs. Definite NPs are again the most problematic

referential anomalies. Cokal et al. (2022) partially reinforced these results. According to their study, population with FTD uses more pronouns and fewer co-referent NPs. However, referential anomalies do not distinguish between the -FTD and +FTD groups, although both groups have a higher error rate if compared to NTs. In summary, the schizophrenic speech is characterised by the overuse of bare NPs, null NPs and overt pronouns. Referential definiteness, therefore, is a linguistic identifier of SZ, and FTD in particular.

Within the broad scope of referential function in SZ, a narrower language dimension was also researched: temporal deictic anchoring of space and time. Crow (2010) analysed psychotic episodes as a disruption of the deictic frame. Let us examine the scenario proposed by Zimmerer et al. (2017) to illustrate the importance of this concept in the analysis of language disturbance in SZ:

> 'Considerer a news report about a road traffic incident in which the reporter says: 'A jeep crashed into a barrier.' The meaning is deictically anchored firstly by references to entities, e.g., 'a jeep' or 'a barrier,' and we know from the context that it is one particular jeep and one particular barrier (as opposed to the same phrase in the generic statement 'a jeep is a type of car'). The event ('crashed') is anchored in time as being in the past relative to the speech act. Healthy deictic anchoring further expands to having a sense of who the speaker is and who that speaker is addressing. As a listener, I know that the reporter is not talking about a car crash I had last year and that she is not talking exclusively to me.' (p.2)

However, a subject with an altered deictic anchor might believe that a TV presenter speaks directly to them, and the presenter refers to a specific car accident the subject suffered in the past. These self-referential beliefs are characteristic of the SZ. Following this line, Hinzen et al. (2016) reinterpreted typical SZ delusions (e.g., stating 'I am Jesus Christ') as deictic confusions: the speaker loses the meaning of their deictic location by understanding a third person description (e.g., 'Jesus') as self-referential.

In short, deictic alterations clearly extend beyond personal pronouns, reflecting difficulties in anchoring events in time or space, or even themselves as participants. Therefore, the notion of reference plays a crucial role in understanding language impairment in SZ, although it was often generically labelled as 'cohesive mechanisms' in pioneering studies within this field of research (Rochester & Martin, 1979).

2.3.1.4 Auditory verbal hallucinations

Along with FTD and delusions, auditory verbal hallucinations (AVHs) are the third central criterion symptom of SZ. AVHs consist of hearing voices in the absence of external stimuli, which are perceived as real and different from one's own thoughts. AVHs are a common symptom in several mental and neurological disorders, such as dementia or bipolar disorder. They are also found in non-clinical populations. Nevertheless, it is reported by 3 out of 4 SZ patients (Thomas et al., 2007), meaning that it is a very frequent symptom within the clinical picture of this disease, as already pointed out by Bleuler in his seminal study (1911).

Although hallucinations are not necessarily auditory or verbal, a large-scale study (Baethge et al., 2005) compared the frequency and type of hallucinations among patients with bipolar disorder and other major psychiatric disorders, and results showed that auditory hallucinations outnumber somatic and visual hallucinations in all patients. Additionally, compared to patients diagnosed with bipolar disorder, the SZ group has more severe and verbal hallucinations in most cases.

Therefore, language plays a key role in the type of hallucinations that characterise SZ. Hallucinated speech goes beyond the mere atypical perception of language content: it involves hearing voices directly addressing the receiver or conversations commenting on or criticising their actions and behaviour (McCarthy-Jones, 2012, p.84). According to Hinzen (2017, p. 209), the main characteristic of AVHs is the fact that the listener perceives such thoughts/voices as acts of linguistic communication, which, descriptively speaking, imply a false perception of language: the production of the patient's own mind is perceived as someone else's.

Although language is a crucial feature for the notion of AVH, little is known about its formal linguistic characteristics. Boer et al. (2016) compared voices reported by psychotic and non-psychotic patients and found that syntactic complexity is useful to differentiate the two groups. The linguistic profile of AVHs in patients with psychosis is characterised by a reduction in average utterance length and a decrease in verbal complexity. They also show higher levels of linguistic repetition and more abusive content than the AVHs of non-psychotic individuals. Although this work represents a first

approach to the language characteristics of AVHs, an exhaustive study on the linguistic profile of this phenomenon is still needed.

On the other hand, the neurophysiological mechanisms underlying AVHs created an increased interest in the literature. One of the predominant theoretical approaches maintains that AVHs are perceptual in nature, as they originate from pathological neuronal activity in the auditory cortex. Several neuroimaging studies suggested that AVHs lead to hyperexcitability of the auditory cortex *(see* Kompus et al. 2011). On the other hand, a recent study (Fuentes-Claramonte et al., 2021) found no signs of such increased activity in the auditory cortex during AVH episodes. However, they did report activations of regions related to language and verbal short-term memory. Following this line of research, Fuentes-Claramonte et al. (2022) went a step further and studied the neural correlates of deictic processing in patients with and without AVH. Considering that deixis is a problematic referential domain for SZ patients (*see* section 2.4.1.3), the authors sought to discover whether it was a defining feature of AVH subjects. Results showed that deictic processing abnormalities affect people with SZ in general, with no differences observed between the groups with and without AVHs. In terms of neural activation, it was suggested that the inferior parietal lobule is a key region in the study of referential function.

In summary, there are two lines of research open in the field of AVHs. On the one hand, the creation of an exhaustive linguistic profile including the formal characteristics of the AVHs. If language impairment plays a key role in the detection and progression of SZ, it is logical to assume that it will play an equally important role in one of the most widespread symptoms of this disease. On the other hand, research into the neural circuits activated during psychotic episodes in order to determine the biological basis of AVHs. The two goals are closely related: discovering the main features of linguistic impairment leads to the development of linguistic paradigms that are tested in neuroimaging studies. These studies could help to detect the brain regions involved in language comprehension and production.

2.3.2. Summary: the relevance of SZ as a disease model of language

Unlike in HD, language is part of the SZ clinical picture and has a direct impact on the three main positive symptoms of this disorder. Traditionally, the language disturbance present in SZ has been analysed as an anomaly of thought or a general intellectual decline

(Oh et al., 2002). However, more recent studies (Hinzen & Rosselló, 2016) suggested that linguistic impairment could explain thought disturbances in this disorder, on the assumption that thought is -at least partially- dependent on language (section 2.4.1).

In summary, the linguistic profile of SZ is broadly characterised by disorganised discourse, with reduced syntactic structure (section 2.4.1.1) and anomalous semantic content (section 2.4.1.2). This discursive disorganisation researched through referential problems (Ditman & Kuperberg, 2010; Docherty et al., 2013; Hinzen & Rosselló, 2015; Hinzen, 2017), particularly the vague use of the pronominal system (Sevilla et al. 2018, Cokal et al. 2018, 2022) (section 2.4.1.3). Therefore, SZ is the perfect model for analysing the disintegration of the referential function. The study of the neural circuits activated during AVH has also made it possible to detect the brain regions involved in linguistic processing and to shed light on the neural basis of reference (section 2.4.1.4).

2.4. General summary: mental disorders as models of language disturbance

Although clinical linguistics started researching linguistic impairment in aphasia, the relevance of language within the clinical picture of mental disorders has become more evident over the years. Nonetheless, in both HD and SZ, the impact of language impairment on the neurocognitive profile of patients has been relatively neglected. Clinically, HD has been characterised as a primarily motor disorder, and the existence of a problem in language ability *per se* has been dismissed. On the other hand, language deficit in SZ has been described as a thought distortion, disregarding a potentially close relationship between language and thought, or as the result of a generalised intellectual reduction.

However, as the literature review in this chapter already shows, both diseases manifest specific patterns of language distortion. SZ, on the one hand, exhibits referential abnormalities that are not present in other disorders, such as aphasia. The study of schizophrenic speech, including AVHs and FTD, provided the neural basis of reference in particular, and allowed to uncover which neural regions have the greatest impact on language processing in general. On the other hand, the case of HD is particularly relevant because of its pattern of neurodegeneration. This disorder is thus presented as the ideal model of language disturbance under the effects of striatal neurodegeneration, and the development of a specific linguistic profile for HD helps to elucidate the role of the striatum in language processing.

In short, as the studies described in this introduction show, investigating language disturbance in both HD and SZ allows us to locate anomalies that go unnoticed by traditional clinical batteries. It also allows for the detection of cognitive change concomitant with language disturbance, thus showing the relationship between cognition and language. Finally, by developing specific linguistic paradigms and testing them in neuroimaging studies, it is possible to detect which brain regions play a relevant role in language comprehension and production. Therein lies the power of language as an object of study under the models of mental disorders.

3. STUDY 1: LANGUAGE DISINTEGRATION IN SPONTANEOUS SPEECH IN HUNTINGTON'S DISEASE: A MORE FINE-GRAINED ANALYSIS

Abstract. Huntington's disease (HD) is a neurodegenerative disease causing motor symptoms along with cognitive and affective problems. Recent evidence suggests that HD also affects language across core levels of linguistic organization, including at stages of the disease when standardized neuropsychological test profiles are still normal and motor symptoms do not yet reach clinical thresholds ('pre-manifest HD'). The present study aimed to subject spontaneous speech to a more fine-grained linguistic analysis in a sample of 20 identified HD gene-carriers, 10 with pre-manifest and 10 with early manifest HD. We further explored how language performance related to non-linguistic cognitive impairment, using standardized neuropsychological measures. A distinctive pattern of linguistic impairments marked off participants with both pre-manifest and manifest HD from healthy controls and each other. Fluency patterns in premanifest HD were marked by prolongations, filled pauses, and repetitions, which shifted to a pattern marked by empty (unfilled) pauses, re-phrasings, and truncations in manifest HD. Both HD groups also significantly differed from controls and each other in how they grammatically connected clauses and used noun phrases referentially. Functional deficits in language occurred in pre-manifest HD in the absence of any non-linguistic neuropsychological impairment and did largely not correlate with standardized neuropsychological measures in manifest HD. These results further corroborate that language can act as a fine-grained clinical marker in HD, which can track disease progression from the pre-manifest stage, define critical remediation targets, and inform the role of the basal ganglia in language processing.

Keywords: Huntington's disease, language impairment, grammatical deficits, basal ganglia

1. Introduction

Huntington's disease (HD) is an autosomal dominant genetic neurodegenerative disease that involves neural death particularly in the striatum (caudate and putamen) (Bano, Zanetti, Mende, & Nicotera, 2011) and causes motor impairments (involuntary movements, chorea). Cognitive and psychiatric symptoms accompany the clinically primary motor symptoms. Cognitive impairments include deficits in executive functions, working and episodic memory, processing speed, and social cognition (Ho et al., 2003;

Papoutsi, Labuschagne, Tabrizi, & Stout, 2014; Foroud et al., 1995). In milder forms, these can characterize a prodromal stage of the disease, 15 years or more prior to motor symptoms becoming clinically manifest and crossing diagnostic thresholds (Stout et al., 2011; Paulsen et al. 2014; Bora, Velakoulis, & Walterfang, 2016). As this prodromal period is a critical one for therapies delaying or even preventing symptomatic disease onset, much attention has been devoted to detecting and evaluating the most promising biomarkers that can predict and track disease progression in this phase, including neurocognitive performance in domains such as executive functioning (Paulsen et al., 2014; Wiecki et al., 2016), verbal episodic memory (Solomon et al., 2007), and working memory (Poudel et al., 2015). In the domain of language, too, prodromal changes have been detected, including in the domains of word morphology (regular but not irregular verb and noun inflection: Nemeth et al., 2012), action semantics and sentences with embedded clauses (García et al., 2017; Hinzen et al., 2018). Speech-acoustic aspects such as vowel phonation or speech rate and alterations in steadiness in syllable repetition tasks have also been documented (Vogel, Shirbin, Churchyard, & Stout, 2012; Rusz, Saft, Schlegel, Hoffman, & Skodda, 2014; Skodda, Grönheit, Lukas et al., 2016).

Language impairment is expected from neural atrophy in the basal ganglia, which have been argued to play an important role in non-motor cognitive functions including language (Graybiel, 1995; Ullman et al., 1997; Friederici & Kotz, 2003; Kotz & Schwartze, 2010; Moro et al., 2001). Fronto-striatal circuits may specifically support the building and sequencing of hierarchical structures in language, with phrases embedded in other phrases (Lieberman, 2007), though this process may also depend on more specialized and evolutionary more recent cortical mechanisms (Friederici, 2017). Systematic and detailed behavioural linguistic profiles could inform debate of the role of the basal ganglia in language, yet are still missing at any phase of the disease. To this purpose, language needs to be assessed as a multi-dimensional construct organized at multiple levels (phonology, morphology, syntax, semantics, discourse), which are re-fitted together into an integrated functional whole. The few behavioural linguistic studies of spontaneous connected speech in HD have typically found a pattern of reduced syntactic complexity, with fewer words and syntactic structures formed in short, simple sentence constructions, more paraphasic and grammatical errors, and sentence truncations (Podoll, Caspary, Lange, & Noth, 1988; Murray & Lenz, 2001; Gordon, & Illes, 1987; Illes, 1989; Chenery, Copland, & Murdoch, 2002; Jensen, Chenery Copland, 2006).

These language deficits may form a distinctive signature profile of HD as compared with Parkinson's disease (Murray & Lenz, 2001; Illes, 1989) and people with non-thalamic subcortical lesions (Jensen et al., 2006). Compared to both of these other groups, in particular, HD (at least in early stages) may affect syntactic abilities more than lexical-sematic ones. In later stages of the disease, naming and lexical retrieving difficulties have also been documented in HD (Gordon & Illes, 1987; Illes, 1989; Caine, Bamford, Schiffer, Shoulson, & Levy, 1986), but the origin of these difficulties is unclear and may not relate to semantic memory per se, as opposed to retrieval difficulties and difficulties of visual analysis (Hodges, Salmon, & Butters,1990; 1991). In the case of action words, semantic deficits can characterize prodromal and early symptomatic stages as well (García et al., 2018).

Progress in investigating spontaneous speech production in HD depends on addressing a number of limitations. Thus, sample sizes of the above studies have been small (typically fewer than 12 HD participants), participants with HD at different stages of the disease have often been mixed, and no prodromal cases matched with non-gene carrying neurotypical controls have been included. Moreover, when measuring syntactic complexity, generic measures of complexity (e.g. utterance length or number of embedded clauses, without distinguishing specific forms of embedding, as in Murray & Lenz, 2001; Gordon & Illes, 1987) have typically been used, so that it is unclear which specific aspect of syntactic structuring is compromised. Hinzen et al. (2018) recently set out to address some of these limitations, seeking to profile spontaneous speech production in HD more comprehensively and to identify those aspects of language structure and function that might differentiate the narrative speech of two groups of identified HD gene-carriers, one prodromal and the other in the early stages of the disease. These groups were compared against each other and that of age- and education-matched neurotypical controls. Speech was elicited through a fairytale retelling task and annotated for a large set of 57 fine-grained linguistic variables (e.g. 'multiple functional word repetition', 'truncation within a word with morpheme integrity preserved', 'hanging determiners', etc.). To create a comprehensive linguistic 'map', these individual variables were then grouped into five broad linguistic 'domains', for which composite measures were computed: 1. *Quantitative* (e.g. number of words produced, mean length of utterance), 2. *Fluency* (e.g. repetitions, pauses, truncations), 3. *Clausal Connectivity* (e.g. use of coordinations such as *and* vs. subordinations such as *(said) that* to connect clauses), 4.

Reference (use of noun phrases to pick out story characters and maintain topics), and 5. *Concordance* (e.g., marking of grammatical agreement and other morpho-syntactic aspects). Results revealed that narrative speech in early-manifest HD was different in all of these domains relative to the matched controls. Two domains (Reference and Connectivity) showed impairments in pre-manifest HD relative to controls, at a stage of the disease when standardized neuropsychological test profiles were still normal. Scores in the Quantitative but no other domain significantly correlated with the overall Unified HD Rating Scale (UHDRS) motor scores, with working memory scores (Digit Span Backwards; Wechsler, 1981), and with gray matter volume bilaterally in the dorsal basal ganglia (putamen/pallidum). No other domain than Quantitative showed any significant correlations with measures of neurodegeneration. Fluency and Reference correlated with an executive functioning task (the Trail Making Test; Tombaugh, 2004). The remaining two domains, Connectivity and Concordance, did not correlate with any non-linguistic neuropsychological or volumetric measures.

These findings from spontaneous speech production stand in the context of several studies of linguistic comprehension or perception in controlled experimental settings, which have also shown specific linguistic functions to be affected in HD. Sambin et al. (2012) documented this for aspects of the Binding Theory, i.e. syntactic rules governing co-referentiality between two noun phrases, independently of working memory demands. Teichman et al. (2005) argued for a specific role of the striatum in the application of syntactic and morphological rules, but not lexical knowledge (but see Longworth, Keenan, Barker, Marslen-Wilson, & Tyler, 2008). Teichmann, Dupoux, Kouider, & Bachoud-Lévi (2006) generalized this dissociation to the perception of morphological rules and showed it to be uncorrelated with executive functions. Teichmann, Dupoux, Cesaro, & Bachoud-Lévi (2008) further refined this account through evidence that specific syntactic rules (i.e. syntactic movement), rather than syntactic or combinatorial rules in general, are affected by striatal degeneration; and Teichmann et al. (2008) showed that while syntactic rules are affected over lexical rules, there are impairments in both, traceable to distinct striatal sub-regions and disease stages (see also De Diego-Balaguer et al., 2008).

Together, these findings suggest that language is affected over and above aspects of speech-motor articulation in early HD, and they cast significant doubt on the traditional view that there are no primary language deficits in HD, i.e. these only ‘develop secondary

to other neurobiological and neuropsychological changes' (Podoll et al., 1988; see also Murray & Lenz, 2001; Gagnon, Barrette, J., & Macoir, 2018). Normal language processing requires and integrates domain-general cognitive functions such as working memory or cognitive control (Just & Carpenter, 1992; Caplan & Waters, 1999; Walker, 1996), hence deficits in these are also expected to bear on language function. However, language deficits documented can concern rather specific linguistic variables; moreover, they are seen in early and even pre-manifest gene-carriers without any other neuropsychological impairment, and there is a lack of correlations with neuropsychological measures in some core aspects of linguistic function (e.g. connectivity). This reinforces the idea of a primary language impairment in HD caused by the neurodegeneration involved. They also strengthen the case for language performance and processing as a potential cognitive biomarker (García et al., 2017; Vogel et al., 2012), in addition to its being an important target for remediation and protective measures, given the importance of language in daily social functioning (Klasner & Yorkston, 2001; Hamilton et al., 2012; Hartelius, Jonsson, Rickeberg, & Laakso, 2010).

Several questions, however, arise. First, given the methodology of Hinzen et al. (2018), their results reveal little about differences between groups at the level of the fine-grained, non-composite linguistic variables that were factored into the overall domain-level composite scores. This may also have been the reason that these authors failed to find neural correlates of language dysfunction in any except the Quantitative domain. More fine-grained behavioral profiling of linguistic functions is needed to identify language patterns in HD at different stages and to inform future structural and functional neuroimaging studies. These could then also further address the role of the basal ganglia in language processing (Moro et al., 2001; Friederici & Kotz, 2003; Friederici, Steinhauer, & Frisch, 1999), and the link between motor and language functions in the brain more broadly (Lieberman, 2007). A second question concerns the relation and possible interactions between language and cognition in HD. Aphasia-based models of the interface between these two have often stressed the independence of linguistic from general cognitive functioning (Fedorenko & Varley, 2016). However, there is evidence that cognitive decline in putatively nonverbal tasks in fact systematically accompanies language impairment in aphasia (Fonseca, Ferreiras, & Martins., 2016; Baldo Dronkers, Wilkins et al., 2005). In turn, impairment in non-linguistic cognitive domains can contribute to aphasic language performance (Swinney, Zurif, Prather, & Love,1996;

Wright, Downey, Gravier, Love, & Shapiro, 2007), stressing the interdependence of language and cognition. Linguistic and cognitive development are closely intertwined as well (Arunachalam & Waxman, 2010), and there is strong evidence for correlations between language performance and performance on standardized ToM tasks in particular (De Villiers, 2007; Paynter & Peterson 2010). Like aphasia and development, HD is an important model for studying this interdependence between language and nonverbal cognition further.

To begin addressing these questions, we had three aims. The first aim was to reproduce the pattern of domain-level results from Hinzen et al. (2018) in an independent cohort with speech samples obtained from different tasks and in more quantity. The second aim was to move from the composite measures of that study to a more specific and fine-grained level of linguistic analysis with non-composite variables. The third aim was to cast further light on the relation between linguistic performance and cognitive performance as assessed through standardized neuropsychological tests.

2. Materials and Methods

2.1 Participants

This cohort consisted of 20 participants identified as carrying an abnormal polyglutamine expansion in the N-terminal region of the huntingtin protein (HTT) caused by a mutation in the IT15 gene located in chromosome 4, who were matched to 20 neurotypical controls on age, gender, educational background, and (pre-morbid) IQ. Pre-morbid Intelligence Quotient (IQ) was evaluated by the Word Accentuation Test, Spanish version (TAP, Test de Acentuación de Palabras, Gomar, Ortiz-Gil, McKenna et al., 2011). Ten of the 20 HD gene-carriers (which will be referred to below as the 'pre-manifest' HD-group) presented with a score of less than 4 in the Diagnostic Confidence Level (DCL) of the Unified Huntington's Disease Rating Scale (UHDRS). The remaining 10 gene-carriers were at early stages of the disease (henceforth referred to as 'manifest HD'), identified technically by a score between 7-13 on the Total Functional Capacity Scale. All participants were native Spanish speakers. Table 1 summarizes the demographic, genetic and clinical data from the subjects. All participants received the relevant information about the study and the methods and signed an informed consent to participate in this investigation. This informed consent was approved by the ethics committee of the Universitat de Barcelona and the Hospital Mare de Déu de la Mercè (Germanes Hospitalàries).

Table 1: Demographic, genetic and clinical data

	Pre-manifest (N=10)	**Manifest (N=10)**	**Controls (N=20)**
Gender (M/F)	3/7	2/8	5/15
Age (mean/SD)	38.10 (6.82)	48.70 (10.88)	43.30 (9.72)
IQ (mean/SD)	104.6 (5.72)	102.7 (5.87)	105.65 (5.74)
Education in years (mean/SD)	13.5 (3.62)	11.5 (4.06)	12.2 (3.35)
Number of CAG repeats in the larger HTT allele (mean/SD)	41.90 (3.87)	43.40 (3.13)	-
UHDRS TMS* (mean/SD)	1.20 (1.93)	27.10 (13.20)	-
TFC** (mean/SD)	13 (0)	9.50 (1.34)	-

*UHDRS TMS: Unified Huntington Disease Rating Scale Total Motor Score
**TFC: Total Functional Capacity

2.2 Clinical neuropsychological assessment

All participants with HD were evaluated using the motor and functional sections of the Unified Huntington's Disease Rating Scale (UHDRS; The Huntington Study Group, 1996). Their motor performance was described by the total motor scale score (UHDRS-TMS), which was calculated by adding the scores on each of the 31 items of the motor function section of the UHDRS. Each item is rated on a 0 to 4 points scale with 4 indicating the most severe impairment (range 0-124 points). Functional assessment was made using the Total Functional Capacity scale (TFC). Scores on the TFC represent five stages in the neurodegenerative disease process. Lower scores represent greater functional impairment: stage I represents scores of 13–11; stage II, scores of 10–7; stage III, scores of 6–3; stage IV, scores of 2–1; and stage V, a score of 0. The Mini–Mental State Examination (MMSE; Folstein, Folstein, & McHugh, 1975) was used to assess mental status. This is a 30-point questionnaire extensively used in clinical evaluation to measure cognitive impairment. In addition, a cognitive battery was administered consisting of: the Stroop Test (Golden & Freshwater, 1978) assessing inhibition; the Digit Symbol Substitution Test (DSST; Wechsler, 1981) evaluating alternating attention; the Trail Making Test (Tombaugh, 2004) Part A (Trail A), assessing processing speed and

sustained visual attention; and Part B (Trail B), evaluating cognitive flexibility. In addition, subtests of the Boston Diagnostic Aphasia Examination test (BDAE; Goodglass & Kaplan, 1972, 1983) were administered in order to assess oral and reading comprehension and naming. Participants also completed one verbal fluency task (naming of animals). Controls were tested with the same neuropsychological battery to have cognitive indexes characterizing this specific sample and to compare their results with the performance of HD subjects.

2.3 Procedure

Spontaneous speech samples were firstly obtained based on an open, 15-minute interview, in which a list of general questions was specified to structure the conversation (e.g. *Which is your favourite book? Where did you go on holidays?*). Secondly, participants were presented with two video clips, one non-verbal or wordless (1:58 min.) and the other verbal (2:22 min), and asked to retell their story contents to the experimenter. The wordless video showed Mr. Bean faking an accident of falling from a window in order to receive medical attention and care from an attractive nurse. The verbal video presented a discussion between a mother and her daughter in a Chinese restaurant where the waitress casts a spell, causing the mother and the daughter to exchange their bodies the next day. Participants were informed that their speech would be recorded. Speech samples were transcribed and then analysed utterance by utterance utilizing CLAN (MacWhinney, 2000). The linguistic manual of Hinzen et al. (2018) was used for the linguistic analysis, slightly adapted for the present study; it is added here in Supplementary Materials. Recordings were anonymized.

2.4 Linguistic and statistical analysis

Following the method of Hinzen et al. (2018), a set of 56 individual linguistic variables was chosen for purposes of a comprehensive annotation of spontaneous speech at all levels of linguistic organization, excluding only more peripheral phonetic and articulatory aspects of speech. For analysis purposes, these individual variables were grouped into the same five domains as in Hinzen et al. (2018), capturing different dimensions of linguistic organization. This led to five composite variables, named Fluency, Reference, Connectivity, Concordance and Quantitative. The first four comprise variables capturing different types of errors or anomalies, while the last comprises variables relating to purely quantitative aspects of speech. Specifically, **Fluency** was made up of prolongations and

repetitions, pauses, and truncations, indexed by the syntactic positions in which these appeared (e.g. pauses between clauses or within noun phrases). **Reference** targeted referential problems inside clauses, such as introducing referents in the discourse that a hearer cannot track, e.g. *Pues no sé por qué. Porque el hermano tenía un piso allí, porque él trabajaba allí* (*I do not know why. Because the brother has an apartment there, because he worked there*, where no such brother has previously been mentioned). **Clausal Connectivity** concerned how clauses were grammatically connected with others, e.g. through coordinations with *and* or subordination with *(said) that*… A characteristic example of a problem of Clausal Connectivity is overuse of coordination in narration, as e.g. in *Y que se lo escuchen más, y que le hagan más caso. Y al final después de insistir tanto y llamar bueno y pues acaba accidentado ¿no?* (*And so that they listen to him and notice him. And at the end, after insisting so much and calling, well, and he ends up injured*). **Concordance** targeted agreement (morphosyntax). Within the domain of Concordance, characteristic examples are agreement failures, like for example in the DP *Las hermanos* (*the brothers*), where the determiner is in feminine gender and the noun in masculine. **Quantitative**, finally, comprised purely quantitative features of speech, such as total number of word/utterances or Mean Length of Utterances/Words. A complete list of variables for each domain is provided in Table 2.

Table 2: List of all variables in each linguistic domain

Fluency	Prolongations (Prol)
	Filled pauses (FilP)
	Lexical word repetition (LWR)
	Single functional word repetition (sFWR)
	Multiple functional word repetition (mFWR)
	Partial functional word repetition (pFWR)
	Partial lexical word repetition (pLWR)
	Partial repetition of a CP (XPR:CPR)
	Repetition of a determiner phrase (XPR:DPR)
	Repetition of a prepositional phrase (XPR:PPR)
	Repetition of a verb phrase (XPR:VPR)
	Partial repetition of a phrase (pXPR)
	Pause between determiner and noun phrase (D-NP.P)
	Pause between verb and its complement or another clause (V-TP.P:V-CP.P)
	Pause between clause and tense (C-TP.P)
	Pause between auxiliary verb and main verb (T-VP.P)
	Pause between clauses (CP-CP.P)
	Pause between phrases (XP-YP.P)
	Pause between a preposition and the following phrase (P-XP.P)
	Pause between discourse marker and a clause (DM.P)
	Truncation within a word with morpheme integrity preserved (-W/T)
	Truncation within a word with morpheme integrity violated (*–W/T)

	Truncation of a phrase after the complementizer (CP/T) Truncation of a quantifier phrase (QP/T) Truncation of a determiner phrase (DP/T) Truncation of a prepositional phrase (PP/T) Truncation of a verb phrase (VP/T) Truncation of a phrase after the auxiliary verb or nexus is uttered (TP/T) Pause within words with morpheme integrity preserved (–WP) Rephrasing (Rephrasing)
Reference	Hanging topic (/top) Abnormal topic shift (#top) Vagueness or lack of topic (*0top) Ambivalence (+/–ref) Hanging determiners (/D) Vague referent (VagRef) Definiteness repair (DefRep) Missing determiner (MX:MD) Missing preposition (MX:MP) Failures in temporal reference (*refT) Incorrect self-correction of determiners (*corXP) Number of mental verbs (*v) Paraphasia (Paraphasia)
Connectivity	Missing discourse markers (links) (*0D-link) Incorrect discourse marker (*D-link) Intrusive parenthetical (#X) Coordination wrong (CRD WRONG) Subordination wrong (SUB WRONG) Coordination total (CRD TOTAL) Subordination total (SUB TOTAL) Failures in consecutio temporum (*Tcons.temp)
Concordance	Agreement failure in the auxiliary verb (*AgrX:IAgrT) Agreement failure in the main verb (*AgrX:IAgrV) Government (*GovV) Infelicitous verb (#V)
Quantitative	Utterances (Utterances) Mean length of utterance in morphemes (MLUm) Total words (WORDS)

Statistical analysis proceeded first at a domain level, then at the level of the fine-grained linguistic variables that made up these domains themselves. Before applying any statistical test, values for every participant were normalized by their number of words or utterances. The choice between normalizing by the total number of words or by the total number of utterances was made based on the nature of the variable. Normalization was not applied to variables in the Quantitative domain. In order to create the composite scores for each domain, variables were first scaled by dividing them by their standard deviation, so as to equate the weight of each variable in the composite score. Composite scores were

then obtained by adding the corresponding rescaled variables up. Groups were compared for the composite variables with ANOVA or Kruskal-Wallis tests and the corresponding post hoc tests (Tukey's HSD test and Bonferroni-corrected Dunn test, respectively). ANOVA was used when a normal distribution was present in all groups, as determined by Shapiro-Wilk normality tests. Otherwise, Kruskal-Wallis was applied. Corrections for multiple comparisons were applied to post-hoc group comparisons by means of the Tukey's HSD test itself or with a Bonferroni correction of Dunn test p-values. All p-values of post-hoc tests are reported in their corrected form and can be interpreted with a significance threshold (α) of .05.

Further comparisons were carried out for the individual (non-composite) speech variables. Similarly to the analysis by domain, groups were compared for each variable by means of ANOVA or Kruskal-Wallis tests followed by a post-hoc Tukey's HSD tests or a Dunn test, except in the cases of variables with 50% or more null values (see further below). Corrections for multiple comparisons were again applied only to post-hoc group comparisons. P-values of post-hoc tests are reported already corrected, as described above. Additional corrections by domain were not applied, in order to avoid inflating type II error as well as for comparability with the previous study (Hinzen et al., 2018). We acknowledge the possibility of an increased type I error due to domains being made up of related variables. We provide tables showing Bonferroni-corrected intercorrelations between variables by domain in the Supplementary Materials (S4-S8). Variables with 50% or more null values were dichotomized in terms of absence and presence, and differences between groups were analysed with Bonferroni-corrected Fisher's Exact tests. Fisher's test was preferred over χ^2 tests because expected values per group were small.

Comparisons between groups were also explored for the neuropsychological variables by means of ANOVA or Kruskal-Wallis tests. Next, correlation analyses were run between several variables, including speech domain variables and neuropsychological variables. Since the interest was focused on relations between variables in the participants with HD, the group of controls was left out of the correlational analysis. For linguistic variables, correlations in separate groups (pre-manifest and manifest) were also explored. Pearson's or Spearman's coefficients were computed when linearity and monotonicity allowed this. A false discovery rate (FDR) correction was applied to account for multiple comparisons. FDR is the expected proportion of false discoveries amongst all rejected

null hypotheses. All reported corrected p-values can be interpreted with a significance threshold of .05.

In all analyses carried out, non-parametric equivalents of parametric tests were applied in cases where variables did not meet the normality assumptions as determined by Shapiro-Wilk tests. Effect sizes of significant results are reported as η2 or Cramer's V as appropriate. The significance threshold was set at .05 for all tests.

2.5 Reliability analysis

As blindness to medical diagnosis of the participants could only be incompletely ensured, a subset of the sample was re-rated by two independent raters not involved in the study, to check reliability and replicability of rating for the linguistic analysis and the two narrative tasks. Both raters were trained, but as both were linguists, they were largely familiar with the linguistic notions used. Transcriptions not involved in the reliability assessment were used for training, which was minimal in both cases. The Intraclass Correlation Coefficient (ICC) was used to measure agreement. In the linguistic analysis, on two out of 56 variables, namely 'Factives' and 'pause within words with morpheme integrity preserved', the ICC could not be calculated due to the lack of variability in the ratings or instances of the relevant variable. Low or null variance does not imply low or null agreement; in fact, most of the ratings in the first of these two variables coincided. We therefore decided to omit these two variables in the agreement analysis. The resulting ICC showed agreement to be very high in general (M=0.950, Median=0.984, SD=0.112). The minimum value was 0.276 in the variable 'agreement failure', very far from the second lowest value (0.774, present in three variables). This variable was omitted in subsequent analyses.

3. Results

Results will be presented in three parts: 1. Domain-level analysis, 2. Individual variables analysis, 3. Neuropsychological results and correlations. Only tables with results of post-hoc comparisons are included here; group-level results can be found in the Supplementary Materials. In the tables, we only include variables for which significant p-values were obtained.

3.1 Domain-level analysis

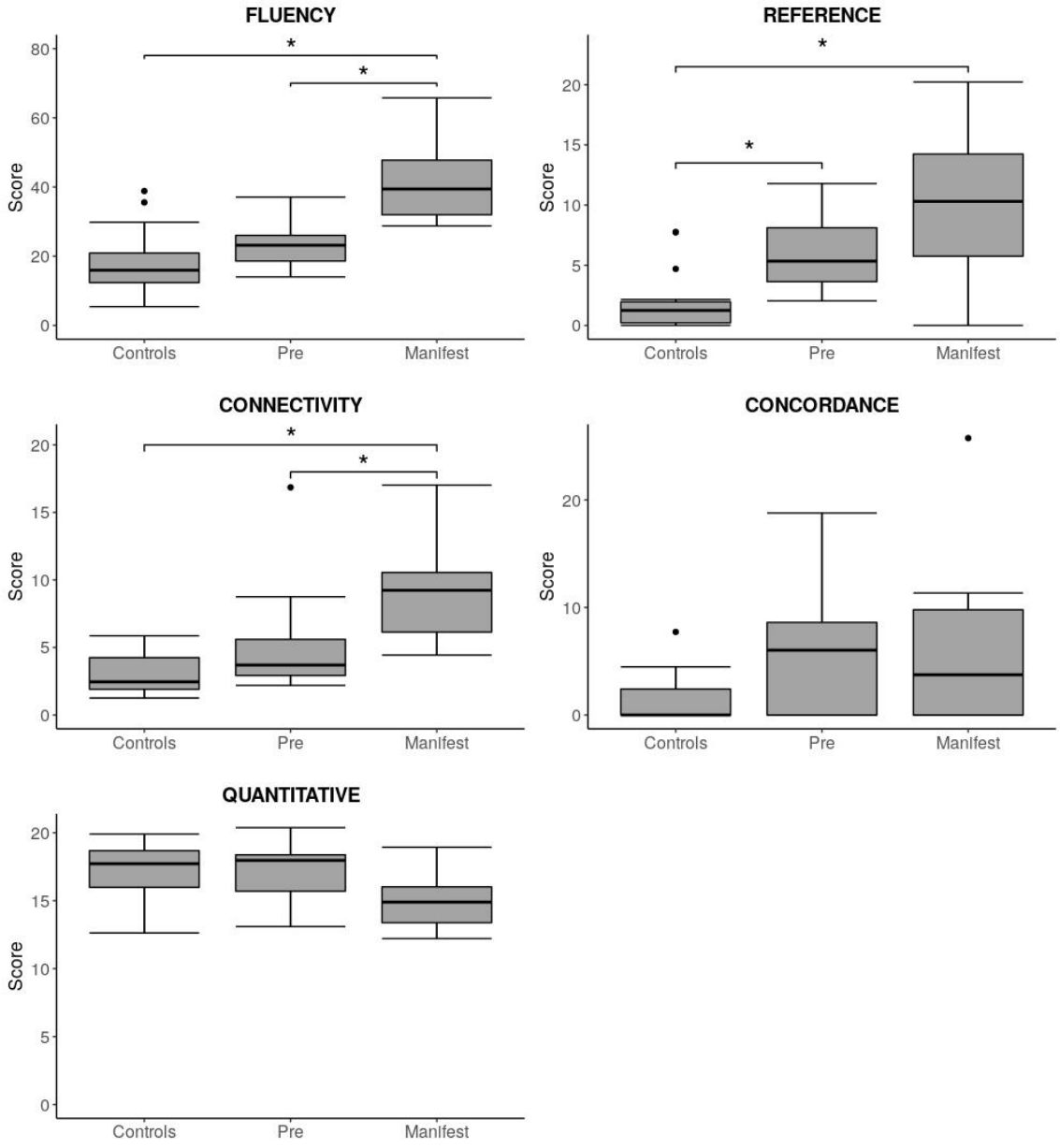
One-way ANOVA and Kruskal–Wallis tests showed there were statistically significant differences between groups in 3 of the error-based domain variables (Fluency: $\eta 2 = 0.530$, $p < .001$; Reference: $\eta 2 = 0.488$; $p < .001$; Connectivity: $\eta 2 = 0.479$, $p < .001$); but not in Concordance: ($p = .068$), nor in Quantitative ($p = .073$) (see also Table S1). Tukey's HSD and Bonferroni-corrected Dunn post hoc tests were respectively applied in order to determine the differences between specific groups showing that there were significant differences in Fluency (control vs. manifest HD: $p < .001$; pre-manifest vs. manifest HD: $p < .001$), Reference (control vs. pre-manifest: $p < .005$; control vs. manifest HD: $p < .001$), and Connectivity (control vs. manifest: $p < .001$; control vs. pre-manifest: $p < .001$; pre-manifest vs. manifest HD: $p < .043$) (see Table 3 and Figure 1 below).

Table 3: Domain-level analysis: post-hoc pairwise group comparisons

Variable (domain)	**Mean ± SD**	**Test**	**P-values**		
			C-P	C-M	P-M
Fluency	C: 17.584 ± 9.195 P: 23.992 ± 7.419 S: 41.108 ± 11.466	Tukey's HSD	.198	< .001*	< .001*
Reference	C: 1.781 ± 2.324 P: 6.011 ± 3.244 S: 10.529 ± 6.483	Dunn's test	.005*	<.001*	.544
Connectivity	C: 3.017 ± 1.424 P: 5.463 ± 4.452 S: 9.089 ± 3.709	Dunn's test	.110	< .001*	.043*

Abbreviations (applying to all result tables): C = controls, P = pre-manifest, M = early manifest. In all result tables, an asterisk (*) indicates significance at $\alpha = 0.05$.

Figure 1: Boxplots for differences between groups in the analysis of linguistic variables grouped into five linguistic domains



3.2 Individual variable analysis

Results of the analysis of non-dichotomized individual variables can be found in Table S2 (general group comparisons) and Table 4 below (pairwise comparisons). Results for dichotomized variables can be found in Table 5 below. In Fluency, the following variables were significant in the general group comparisons: Prolongations (Prol, η2=.192;

p=.019), Filled pauses (FilP, η2=.266; p=.006), single functional word repetitions (sFWR, η2=.277; p=.005), Pause between clauses (CP-CP.P, η2=.169; p=.037), Pause between discourse markers and/or XP (DM.P, η2=.210, p=.017), Truncation with morpheme integrity preserved (-W/T, η2=.234, p=.010), Truncation of DP (DP/T, η2=.201, p=.020), Truncation of TP (TP/T, η2=.292, p=.003), Pauses (PAUSES, η2=.221, p=.013), Rephrasing (Rephrasing, η2=.386, p=.001), Hanging topic (/top, η2=.168, p=.038), Coordination wrong (CRD WRONG, η2=.554, p=.001), Subordination wrong (SUB WRONG, η2=.527, p=.001). Pairwise comparisons revealed significant differences between both manifest and pre-manifest HD relative to controls, with both HD groups showing more Filled pauses (FilP) than controls (pre-manifest: p=.005, manifest: p=.039). On the other hand, only pre-manifest HD showed more Prolongations (Prol) relative to controls (p=.028), and only manifest HD showed more Empty pauses (PAUSES) than controls (p=.005). When indexed by syntactic position, these silent lapses occurred in clausal boundary positions, i.e. pauses between or before (as opposed to within) clauses (in formal linguistic terms, either complementizer phrases, CPs, or Tense Phrases, TPs), as reflected in the following variables: Pause between clauses (CP-CP-P: manifest vs. control: p=.016); Pause after a discourse marker, (DM.P (manifest vs. control: p=.013, pre-manifest vs. manifest: p=.022); Pause between V and CP or TP (V-CP/TP.P: manifest vs. control: p=.038, V='verb'), and Pause between full (as opposed to within) phrases (XP-YP.P: manifest vs. control: p=.029). Finally, participants with manifest HD produced more Truncations within words (-W/T, manifest vs. control: p=.007; vs. pre-manifest: p=.017), and within non-clausal phrases (CP/T, manifest vs. control: p=.043, DP/T: manifest vs. control: p=.013, TP/T: manifest vs. control: p=.002). In pre-manifest HD, Single functional word repetitions (sFWR), unlike lexical repetitions, were also increased in relation to controls (p=.002).

In Reference, the following variable was significant in the general group comparisons: Hanging topic (/top: η2=168, p=.038). This variable also distinguished manifest and pre-manifest HD (p=.028). In the dichotomized variables, both Abnormal topic shift (#top) and Vagueness or lack of topic (*0top) distinguished controls and manifest HD (Cramer's V=.595, p=.002 and Cramer's V=.557, p=.009, respectively). The same two groups differed in Ambivalence (+/-ref) (Cramer's V=.612, p=.002), Vague referent (VagRef) (Cramer's V=.515, p=.012), while Definiteness repair (DefRep) (Cramer's V=.596, p=.002) only distinguished controls from pre-manifest HD.

In Connectivity, the general group comparisons revealed significant differences in the variables Total coordination (CRD TOTAL: η2=.248, p=.005), Total subordination (SUB TOTAL: η2=.201, p=.016), Coordination wrong (CRD WRONG, η2=.554, p<.001), Subordination wrong (SUB WRONG: η2=.527, p<.001), and Intrusive parentheticals (#X: η2=.205, p=.018). In the pairwise comparisons, total coordination (CRD TOTAL) distinguished controls from both pre-manifest HD (p=.035) and manifest HD (p=.005); while Total subordination (SUB TOTAL) distinguished manifest HD from both controls (p=.043) and pre-manifest HD (p=.018). Manifest HD used both coordinations and subordinations least, i.e. had more isolated sentences with no grammatical connections between them, as mediated through coordinating and subordinating devices. HD groups also misused these patterns of grammatical connectivity between clauses most. Thus, pre-manifest HD misused coordinations (CRD WRONG) relative to controls (p=.005) and manifest HD (p=.017), as well as misusing subordinations (SUB WRONG, pre-manifest HD vs. controls: p=.033). Manifest HD also had more misuses of both coordinations and subordinations in relation to controls: both p<.001.

In the Quantitative domain, the following variables were significant in the general group comparisons: number of words (WORDS: η2=.154; p=.049), mean length of utterance in morphemes (MLUm: η2=.195; p=.018). In pairwise comparisons, participants with manifest HD produced fewer words than participants with pre-manifest HD (p=.038), and they produced shorter utterances than either participants with pre-manifest HD (p=.037) or controls (p=.025).

In Concordance, the variable Agreement failure in the verbal domain (*AgrX:IAgrT) was significant between controls and pre-manifest HD (V=0.452, p=.049).

Table 4: Non-dichotomized individual variables: post-hoc pairwise group comparisons

Variable	**Mean ± SD**	**Post hoc test**	**p-value**		
			C-P	C-M	P-M
		Fluency			
Prolongations (Prol)	C: 0.634 ± 0.255 P: 0.921 ± 0.296 M: 0.856 ± 0.297	Tukey's HSD	.028*	.108	.859

Filled pauses (FilP)	C: 0.078 ± 0.042 P: 0.198 ± 0.132 M: 0.171 ± 0.151	Dunn's test	.005*	.039*	.811
Single functional word repetition (sFWR)	C: 0.007 ± 0.006 P: 0.012 ± 0.003 M: 0.009 ± 0.004	Dunn's test	.002*	.306	.122
Pauses between clauses (CP-CP.P)	C: 0.075 ± 0.073 P: 0.092 ± 0.084 M: 0.173 ± 0.118	Dunn's test	.837	.016*	.133
Pause between XP and YP (XP-YP.P)	C: 0.038 ± 0.047 P: 0.042 ± 0.035 M: 0.118 ± 0.108	Dunn's test	.776	.029*	.214
Pause between discourse markers and/or XP (DM.P)	C: 0.013 ± 0.024 P: 0.017± 0.037 M: 0.091 ± 0.141	Dunn's test	1	.013*	.022*
Truncation with morpheme integrity preserved (-W/T)	C: 0.001 ± 0.001 P: 0.002 ± 0.003 M: 0.004 ± 0.003	Dunn's test	1	.007*	.017*
Truncation of CP (CP/T)	C: 0.014 ± 0.013 P: 0.022 ± 0.021 M: 0.040 ± 0.031	Dunn's test	.767	.043*	.275
Truncation of DP (DP/T)	C: 0.018 ± 0.015 P: 0.020 ± 0.021 M: 0.039 ± 0.016	Dunn's test	1	.013*	0.033*
Truncation of TP (TP/T)	C: 0.022 ± 0.021 P: 0.026 ± 0.019 M: 0.059 ± 0.028	Dunn's test	1	.002*	.018*
Total pauses (PAUSES)	C: 0.159 ± 0.159 P:0.195 ± 0.173 M: 0.462 ± 0.370	Dunn's test	.761	.005*	.077
Rephrasing (Rephrasing)	C: 0.112 ± 0.057 P: 0.121 ± 0.073 M: 0.234 ± 0.081	Tukey's HSD	.944	< .001*	<.001*
Reference					
Hanging topic (/top)	C: 0.012 ± 0.020 P: 0.023 ± 0.017 M: 0.033 ± 0.026	Dunn's test	.125	.028*	.886
Connectivity					
Coordination wrong (CRD WRONG)	C: 0.262 ± 0.101 P: 0.382 ±0.091 M: 0.502 ± 0.075	Tukey's HSD	.005*	< .001*	.017*

Subordination wrong (SUB WRONG)	C: 0.021 ± 0.016 P: 0.083 ± 0.107 M: 0.127 ± 0.078	Dunn's test	.033*	< .001*	.092
Intrusive parenthetical (#X)	C: 0.009 ± 0.016 P: 0.028 ± 0.040 M: 0.037 ± 0.036	Dunn's test	.127	.011*	.605
Total coordination (CRDTOTAL)	C: 0.534 ± 0.137 P: 0.680 ± 0.145 M: 0.460 ± 0.157	Tukey's HSD	.035*	.389	.005*
Total subordination (SUBTOTAL)	C: 0.751 ± 0.253 P: 0.832 ± 0.335 M: 0.495 ± 0.192	Tukey's HSD	.709	.043*	.018*
Quantitative					
Number of words (WORDS)	C: 1787.950 ± 333.750 P: 1841.400 ± 371.345 M: 1438.900 ± 360.179	Dunn's test	.952	.052	.038*
MLU morphemes (MLUm)	C: 19.796 ± 3.398 P: 20.108 ± 3.802 M: 16.149 ± 3.136	Tukey's HSD	.970	.025*	.037*

Table 5: Dichotomized individual variables: pairwise group comparisons
Percentages are of subjects exhibiting instances of the relevant variable in each group. The effect size is an omnibus group effect.

Variables	**Percentages**			**P-values**			**Effect size (Cramer's V)**
	C	P	M	C-P	C-M	P-M	
Fluency							
Pause between V and CP/TP (V-TP.P;V-CP.P)	25%	60%	80%	.297	.038*	1	0.472
Reference							
Abnormal topic shift (#top)	5%	30%	70%	.262	.002*	.328	0.595
Vagueness or lack of topic (*0top)	10%	20%	70%	1	.009*	.192	0.557
Ambivalence (+/-ref)	0%	20%	60%	.284	.002*	.311	0.612
Vague referent (VagRef)	5%	40%	60%	.086	.012*	1	0.515
Definiteness repair (DefRep)	0%	60%	30%	.002*	.081	.678	0.596

			Concordance				
Agreement failure in the verbal domain (*AgrX:IAgrT)	5%	50%	30%	.049*	.262	1	0.452

3.3 Neuropsychological variables and correlations

Results revealed that there were statistically significant differences between groups in all but two of the neuropsychological variables (no differences were found between manifest and pre-manifest groups for the BDAE subtest of Reading Comprehension (sentences) and between all the groups for the TAP/NART); see Tables S3 (for results at the group level) and Table 6 (pairwise comparisons). However, the manifest HD group crossed the threshold for clinical impairment (at a 'moderate' level) only in three neuropsychological tasks, namely Stroop (Reading and Denomination), Trail Making Test (TMT, part A and B), and the Digit Symbol Substitution Test.

Table 6: Neuropsychological variables: Post-hoc pairwise group comparisons

Variable	**Mean ± SD**	**Post hoc test**	**p-value**		
			C-P	C-M	P-M
Boston Listening Comprehension test: commands	C: 10.0± 0.00 P: 9.9 ± 0.32 M: 9.4 ± 0.70	Dunn's test	.731	< .001*	.019*
Boston Listening Comprehension test: Complex Ideative Material	C: 6.0 ± 0.00 P: 5.5 ± 0.71 M: 4.3 ± 1.16	Dunn's test	.086	< .001*	.025*
Boston Naming test: Naming Response (questions)	C: 10.0 ± 0.00 P: 10.0 ± 0.00 M: 9.5 ± 0.53	Dunn's test	1	< .001*	.001*
Boston Naming test: Visual Confrontation (images)	C: 14.3 ± 0.66 P: 14.0 ± 1.33 M: 11.3 ± 1.64	Dunn's test	1	<.001*	.001*
Boston Reading + Comprehension	C: 5.0 ± 0.00 P: 5.0± 0.00 M: 4.6 ± 0.70	Dunn's test	1	.005*	.018*
Boston Reading Comprehension: sentences	C: 3.0 ± 0.22 P: 2.8 ± 0.42 M: 2.3 ± 0.95	Dunn's test	.529	.005*	.128
Verbal Fluency test (animal naming)	C: 26.5 ± 6.25 P: 24.1 ± 6.24	Tukey's HSD	.566	.001*	.046*

	M: 17.4 ± 5.38				
Stroop Reading test	C: 111.3 ± 12.95 P: 99.4 ± 20.97 M: 60.4 ± 15.94	Tukey's HSD	.146	< .001*	<.001*
Stroop Denomination test	C: 77.2 ± 10.09 P: 69.9 ± 12.34 M: 38.4 ± 13.29	Tukey's HSD	.242	< .001*	<.001*
Stroop Interference test	C: 43.5 ± 9.03 P: 43.5 ± 9.88 M: 24.2 ± 9.65	Tukey's HSD	.999	< .001*	< .001*
Trail Making test Part A	C: 36.9 ± 34.78 P: 33.0 ± 9.55 M: 84.0 ± 48.01	Dunn's test	.597	<.001*	.012*
Trail Making test Part B	C: 92.1 ± 61.07 P: 94.9 ± 47.91 M: 160.5 ± 89.43	Dunn's test	1	.022*	.089
Digit Symbol Substitution Test	C: 50.6 ± 14.52 P: 51.3 ± 9.78 M: 26.3 ± 11.66	Dunn's test	1	< .001*	< .001*
Mini–Mental State Examination	C: 29.8 ± 0.52 P: 28.6 ± 1.58 M: 26.5 ± 2.27	Dunn's test	.042*	< .001*	.039*

After FDR correction, only two significant correlations between linguistic and neuropsychological domains remained: (i) between TMT-A and the domain of Fluency ($r = 0.80$, $p = <.001$), and (ii) between TMT-A and the domain of Reference ($r = 0.611$, $p = .041$).

4. Discussion

Results of the present study showed that, at the domain-level, groups differed in the domains of Fluency, Reference and Connectivity, but neither Quantitative nor Concordance. A progressive decline from controls to pre-manifest to manifest HD was seen in all of the three former domains, with significance thresholds crossed in the comparison of manifest HD and controls in all three cases, while pre-manifest HD differed from controls only in Reference. This pattern exhibits important overlaps but also differences with the previous study of Hinzen et al. (2018), where significant differences between manifest HD and controls were seen in both Quantitative and Concordance as well. Results in Fluency, Connectivity and Reference, on the other hand, were broadly

similar. Specifically, they were identical in terms of comparisons between manifest HD and controls, and like in the previous study, also pre-manifest HD differed from controls in Reference. However, in Connectivity they only differed from controls in the previous but not the present study.

A comparison of the demographics of the two samples involved in these two studies, which were recruited in the same region, revealed no significant differences in age, gender, or education. Difference in the results of the two studies suggest that purely quantitative or else formal grammatical measures (e.g. number or length of utterances or grammatical agreement) may be less sensitive, at a domain-level, in capturing the neuropathology in question at a linguistic level: measures in the domains of fluency, connectivity and reference may reveal language decline more reliably and earlier in the disease process. Note that, on the other hand, some of the individual linguistic variables within Quantitative and Concordance showed significant group differences also in the present study. In particular, manifest HD differed from both pre-manifest and controls in the Mean Length of Utterance. A larger sample size may have shown significant group differences in Quantitative at the domain level as well. As for the loss of significance in Connectivity when comparing pre-manifest HD and controls, this difference may in part be due to a difference in how relevant variables were normalized in both studies (in particular, anomalous uses of coordinations and subordinations were normalized relative to total coordinations and subordinations, respectively, in the present study).

This combined outcome from two studies and independent samples and different tasks used for elicitation, provides renewed support that neurodegeneration in HD affects core domains of language functioning in spontaneous speech as well, from the pre-manifest stage. This is in line with reports of neurodegeneration in HD in language-relevant subcortical areas long before clinical symptoms are seen (Aylward et al., 2012; Bano et al., 2011). In the present study, moreover, this again occurred when neuropsychological tests revealed no decline in pre-manifest gene carriers except in the case of the MMSE (see Table 6). Even in manifest HD, MMSE scores were not in the range of impairment and turned out not to correlate with any linguistic variables or domains. We do not interpret language decline ahead of motor symptomatology as suggesting that language function is unrelated to motor function. Rather, motor deficits may be too subtle at the pre-manifest stage to show in domains other than language, which is the most complex and rapid motor action that humans perform (Lipski et al., 2017;

Simonyan & Fuertinger, 2015; Simonyan, Ackermann, Chang, & Greenlee, 2016). This underlines the potential role of language as a clinical marker of disease progression (García et al., 2017; Vogel et al., 2012) and calls for longitudinal studies.

Our second aim was to move the domain-level analysis of Hinzen et al. (2018) down to the level of individual variables. Here a telling pattern arose in the domain of Fluency, where participants with manifest but not pre-manifest HD produced more empty pauses ('speech left blank', without fillers) than controls (Table 4). Pauses, in the definition of Silverman (1973), are 'intermittent feedback delay operations, allowing the momentum of semi-automatic speech generation to be halted while information is processed for the appropriate planning of subsequent utterances'. This suggests that differences in such planning are not yet visible at the pre-manifest stage. Pauses, however, can also be 'filled', where filling a pause suggests awareness of the break in the flow of speech, along with interpersonal social signalling that the flow of thought continues. While both HD groups had more filled pauses than controls, only pre-manifest HD showed more prolongations and repetitions in relation to controls. These, too, can be ways of bridging a gap and manifesting awareness of its existence. Further in line with this pattern, *definiteness repair* (DefRep: self-correction of anomalously introduced referents), also indicative of insight into communication failure, was only seen in the pre-manifest group, but not the manifest one. In short, while manifest patients present 'gaps' in their speech (pauses and truncations of utterances and words), pre-manifest HD tend to fill these gaps using prolongations and repetitions.

Importantly, such breakage patterns showed up along clausal boundaries, suggesting the importance of indexing dysfluency patterns by the syntactic positions in which they occur. Clauses are units of structure where relatively complete units of thoughts are encoded. It would be pauses within simple phrases, such as between an article and a noun, or truncations of them, which would point to a problem at the level of lexical retrieval. This pattern, which has been documented for spontaneous speech in both the cases of Alzheimer's disease (Gayraud, Lee, & Barkat-Defradas, 2011) and post-stroke aphasia (Angelopoulou et al., 2018), was not observed here. Instead, the pattern points to a problem in configuring thought-sized units, i.e. units of structure in language encapsulating a complete thought. Further supportive of this conclusion against a specifically lexical problem, word repetition patterns were confined to repetitions of grammatical function words, not lexical items. These results can be contextualized

against those of Vogel et al. (2012) on fluency patterns in people with manifest HD, who differed from both controls and a pre-manifest group in speech rate (syllables/total signal time), total speech time, and total silence time, with manifest HD having a lower speech rate and higher total silence time. These authors, however, did not index pauses by syntactic position, nor distinguished empty and filled pauses. As for the pattern of prolongations, fillers, repetitions, and repairs seen in pre-manifest HD, we tentatively interpret this as reflecting 'adaptive strategies' in the sense of Illes (1989: p. 636), i.e. coping strategies in the face of a functional deficit in language. This functional deficit is centred on the construction of appropriate units of language for purposes of thought and reference.

Results in Connectivity cohere with the significance of the clausal boundary in HD just noted. Grammar across human languages avails us of two key ways in which clauses can be combined: one clause can be embedded in another, in which case one is *sub*ordinated to the other; or they can be *co*ordinated, in which case they are both grammatically independent and the relation between them is symmetric (i.e., none is subordinated under the other). With this difference goes a difference in the thought expressed: a sentence with a subordinated clause embedded under a verb will ipso facto represent how someone represents the world (what he thinks, says, believes, or wants), and hence express a meta-representation. Failure to use subordinations will make it more difficult to express meta-representations, i.e. thoughts about thoughts, and hence reasoning about mental states (ToM). In line with this, clausal embedding of the subordinating type has been widely argued to be a potential mechanism for accomplishing classical ToM tasks (Paynter & Peterson, 2010; Astington & Jenkins, 1999; Steele, Joseph, & Tager-Flusberg 2003). Key group differences that emerged in this study regarding subordination and coordination patterns could thus be cognitively significant, manifesting difficulties in reasoning about mental states. In particular, participants with manifest HD used both of coordination and subordination less than pre-manifest HD, which suggests that, as the diagnostically criterial motor symptoms emerge, grammar also shifts in its organization, becoming more mono-clausal or grammatically unconnected. Moreover, manifest HD had more anomalous uses of coordinations (CRD WRONG) in relation to both pre-manifest HD and controls, and the pre-manifest group produced more wrong coordinations and subordinations than controls. This finding calls for studies in which independently noted impairments in ToM in HD as assessed by standardized ToM

tests (Brüne, Blank, Witthaus, & Saft, 2011; Eddy, Mahalingappa, & Rickards 2012; Saft et al., 2013; Adenzato, & Poletti, 2013; Bora et al., 2016), would be correlated with language measures directly, and specifically with coordinating and subordinating clause types. It also underlines the need to differentiate earlier composite measures of syntactic complexity (e.g. Illes, 1989; Murray & Lenz, 2001), in which different forms of syntactic complexity are often amalgamated into a single overall measure of complexity. Utterances with coordinated or subordinated clauses are both 'complex', yet very different kinds of complexity, corresponding to different cognitive mechanisms and types of thoughts expressed, are involved.

Results in the Reference domain suggest that language decline not only shows in how clauses are combined and the loss of semantic richness and complexity resulting from this, but also in the use of language for purposes of reference, which relates to discourse. The main shifts here, at the level of individual variables, were seen in manifest, but not pre-manifest HD, through a pattern of abnormal topic shifting, setting up a topic without pursuing it (truncated topics), or vagueness and ambivalence of reference, which is still noted and thus 'repaired' only in the pre-manifest group ('definiteness repair' mentioned above).

Turning to our third aim, all of the neuropsychological measures showed significant differences between controls and the manifest HD group, while only the MMSE showed a difference between controls and pre-manifest HD (Table 6). Only the manifest HD group ever crossed clinical thresholds to cognitive impairment, though in no case, impairment was severe. Moreover, they only did so in three measures, namely the Stroop (only Reading and Denomination), Trail Making Test, and Digit Symbol Substitution Test (Table 6). This suggests problems of attention, working memory, and executive functioning, as well as a potential difficulty with visual processing (since all of these tasks are administered via visual stimuli), though performance on other, also visually based tests was close to normal (e.g. Stroop Interference or BDAE Boston Visual Confrontation). However, only one of these tests (the Trail Making Test Part A) correlated with the linguistic domains (Fluency and Reference). The correlation with Fluency would make sense in light of reductions in processing speed which would affect both speech Fluency and the Trail Making Test. Whether the same link explains the correlations with Reference is less clear. Whether the same link could explain the correlations with Reference is less clear, though post hoc analysis to clarify this point revealed that the

domains of Fluency and Reference correlated with each other (r = 0.606, p=.041). As an anonymous referee notes, the Trail Making Test Part B is more challenging cognitively and requires processing speed along with attention and working memory. But it did not correlate with either Fluency or Reference, making the previous correlations difficult to interpret. Unfortunately, direct measures of working memory were not available in the present study, but they correlated only with the Quantitative domain in the previous study of Hinzen et al. (2018).

It is also noteworthy that, although the manifest group differed from both other groups in language tests designed for patients with aphasia (BDAE), performance on these tests was still generally high; and it did not correlate with the linguistic measures introduced in our study. Language is a complex domain that can disintegrate in a large number of different ways and at different levels: language impairment across clinical groups will rarely be the same. This has the important clinical implication that language tests that can detect and assess language patterns in HD should be devised. In this regard, the present results suggest that patterns of linguistic impairment in HD cut across the traditional linguistic levels of 'syntax' and 'semantics', so that this traditional divide would have been unlikely to capture the clinical patterns seen. Indeed, these level descriptors have become problematic within linguistic theory itself (Wiltschko, 2018), and may be of questionable utility clinically.

Apart from correlations with working memory and direct ToM tests, future work calls for replications of our findings in languages other than Spanish and for linking them to patterns of neural degeneration. Hinzen et al. (2018) failed in identifying structural neural correlates for domain-level linguistic impairments except in the Quantitative domain. Although data about neural atrophy were not available in the present study, the significance of clausal connectivity both at the level of Fluency and Connectivity documented here informs the debate on the significance of the striatum and frontal-striatal loops for syntactic structuring, recursion, and the 'chunking' of linguistic information into clausal informational units (Bornkessel & Schlesewsky, 2013; Graybiel, 1995; Lieberman, 2007) under temporal constraints (Kotz & Schwarze, 2010). Clausal embedding would be a particularly useful focus in future functional neuroimaging studies.

In sum, this study has provided further support for language degeneration in early and pre-manifest HD and contributes to a more fine-grained and differentiated profile of the linguistic phenotype of this disease. In HD, language changes precede other cognitive and motor impairment. These clearly lie outside of the speech-articulatory domain, in core domains of grammatical organization, and they are not easily accounted for by non-linguistic cognitive impairment, whether occurring in participants with pre-manifest or manifest HD. As language capacities fundamentally impact on communicative abilities, careful attention should be devoted to their early detection, to clinical linguistic tests appropriate for this population, and to protective treatments. With regard to the potential role of language as a marker of disease progression, language has already shown distinctive signature profiles and potential as a predictive and diagnostic measure in a number of other neuropathologies, including Alzheimer's disease (Ahmed, Haigh, de Jager, & Garrard, 2013), Parkinson's disease (García et al., 2017), autism (Eyler Pierce, & Courchesne, 2012; Lombardo et al., 2015), and schizophrenia (Bedi et al., 2015; Rosenstein Foltz, DeLisi, & Elvevåg,, 2015; Cokal et al., 2018). As language disintegration systematically differs across all of these neuropathologies, further comparative work should systematically investigate the sensitivity and specificity of language as a marker of disease progression.

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Supplementary materials

Table S1: Group-level domain analysis

Variable (domain)	Test	Statistic	P-value	Effect size (η^2)
Fluency	ANOVA	F(2,37)=20.86	<.001	0.530
Reference	Kruskal-Wallis	$\chi^2(2) = 19.04$	<.001	0.488
Connectivity	Kruskal-Wallis	$\chi^2(2) = 18.69$	<.001	0.479

Table S2: Non-dichotomized individual variables: group-level results

Variable (individual)	Test	Statistic	P-values	Effect size (η^2)
Fluency				
Prolongations (Prol)	ANOVA	F(2,37) = 4.41	.019*	.192
Filled pauses (FilP)	Kruskal-Wallis	$\chi^2(2) = 10.39$	.006*	.266
Single functional word repetition (sFWR)	Kruskal-Wallis	$\chi^2(2) = 10.79$	.005*	.277
Pause between clauses (CP-CP.P)	Kruskal-Wallis	$\chi^2(2) = 6.60$	.037*	.169
Pauses between XP and YP (XP-YP.P)	Kruskal-Wallis	$\chi^2(2) = 5.51$	.064	.141
Pause between discourse markers and/or XP (DM.P)	Kruskal-Wallis	$\chi^2(2) = 8.19$	.017*	.210
Truncation with morpheme integrity preserved (-W/T)	Kruskal-Wallis	$\chi^2(2) = 9.13$	.010*	.234
Truncation of CP (CP/T)	Kruskal-Wallis	$\chi^2(2) = 4.81$	.090	.123
Truncation of DP (DP/T)	Kruskal-Wallis	$\chi^2(2) = 7.84$	.020*	.201
Truncation of TP (TP/T)	Kruskal-Wallis	$\chi^2(2) = 11.38$	.003*	.292
Pauses (PAUSES)	Kruskal-Wallis	$\chi^2(2) = 8.61$	.013*	.221
Rephrasing (Rephrasing)	ANOVA	F(2,37) = 11.64	< .001*	.386
Reference				

Hanging topic (/top)	Kruskal-Wallis	$\chi^2(2) = 6.54$	.038*	.168
Connectivity				
Coordination wrong (CRD WRONG)	ANOVA	F(2,37) = 23	<.001*	.554
Subordination wrong (SUB WRONG)	Kruskal Wallis	$\chi^2(2) = 20.56$	< .001*	.527
Total coordination (CRDTOTAL)	ANOVA	F(2,37) = 6.10	.005*	.248
Total subordination (SUBTOTAL)	ANOVA	F(2,37) = 4.65	.016*	.201
Intrusive parentheticals (#X)	Kruskal-Wallis	$\chi^2 = 8.00$	.018*	.205
Quantitative				
Number of words (WORDS)	Kruskal-Wallis	$\chi^2(2) = 6.01$	.049*	.154
MLU morphemes (MLUm)	ANOVA	F(2,37) = 4.48	.018*	.195

Note: All variables were normalized with respect to words or utterances, with the exception of variables in the Quantitative domain (WORDS, MORPHEMES, MLUm), which were not normalized; and CRD WRONG and SUB WRONG, which were normalized over CRD TOTAL and SUB TOTAL, respectively.

Table S3: Neuropsychological variables: Group-level results

Variable	**Test**	**Statistic**	**P-value**	**Effect size (η2)**
Boston Listening Comprehension test: commands	Kruskal-Wallis	$\chi^2(2) = 13.11$	.001*	0.336
Boston Listening comprehension test: Complex Ideational Material	Kruskal-Wallis	$\chi^2(2) = 21.92$	< .001*	0.562
Boston Naming test: Naming Response (questions)	Kruskal-Wallis	$\chi^2(2) = 16.71$	< .001*	0.429
Boston Naming test: Visual Confrontation (images)	Kruskal-Wallis	$\chi^2(2) = 18.12$	< .001*	0.565
Boston Reading + Comprehension	Kruskal-Wallis	$\chi^2(2) = 9.47$	.009*	0.243
Boston Reading Comprehension: sentences	Kruskal-Wallis	$\chi^2(2) = 8.51$	.014*	0.218
Verbal Fluency Test (animal naming)	ANOVA	F(2,37) = 7.58	.002*	0.291

Stroop Reading test	ANOVA	F(2,37) = 34.27	< .001*	0.649
Stroop Denomination test	ANOVA	F(2,37) = 38.85	< .001*	0.677
Stroop Interference test	ANOVA	F(2,37) = 15.78	< .001*	0.460
Trail Making test Part A	Kruskal-Wallis	$\chi^2(2) = 15.49$	< .001*	0.397
Trail Making test Part B	Kruskal-Wallis	$\chi^2(2) = 6.32$	.042*	0.162
Digit Symbol Substitution Test	Kruskal-Wallis	$\chi^2(2) = 15.57$	< .001*	0.399
Mini–Mental State Examination	Kruskal-Wallis	$\chi^2(2) = 23.22$	< .001*	0.595

Tables S4 to S8 contain Pearson correlations between non-dichotomized variables within domains**.** Only variables with significant results in the main analysis (group-level comparisons) were included. Variable acronyms are explained in Table 2 and the manual of speech and language below.

Table S4: Inter-correlations (and p-values) between variables of the Fluency domain.

Bonferroni-corrected significance threshold (α): 0.003

	Prol	FilP	LWR	sFWR	mFWR	C.TP.P	CP.CP.P	XP.YP.P	WT	CP.T	DP.T	PP.T	VP.T	TP.T	DM.P	Rephrasing
Prol	1.00															
FilP	0.32 (.044)	1.00														
LWR	0.09 (.563)	-0.07 (.689)	1.00													
sFWR	0.29 (.065)	0.32 (.044)	0.48 (.002)*	1.00												
mFWR	0.2 (.206)	0.2 (.228)	0.44 (.004)	0.52 (<.001)	1.00											
C-TP.P	0.19 (.236)	0.11 (.505)	0.06 (.700)	-0.07 (.676)	0.15 (.353)	1.00										
CP-CP.P	0.38 (.015)	0.34 (.029)	-0.04 (.785)	-0.05 (.739)	0.06 (.714)	0.54 (<.001) *	1.00									
XP.YP.P	0.23 (.156)	0.37 (.018)	-0.01 (.933)	-0.14 (.382)	0.05 (.746)	0.73 (<.001)*	0.75 (<.001) *	1.00								
CP/T	0.4 (.012)	0.25 (.116)	0.17 (.283)	0.07 (.683)	0.31 (.051)	0.4 (.011)	0.53 (<.001) *	0.51 (<.001) *	0.36 (.022)	1.00						
DP/T	0.24 (.139)	0.34 (.031)	0.02 (.898)	0.02 (.926)	0.13 (.436)	0.25 (.125)	0.59 (<.001) *	0.57 (<.001) *	0.49 (.001) *	0.48 (.002) *	1.00					
PP/T	0.48 (.002)*	0.08 (.606)	-0.0 (.987)	0.02 (.883)	-0.01 (.932)	0.01 (.937)	0.54 (<.001) *	0.17 (.306)	0.23 (.147)	0.45 (.004)	0.47 (.002) *	1.00				
VP/T	-0.09 (.587)	0.0 (.986)	-0.31 (.054)	-0.15 (.354)	-0.07 (.660)	-0.15 (.359)	0.02 (.891)	-0.13 (.417)	0.28 (.075)	0.01 (.933)	0.37 (.019)	0.14 (.399)	1.00			
TP/T	0.31 (.055)	0.41 (.008)	0.14 (.402)	0.1 (.528)	0.35 (.025)	0.3 (.064)	0.42 (.006)	0.55 (<.001)*	0.47 (.002) *	0.57 (<.001)*	0.53 (<.001)*	0.19 (.239)	0.04 (.825)	1.00		
DM.P	0.17 (.286)	0.63 (<.001)*	-0.13 (.430)	-0.09 (.568)	-0.02 (.899)	0.39 (.012)	0.7 (<.001) *	0.78 (<.001)*	0.52 (<.001) *	0.43 (.006)	0.59 (<.001)*	0.27 (.097)	0.03 (.876)	0.47 (.002)*	1.00	
Rephrasing	0.41 (.008)	0.39 (.012)	0.09 (.591)	0.16 (.322)	0.16 (.332)	0.15 (.347)	0.49 (.001)*	0.33 (.036)	0.64 (<.001) *	0.58 (<.001) *	0.71 (<.001) *	0.48 (.002)*	0.44 (.004)	0.68 (<.001)*	0.33 (.0400)	1.00

Table S5: Inter-correlations (and p-values) between variables of the Reference domain.
Corrected significance threshold (α): 0.008

	/top	MX:MD	MX:MP	corXP	refT	v
/top	1.00					
MX:MD	0.28 (.075)	1.00				
MX:MP	0.18 (.276)	0.21 (.193)	1.00			
corXP	0.05 (.764)	-0.14 (.391)	-0.14 (.375)	1.00		
refT	0.10 (.546)	-0.12 (.465)	0.01 (.956)	-0.06 (.711)	1.00	
v	0.00 (.988)	-0.08 (.607)	-0.09 (.595)	-0.04 (.794)	0.58 (<.001)*	1.00

Table S6: Inter-correlations (and p-values) between variables of the Connectivity domain
Bonferroni-corrected significance threshold (α): 0.013

	CRD WRONG	SUB WRONG	#X	Tcons.temp
CRD WRONG	1.00			
SUB WRONG	0.65 (<.001)*	1.00		
#X	0.49 (.001)*	0.7 (<.001)*	1.00	
Tcons.temp	0.37 (.018)	0.53 (<.001)*	0.67 (<.001)*	1.00

Table S7: Inter-correlations (and p-values) between variables of the Concordance domain
Bonferroni-corrected significance threshold (α): 0.017

	AgrX AgrI	GovV	#V
AgrX AgrI	1.00		
GovV	0.57 (<.001)*	1.00	
#V	-0.13 (.439)	-0.15 (.361)	1.00

Table S8: Inter-correlations (and p-values) between variables of the Quantitative domain
Bonferroni-corrected significance threshold (α): 0.017

	Utterances	Words	MLU morphemes

Utterances	1.00		
WORDS	0.56 (<.001)*	1.00	
MLU morphemes	-0.08 (.634)	0.78 (<.001)*	1.00

Manual for speech and language annotation

DEFINITIONS

Utterance: Syntactically independent unit of discourse providing new information. An utterance may be formed by one clause (e.g. *I like this place*), by several clauses including one or more dependent ones (e.g. *I like the place where I live*), or by non-clausal units of discourse (e.g. *Hello / Yes / Peter.*). Independent clauses, even if informationally or grammatically related (e.g. through coordination), will be interpreted as different utterances, with one exception: when they appear inside a dependent clause (e.g. *I like the place where I live and have always worked*).

Clause: Configuration with a predication (verbs, adjectives, copular constructions, small clauses).

Variables

1. **Fluency**

1.1 Rephrasing

Rephrasing (Rephrasing): the speaker reformulates an utterance **'**pues mi padre tenía un amigo *que trab* que era pastelero y me mandaron a mí allí.'

1.2. Additions

Prolongations (Prol): a vowel or syllable is elongated. For example: 'queeeee:'.

Filled pauses: uh, um (FilP): hesitation sounds that speakers employ to indicate uncertainty or to fill conversation while thinking about what to say next. For example: 'uh, em, eh'.

1.2. Repetitions

Word domain

Lexical word repetition (LWR): repetition of a content word. For example: 'coge…coge'.

Functional words (FWR):

- ➢ Single (sFWR): repetition of one functional word, i.e. a word that serves to express grammatical relationships with other words within a sentence. For example: 'y…y'.
- ➢ Multiple (mFWR): repetition of one functional word, i.e. a word that serves to express grammatical relationships with other words within a sentence. For example: 'es que lo, es que lo'.

Part-word repetitions (p-FWR) or (p-LWR): repetition of a word that was left unfinished when first uttered. For example: 'quer…querría'.

Phrasal domain

XP (XPR) [*where X is a category, different from D, to be potentially specified*]: repetition of a phrase where X can be D, P, V, C. For example: 'le hacen limpiar...le hacen limpiar'.

Part-XP repetitions (p-XPR): repetition of a phrase that had an unfinished word when first uttered. For example: 'al p…al príncipe'.

1.3. Pauses

Within words

i. Morpheme integrity preserved (-WP): pause inside the word, where the morpheme structure is preserved. For example: 'ceni...cienta'.

ii. Morpheme integrity violated (–WP): pause inside the word, where the morpheme structure is not preserved.

Within non-clausal phrases

i. Between Determiner and Noun Phrase (D-NP.P): pause in a non-clausal phrase between the determiner and the noun. For example: 'y: en ese momento bajo el (.) príncipe'.

ii. Between V and CP/TP (V-TP.P; V-CP.P): pause in a non-clausal phrase, between the verb and its complement or between the verb and other clause. For example: 'ella pensaba (..) en convertirse (..) en princesa'.

iii. Between C and TP (C-TP.P): pause between C and Tense. For example: 'y: (…) &eh luego estuvo pidiendo'.

iv. Between P and XP (P-XP.P): pause between a preposition and the following phrase. For example: 'un traje con [///] unos pajaritos'.

v. Between T and VP (T-VP.P): pause between the auxiliary verb and the main verb. For example: 'sinose [//] acabará el hechizo'.

vi. Between XP and YP (XP-YP.P): pause between two phrases. For example: 'el príncipe [/?] se pasa todo el rato bailando con ella'.

vii. Between clauses (CP-CP.P): pause between two clauses. For example: 'luego ya si que pudo encontrar el zapato de la cenicienta # y entonces si que era de ella'.

viii. Between discourse markers and/or XP (DM.P): pause between two discourse markers or between a discourse marker and the following XP. For example: 'y nada [/] al final hace trabajos forzados'.

1.4. Truncations

When a phrase is left unfinished but the topic hasn't been dropped.

Within words

i. Morpheme integrity preserved (-W/T): a word is left unfinished but the morpheme integrity is preserved. For example: ‘mont(ó)’.

ii. Morpheme integrity violated (–W/T): a word is left unfinished and the morpheme integrity is violated, i.e. the smallest meaningful unit of language is not preserved. For example: ‘lim(piar)’.

Within non-clausal phrases

i. QP (QP/T): a quantifier phrase is left unfinished after the quantifier determiner is uttered. For example: ‘no había más...’.

ii. DP (DP/T): a determiner phrase is left unfinished after the determiner is uttered. For example: ‘hace el...’.

iii. PP (PP/T): a prepositional phrase is left unfinished after the preposition is uttered. For example: ‘que tenia para...’.

iv. VP (VP/T): a verb phrase is left unfinished after the verb is uttered. For example: ‘le hacen...’.

v. TP (TP/T): a phrase is left unfinished after the auxiliary verb or nexus is uttered. For example: ‘no sabe qué hacer y...’.

vi. CP (CP/T): a phrase is left unfinished after the complementizer: ‘me dijo que...’

2. Reference

2.1. Discourse organization

Hanging topic (/top): the speaker leaves the topic unfinished and the next topic is not associated with the previous one.

Abnormal topic shift (#top): the speaker is talking about a topic and then abnormally changes it in the same utterance.

Vagueness or lack of topic (Øtop): the speaker does not specify what she/he is speaking about. There is no reference in the utterance to understand the topic.

Ambivalence (contradictory statements) (+/-ref): the speaker produces two utterances that are contradictory.

Failures in temporal reference (refT): the discourse does not follow the temporal line. For example, first the story starts in the present and then it switches inappropriately to past.

Mental verbs (v): total number of mental verbs (i.e. to believe, to think). For example: ‘te creo siempre’.

2.2. Aspects of definiteness

Vague referent (VagRef): the speaker introduces a referent with insufficient specification.

Definiteness repair (DefRep): a presupposition failure is produced and the speaker tries to resolve it after the sentence is uttered; e.g. **'**y como Meritxell va a ir allí para estudiar, Meritxell es mi hija' (including infelicitous definites).

Paraphasia: the speaker uses a general word instead the specific noun that is more appropriate to the particular context. 'se cae por la ventana, le atiende un señor, se pone de mal humor' (*señor* instead of *doctor*).

2.3. Determiner phrases

Self-correction of determiners (corXP). The speaker utters a wrong determiner or a wrong preposition and she/he corrects it at the moment.

i. Incorrect (corXP): the correction is carried out wrongly. For example: 'unas...una hermanastras'.

ii. Correct (okcorXP): the correction is carried out correctly. For example: 'la...el zapato'.

Hanging determiners (/D): a determiner is uttered and then left standing alone (i.e. not forming part of a sentence, clause or phrase).

Missing X (ØX/XP): the preposition (MX:MP) or the determiner (MX:MD) is missing in the phrase. For example: 'que es (a) la que le viene bien el zapato'.

Infelicitous wildcard DP (-$DP): an intrusive determiner phrase with no referent is uttered. For example: 'esa, eso, aquel sitio'.

Infelicitous indefinite (#non-defDp): an indefinite determiner is used with a noun/referent that has been previously introduced in the context.

Ambivalence (+/-DP): contradictory referentiality in the DP domain. For example: 'la…una camiseta.'.

3. Connectivity

Coordinated clause misuse (CRDWRONG): two or more clauses are not syntactically dependent on another and are anomalously connected by a coordinated conjunction such as y, o, pero, sino, etc. Idioms and enumerations are not taken into account.

Subordinated clause misuse (SUBWRONG): any error in a clause that is syntactically dependent on another, i.e. the two clauses are in a hierarchical relation. Fixed structures (i.e 'es que') are not taken into account.

Missing discourse markers (links) (ØD-link): There is a missing discourse marker to introduce the topic. For example: 'y el encuentro, hay dificultades'.

Incorrect discourse marker (D-link): the discourse marker used is infelicitous with the utterance meaning.

Intrusive parenthetical (#X): the speaker produces an utterance that is not related to the storytelling. It can be to express a doubt she/he has about the story or just to express a thought.

Failures in consecutio temporum (Tcons.temp): tenses within an utterance are not coherent (e.g. two grammatical tenses inside the same utterance).

4. Concordance

Agreement failures (AgrDP): Incorrect agreement of number and/or gender between the determiner and the noun. For example: ‘los hermanas’.

Agreement (AgrX): incorrect agreement of tense and/or aspect in the auxiliary verb and/or the main verb. For example: ‘hace una carroza que la llevan’.

Government (GovV): incorrect selection of the particle that accompanies the verb. For example: ‘empieza de...’.

Infelicitous verb (#V): misuse of a verb in linking subject and predicate.

5. Quantitative

Mean length of utterance (MLUm): MLUm is the mean length utterance in morphemes.

Utterances: total number of utterances produced (coordinated utterances are entered in separate lines).

Words: total number of words uttered.

4. STUDY 2: DETECTION OF ILLICIT PHRASAL MOVEMENT IN HUNTINGTON'S DISEASE

Abstract. The role of the basal ganglia has been a longstanding issue in neural language models. Huntington's disease (HD) shows primary impairment in the striatum and has previously been shown to affect the processing of phrase-structural hierarchies that are built by phrasal movement (e.g. in passives). Here we asked patients with HD to judge the acceptability of sentences containing different types of illicit phrasal movement, which were contrasted with semantic violations involving no movement. A logistic mixed-effects regression showed that patients had a profound impairment in judging incorrect but not correct sentences across all types of illicit movement, while the semantic condition was also but significantly less affected. Adding neuropsychological variables to the model did not improve predictions. These results demonstrate a loss of cognitive control, worsening with disease progression, over phrase-structural hierarchies, which extends to the forms of meaning built at sentential levels.

Keywords: Huntington's disease; striatum; phrasal movement; locality; semantics

1. Introduction

A sentence exhibits *linear* order in terms of one word following another (e.g., pushed-the-fat-cat), as well as *hierarchical* order, in terms of phrases containing other phrases as parts. For example, the noun phrase (NP) [NP the [fat cat]] occurs as a part of the verb phrase (VP) [VP pushed [NP the [fat cat]]]. Constituents of phrasal hierarchies represent units of structure that function relatively independently in terms of the meaning and grammatical categories they encode. Thus, the above noun phrase can be moved across syntactic positions preserving its meaning and grammatical function, as when moving it to subject position (e.g., [$_{NP}$ the [fat cat]] was [$_{VP}$ pushed [$_{NP}$ …]]), or to a contrastive focus position (e.g. [$_{NP}$ the [fat cat]], he [$_{VP}$ pushed [$_{NP}$ …]]). The same applies pervasively to wh-phrases such as *what*, which move across the sentential subject in English (e.g. [$_{NP}$ what] (did) he [$_{VP}$ push [$_{NP}$ …]]). The neural basis for such 'chunking' of linear sequences of words into phrases and manipulating these through movement is unknown. Claims for the left inferior frontal gyrus to be involved in phrasal movement have long been made based on lesion and functional MRI (fMRI) studies (Ben-Shachar et al., 2004), but more recent evidence from fMRI based on movement vs. non-movement contrasts is less clear (Rogalsky et al., 2015; Europa et al., 2019). Longstanding evidence suggests that cortical-

subcortical loops involving the basal ganglia and more specifically the caudate nucleus of the striatum are also involved (Teichmann et al., 2005; Szalisznyo et al., 2017).

The present study aimed to use a disease-model of the striatum, Huntington's disease (HD), to shed new light on the cognitive control of phrase-structural hierarchies as built through movement. HD involves striatal degeneration as one of its earliest manifestations at the neural level (Bano et al., 2011) and has already featured prominently in studies of the role of the striatum in language (Jacquemot & Bachoud-Lévi, 2021). Early evidence showed that not lexical knowledge is affected in HD, but aspects of complex syntax such as passive constructions involving movement (Teichmann et al., 2005; Teichmann et al., 2008). Teichmann et al. (2008) specifically suggested that 'the role of the striatum in sentence processing specifically pertains to the application of syntactic movement rules' (p.174). In a later study, Teichmann et al. (2015) tested a group of 12 patients with frontal-striatal damage and found that impairments in comprehending non-canonical sentences (comprising passive sentences and object relative clauses) correlated with lesions and white matter connectivity in a fronto-striatal circuit. Szalisznyo et al. (2017) report a case series of patients with fronto-striatal damage following tumour resections who again showed significant impairment on non-canonical sentences (passive constructions and object relatives) relative to healthy controls. Within the patient group, non-canonical syntactic capacity correlated significantly with lesion load values of the Broca-caudate tracts, but not cortico-cortical tracts. Probing into a different but crucial aspect of syntactic hierarchies, García et al. (2018) found impairment on comprehension of sentences with embedded clauses, not only in symptomatic HD gene carriers, but even clinically unaffected first-degree relatives, who had not received genetic testing. Together, this evidence shows that certain syntactic tasks, such as those tapping into constituent structure hierarchies and their manipulation, show high sensitivity to conditions of striatal degeneration in HD. At the same time, syntax is not uniformly seen to be impaired in HD. Thus, in a control condition used by Sambin et al. (2007) involving long-distance morphological agreement, no impairment was seen.

In spontaneous narrative speech in HD, too, a pattern of 'flattening' the syntactic hierarchy has been observed, leading to a preference for mono-clausal units and coordination of clausal units as opposed to structures with a syntactic hierarchy (e.g. subordinated clauses), including in pre-symptomatic gene-carriers (Hinzen et al., 2018; Tovar et al., 2020). At the same time, in Tovar et al. (2020), a syntactic domain identified

as ‘concordance’ (capturing morphosyntactic agreement patterns) showed no significant group differences, demonstrating again that syntactic impairment in HD is quite selective: in spontaneous speech, agreement and other formal-syntactic errors are by no means prominent in early or even moderate HD.

Based on the linguistic profile of HD as reviewed above, we centered the present study on the notion of syntactic movement. We hypothesized that chunking sets of lexical items into meaningful chunks, embedding them as parts in a hierarchical structure, and moving such constituents within a given syntactic hierarchy, would be key to what language functions are under striatal control. We investigated this through an acceptability judgement task in which we manipulated whether sentential stimuli violated constraints on movement across phrasal boundaries. These constraints are arguably universal and have long formed a core subject of linguistic theory (Lasnik, 2017). We reasoned that sensitivity to such constraints would signal awareness of boundaries of such units of structure and what can be moved across them, and we predicted failure of such sensitivity in HD. To illustrate the constraints in question, in (1a), *the fridge* is the direct object of *fix*, while in (1b), the object is replaced by a *wh*-expression, which in (1c) is moved across a clausal boundary to the front:

(1) a. He told his worker [$_{\text{CLAUSE}}$ to fix [$_{\text{NP}}$ the fridge]]

b. He told his worker [$_{\text{CLAUSE}}$ to fix [what]]

c. *What* did [$_{\text{CLAUSE}}$ he tell his worker [$_{\text{CLAUSE}}$ to fix [...]]]

Clausal boundaries, however, cannot always be crossed in this fashion. Thus, in (2) and (3), the exact same procedure leads to the expressions (2c) and (3c), which are anomalous as indicated by the star:

(2) a. He left the flat [$_{\text{CLAUSE}}$ before fixing the [$_{\text{NP}}$ the fridge]]

b. He left the flat [$_{\text{CLAUSE}}$ before fixing [$_{\text{NP}}$ what]]

c. *What did [$_{\text{CLAUSE}}$ he leave the flat [$_{\text{CLAUSE}}$ before fixing [...]]]

(3) a. He left [before fixing the [$_{\text{NP}}$ the fridge]]

b. He left [before fixing the [$_{\text{NP}}$ what]]

c. *What did he leave [before fixing [...]]?

These data show that some phrasal configurations, but not others, constitute circumscribed domains beyond which constituents within them cannot be extracted or moved. Such 'locality' constraints (Lasnik, 2017) are not confined to wh-constructions. Thus, in Spanish, clitic pronouns are confined to a local domain with the head on which they depend. While in (4a), the clitic *la* is in an appropriately local configuration with the verb on which it depends, in (4b) it illegitimately 'climbs' to a higher position where it is not interpretable anymore:

(4a) No [$_{VP}$ supe que ya [$_{VP}$ **la** habías encontrado]]

not it knew that already you-have found

'I didn't know that you had already found it'

(4b) *No **la** [$_{VP}$ supe que ya [$_{VP}$ … habías encontrado]]

A more local exemplification of the same constraint is shown in (5), where the clitic pronoun in (5a) cannot be moved across the VP boundary, as shown in the unacceptable (5b):

(5a) Al [$_{VP}$ haber**lo** confesado], el delincuente ha sido juzgado

(5b) *Al **lo** [$_{VP}$ haber confesado], el delincuente ha sido juzgado

by it to-have confessed, the offender has been prosecuted

'Having confessed it, the offender has been prosecuted.

Finally, in Spanish, adjectives typically follow the noun phrases they modify, as in (6a), and they cannot cross over the NP boundary as in (6b):

(6a) la [$_{NP}$ casa [azul]]

the house blue

'The blue house'

(6b) la [azul] [$_{NP}$ casa …]

The four cases of movements illustrated above (i.e., wh-movement, clitic-climbing, verb-clitic and noun-adjective combinations), differ in how local the configuration is across which a constituent is moved: a whole clause in (1-3), a VP in (4-5), an NP in (6). Based on this difference, we further hypothesized that performance in

HD may show a gradient, with judgements on the more local structures being less affected. This pattern would be in line with a prominent view today that the role of the striatum in language processing depends on task demands and the amount of cognitive control required, with the caudate more activated when control demands and syntactic load are high and linguistic processing cannot be automatic (Copland et al., 2021; Giavazzi et al., 2018; Longworth et al., 2005; Progovac et al., 2018; Ye et al., 2012; Crinion et al., 2006). Mestres-Missé et al. (2012) specifically argued for an anterior-to-posterior gradient in the caudate reflecting the varying needs for cognitive control as a domain-general capacity, in which the more anterior/ventral portions of the caudate are only recruited as general cognitive demands increase (see also Mestres-Missé et al., 2017).

To assess the specificity of the linguistic profile of HD, we grouped all movement condition as 'syntactic', and contrasted these with sentential stimuli that only violated constraints of semantics, e.g. *I dried my shirt with water after running the marathon*. Such stimuli require processing meaning at the grammatical level of a full sentence, and do not involve a direct clash driven by incompatible lexical features, as e.g. in *The shoes ate*. Crucially, anomalies of this kind do not require any need to grasp constraints on phrasal movement. Although semantic processing under conditions of striatal damage has been less studied, there is some evidence of impairment in both verbal and nonverbal forms of semantics. Thus, García et al. (2018) found impairment in picture-based semantic association tasks in both patients and unaffected and genetically untested first-degree relatives. Crinion et al. (2006) reported activations in the left caudate during purely lexical semantic priming paradigms, when such priming exceeds a purely automatic level (i.e., when switches between languages are involved or with semantically unrelated words). At the level of sentential meaning, Ye et al. (2012) compared the processing of 'before-clauses' (e.g., *before the female dancer cancelled the show, the director fired the conductor*) with that of 'after'-clauses (e.g., *after the magician removed the bunny, the cameraman changed the film*). The former but not the latter require to reconstruct the temporal order of events as not matching the linear order of embedded and main clauses, and the contrast between them showed greater activation within the caudate nucleus. Semantic anomalies at the sentential level of the above kind have not previously been studied in HD to our knowledge.

Previous results in the domain of spontaneous speech did not suggest that domain-general cognitive deficits play a crucial explanatory role in linguistic anomalies seen (Hinzen et al., 2018; Tovar et al., 2020; see also Sambin et al., 2012). This suggests they are linguistically more specific. To explore this issue further, we also probed here into whether a range of neuropsychological test scores contributed to the predictions of performance on the acceptability judgement task. These came specifically from the domains of working memory, naming, comprehension, alternating attention, inhibition, and verbal, phonetic and semantic fluency.

In sum, the present study sought to illuminate the neural basis of building phrase-structural hierarchies through movements of constituents affecting the internal architecture of the hierarchical phrases that contain them. We predicted a loss of sensitivity to constraints on phrasal movement in people with HD, with a gradient seen ranging from less impairment on violations in more local phrase-structural configurations; and less or no impairment in semantically anomalous sentences in which there were no phrase-structural manipulations through movement. We hoped to illuminate the neural basis of phrasal hierarchies and movement, as well as shedding light on the specificity of the linguistic profile of HD, and its relation to non-linguistic neuropsychological domains.

2. Materials and methods

2.1 Participants

This is a multicentre cross-sectional study conducted between 2019 and 2021. Subjects were recruited from Hospital Mare de Déu de la Mercé and Hospital Clínic of Barcelona. This cohort consists of 31 identified gene-carriers patients matched to 31 neurotypical controls on age, gender, IQ (TAP, Test de Acentuación de Palabras, Gomar et al., 2011) and level of education (International Standard Classification of Education, ISCED). Healthy controls were recruited from volunteers. Controls had no reported neurological or psychiatric conditions, and did not use neuropsychiatric medications. All participants were native Spanish or Spanish-Catalan bilingual speakers.

Patients carried a diagnosis of HD and were participants of the ENROLL-HD study (CHDI Foundation, Inc.). Enroll-HD is a global clinical research platform designed to facilitate clinical research in HD. Core datasets are collected annually from all research participants as part of this multi-centre longitudinal observational study. Participants were

classified according to the motor subscale of Unified Huntington Disease Rating Scale (UHDRS-m, The Huntington Study Group, 1996) in premanifest individuals (UHDRS score below 10) and manifest HD patients (UHDRS ≥10). Using the Total Functional Capacity scale scores (TFC, The Huntington Study Group, 1996), manifest HD patients were subdivided into early-mid stages (TFC=13−7) and advanced stages (TFC = <7). In this cohort of 31 HD gene-carriers, 8 subjects were asymptomatic carriers, 16 patients were in an early-mid disease stage. The remaining 6 subjects were considered to be in an advanced stage of the disease.

All participants signed the informed consent before being included. This study was approved by the ethics committee of Universitat Pompeu Fabra, Hospital Mare de Déu de la Mercè (Germanes Hospitalàries) and Hospital Clínic.

Table 1. Demographic, genetic and clinical data

	Pre-manifest (N=8)	**Early-HD (N=16)**	**Advanced-HD (N=6)**	**Controls (N=31)**
Gender (M/F)	6/2	8/8	1/5	15/16
Age (mean/SD)	42.9 (5.9)	52.4 (13.1)	57.8 (6.8)	50.9 (11.6)
IQ (mean/SD)	103.2 (6.5)	102.3 (4.8)	100.3 (5.3)	106.75 (7.1)
Education in years (mean/SD)	14.2 (7.4)	11.7 (5.8)	10.8 (3.2)	13.9 (9.3)
CAG repetitions (mean/SD)	41.92 (1. 7)	42.1 (5.2)	43.6 (2.1)	-
UHDRS TMS* (mean/SD)	3.2. (4.5)	22.3 (7.3)	28.1 (8.2)	-
TFC** (mean/SD)	12.75 (0.6)	9.3 (2.4)	4.4 (1.6)	-

*UHDRS TMS: Unified Huntington Disease Rating Scale Total Motor Score

**TFC: Total Functional Capacity

2.2 Clinical and neuropsychological assessment

Demographic and clinical data were collected at the same time of sample collection, including age, gender, IQ, level of education, CAG repeat length, TFC and age of onset. Table 1 summarizes the demographic, genetic and clinical data from the subjects.

Participants were evaluated by both a trained neurologist and a psychologist. The following scales were administrated:

(1) The Total Motor Score (TMS) (score range 0-124) of the Unified Huntington's Disease Rating Scale (UHDRS; The Huntington Study Group, 1996).

(2) The Total Functional Capacity scale (TFC) (score range 0–13) of the UHDRS (The Huntington Study Group, 1996).

(3) Behavioral and psychiatric disturbances were evaluated using the short form of the Problem Behaviors Assessment scale (PBA-s) (McNally et al. 2015).

(4) The cognitive battery included: the Digit Span test forward and backward (Wechsler, 1981) measuring working memory, the Stroop Test (Golden & Freshwater, 2002) assessing naming and inhibition, the Digit Symbol Substitution Test (DSST; Wechsler, 1981) evaluating alternating attention. Participants also completed two verbal fluency tasks, one semantic fluency test (naming animals) and one phonetic fluency test (COWAT with letters FAS, from the Multilingual Aphasia Examination, Benton & Hamsher, 1976).

(5) Three subtests of the Boston Diagnostic Aphasia Examination test (BDAE; Goodglass & Kaplan, 1972, 1983) were administered to evaluate naming skills, and oral comprehension.

(6) Pre-morbid Intelligence Quotient (TAP) was evaluated by the Word Accentuation Test, with the Spanish version (Gomar et al., 2011).

Controls were tested with the same neuropsychological battery to compare their performance with the results of the HD sample.

2.3 Procedure and materials

Clinical and neuropsychological data were collected clinically at both hospitals under conditions of the ENROLL-HD study. Experimental linguistic data collection was conducted, whenever possible, at the Hospital Mare de Déu de la Mercè and the Hospital Clínic, Barcelona, otherwise in participants' homes or residences; particularly in participants with advanced HD with mobility difficulties. The experiment was performed in a quiet environment without distractions. Controls were tested in their own homes.

Stimuli 72 Spanish sentences in total, half of which (36) were anomalous. Anomalous sentences contained violations that were classified as either syntactic (24) or semantic (12). Syntactic violations were further subdivided into violations of locality principles on long-distance wh-movement (6), clitic climbing (6), verb-clitic configurations (6), and noun-adjective configurations (6). In the first of these, a clausal boundary blocks extraction of a *wh*-element from its original position as a verbal object (e.g.*¿*Qué se fue antes de arreglar?* tr. **What did he leave before fixing?*). In the second, e.g. **Lo intentó que comprara* (literal translation **It he tried that he buys*), lo as associated to the verb buy cannot climb to the initial position before the matrix verb intentó. Noun-adjective violations involved crossing over the NP, e.g.**Juan quería una negra chaqueta* (**Juan wanted a jacket black*), instead of *Juan quería una chaqueta negra*. Finally, in clitic+verb configurations, the clitic crosses over the VP **Se metieron en líos por lo cantar* (**They got in trouble for it singing*). Semantic errors (12) were syntactically well-formed sentences anomalous in sentence-level meaning, e.g.**Me sequé la camiseta con agua* (tr. **I dried my shirt with water*). Each incorrect item was matched with a correct item on the parameters of number of words and word frequency (Corpus del Español del siglo XXI, CORPES XXI, RAE). Complete stimulus material is provided in Appendix 2.

The task was designed as an anomaly detection task. Participants were instructed to listen to the sentences and say if they were anomalous or not. To explain the task, four training items were provided, two of them anomalous and two not. Sentences were presented orally and in two randomized orders.

2.4 Statistical analyses

In order to analyse the probability of correct responses across conditions and experimental groups, we fit a logistic mixed-effects regression using the 'lme4' package (version 1.1-27.1; Bates et al., 2014) in R (version 4.1.1; R Core Team, 2021). The random-effects structure included intercepts for Item and Participant. More complicated structures, including uncorrelated random slopes, led to singular fits. The fixed-effects structure included the sentence Condition and participant Group, as well as the interaction of the two variables. Condition was a categorical predictor with three levels: Correct, Semantic violation, and Syntactic violation. Group was a categorical predictor with three levels: healthy controls, early-symptomatic, and advanced-symptomatic. The pre-symptomatic group had to be dropped from the analysis, as they created separation in the model due to

a lack of incorrect responses. All categorical predictors were dummy coded, with the reference levels being healthy controls and correct sentences. Pairwise comparisons in the context of interactions were computed using the 'emmeans' package (version 1.6.3; Lenth, 2021), with Tukey adjustments being made for multiple comparisons.

A follow-up analysis of a subset of the items pertaining to the syntactic condition was carried out. This analysis contained items with syntactic violations as well as well-formed sentences that followed the same sentence structure. The four types of syntactic structures were modeled as four separate categories: Clitic-climbing, Noun-adjective, Verb-clitic, and wh-movement. Since these categories contained fewer items, the healthy controls could not be included due to issues of complete separation, and the early symptomatic group was represented by the global intercept in dummy coding. This model contained Group (Early vs advanced symptomatic), and the interaction between Condition (correct vs. incorrect) and the syntactic category (Clitic-climbing, Noun-adjective, Verb-clitic, and wh-movement). Pairwise comparisons between syntactic category and group were also computed in the same manner as described above.

To analyse whether the results obtained in the original analysis were influenced by a bias towards rejecting or accepting items as grammatical, we calculated the criterion location for each clinical participant. Criterion location is a construct from signal detection theory and can be used to characterize whether participants have a bias towards accepting or rejecting items in the experiment overall. A criterion location of 0 means that a participant has no bias, a negative value means the participant is biased towards accepting items and a positive value means participants are biased towards rejecting items. See Huang and Ferreira (2020) for an in-depth discussion of applications of signal detection theory to acceptability judgements. The criterion location was added to the model as a fixed effect along with Condition and Group. The interaction between criterion location and Condition was fit, as we expected any potential bias to differ among the levels of Condition. The interaction between Condition and Group was dropped in this model, as it did not contribute significantly to model fit as evidenced by a likelihood ratio test comparing the full model to a reduced model ($\chi 2(2)=1.67$, $p=0.4324$).

In addition to modelling the effects of condition and group with logistic regression, the second part of this analysis constituted an exploratory investigation into how the variables measured from the neuropsychological assessment influenced the outcome. As there were many potential variables of interest and a modest amount of data,

we elected to explore the relationship between these variables and the experimental outcome with random forest analysis employing conditional inference trees using the 'party' package (version 1.3-9; Strobl et al., 2007). This method is more appropriate to use when there are many explanatory variables and a small amount of data. The results of a random forest include a measure of importance to each variable, indicating the decrease in classification accuracy when that variable is removed from the model. For a discussion of the advantages of random forests, the reader is directed to Tagliamonte and Baayen (2012).

3. Results

Only significant differences in the pairwise comparisons are reported in the text that follows, with the full information for all models and pairwise comparisons being listed in Appendix 1.

3.1 Full experiment

The results of the model predicting correct responses from all groups by experimental conditions are reported in Table 2, and plotted in Figure 1. The intercept represents the log-odds of an accurate response for the healthy control group on correct sentences. As we used treatment coding, 'Condition: Semantically incorrect' and 'Condition: Syntactically incorrect' represent the estimated difference in log-odds of the probability of an accurate response for these two conditions sentences compared to correct sentences. As there is an interaction specified, this is only the difference for healthy controls. The next lines, 'Group: Early-sym' and 'Group: Advanced-sym' likewise represent the estimated difference between the healthy controls and these two clinical groups only for correct sentences. The interaction terms listed next represent the difference in the effect of semantic and syntactic errors for the two clinical groups compared to the difference estimated for the healthy controls. For example, to get the average log-odds probability of an accurate response to a syntactically incorrect sentence for the advanced symptomatic group, one would have to take the sum of the following terms: 'Intercept', 'Condition: Syntactically incorrect', 'Group: Advanced-sym', and 'Condition: Syntactically incorrect * Group: Advanced-sym'. Note that this table does not provide all of the comparisons that are of interest, so the comparisons provided in the following paragraph were calculated using the emmeans package with the Tukey adjustment as

described in section detailing the statistical analysis above. A full output of these following pairwise comparisons is presented in the Appendix.

Results show that for healthy controls, there were no significant differences between the different levels of Condition, as expected. For the early-symptomatic group, there were significantly more correct responses to correct sentences than those with syntactic violations ($p < .0001$) and semantic violations ($p = .0456$), and more correct responses to semantically incorrect sentences than syntactically incorrect sentences ($p < .0001$). For the advanced symptomatic group, correct sentences were responded to more accurately than those with syntactic violations ($p < .0001$) but not semantic violations ($p = .2347$), and sentences with semantic violations were responded to more accurately than syntactic violations ($p < .0001$).

Across groups, comparisons show that healthy controls were more accurate responding to correct sentences than the early-symptomatic group ($p = .0001$), and the advanced-symptomatic group ($p < .0001$). They were also more accurate on items with semantic violations than the early-symptomatic ($p < .0001$) and advanced-symptomatic groups ($p < .0001$). For syntactic violations, the control group was more accurate than the early-symptomatic ($p < .0001$) and advanced-symptomatic ($p < .0001$) groups. The only significant difference between the two clinical groups was that the early-symptomatic group was more accurate than the advanced-symptomatic group on items with syntactic violations ($p = .0017$).

Table 2. Regression table for the logistic mixed-effect regression modelling accuracy by experimental Condition interacting with Group. Correct sentences and healthy controls are represented by the intercept.

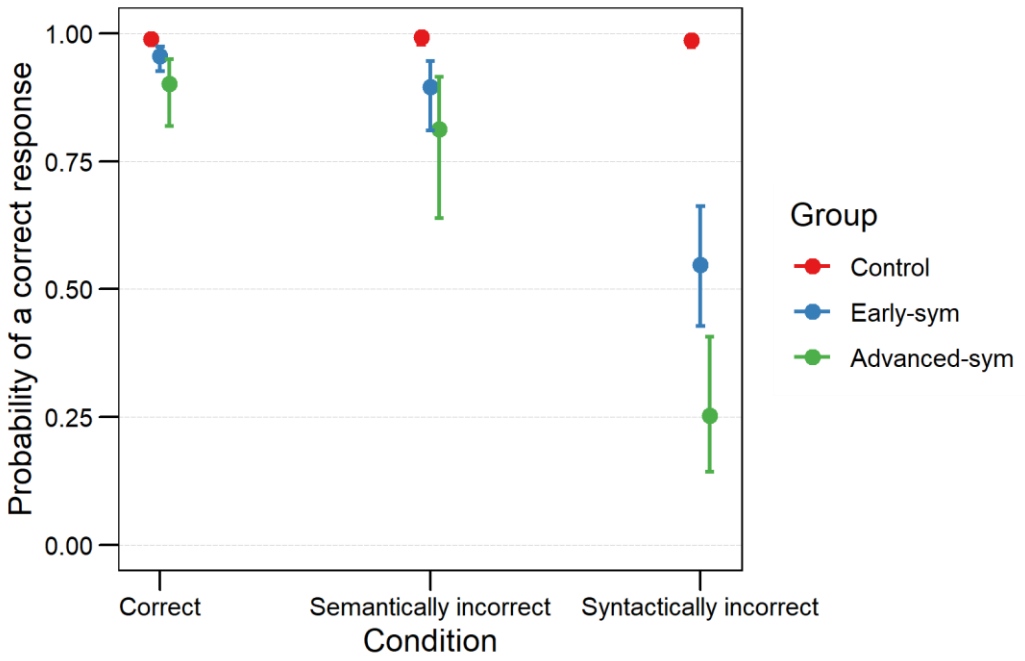
Predictors	*Estimate*	*Std. Error*	*95% CI*	*p*-value
(Intercept)	4.45	0.30	3.86 – 5.04	1.82e-49 (<.0001)
Condition: Semantically incorrect	0.37	0.58	-0.77 – 1.52	5.21e-01 **(.521)**
Condition: Syntactically incorrect	-0.23	0.41	-1.04 – 0.58	5.74e-01 (.574)
Group: Early-sym	-1.39	0.34	-2.05 – -0.73	4.03e-05 (<.0001)
Group: Advanced-sym	-2.24	0.41	-3.05 – -1.43	6.52e-08 (<.0001)

Condition: Semantically incorrect * Group: Early-sym	-1.29	0.58	-2.42 – -0.16	2.47e-02 (.025)
Condition: Syntactically incorrect * Group: Early-sym	-2.64	0.40	-3.43 – -1.85	6.79e-11 (<.0001)
Condition: Semantically incorrect * Group: Advanced-sym	-1.12	0.63	-2.35 – 0.11	7.55e-02 (.076)
Condition: Syntactically incorrect * Group: Advanced-sym	-3.06	0.46	-3.96 – -2.16	2.65e-11 (<.0001)

Random Effects

σ^2_{ItemID}	0.63
σ^2_{SubjID}	0.37

Figure 1. Predicted probabilities of a correct response for average participants and items by experimental condition and participant group. Whiskers correspond to 95% confidence intervals.



3.2 Bias-corrected analysis

To analyse how bias, operationalized as criterion location (CL), influenced participant responses, we fit a logistic mixed-effect regression with only clinical participants. The control group was not included, as the CL cannot be calculated for participants who do not make errors, which was common in the control group. As such, for the low and average levels of bias in the sample, correct sentences were responded to more accurately than syntactically incorrect sentences ($p < .0001$). At higher levels of bias, however, there were no statistically significant differences between the conditions.

In this model, there was no interaction of Group and Condition, as that did not contribute to model fit as evidenced by a likelihood ratio test ($\chi2(2)=1.677$, $p = .4324$). As such, the intercept represents the predicted probability in log-odds of an accurate response to a correct sentence for all participants. The estimate for 'Group: Advanced-sym' now represents the difference between the advanced- and early-symptomatic groups averaged across all conditions, while 'Condition: Semantically incorrect' and 'Condition: Syntactically incorrect' represent the difference between those two conditions and correct sentences for both clinical groups. The estimate for CL is the estimate for how average bias in responses affects the probability of accurate responses only to correct sentences, while the interaction of 'Condition: Semantically incorrect' and 'Condition: Syntactically incorrect' with CL is the difference in that effect for the other two experimental conditions.

Table 3. Regression table for the logistic mixed-effect regression modelling accuracy by experimental Condition interacting with Group controlling for bias, fitted to only the data from the two clinical groups. Correct sentences across all participants are represented by the intercept.

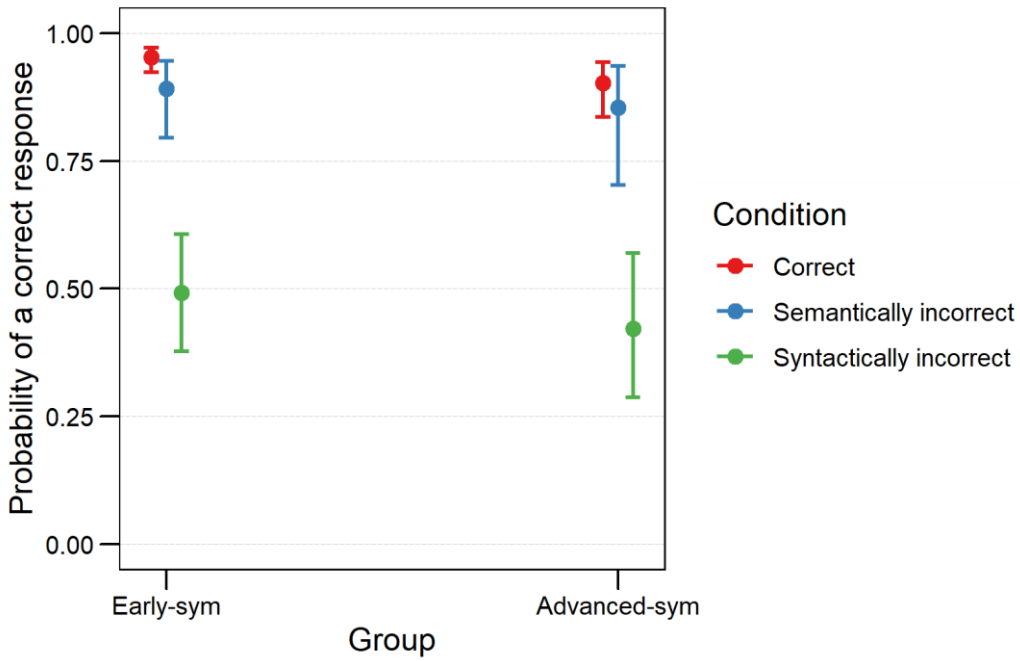
Predictors	*Estimate*	*Std. Error*	*95% CI*	*p*-value
(Intercept)	2.68	0.43	1.84 – 3.52	3.70e-10 (<.0001)
Group: Advanced-sym	-0.46	0.18	-0.81 – -0.11	9.85e-03 (.009)
Condition: Semantically incorrect	-1.51	0.69	-2.87 – -0.15	2.96e-02 (.029)
Condition: Syntactically incorrect	-6.34	0.58	-7.49 – -5.20	1.74e-27 (<.0001)

CL	0.41	0.72	-0.99 – 1.81	5.68e-01 (.568)
Condition: Semantically incorrect * CL	1.52	1.21	-0.86 – 3.89	2.10e-01 (.210)
Condition: Syntactically incorrect * CL	6.92	0.99	4.98 – 8.87	3.04e-12 (<.0001)

Random Effects

σ^2 ItemID	0.99
σ^2 SubjID	0.00

Figure 2. Predicted probabilities of a correct response for average participants and items for the three experimental conditions according to participant group, after accounting for bias. Whiskers represent 95% confidence intervals.



3.3 Syntactic sub-analysis

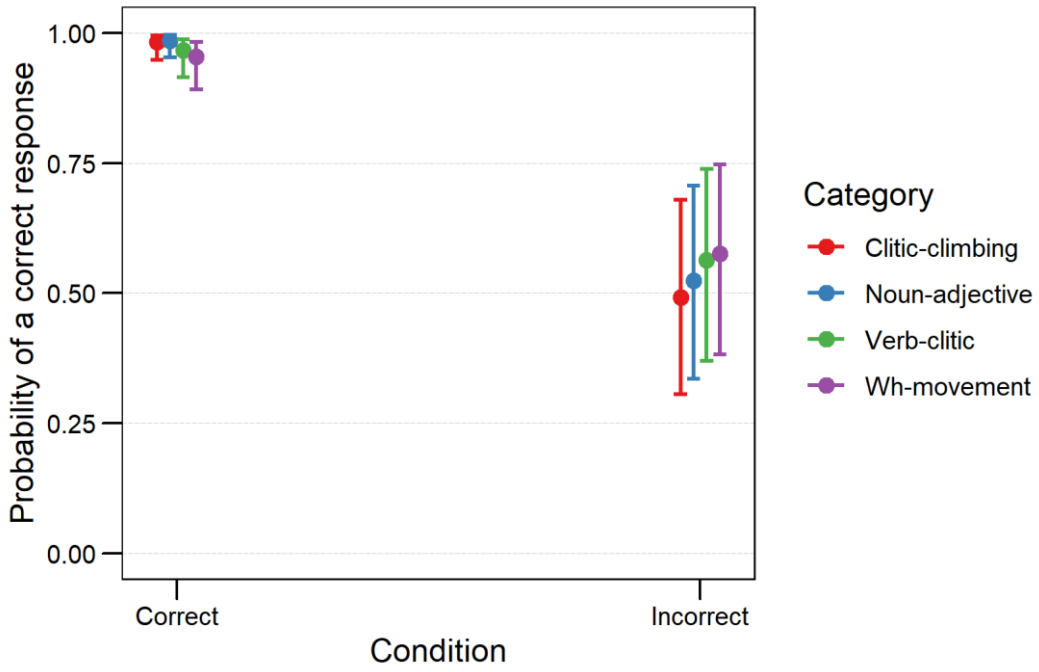
For all sub-categories of syntactic structure, correct sentences were responded to more accurately than sentences with a syntactic violation ($p < .0001$). For both correct and incorrect sentences, there were no significant differences between the sub-categories. Full pairwise comparisons between these conditions are again presented in the Appendix.

For this model, the intercept is representing the predicted accuracy for correct sentences that have a clitic-climbing structure. Due to the interaction, 'Condition: Incorrect' is the estimated difference in accuracy between correct and incorrect sentences for the clitic-climbing condition only. 'Group: Advanced-sym' is the predicted difference between the two clinical groups for all sentence types for correct and incorrect. The interaction terms are the difference in the estimated effects for incorrect as opposed to correct sentences.

Table 4. Regression table for the logistic mixed-effect regression modelling accuracy for syntactic sub-analysis, fitted to only the data from the two clinical groups. Correct sentences and clitic-climbing syntactic category are represented by the intercept.

Predictors	*Estimate*	*Std. Error*	*95% CI*	*p*-value
(Intercept)	4.01	0.57	2.89 – 5.13	2.36e-12 (<.0001)
Condition: Incorrect	-4.04	0.61	-5.24 – -2.85	3.52e-11 (<.0001)
Group: Advanced-sym	-1.19	0.50	-2.17 – -0.22	1.59e-02 (.016)
Condition: Incorrect * Category: Noun-adjective	-0.05	0.86	-1.74 – 1.65	9.57e-01 (.957)
Condition: Incorrect * Category: Verb-clitic	0.95	0.80	-0.62 – 252	2.34e-01 (.234)
Condition: Incorrect * Category: *Wh*-movement	1.31	0.79	-0.23 – 2.85	9.63e-02 (.096)
Random Effects				
σ^2 ItemID	0.38			
σ^2 SubjID	0.91			

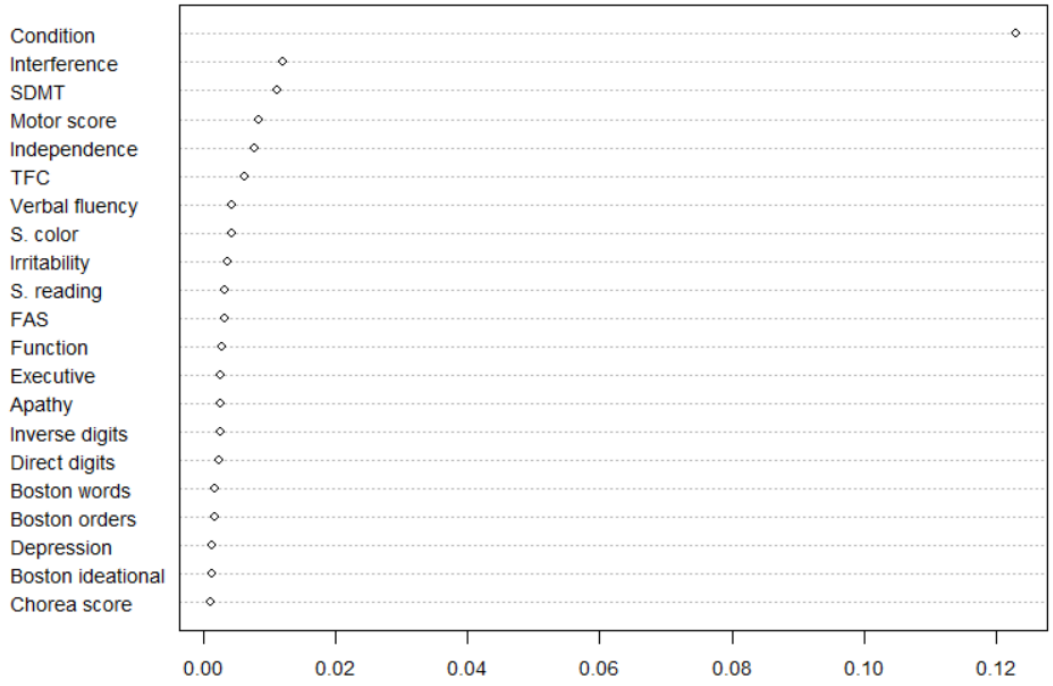
Figure 3. Predicted probabilities of a correct response for average participants and items for the four syntactic sub-conditions. Whiskers represent 95% confidence intervals.



3.4 Random Forest exploratory analysis

Results of the random forest analysis indicated that the experimental condition, found to be predictive of responses in the logistic regression analysis, was the most important variable in predicting correct responses. There were a small handful of other variables that may have contributed to distinguishing accurate from inaccurate responses (Figure 4). As we were interested in the effect of these variables, we visualize here unconditional inference trees, where the effect of other predictors is not accounted for when evaluating the importance of the variable in question. When conditional inference trees were employed, where the importance is calculated after accounting for all other variables in the model, all variables except for Condition dropped to negligible values (see Appendix).

Figure 4. Variable importance plot visualizing a measurement of how important predictors are to model performance. Larger values indicate larger decreases in predictive



4. Discussion

In this study we tested with a violation detection paradigm whether patients with HD and HD-gene carriers are sensitive to violations of locality constraints on movement, which govern when a constituent can be moved across a particular phrasal boundary. Our results demonstrate that such sensitivity is profoundly lost in patients with manifest HD at both disease stages distinguished here, and across all sub-conditions, including both the more and the less local violations and the associated forms of structural complexity. While a difficulty in processing noncanonical sentences (such as passives) in HD has been reported previously (Teichmann et al., 2015; Szalisznyo et al., 2017), the stimuli used here were crucially differed. These differences do not only relate to task demands (involving a metalinguistic judgement in our case), but also to the fact that violations of a syntactic constraint were involved in the present case. Very high performance on all of our stimuli in controls with little variation across this group shows that these results do not reflect any ambiguity or difficulty in these stimuli from the viewpoint of a neurotypical brain. In this sense, our result reflects the conscious control of grammar in the HD population, more than task difficulty. Moreover, no generic problem of judging complex linguistic stimuli per se, or performing metalinguistic judgements, was seen,

since correct sentences were largely judged correctly; even anomalous sentences, moreover, were not judged inaccurately across the board, since as long as the anomalies were semantic, they were judged significantly more accurately than those with syntactic anomalies. In short, it is more specifically when sentences are manipulated so as to violate a syntactic constraint of movement, that such violations are incorrectly tolerated. A basic apprehension of phrase-structural hierarchies is clearly maintained, but this apprehension crumbles once these are internally and illicitly manipulated so as to deviate from their expected configurations.

Despite this specificity, it would nonetheless be wrong to characterize this deficit as being specific to syntax (non-canonicity and movement). This is because semantics, though significantly less affected, was affected in both HD groups, though differences reached statistical significance only in the early symptomatic group. We conclude that the problem caused by damage to the cortico-striatal circuits in HD concerns the integration of information at the sentential level, both at the level of integrating a number of lexical concepts into a proposition, and in terms of manipulating phrasal structures syntactically through movement. Loss of control over the latter is both worse and worsens with disease progression; while the former problem was less and non-distinct across the disease stages distinguished here. Why does a semantic problem exist, even at early stages of HD? While this is a significant problem for further exploration, we suggest that it is because the semantic problem is not a lexical semantic one, but a problem of combining such concepts using syntactic means, and the sentence-level (propositional) meanings that arise from this.

Against our predictions of a gradient in performance, failure to notice when a particular phrasal boundary is illicitly crossed was global, affecting the manipulations of syntactic hierarchies through movement regardless of how local the violations are. If having to evaluate larger chunks of structure indicates an increase in cognitive cost, this result therefore again suggests that failure to recognize illicit phrasal movement is more specific and independent of general cognitive factors. Our exploratory random forests analysis further supports the conclusion of a more specific linguistic effect, to which non-linguistic neuropsychological predictors made no significant contributions. While our experimental design was not aimed to address the current controversy of whether the role of the striatum is language-specific in some sense, the pattern of the above findings does not support the idea that the caudate is more activated when control demands and

syntactic load are higher and linguistic processing cannot be automatic (Copland et al., 2021; Giavazzi et al., 2018; Longworth et al., 2005; Progovac et al., 2018; Ye et al., 2012; Crinion et al., 2006; Mestres-Missé et al., 2012). Indeed, in controls, judgements of the ungrammaticalities involved in our study are, if anything, highly automatic, and our stimuli did not exhibit semantic ambiguities or anomalous sentences with marginal grammaticality consisting of different judgements on whether they were really grammatical or not. It is arguable that our syntactic violations in some sense require more cognitive control than the semantic ones, yet this difference also co-varies with the structural linguistic differences involved, suggesting that the form of cognitive control required is linguistically indexed (Jacquemot & Bachoud-Lévi, 2021).

Our results are consistent with observations from spontaneous speech in HD of a loss of specific forms of hierarchical syntactic complexity (Gordon, & Illes, 1987; Murray 2000, Murray & Lenz, 2001; Hinzen et al., 2017; Tovar et al., 2020). As reviewed in the introduction, the last two of these studies noted a pattern of over-use of clausal coordination over clausal subordination beginning in the pre-symptomatic phase, which the authors in both cases interpreted as signaling a loss of hierarchical syntactic complexity through embedding. In both of these studies, pre-symptomatic patients also significantly differed in a domain of linguistic variables termed 'Reference', which covered anomalies in the referential use of noun phrases (NPs) as manifest in vague reference, hanging topics, abnormal topic sifts, missing determiners, and failed definite reference. Referential NPs are paradigmatic units of structure having an independent meaning and entering into phrase-structural hierarchies as arguments of predicates. In the fluency domain, this profile of lesser grammatical connectedness of clauses and referential failures was seen to correspond to a pattern of unfilled pauses, re-phrasings, and truncations in manifest HD patients, and of prolongations, filled pauses, and repetitions in pre-manifest gene carriers. Such breakage patterns appeared along clausal boundaries, suggesting again and at this level that sequencing and structuring syntactic information in such independently meaningful units was clearly affected in HD. Unlike these previous studies of spontaneous speech, which showed linguistic effects at several levels in pre-symptomatic gene carriers, the present study found no effect of HD at all in this group, whose patterns of judgement on both correct and incorrect patterns were neurotypical. It is noteworthy that, for this reason, subtle changes in spontaneous speech may carry an earlier signal of the disease, which is clinically important.

In sum, our results shed some light on the neural basis of building phrase hierarchies through the movement of constituents, confirming a crucial role of the striatum in the awareness of constraints on such hierarchies. Contrary to predictions, no gradience was seen depending on how local the syntactic violations of movement were, affecting movement-based anomaly detection generally and uniformly, while leaving semantic processing unaffected. Future studies should use syntactic movement paradigms in conjunction with neuroimaging, so as to further illuminate the brain basis of this ubiquitous phenomenon in language.

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Supplementary materials

Appendix 1

Full model reported in the Results section

```
Generalized linear mixed model fit by maximum likelihood (Laplace
Approximation) ['glmerMod']

 Family: binomial  ( logit )

Formula: Reponse ~ Condition * Group + (1 | ItemID) + (1 | SubjID)

   Data: d2

Control: glmerControl(optimizer = "bobyqa")


     AIC      BIC   logLik deviance df.resid

  1790.0   1858.9   -884.0   1768.0     3877


Scaled residuals:

     Min       1Q   Median       3Q      Max

-13.7179   0.0904   0.1232   0.2221   2.4288


Random effects:

 Groups Name        Variance Std.Dev.

 ItemID (Intercept) 0.6266   0.7916

 SubjID (Intercept) 0.3723   0.6102

Number of obs: 3888, groups:  ItemID, 72; SubjID, 54


Fixed effects:

                                                 Estimate Std. Error
z value Pr(>|z|)

(Intercept)                                       4.4488     0.3008
14.788  < 2e-16 ***

ConditionSemantically incorrect                   0.3748     0.5843
0.641   0.5212

ConditionSyntactically incorrect                 -0.2322     0.4133
-0.562   0.5743

GroupEarly-sym                                   -1.3875     0.3379
-4.106 4.02e-05 ***

GroupAdvanced-sym                                -2.2385     0.4142
-5.405 6.49e-08 ***
```

```
ConditionSemantically incorrect:GroupEarly-sym      -1.2922     0.5753
-2.246   0.0247 *

ConditionSyntactically incorrect:GroupEarly-sym     -2.6376     0.4042
-6.526 6.77e-11 ***

ConditionSemantically incorrect:GroupAdvanced-sym   -1.1182     0.6291
-1.777   0.0755 .

ConditionSyntactically incorrect:GroupAdvanced-sym  -3.0583     0.4588
-6.666 2.64e-11 ***

---

Signif. codes:  0 ‘***’ 0.001 ‘**’ 0.01 ‘*’ 0.05 ‘.’ 0.1 ‘ ’ 1
```

Full model pairwise comparisons for Condition

```
Group = Control:

 contrast                                          estimate    SE  df
z.ratio p.value

 Correct - Semantically incorrect                    -0.375 0.584 Inf
-0.641  0.7972

 Correct - Syntactically incorrect                    0.232 0.413 Inf
0.562  0.8404

 Semantically incorrect - Syntactically incorrect     0.607 0.608 Inf
0.999  0.5775


Group = Early-sym:

 contrast                                          estimate    SE  df
z.ratio p.value

 Correct - Semantically incorrect                     0.917 0.385 Inf
2.380  0.0456

 Correct - Syntactically incorrect                    2.870 0.300 Inf
9.572  <.0001

 Semantically incorrect - Syntactically incorrect     1.952 0.378 Inf
5.167  <.0001


Group = Advanced-sym:

 contrast                                          estimate    SE  df
z.ratio p.value

 Correct - Semantically incorrect                     0.743 0.457 Inf
1.626  0.2347

 Correct - Syntactically incorrect                    3.290 0.366 Inf
8.982  <.0001

 Semantically incorrect - Syntactically incorrect     2.547 0.463 Inf
5.499  <.0001
```

```
Results are given on the log odds ratio (not the response) scale.
P value adjustment: Tukey method for comparing a family of 3 estimates
```

Full model pairwise comparisons for Group

```
Condition = Correct:
 contrast                    estimate    SE  df z.ratio p.value
 Control - (Early-sym)          1.387 0.338 Inf   4.106  0.0001
 Control - (Advanced-sym)       2.239 0.414 Inf   5.405  <.0001
 (Early-sym) - (Advanced-sym)   0.851 0.397 Inf   2.143  0.0814

Condition = Semantically incorrect:
 contrast                    estimate    SE  df z.ratio p.value
 Control - (Early-sym)          2.680 0.538 Inf   4.979  <.0001
 Control - (Advanced-sym)       3.357 0.614 Inf   5.470  <.0001
 (Early-sym) - (Advanced-sym)   0.677 0.469 Inf   1.443  0.3187

Condition = Syntactically incorrect:
 contrast                    estimate    SE  df z.ratio p.value
 Control - (Early-sym)          4.025 0.347 Inf  11.612  <.0001
 Control - (Advanced-sym)       5.297 0.436 Inf  12.153  <.0001
 (Early-sym) - (Advanced-sym)   1.272 0.369 Inf   3.446  0.0017

Results are given on the log odds ratio (not the response) scale.
P value adjustment: tukey method for comparing a family of 3 estimates
```

Bias-control model

```
Generalized linear mixed model fit by maximum likelihood (Laplace
Approximation) ['glmerMod']
 Family: binomial  ( logit )
Formula: Reponse ~ Group + Condition + CL + CL:Condition + (1 |
SubjID) +      (1 | ItemID)
   Data: clinical
Control: glmerControl(optimizer = "bobyqa")
```

```
     AIC      BIC   logLik deviance df.resid
  1265.8   1314.5   -623.9   1247.8     1645

Scaled residuals:
    Min      1Q  Median      3Q     Max
-4.8750 -0.0541  0.2230  0.3425  4.9265

Random effects:
 Groups Name        Variance Std.Dev.
 ItemID (Intercept) 0.987083 0.99352
 SubjID (Intercept) 0.002612 0.05111
Number of obs: 1654, groups:  ItemID, 72; SubjID, 23

Fixed effects:
                                    Estimate Std. Error z value
Pr(>|z|)
(Intercept)                          2.6815     0.4279   6.266 3.70e-
10 ***
GroupAdvanced-sym                   -0.4612     0.1787  -2.581
0.00985 **
ConditionSemantically incorrect     -1.5094     0.6940  -2.175
0.02963 *
ConditionSyntactically incorrect    -6.3414     0.5838 -10.863  < 2e-
16 ***
CL                                   0.4087     0.7152   0.572
0.56764
ConditionSemantically incorrect:CL   1.5184     1.2114   1.253
0.21008
ConditionSyntactically incorrect:CL  6.9230     0.9924   6.976 3.04e-
12 ***
---
Signif. codes:  0 ‘***’ 0.001 ‘**’ 0.01 ‘*’ 0.05 ‘.’ 0.1 ‘ ’ 1
```

Bias-control models pairwise for Condition by Criterion location

```
Criterion Location = 0.213:
 contrast                                          estimate    SE  df
z.ratio p.value
```

```
 Correct - Semantically incorrect                    1.19 0.511 Inf
2.321  0.0529

 Correct - Syntactically incorrect                   4.87 0.426 Inf
11.437  <.0001

 Semantically incorrect - Syntactically incorrect    3.68 0.525 Inf
7.016  <.0001


Criterion Location = 0.5:

 contrast                                          estimate    SE  df
z.ratio p.value

 Correct - Semantically incorrect                    0.75 0.418 Inf
1.796  0.1710

 Correct - Syntactically incorrect                   2.88 0.327 Inf
8.788  <.0001

 Semantically incorrect - Syntactically incorrect    2.13 0.427 Inf
4.982  <.0001


Criterion Location = 0.975:

 contrast                                          estimate    SE  df
z.ratio p.value

 Correct - Semantically incorrect                  0.0295 0.749 Inf
0.039  0.9991

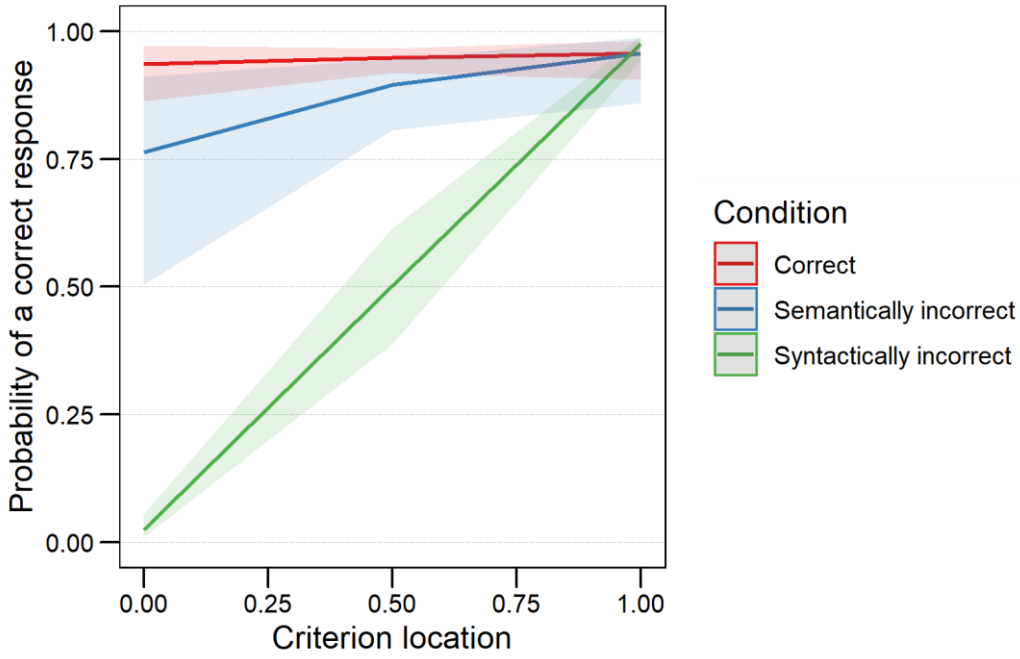
 Correct - Syntactically incorrect                -0.4062 0.584 Inf
-0.696  0.7661

 Semantically incorrect - Syntactically incorrect -0.4358 0.754 Inf
-0.578  0.8321


Results are averaged over the levels of: Group

Results are given on the log odds ratio (not the response) scale.

P value adjustment: tukey method for comparing a family of 3 estimates
```



Bias-control models pairwise for Group by Condition

```
Condition = Correct:
 contrast                      estimate    SE  df z.ratio p.value
 (Early-sym) - (Advanced-sym)     0.774 0.304 Inf   2.548  0.0108

Condition = Locality violation:
 contrast                      estimate    SE  df z.ratio p.value
 (Early-sym) - (Advanced-sym)     0.265 0.353 Inf   0.751  0.4524

Condition = Semantic violation:
 contrast                      estimate    SE  df z.ratio p.value
 (Early-sym) - (Advanced-sym)     0.340 0.405 Inf   0.840  0.4007

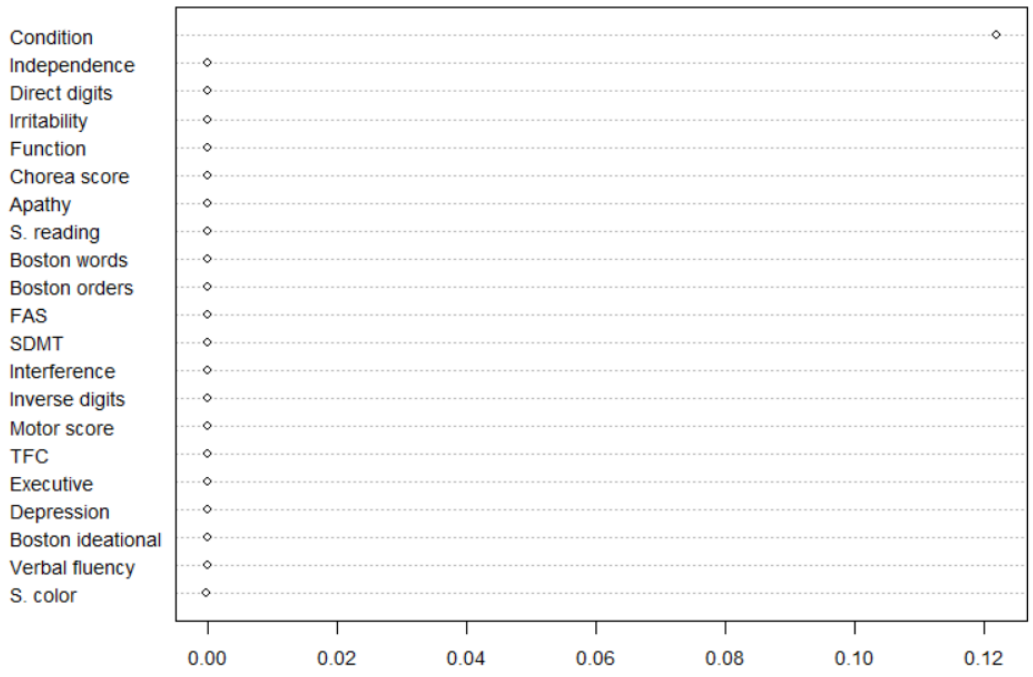
Condition = Word order violation:
 contrast                      estimate    SE  df z.ratio p.value
 (Early-sym) - (Advanced-sym)     0.307 0.378 Inf   0.811  0.4174

Results are given on the log odds ratio (not the response) scale.
```

Random forest importance values

The following are the importance values and the corresponding plot for the random forest using conditional vs. unconditional inference trees:

Predictor	Conditional tree importance	Unconditional tree importance
Condition	1.247835e-01	0.122876289
Independence	4.467354e-05	0.007869416
Direct digits	0.000000e+00	0.002426117
Irritability	1.030928e-05	0.003618557
Function	0.000000e+00	0.002924399
Chorea score	0.000000e+00	0.001226804
Apathy	5.498282e-05	0.002628866
S. reading	-6.872852e-06	0.003240550
Boston words	0.000000e+00	0.001814433
Boston orders	2.405498e-05	0.001714777
FAS	0.000000e+00	0.003182131
SDMT	5.841924e-05	0.011103093
Interference	2.405498e-05	0.012065292
Inverse digits	1.718213e-05	0.002587629
Motor score	1.374570e-05	0.008395189
TFC	3.092784e-05	0.006243986
Executive	0.000000e+00	0.002725086
Depression	-6.872852e-06	0.001408935
Boston ideational	0.000000e+00	0.001319588
Verbal fluency	3.780069e-05	0.004367698
S. color	0.000000e+00	0.004268041



Appendix 2

Experimental setting: anomalies detection task

Training stimuli

(1) *¿Qué plato cocinó el chef y Juan puso la mesa?

(2) Limpió la mancha con detergente.

(3) ***Compró la roja sandía.

(4) Ana sugirió que lo comprara para su cumpleaños.

Condition	Subcondition	Grammatical	Ungrammatical
Syntactic violations (24)	**Long distance *wh*-movement**	¿De qué dijo Ana que vendría disfrazada hoy?	*¿A quién que tengan mucho miedo te sorprende mucho?
		¿Cómo supieron que el ladrón entró en la casa?	*¿Qué se fue a casa antes de arreglar?
		¿Por qué la enfermera le acusó de mentir?	*¿Dónde no sabes exactamente quién compró el pan?
		¿Dónde pensó que la profesora guardaría los exámenes?	*¿Qué monumento fotografió Ana y Juan visitó el castillo?
		¿Cuándo sugieren que compremos los billetes a Portugal?	*¿Qué coche abofeteó Laura al hombre que estaba conduciendo?
		¿Qué creía Juan que María querría como regalo?	*¿A dónde votó Ana la propuesta de que nos fuéramos?
	Clitic climbing	Juan lo intentó comprar como regalo de cumpleaños.	*Lo intentó que comprara como regalo de cumpleaños.
		Ella la quiere ver en el cine de abajo.	*La quiso que viera en el cine de abajo.
		Su padre lo consiguió encontrar en tres segundos.	*Su padre lo consiguió que encontrara en tres segundos y sin quejas.
		La busqué y no la supe encontrar nunca más.	*La busqué y no la supe que ya habías encontrado.
		Los policías lo consiguieron atrapar gracias a las pruebas.	*Los policías lo consiguieron que atraparan gracias a las pruebas.
		Ana lo quiso besar en la primera cita.	

			*Ana lo quiso que besara en la primera cita.
	Noun-adjective	A Ana le gusta esa camiseta azul que le dieron para Navidad. La madre prefiere encontrar un vestido verde para ir de fiesta. El bolso naranja está sobre la estantería que compré ayer. Busca una botella verde para el regalo de tu madre. Le gusta leer libros de adolescentes durante las vacaciones de Navidad. Mira la camiseta rosa qué bonita es.	*La azul casa que hay en la calle en la que te conocí. *Hay un tan gris cielo en este momento que me gustaría fotografiarlo. *Siempre quise tener una plateada silla para sentirme como los ricos. *Laura ansiaba comprar una negra chaqueta con su propio dinero. *El niño quería que su padre le regalara un rojo coche.
	Verb-clitic	Los detectives buscan al hombre para poder acusarlo. Lo compraré para nuestra nueva casa en la playa. Al haberlo confesado, el delincuente ha sido juzgado por la justicia. Por haberla amenazado, el empleado seguramente perderá el trabajo. Al decirlo, el deseo no se va a cumplir. María lo necesita para ir al colegio mañana.	*Por lo haber hecho, Ana fue castigada. *Se metieron en líos por lo cantar. *Las profesoras quiérenlo para enseñar mejor. *Al lo haber contado, llamaron a Luis chivato durante meses. *Los policías búscanlo para poder meterlo en la cárcel. *La rosa pared de mi habitación ya ha sido repintada. *Nunca supieron dónde encontrar la gris libreta.
Semantic violations (12)		Cosí el disfraz con hilo rojo para disimular el horrible agujero. Laura borró la falta con típex tras comprobar que no se notaba. Ayer cortó la cartulina con unas tijeras que había robado de la oficina. La abuela calentó la comida de anteayer en el microondas nuevo.	*Sequé mi camiseta usando agua después de correr la maratón. *Escribió la carta con una goma porque quería dársela en mano. *Ana decidió peinar a su hermano con un cúter. *Corté la hoja que me dieron en el médico en un trozo.

	Pegó los dibujos en el álbum con ese pegamento. Limpiaron la cocina con agua y jabón. Cortan pescado con el cuchillo jamonero. Apagaron el fuego con agua. Sequé la ropa en la secadora. Endulzaron el pastel con azúcar. Rompió mi corazón en mil pedazos. Se mojaron la ropa en el río.	*El jarrón que compraste se rompió en un pedazo. *Los bomberos sofocaron el fuego con gasolina. *El barco navega por el asfalto. *Apagaron la vela con fuego . *Cosieron la camiseta con unas tijeras. *Limpié la casa con suciedad. *El avión aterrizó en el aire.

5. STUDY 3: UNDERSTANDING OF REFERENTIAL STRUCTURES IN HUNTINGTON'S DISEASE

Abstract. Whether one referential noun phrase (NP) refers to the same entity as another NP is well-known to be subject to a number of linguistic constraints. As classically formalized in the binding theory (BT), these constraints make reference to the larger phrases ('domains') of which these NPs form parts and confine referential dependencies to them ('locality'). To explore a potential subcortical neural basis for reference, we tested sensitivity to the three classical principles of the BT in early and advanced patients with Huntington's disease (HD), which involves early neural degeneration in the basal ganglia. A sentence-picture matching task was designed in which simple (e.g. *Mary washed him*) or complex sentences (e.g. *He smiled before John entered*) either matched pictures or not, depending on whether constraints of the BT were violated or not. Results from mixed effect logistic regression models showed that regardless of the specific condition of the BT, the complexity, and the correctness of the stimuli, both HD groups significantly underperformed relative to controls, which were at ceiling. The estimated probability of a correct response specifically on correct (matching) stimuli fell to around 50% in both HD groups. A bias for rejecting stimuli was observed, which increased from early to advanced HD. A new model taking this bias into account suggested that differences in performance between the early and advanced HD groups disappeared, for all stimulus types. These new results confirm earlier evidence for a loss of sensitivity to syntactic locality constraints in patients with basal ganglia damage, but newly suggest that these specifically affect the processing and understanding of referential dependencies, when these are grammatically constrained.

1. Introduction

Establishing referential dependencies is a fundamental precondition for referring to events in language: if an event involves two entities, we need to know whether one is the same as the other, and if we tell a story, it will involve several events, where at least one entity needs to recur and be identifiable as the same. Tracking these dependencies is subject to a number of constraints, both general cognitive ones (e.g., working memory) and apparently more specific formal-syntactic ones. These latter regulate when coreference is grammatically obligatory, permitted, or blocked. Classically these constraints have formed a cornerstone of linguistic theory, where they were systematized through three principles of the 'Binding Theory' (BT, Chomsky, 1981). The first principle

(A) governs the fact that, in *John shaved himself*, the reflexive anaphor *himself* can only refer to John, while in *John said that Mary shaved himself*, it cannot do so, even though *himself* agrees in both gender and number with John and cannot refer to Mary. The second principle (B) regulates that in *John shaved him*, the pronoun him cannot refer to John, but it can refer to Bill in *Bill said John shaved him*, even though Bill is further away in linear terms from the pronoun than John is. Finally, an instance of principle C is that in *He said John shaved him*, the pronoun *he* cannot refer to John: coreference is blocked.

While details for precisely formulating and for explaining these three principles are complex and differ greatly across frameworks (Reuland, 2011), crucial to our present purposes is only the fact that they all arguably relate to, and depend on, a fundamental design feature of human language, namely the *locality* of syntactic operations (which may reduce further to general principles of economy or computational efficiency in language design, see Reuland, 2011; Chomsky, 1995). While there are a number of different attempts to formally characterize the relevant notion of a *local domain*, a simplification sufficient for our purposes is that principle A requires the reflexive anaphor to be bound to its referent *within the clause* it occurs in. This would be why *himself* is bound to *John* in *John shaved himself*, but cannot be bound to *John* in *John said that [Mary shaved himself]* (with the local domain indicated in square brackets). Principle B, by contrast, requires a pronoun to be 'free' in its own local domain, i.e. to be bound *outside* of it. In turn, principle C requires the referential expression occurring as subject in the embedded clause to be free in its own domain, e.g. *John* in *He said [John shaved him]*. By referencing locality in each of its principles, the BT proves to be crucially sensitive to the phrase-structural hierarchies within which referential expressions occur and function.

The basis for the study reported here is that the foregoing facts make a prediction for the processing of referential dependencies in populations whose grasp of phrase-structural hierarchies is weakened. One of these are people with Huntington's disease (HD). At a neural level, HD involves neurodegeneration in the striatum as one of its earliest signs (Bano et al., 2011). As such it has already played a major role in studies of the contribution of the striatum to language processing (Jacquemot & Bachoud-Lévi, 2021). While the nature of this contribution remains controversial, earlier evidence from the neurotypical brain has suggested that the striatum plays a key role in linguistic articulation (Wildgruber et al., 2001; Wise et al., 1999), syntactic processing (Moro et al. 2001), lexical processing (Friederici and Kotz, 2003; Kotz et al. 2002), lexical retrieval

(Rosen et al. 2000) and handwriting (Siebner et al. 2001). Within HD, a replicated finding is that striatal damage affects the capacity to process 'noncanonical' sentences, in which particular phrasal constituents are moved to different places in a given phrase-structural hierarchy, as in the case of passives (Teichmann et al., 2008; Teichmann et al., 2015; Szalisznyo et al., 2017). A more recent view is that the role of the striatum is of a more domain-general nature and its involvement depends on task demands and the level of cognitive control required, with the caudate more activated when control demands and syntactic load are high and processing cannot be automatic (Copland et al., 2021; Giavazzi et al., 2018; Longworth et al., 2005; Progovac et al., 2018; Ye et al., 2012; Crinion et al., 2006; Mestres-Misse et al., 2012). Both views entail, though on different grounds, that the grammatical system is weakened under conditions of striatal damage.

The research question of the present study was whether this weakening affects referential processing as depending on grammatical and phrase-structural constraints. Reference is not a domain so far targeted in studies of HD. Sensitivity to the three constraints of the BT in patients with HD is an appropriate inroad into this issue. Several lines of evidence already support the prediction that sensitivity to the BT could be weakened in HD. First, Sambin et al. (2012) directly tested whether people with HD were sensitive to principle C of the BT. The target of this study was syntactic impairment as compared to working memory, rather than referential processing. Yet the authors found, controlling for working memory limitations, that the patients correctly established name-pronoun coreference when this was grammatically licensed, and at long distances, but failed to block coreference when it contradicted principle C (e.g. coreference of *he* and *Paul* in *He smiled when Paul entered*). In line with the traditional conception of the BT as reflecting narrowly syntactic constraints specific to the linguistic domain, the authors concluded that there is a specific role for the striatum in syntactic processing. This leaves open the question of whether the problem is one with syntax as such or, more specifically, with referential dependencies as subject to grammatical constraints, crucially including locality as reviewed above. In the latter case, failure of sensitivity to condition C should extend to the other conditions of the BT, which we tested here.

A second piece of preliminary evidence is that a recent study directly explored sensitivity in people with HD to violations of locality principles in grammar, using a grammaticality judgement task (Tovar et al., 2022). As Teichmann et al. (2018) had specifically suggested, 'the role of the striatum in sentence processing specifically

pertains to the application of syntactic movement rules' (p.174). But such movement rules, like binding, are universally subject to locality constraints. In their study, therefore, Tovar et al. (submitted, June 2022) manipulated sentential stimuli so as to contain violations of locality. They found that patients with HD (N=31) at all disease stages lacked sensitivity to such manipulations. The third and final piece of evidence comes from linguistic patterns in spontaneous speech, which show elevated levels of referential anomalies even in pre-symptomatic gene carriers (Hinzen et al., 2018; Tovar et al., 2020). Together, these three pieces of evidence motivate the exploration of the BT in HD and they provide a basis for the prediction that a generalized referential processing problem will be encountered in this group.

2. Materials and Methods

2.1 Participants

This is a multicentre cross-sectional study conducted between 2019 and 2021. Subjects were recruited from Hospital Mare de Déu de la Mercé and Hospital Clínic. This cohort consists of 31 identified gene-carriers patients matched to 31 neurotypical controls on age, gender, IQ (TAP, Test de Acentuación de Palabras, Gomar et al., 2011) and level of education (International Standard Classification of Education, ISCED). Healthy controls were recruited from volunteers. Controls had no reported neurological or psychiatric conditions, and did not use medications. All participants were native Spanish or Spanish-Catalan bilingual speakers.

Patients carried a diagnosis of HD and were participants of the ENROLL-HD study (CHDI Foundation, Inc.). Participants were classified according to the Total Functional Capacity Scale (UHDRS; The Huntington Study Group, 1996). HD stages were established as follows: pre-manifest stage: TFC=13−11; early stage: TFC=10−7; advanced stage: TFC = <7. In this cohort of 31 HD gene-carriers, 8 subjects presented with a score of >11 in the TFC scale (henceforth referred to as 'Pre-manifest'), 16 of the 31 patients (which will be referred to below as 'Early-HD') identified technically by a score between 7-11 on the TFC scale. The remaining 6 subjects (below referred to as 'Advanced-HD') were in later stages of the disease (<7 on the TFC scale).

Demographic and clinical data were collected at the same time of sample collection, including age, gender, IQ, level of education, CAG repeat length, TFC and

age of onset. Table 1 summarizes the demographic, genetic and clinical data from the subjects.

The relevant information about the study and the methodology was provided to all subjects. In addition, all participants signed the informed consent before being included. This informed consent was approved by the ethics committee of Universitat Pompeu Fabra, Hospital Mare de Déu de la Mercè (Germanes Hospitalàries) and Hospital Clínic.

Table 1: Demographic, genetic and clinical data

	Pre-manifest (N=8)	**Early-HD (N=16)**	**Advanced-HD (N=6)**	**Controls (N=31)**
Gender (M/F)	6/2	8/8	1/5	15/16
Age (mean/SD)	42.9 (5.9)	52.4 (13.1)	57.8 (6.8)	50.9 (11.6)
IQ (mean/SD)	103.2 (6.5)	102.3 (4.8)	100.3 (5.3)	106.75 (7.1)
Education in years (mean/SD)	14.2 (7.4)	11.7 (5.8)	10.8 (3.2)	13.9 (9.3)
CAG repetitions (mean/SD)	41.92 (1. 7)	42.1 (5.2)	43.6 (2.1)	-
UHDRS TMS* (mean/SD)	3.2. (4.5)	22.3 (7.3)	28.1 (8.2)	-
TFC** (mean/SD)	12.75 (0.6)	9.3 (2.4)	4.4 (1.6)	-

*UHDRS TMS: Unified Huntington Disease Rating Scale Total Motor Score
**TFC: Total Functional Capacity

2.2 Clinical and neuropsychological assessment

A trained specialist evaluated all the HD participants following the parameters of the ENROLL-HD study. The motor condition was assessed through the motor and functional sections of the Unified Huntington's Disease Rating Scale (UHDRS; The Huntington Study Group, 1996). Functional capacity was evaluated using the UHDRS Total Functional Capacity (TFC; score range 0–13) and motor dysfunction was evaluated using the UHDRS Total Motor Score (TMS, score range 1–124). Behavioral and psychiatric

disturbances were evaluated using the short form Problem Behaviors Assessment (PBA-s; maximum score 160). The cognitive battery included: the Digit Span test forward and backward (Wechsler, 1981) measuring working memory, the Stroop Test (Golden & Freshwater, 1978) assessing naming and inhibition, the Digit Symbol Substitution Test (DSST; Wechsler, 1981) evaluating alternating attention. Additionally, three subtests of the Boston Diagnostic Aphasia Examination test (BDAE; Goodglass & Kaplan, 1972, 1983) were administered to evaluate naming skills, and oral comprehension. Participants also completed two verbal fluency tasks, one semantic fluency test (naming animals) and one phonetic fluency test (COWAT with letters FAS, from the Multilingual Aphasia Examination, Benton & Hamsher, 1976). Lastly, pre-morbid Intelligence Quotient (TAP) was evaluated by the Word Accentuation Test, with the Spanish version. Controls were tested with the same neuropsychological battery to compare their performance with the results of the HD sample.

2.3 Procedure and test materials

Clinical and neuropsychological data collection were conducted at the Hospital Mare de Déu de la Mercè and the Hospital Clínic, Barcelona, under conditions of the ENROLL-HD study. Whenever possible, linguistic data was collected in both hospitals, otherwise in participants' homes or residences, particularly in elderly subjects or participants with HD mobility difficulties. In these specific cases, the experiment was performed in a quiet environment without distractions. Controls were tested in their own homes.

Comprehension of BT constraints was assessed through a sentence-picture matching task, broadly following the design used by Sambin et al. (2012) for condition C of the BT. Sentences were presented orally in two randomized orders. Subjects were instructed to listen to the sentences and say if they matched with the pictures that they saw on a computer screen. The three principles of the BT were tested with 8 sentence stimuli each, which resulted in 24 sentence stimuli, each of which was tested three times: (i) When the picture did not match the sentence, corresponding to an interpretation that the sentence did not have due to its syntactic constraints, (ii) When the picture did match the sentence, (iii) When the picture violated the direction of the action, i.e. instead of A doing something to B, B did something to A. The third condition was added as an attentional control condition, in which principles of the BT were not at stake. The total number of trials was therefore 72.

Unlike in Sambin et al., and in order to explore a potential complexity effect, half of the 72 trials (i.e., 36) involved 'simple' sentences, in the technical sense that they involved a single clause with no embedding. These clauses had a mean length of words of 3.6. (e.g. *Ana se estaba bañanado,* tr. *Ana was bathing (herself)*), while 'complex' sentences were built with a mean length of 5.3 words, and contained one matrix verb in the main clause and a subordinated clause with a dependent verb (e.g. *Pensaba que Ana se estaba bañanado*, tr. *He thought Ana was bathing (herself)*). Principle A violations involved contexts where a reflexive anaphor forces a referential dependence between itself and a referential NP in the same clausal domain (e.g. *$John_i$ is combing $himself_j$*, where subscripting indicates coreference and stars indicate ungrammaticality), and hence a picture where John is combed by someone else has to be rejected. Principle B violations involved sentences where the co-referential interpretation of the pronoun must be rejected (e.g. *$Paul_i$ is combing him_i*), and hence a picture where Paul is combing himself has to be rejected. Finally, principle C violations involved sentences where a referential expression cannot be coreferential with a pronoun preceding it (e.g. *He_i talks to $John_i$*). Two of the complex sentences in this condition were ambiguous, i.e. allowed for two possible correct answers (see also Sambin et al., 2012). For example, in *Cuando Juan entró, sonreía* (tr. *When $Juan_i$ entered, (he_i) smiled*) both verbs can share the same subject. *Cuando entró, Juan sonreía* (tr. *When (he_i) entered, $Juan_i$ smiled*) is also an ambiguous sentence where the coreferential interpretation of the noun is grammatical.

At the beginning of the experiment, four characters were presented with their names and the task was explained. All of the characters had the initial of their first names on their shirts: Pedro, Juan, Ana and María. In Figure 1, one example of each principle is provided as tested in three scenarios each:

a) Principle A: *María se maquilla* (tr. *María is putting on her make-up*) was tested three times: (i) in scenario 1, María is putting make-up on Ana [FALSE], (ii) in scenario 2, María is putting on her make-up [TRUE], and (iii) in scenario 3, Ana is putting make-up on María [FALSE]

b) Principle B: *Juan lo afeita* (tr. *Juan is shaving him*) was tested three times: (i) in scenario 1, Juan is shaving himself [FALSE], (ii) in scenario 2, Juan is shaving Pedro [TRUE], and (iii) in scenario 3, Pedro is shaving Juan [FALSE]

c) Principle C: *Ella tapa a María* (tr. *She is covering María (with a blanket)* was tested three times: (i) in scenario 1, Ana is covering herself, [FALSE] (ii) in scenario 2, Ana is covering María [TRUE], and (iii) in scenario 3, Mary is covering Ana [FALSE]

Figure 1: Three sentence stimuli illustrating BT principles A-C and the three conditions under which they were tested:

	Violation	Correct	Control
Principle A			
Principle B			
Principle C			

The complete stimulus paradigm is provided in Table 2. Presentation order was randomized.

Table 2: Complete stimulus paradigm.

Principle/ Condition	Simple	Complex
A	1. Juan se peina. 2. María se maquilla. 3. Juan habla consigo mismo. 4. María se ríe siempre de sí misma.	1.Cuando él entró, Juan se vestía. 2. María se vestía cuando ella entró. 3. María entró cuando Juan se vestía. 4. Pensó que Juan se estaba bañando.
B	1. María la abraza. 2. Juan lo afeita. 3. María la besa. 4. Juan le esconde.	1. Cuando Ana entró, María le gritó 2. María le gritó cuando entró 3. Cuando entró, María le gritó 4. Pensó que Pedro le observaba.
C	1. Ella regaña a María. 2. Él mira a Juan. 3. Ella tapa a María. 4. Él señala a Juan.	1. Cuando Juan entró, sonreía. 2. Cuando entró, Juan sonreía. 3. Sonreía cuando Juan entró. 4. Pensó que María estaba en la playa.

2.4 Statistical analysis

To analyse the probability of a correct response during the SPM task, we fit a logistic mixed-effects regression model with the *lme4* package (v 1.1-29, Bates et al., 2015) in R (v 4.1.1, R Core Team, 2021). Correct responses were coded as 1's and incorrect responses as 0's. All models included varying intercepts for Item and Participant to take into account the repeated-measures nature of the data. More complicated random effects structures always led to convergence errors and singular fits. Pairwise comparisons, when appropriate, were carried out using the package *emmeans* (v 1.6.3, Length, 2021).

The first model analyzed the early and advanced HD groups as well as the healthy controls. The pre-manifest group could not be entered in the regression model due to issues of separation in the logistic regression model (there were not enough incorrect answers for the model to be identifiable). The *Group* variable was treatment-coded into the regression with healthy controls being represented by the intercept. The *Binding* variable was also treatment-coded into the regression, with the A level being represented

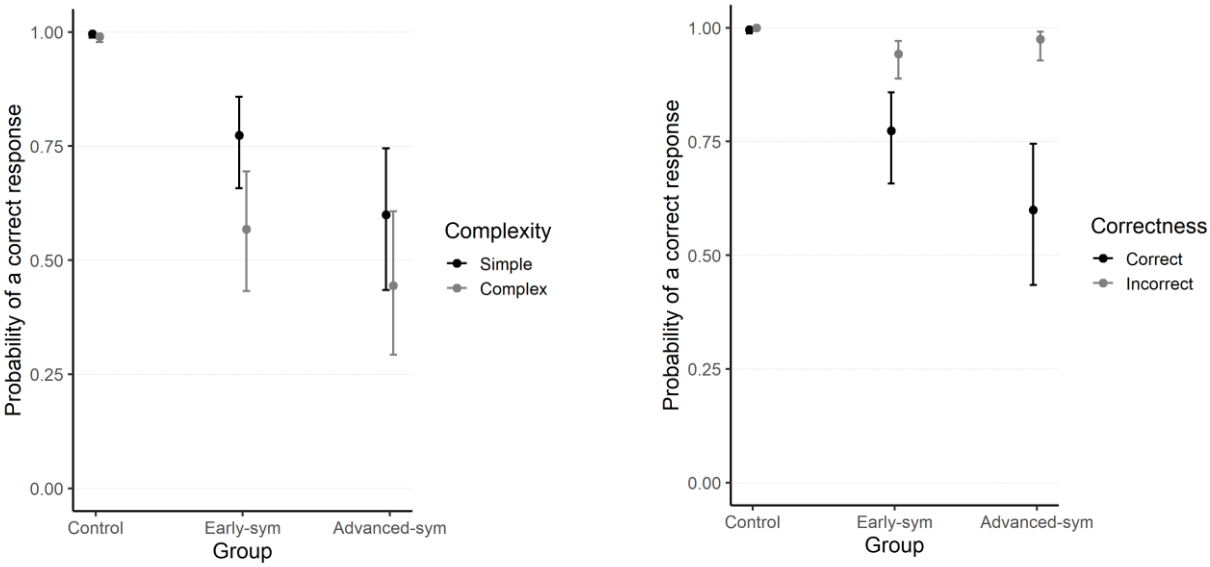
by the intercept. The *Correctness* variable, which coded whether the picture matched the sentence, was treatment-coded into the regression with Correct sentences represented by the intercept. The *Complexity* variable, which coded whether a given item was syntactically complex or not, was treatment-coded into the regression with Simple as the reference level. In addition to these variables, the interactions of Group and Complexity and Group by Correctness were fitted to allow the model to estimate separate probabilities of a correct response according to Complexity and Correctness across the three different groups.

3. Results

The results of the initial model fit to data from the healthy controls and the two symptomatic groups are as follows. The interactions between Group and Complexity and Group and Correctness led to a significantly better model fit as evidenced by a likelihood ratio test against a reduced model without these interactions ($\chi 2(4) = 15.191$, $p = 0.004$).We therefore computed pairwise comparisons within the context of these interactions, with the results as follows. For simple sentences, the Control group was significantly more accurate than the early HD ($p < .0001$) and the advanced ($p < .0001$) HD groups. The HD groups were not significantly different from one another ($p = 0.999$). The same pattern emerged for the Complex sentences, with the Control group being significantly more accurate than the early ($p < .0001$) and the advanced groups ($p < .0001$) and the early and advanced HD groups not being significantly different ($p = 0.622$). Regarding the difference between simple and complex sentences, both the early HD ($p = 0.0002$) and the advanced HD ($p = 0.047$) groups were significantly more accurate on simple sentences than complex, while there was no significant difference for the control group ($p = 0.127$).

Regarding the difference between correct and incorrect sentences, there was no significant difference between them for healthy controls ($p = 0.076$), but both the early ($p < .0001$) and advanced ($p < .0001$) HD groups were more accurate on incorrect sentences than correct ones. For correct sentences, the healthy controls were significantly more accurate than both the early ($p < .0001$) and advanced ($p < .0001$) HD groups. The early HD group were also significantly more accurate than the advanced HD group ($p = 0.049$). For incorrect sentences, the control group was again more accurate than the early ($p < .0001$) and the advanced ($p = 0.004$) HD groups. But there was not a significant difference between the early and advanced HD groups for incorrect sentences ($p = 0.132$).

Figure 2: Probability of correct responses across groups for simple versus complex (left panel) and correct (matching) versus incorrect (right panel) stimuli. The y-axis of both plots contains the back-transformed predicted probability of a correct response for an average participant and item.



3.1 Bias-controlled model

Since both HD groups were more accurate on incorrect than correct sentences, we explored a possible bias and its effect on the difference between the early and advanced HD groups. We fit another model with only clinical participants that included an additional predictor to control for bias. We elected to use the criterion location, which was calculated for each participant. This follow-up analysis was limited to the clinical groups because the criterion location could not be calculated for the majority of participants in the control group due to an absence of incorrect responses where it was said a picture matched a sentence when it did not (i.e., false alarms). A criterion location of zero indicates that a participant is not in general biased towards saying that the sentence and picture matched or did not match. A negative score is indicative of a bias toward responding 'matching' and a positive score indicates that a participant was biased toward responding that the picture did not match the sentence. For a discussion of how signal detection theoretic measures can be applied to linguistic judgments, the reader is directed to Huang and Ferreira (2020).

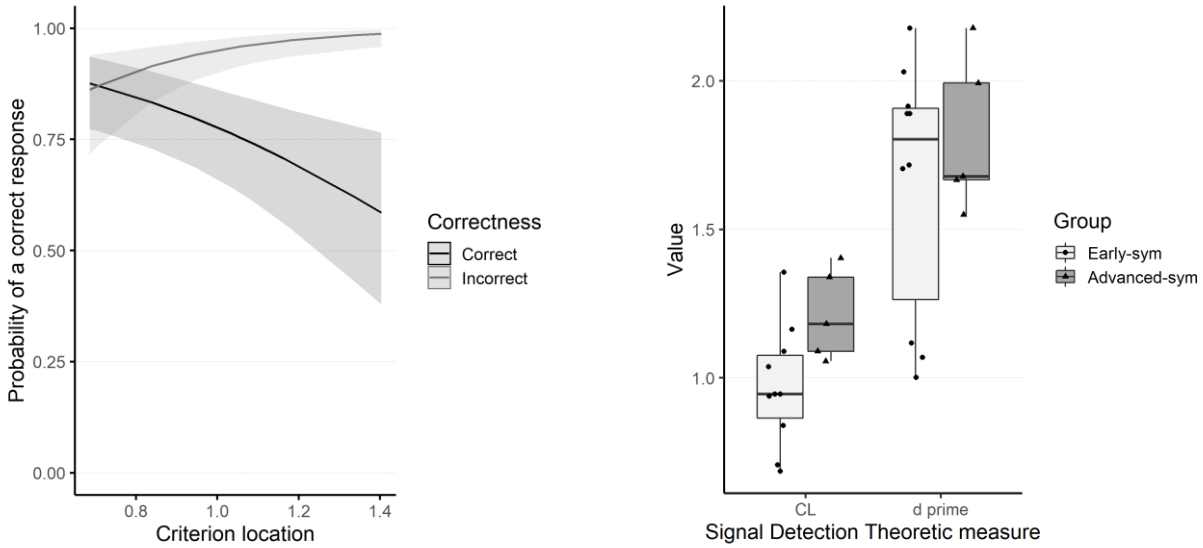
The model fit was similar to the first above model, but with the removal of the control group, the early HD group was now represented by the global intercept in treatment coding. The model also included the criterion location, which was calculated for each participant, and the interaction of criterion location and correctness, as we would expect a bias to influence the two levels of this variable differently. Three participants were dropped from this analysis due to the criterion location not being able to be calculated, two from the early HD group and one from the advanced HD group. As with the first model, we first tested the significance of the interactions between Group and Correctness and Group and Complexity, and found that without the Control group, and accounting for bias, these interactions no longer contributed significantly to model fit ($\chi 2(2) = 0.7148$, $p = 0.670$), and we therefore report the simpler model here.

The two groups did not differ significantly in terms of overall accuracy ($p = 0.787$). Simple sentences were responded to significantly more accurately than complex sentences ($p < 0.001$). Interpreting the interaction between criterion location and correctness, we observe that for the lowest levels of bias in our sample, there was not a significant difference between correct and incorrect sentences, but that higher levels of bias towards saying pictures and sentences did not match resulted in more accurate responses to incorrect items and fewer accurate responses to correct items, as expected. This interaction is visualized on the left side of Figure 3 below.

3.2 Signal detection theoretic group analysis

Results reported so far indicated that there was a difference between the two clinical groups with respect to their average accuracy in correct vs incorrect items and that this difference disappeared after controlling for bias. We next explored whether bias, as well as the sensitivity to the difference between matching and mismatching items, changed as the disease progressed in our clinical groups. To explore the bias found during the task, and to operationalize participants' ability to distinguish matched from mismatched sentences and pictures, we used the already calculated criterion locations and calculated d-prime (d′). As visualized in Figure 3 (right panel), the bias toward saying that a sentence did not match a picture increased from the early to the advanced group (Est.=0.243, SE=0.103). However, d′, the measure of the ability to distinguish between the two conditions, did not seem to show the same pattern (Est.=0.1621, SE=0.2112).

Figure 3: The influence of bias on performance. Left panel: The back-transformed predicted probability of a correct response for the average participant and item according to the criterion location and the two levels of Correctness. Right pane: The two Signal Detection Theoretic measures, criterion location and *d*-prime for the two clinical groups. Individual data points are overlaid onto the boxplot.



4. Discussion

This investigation tested sensitivity to violations of syntactic constraints on referential dependencies under conditions of striatal neurodegeneration. Our results show that such sensitivity sharply declines in patients with manifest HD, both at the early and advanced stages distinguished here. This decline obtains regardless of the specific condition of the BT involved and is seen even for simple (mono-clausal) stimuli, though it exacerbates for complex ones (with a referential dependency to be established across an embedded clausal boundary). This result generalizes the finding the Sambin et al. (2012), which was specific to condition C of the BT. These authors showed that HD subjects are able to correctly establish referential dependencies between NPs over long distances, but fail when they must avoid co-reference as dictated by principle C (e.g. co-reference of *He* and *Paul* in *He entered when Paul was getting dressed*. While in our design we did not specifically control for working memory limitations, the presence of the effect even for simple sentences questions the idea that the impairment seen is secondary to a working memory defect.

Very clearly, it is also not an effect specific to principle C. It could thus reflect a generalized problem with referential dependencies as depending on the hierarchical syntactic configurations in which they appear. As complex stimuli were responded to less accurately by both HD groups, though, syntactic complexity clearly plays a role for performance, beyond the referential dependencies as such. A more generic effect of syntax would be in keeping with prior evidence for a reduction of hierarchical syntactic complexity in language production in this population (Gordon, & Illes, 1987; Murray 2000, Murray & Lenz, 2001; Hinzen et al., 2018; Tovar et al., 2020). On the other hand, as noted in the introduction, a crucial concept to the BT is locality, which is a notion more specific than syntactic complexity as such. It indicates that certain grammatical operations (e.g. the referential binding of one referential noun phrase to another) needs to take into account the local domain (e.g. the clause) of which both noun phrases for parts. Thus, for example, a reflexive anaphor like *himself* needs to be bound locally to this clause. A failure to represent clausal boundaries, or hierarchical structure more generally, would therefore be a possible cause of a failure to recognize when referential dependencies are licit or not. This hypothesis would be consistent with recent evidence (Tovar et al., 2022) of a basic problem of people with HD to recognize ungrammatical sentences that violate principles of locality as applying to syntactic movement (the extraction of a syntactic constituent from its original place and insertion of it elsewhere in the structure, as in *The fridge, he did not fix,* or *The fridge was not fixed*, where the grammatical object 'the fridge' is canonically linearized *after* 'fix'). As with the BT, syntactic movement is governed by locality principles, requiring processing the boundaries of phrases beyond which an item is moved.

A bias we observed for rejecting sentences (stating they don't match the picture), which inflated accuracy for incorrect stimuli (see Figure 3). Taking this bias into account eliminated significant differences in accuracy between the HD groups, suggesting an early deficit that is stable over disease progression. Nonetheless, it is noteworthy that the pre-manifest HD group show no impairment on our task at all, while previous studies of spontaneous speech did observe effects of linguistic weakening in pre-manifest gene carriers (Hinzen et al., 2018; Tovar et al., 2020). As for the bias in question, it is unclear why it obtained and why it was for rejecting stimuli rather being over-tolerant of them. This bias is clinically significant as a possible aspect of the overall HD neuropsychological profile. In the cognitive domain of emotion recognition, some studies

suggested that there is a 'positive bias' in the HD population. In Ille et al.(2011a) results showed that patients rated angry, fearful and sad faces as happier compared to healthy individuals. They also perceived neutral faces as happier. Ille et al. (2011b) went a step further and compared this impaired recognition of negative emotions with brain atrophy. They discovered that there was a correlation with regional atrophy in emotion-relevant areas (insula, orbitofrontal cortex) and in memory-relevant areas (dorsolateral prefrontal cortex, hippocampus).

Our results also shed new light on the role of the striatum in language processing. While our study was not designed to address the issue of its linguistic specificity (Giavazzi et al., 2018; Mestres-Missé et al., 2012; Jacquemot & Bachoud-Lévi, 2021), it does suggest a new subcortical road towards addressing the neural basis of reference. Reference consists in using lexical concepts to identify entities in the world, which are then available for further referencing, giving rise to referential dependencies (Hinzen and Sheehan, 2015). As such reference is a function that bridges between lexical meaning and semantic memory, on the one hand, and grammar, on the other. The neural basis of this function remains unclear; its vulnerability across multiple neurological and neuropsychiatric disorders suggests that this is a both foundationally and clinically important area for future investigation (Hinzen, 2017; Chapin et al., 2022; Tovar et al., 2020; Docherty et al., 2013, Ditman & Kuperberg, 2010)

In sum, our results confirm that basal ganglia damage reduces the sensitivity to syntactic locality constraints on BT principles, affecting the processing of referential dependencies when they are grammatically ruled. These results also shed some light on the neural basis of locality and reference, confirming an important role of the striatum in the awareness of constraints on referential units locally constrained.

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Supplementary materials

Full accuracy model:

```
Generalized linear mixed model fit by maximum likelihood (Laplace
Approximation) ['glmerMod']

 Family: binomial  ( logit )

Formula: Response ~ Group + Binding + Correctness + Complexity +
Group:Complexity +      Group:Correctness + (1 | SubjID) + (1 | Item)

   Data: d2

Control: glmerControl(optimizer = "bobyqa")


     AIC      BIC   logLik deviance df.resid

  1452.6   1532.0   -713.3   1426.6     3319


Scaled residuals:

     Min       1Q   Median       3Q      Max

-23.3144   0.0372   0.0720   0.1710   2.4538


Random effects:

 Groups Name        Variance Std.Dev.

 Item   (Intercept) 0.5473   0.7398

 SubjID (Intercept) 0.2074   0.4554

Number of obs: 3332, groups:  Item, 68; SubjID, 49


Fixed effects:

                                     Estimate Std. Error z value Pr(>|z|)

(Intercept)                           5.49281    0.56511   9.720  < 2e-16
***

GroupEarly-sym                       -4.26642    0.55177  -7.732 1.06e-14
***

GroupAdvanced-sym                    -5.08801    0.57943  -8.781  < 2e-16
***

BindingB                              0.10420    0.27915   0.373   0.7089

BindingC                             -0.05258    0.29200  -0.180   0.8571

CorrectnessIncorrect                  1.87974    1.05909   1.775   0.0759 .

ComplexityComplex                    -0.94264    0.61721  -1.527   0.1267

GroupEarly-sym:ComplexityComplex     -0.01043    0.61208  -0.017   0.9864

GroupAdvanced-sym:ComplexityComplex   0.31319    0.63656   0.492   0.6227

GroupEarly-sym:CorrectnessIncorrect  -0.31063    1.06224  -0.292   0.7700

GroupAdvanced-sym:CorrectnessIncorrect  1.36611    1.13096   1.208   0.2271
```

```
---
Signif. codes:  0 ‘***’ 0.001 ‘**’ 0.01 ‘*’ 0.05 ‘.’ 0.1 ‘ ’ 1

Full accuracy bias model:
Generalized linear mixed model fit by maximum likelihood (Laplace
Approximation) ['glmerMod']
 Family: binomial  ( logit )
Formula: Response ~ Group + Binding + Correctness + Complexity +
Group:Correctness +
    Group:Complexity + CL + CL:Correctness + (1 | SubjID) + (1 |      Item)
   Data: clinical
Control: glmerControl(optimizer = "bobyqa")

     AIC      BIC   logLik deviance df.resid
  1069.9   1129.1   -523.0   1045.9     1008

Scaled residuals:
    Min      1Q  Median      3Q     Max
-5.9949 -0.7259  0.3183  0.5788  2.3399

Random effects:
 Groups Name        Variance Std.Dev.
 Item   (Intercept) 0.5949   0.7713
 SubjID (Intercept) 0.1443   0.3798
Number of obs: 1020, groups:  Item, 68; SubjID, 15

Fixed effects:
                                    Estimate Std. Error z value Pr(>|z|)
(Intercept)                          3.54389    0.78850   4.494 6.97e-06
***
GroupAdvanced-sym                   -0.06263    0.36960  -0.169 0.865445
BindingB                            -0.11587    0.29832  -0.388 0.697712
BindingC                            -0.38189    0.31238  -1.223 0.221511
CorrectnessIncorrect                -4.08415    1.15265  -3.543 0.000395
***
ComplexityComplex                   -0.97783    0.27411  -3.567 0.000361
***
CL                                  -2.24496    0.74117  -3.029 0.002454 **
GroupAdvanced-sym:CorrectnessIncorrect -0.01123    0.61014  -0.018 0.985319
```

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GroupAdvanced-sym:ComplexityComplex     0.28651    0.33645   0.852 0.394455

CorrectnessIncorrect:CL                 5.78000    1.22585   4.715 2.42e-06
***

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Signif. codes:  0 ‘***’ 0.001 ‘**’ 0.01 ‘*’ 0.05 ‘.’ 0.1 ‘ ’ 1
```

6. STUDY 4: LANGUAGE DISINTEGRATION UNDER CONDITIONS OF SEVERE FORMAL THOUGHT DISORDER

Abstract. On current models of the language faculty, the language system is taken to be divided by an interface with systems of thought. However, thought of the type expressed in language is difficult to access in language-independent terms. Potential inter-dependence of the two systems can be addressed by considering language under conditions of pathological changes in the neurotypical thought process. Speech patterns seen in patients with schizophrenia and formal thought disorder (FTD) present an opportunity to do this. Here we reanalysed a corpus of severely thought-disordered speech with a view to capture patterns of linguistic disintegration comparatively across hierarchical layers of linguistic organization: 1. Referential anomalies, subcategorized into NP type involved, 2. Argument structure, 3. Lexis, and 4. Morphosyntax. Results showed significantly higher error proportions in referential anomalies against all other domains. Morphosyntax and lexis were comparatively least affected, while argument structure was intermediate. No differential impairment was seen in definite vs. indefinite NPs, or 3rd Person pronouns vs. lexical NPs. Statistically significant differences in error proportions emerged *within* the domain of pronominals, where covert pronouns were more affected than overt pronouns, and 3rd Person pronouns more than 1st and 2nd Person ones. Moreover, copular clauses were more often anomalous than non-copular ones. These results provide evidence of how language and thought disintegrate together in FTD, with language disintegrating along hierarchical layers of linguistic organization and affecting specific construction types. A relative intactness of language at a procedural, morphosyntactic surface level masks a profound impairment in the referential functioning of language.

1. Introduction

In neurotypical speech no sentence is uttered without a thought expressed in it: the absence of such a link would be sign of a pathology, as for example in the echolalic speech seen in parts of the autism spectrum (Prizant 1983). In line with this basic design feature of language, current architectural models of the language faculty posit an *interface* between two systems identified as *language* and *thought*, respectively (Chomsky 1995; Jackendoff 2002). Addressing the empirical problem of how this interface is structured, however, faces considerable methodological obstacles, including the obvious difficulty of studying the specific kind of thought expressed in language in language-independent

terms. Moreover, which system or theory would account for *thought* itself in its human-specific form, if not language, remains unclear in empirical terms, though a *Language of Thought* (LOT) has long been postulated to this effect (Fodor 1975; 2008; Burton-Roberts 2011). One tradition in linguistic theory has considered language to be the generative principle behind the relevant kind of thought itself: Ancient Indian grammar (Chaturvedi 2009); late Medieval Modistic grammar (Covington 2009); and un-Cartesian linguistics (Hinzen & Sheehan 2015); see also Humboldt (1836) and Mueller (1887). This tradition broadly contrasts with a more *rationalist* or *Cartesian* tradition, in which language is conceptualized as an expressive system, whose essential function is to encode or communicate a rational thought process that is as such given independently and grounded in language-independent principles (Arnauld & Lancelot 1660; Chomsky 1966; Pinker & Jackendoff 2005; Fodor 2008). Considerable light could be cast on this historical and foundational dichotomy by considering patterns of language variation not merely under conditions of cognitive uniformity and neurotypicality, but under conditions of changes in the thought process as seen in neurological and neuropsychiatric disorders, where linguistic diversity co-occurs with clinical cognitive diversity. Delineating *Universal grammar* in the technical sense of a language-specific biological endowment ultimately depends on clarifying its relation to the species-typical thought system. Without considering linguistic changes under conditions of changes in this other system, we would deprive ourselves of variation that could address this relation.

In acquired language disorders such as post-stroke aphasia, the co-existence of cognitive decline with language impairment remains debated. Though cognitive decline is difficult to test when language impairment will typically interfere with task demands in language-based tests, considerable evidence supports that some aspects of nonverbal cognition decline along with language in acquired aphasia (Baldo et al. 2005; Baldo et al. 2010; Fonseca et al. 2016), as well as primary progressive aphasia (Fittipaldi et al. 2019). Nonetheless, clinical impression often suggests that the thinking process is surprisingly preserved in aphasia: patients seem to struggle to get normal thoughts across linguistically, but not with the thoughts themselves (Varley 2014). In line with this clinical impression, single-case studies have documented dissociations in aphasia between language and other cognitive domains such as arithmetic, theory of mind, music, or scientific and spatial reasoning (Fedorenko & Varley 2016), though it remains debatable how much language was preserved in the patients in question, to what extent

some of the tasks could not be solved by lower-level perceptual mechanisms, and to what extent the forms of thinking involved in these non-linguistic tasks and in language are comparable (arithmetic and music in particular involve no referential concepts of the sort seen in language). Aphasia, moreover, affects people that have had normal language for many decades. The degree to which aphasic performance reveals processing limitations rather than the fundamental language deficit (*knowledge* or *competence*) has also long been debated (Linebarger et al. 1983; Bates et al. 1991). In this regard, a more telling case are 25-30 % of individuals on the autism spectrum who never develop language in the first place in either production or comprehension and in any modality (Picket et al. 2009; Tager-Flusberg & Kasari 2013; Slusna et al. 2018). The little evidence that exists about this population suggests that normal intelligence (largely even in nonverbal IQ) and social cognition (including nonverbal communication) effectively collapse, pointing to a fundamental integration of early cognitive and linguistic development (Maljaars et al. 2011; Norrelgen et al. 2015; Slusna et al. 2018). The critical role of language in categorization and learning in preverbal infants independently supports this integration (Perszyk & Waxman 2018).

Here we will consider a different neuropsychiatric condition affecting adults who have had normal language development but are affected by cognitive decline in early adulthood: formal thought disorder (FTD) in patients with schizophrenia (Andreasen 1979). While not exclusive to schizophrenia, FTD is one of schizophrenia's criterial symptoms and objective signs in the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5, American Psychiatric Association 2013). Detected at the level of linguistic form, it contrasts with 'disorders of content' clinically identified as delusions (e.g. a patient's expressed convictions that he is Jesus or that he came to earth in a cosmic bubble). FTD is undoubtedly linked to a language dysfunction insofar as it is diagnosed as such. Moreover, meta-analyses point to dysfunction in language areas as a neural correlate (Wensing et al. 2017; Cavelti et al. 2018). However, the disorder remains conceptualized within psychiatry as being located at the level of thought, of which the clinically manifest language dysfunction is widely regarded as only an overt expression. In line with this Cartesian viewpoint, linguistic studies of spontaneous speech in this syndrome, though inaugurated by Chaika (1974) early on, have remained scarce and they have often been confined to minimal or small samples of patients with FTD (e.g. Chaika 1974, N=1; Rochester & Martin 1979, N=6; Harvey 1983, N=10; Oh et al. 2002, N=10).

Today, productive speech in FTD thus remains largely characterized clinically through terms such as derailment, incoherence, tangentiality, or 'word salad'. Since none of these are linguistic terms, it remains as a challenge to determine the more properly linguistic variables that might identify such speech and distinguish it from both non-thought disordered speech in schizophrenia and from that of neurotypical controls. Current cognitive neuropsychological approaches to FTD still largely seek to identify neurocognitive deficits in non-verbal cognitive domains, particularly in semantic memory and executive functioning, though identifying such deficits specific to FTD has proved elusive (McKenna & Oh 2005).

More linguistic studies of FTD are required to assess the role of language dysfunction in the neurocognitive basis of FTD. Language as a neurocognitive domain plays a role not merely in FTD, but in other core symptoms as well, particularly in auditory verbal hallucinations (Tovar et al. 2018), but arguably also in delusions (Hinzen, Rosselló & McKenna 2016). Recent work in computational linguistics have suggested considerable potential for language as a biomarker in schizophrenia, as automated linguistic measures can predict symptoms of schizophrenia including FTD (Elvevåg et al. 2010; Bedi et al. 2015; Holshausen et al. 2014). Experimental psycholinguistic studies have also revealed numerous language processing anomalies in schizophrenia, largely in comprehension/perception (Titone et al. 2007; Kuperberg 2010; Kuperberg et al. 2017; but see Kuperberg, et al. 2018, for a recent study of semantic priming in a naming task), and in part specific to FTD (Kuperberg et al. 1998).

Studies of FTD inspired by theoretical linguistic models, in the case of language production, fall into two main traditions. In the first of these, starting from Rochester & Martin (1979), the focus has been on the discourse level using the theoretical framework of Halliday & Hassan (1976). The authors targeted the use of various linguistic devices for establishing 'cohesion' across sentences, given the assumption at the time that schizophrenic speech at lexical and single sentence levels was largely normal (McKenna & Oh 2005). The markers of cohesion in question were a mixed set comprising anaphoric pronominal reference, substitution, ellipsis, conjunction and lexical cohesion. Differences between patients with schizophrenia with and without FTD were mainly found in the mis-use of anaphoric pronouns and demonstratives leading to unclear reference to objects or persons, but not in the quantity of such cohesion markers. This broad finding was replicated in several later studies (Wykes & Leff 1982; Harvey 1983; Docherty et al.

1996; Docherty et al. 2003). This tradition conceptualized such anomalies as communication/discourse disturbances, with the exact link to the linguistic substrate in which they occur still unclear. A second linguistic tradition in the study of schizophrenic speech has documented less syntactic complexity and more syntactic errors (Faber & Reichstein 1981, Morice & Ingram 1982; Morice & McNicol 1986; Hoffman & Sledge 1988;), but with some evidence that syntactic anomalies may characterize language in schizophrenia generally, i.e. without being specific to FTD (Oh et al. 2002; Stirling et al. 2006; Moro et al. 2015; but see Cokal et al. 2018 for evidence that they are more pronounced in FTD as compared with either patients without FTD or controls). Oh et al. (2002) argued, though based on a small sample of six patients with FTD, that it is semantic anomalies at a sentence-level which are characteristic of FTD.

Our goal here was to investigate language in FTD with a particular view to how it may illuminate the thought-language relation. From this point of view, the referential use of Noun Phrases (NPs) is a natural focus. At this referential level, language inherently connects to thought: normal language use always is referential, with speakers picking out objects and events and saying something about them; just as referential thinking is always expressible in language (but only partially in music or imagery). Use of NPs also connects to the first of the above traditions of the study of language in FTD, since different types of NPs naturally serve different functions in discourse, with definite NPs in particular often being anaphoric, i.e. picking up on a referent identified before. It also connects to the second tradition, since NPs are a particular instance of syntactic complexity and NPs that serve different referential functions also exhibit different forms of syntactic complexity (Hinzen & Sheehan 2015; Martín & Hinzen 2016). Recent linguistic work on FTD further supports a focus on NPs. One recent study (Sevilla et al. 2018) compared the proportions of anomalous NPs in a group of Spanish-speaking patients with FTD (N=20) against a second group with schizophrenia without FTD (N=20) and neurotypical controls (N=14), with data obtained from a fairy-tale retelling task. This study reported a significant difference between groups when anomalies in the referential NPs were annotated as occurring in definite NPs and pronouns, but not when annotated as occurring in indefinite NPs and lexical NPs (NPs containing a lexical noun), suggesting a specific linguistic signature of FTD speech. Although the grammatical categories ‘lexical NP’ and ‘definite NPs’ overlap (a lexical NP like *the man* is definite, but need not be, as in *a man*, while a definite NP can be a lexical NP or pronominal), the exact linguistic distinction

involved thus matters when seeking to linguistically distinguish these groups. The result of Sevilla et al. is consistent with the fact that *unclear reference* and *poverty of content* are among the terms clinically identifying FTD (Andreasen 1979; 1986): although these terms reflect clinical judgements, at a linguistic level they naturally correspond to an anomalous indefiniteness (or lack of specificity) of referential phrases: either it is unclear what object, person or event is being referred to (unclear reference), or it is so indefinite that the impression of a lack of proper content arises (poverty of content). Quantity and quality of use of definite NPs is thus an appropriate and promising focus for linguistic studies of FTD. The results of Sevilla et al. (2018) on misuses of definite vs. indefinite NPs, furthermore, are broadly in line with another study in an English-speaking sample of patients with and without FTD (Cokal et al. 2018).

We also aimed to illuminate the language-thought relation by contextualizing deficits in NP use against anomalies in other levels of linguistic organization. In language, a complete thought is built in layers, starting from a selection of lexical concepts and then some initial structure-building that integrates objects or persons into verb phrases: argument structure, which reflects a layer of meaning intermediate in hierarchical complexity between lexis and full propositional information at the level of utterances that come with referential meaning. While anomalies at the lexical level (paraphasias and neologisms) are well-established in schizophrenia and FTD in particular (McKenna & Oh 2005), as are syntactic anomalies as per the second linguistic tradition above, degrees of impairments across these levels have not yet been systematically compared. Our annotation scheme thus covers (i) referential anomalies as linked to their linguistic substrates (NP types in which they occur), (ii) argument structure, (iii) lexis, and (iv) (morpho-) syntax. Based on Sevilla et al. (2018) and Cokal et al. (2018) we predicted that:

- Proportions of anomalies in definite NPs and pronouns would outweigh those in indefinites and lexical (non-pronominal) NPs;
- Despite evidence for lexical (word) -level and formal syntactic anomalies in FTD in the literature, referential anomalies would be a more indicative marker of language impairment in FTD when these respective layers of linguistic organization are compared with one another.

We further explored with post-hoc analyses whether a more fine-grained sub-classification of NP types involved in referential anomalies and of clause types could further illuminate the patterns found in the main analysis. We specifically explored the following linguistic distinctions: (i) covert-vs-overt pronouns, (ii) 1st person-vs- non-1st Person, (iii) animate-vs-inanimate pronoun; and finally, (iv) copular-vs-non-copular clause types. This was motivated, in the case of (i), by different functions of covert and overt pronouns in Romance (particularly discourse and anaphoric functions in the former case, see Sorace et al. 2009; Camacho 2013; Jiménez-Fernández 2016); in the case of (ii), a potential influence of self-referential (1st Personal) discourse on referential anomalies, given the importance that the 1st Person plays across other core symptoms of schizophrenia, including auditory verbal hallucinations (Tovar et al. 2018) and delusions (Hinzen, Rossello & McKenna 2016); and, in the case of (iii), whether language disintegration is 'content-sensitive' in the sense that it plays a role whether the NP in question denotes animate entities or not. In the case of (iv), finally, we inquired whether sentence type plays a role in how anomalous sentences are: copular clauses like She is my mother are more based on grammar than on lexical information: they do not contain a lexical verb and often express identities (of one thing with another), about which patients in our sample appear to be very often confused.

2. Methods

2.1 Participants and corpus

The basis for this study was a historical corpus collected from 38 Spanish- and Catalan-speaking stable in-patients with schizophrenia by a local psychiatrist, Dr. Moya, for purposes of a PhD dissertation on the language of formal thought disorder (Moya, 1989). Speech samples consisted of free conversations with an interviewing doctor. To make an extremely time-intensive annotation procedure manageable and avoid confounds between Catalan and Spanish-speaking patients, annotations were restricted to a total of 15 Spanish-speaking but otherwise randomly selected participants and the first five pages of transcriptions from their speech, resulting in a mean number of 888,6 words per participant (standard deviation: SD=384,9). Audios on which the original transcriptions were based were still available and provided to us. Existing transcriptions were checked against the audios, and in many cases completed. The mean age of these 15 patients was 46.13 (SD=16.2), the mean length of illness in years was 22.4 (SD=11.17). 7 were male. Clinical records were still available for each patient, capturing family history, clinical

history, disease progression, medication, speech samples, and justification of clinical diagnosis through DSM-III criteria. DSM-III diagnostic codes were: "Paranoid schizophrenia" (295.3, 8 patients), "Undifferentiated schizophrenia" (295.9/92, 2 patients), "Disorganized schizophrenia" (3 patients), "Residual" (1 patients). 1 last patient had no diagnostic code. The DSM-III A-criterion of "incoherence and notable loss of the associative capacity" was noted to be fulfilled in each case; disorganization of speech, in many cases with detailed examples, was mentioned in most of the case reports.

To allow comparability with other studies of FTD, participants were formally rated by a psychiatrist not involved in this study (Dr. Edith Pomarol-Clotet, FIDMAG Hermanas Hospitalarias. Barcelona) using the canonical Thought, Language, and Communication (TLC) scale of Andreasen (1986). TLC ratings confirmed the FTD diagnosis in all cases. According to the TLC scale, 12 of the 14 participants scored an extreme (4) on the "Global" rating, defined as "TLC disorder so severe that communication is difficult or impossible most of the time". In computing the Global score, the TLC suggests that "some TLC disorders are more pathological than others", in the sense of "likely to suggest severe psychopathology" (Andreasen 1986: 481). The above 12 participants all scored either "moderately severe" or "severe" on two or more of the "more pathological" items, i.e. incoherence, derailment, tangentiality and loss of goal. One participant was given a TLC-Global rating of "moderate" (2) – determination of the degree of FTD in this patient was problematic because the main abnormality he showed was delusional confabulation. The final participant was given a TLC-global score of (1) as she principally showed so-called negative FTD or alogia and scored mostly on the criteria of poverty of speech and perseveration. Given the different FTD profiles of these last two participants, post hoc analyses were performed to determine whether results changed when linguistic variables were compared across the group without including these participants.

2.2 Annotation scheme

Annotations proceeded first at the level of clauses, secondly at the level of the four different linguistic strata distinguished here, namely use of different types of referential nominals, argument structure, lexis, and morpho-syntax. As a first step, at the clausal level, annotators were instructed to first identify all clauses with a finite verb (in matrix or embedded positions) and to identify them as either copular clauses (with a predicate *to be*) or non-copular clauses (codes [cop] and [ncop]). They then had to make a first-pass

judgement on whether each of these contained an anomaly or not (codes [g] or [b] for 'good 'or 'bad', resulting in codes such as [copg] or [ncopb] annotated directly after the relevant finite verbs). Three criteria were individually or jointly sufficient for an anomaly rating: (i) the clauses contained any kind of formal-grammatical errors, e.g. *Él es ángeles* (lit. '*he is angels'*), which involves a violation of grammatical agreement; (ii) they involved three or more repetitions or were echolalic, e.g. a participant repeating the phrase *no hay dinero en la casa* ('there is no money in the house') at the end of his utterances as a kind of meaningless stock phrase; (iii) they had NPs with referents that could not be identified or were misplaced and contributed to false or plainly nonsensical statements, e.g. *nací por aquí, por este mundo* (lit. 'I was born around here, around this world', where the place indication (*este mundo*) is vague; or *Bulle a mi alrededor una distracción* (lit. 'Boils around me a distraction'), where it is unclear what the noun *distracción*refer to, in the context of the verb *bulle*; or one participant's claim *Usted estaba allí* ('you were there'), which misplaces the interviewing doctor (*usted*) as participating in a scene that took place years ago in her house (with the consternation of the doctor indicating that this was not the case). See detailed annotation samples for further examples of referential anomalies. Annotations were based on the crucial insight that reference in human language is always a relational phenomenon, in the sense that a word or NP in isolation (e.g. *man*, or the *doctor*) would never refer to anything: reference is always a sentence-(and indeed utterance-) level phenomenon, which depends on the lexical description of the referent provided, the grammatical relations in which the NP stands, and context (Hinzen & Sheehan 2015). Referential anomalies were therefore determined for NPs as occurring in their utterance-context. This point is also relevant to reference in the first Person. Although it may appear that *I* as used in isolation cannot possibly be mis-used referentially (it cannot fail to pick out the speaker), it arguably can become anomalous e.g. when used in a copular clause in which the referent of *I* is identified with another person, as in a female speaker's assertion Yo era mi marido (lit.' I was my husband'), where it is unclear who the speaker actually refers to, himself or another (male) person, i.e. her (his?) husband; or another speaker's assertion *Me mataron a mí en el psiquiátrico* (lit., 'They killed me in the psychiatric'), which clearly cannot have happened to the referent of 'me' except metaphorically.

In the case of an anomaly based on (iii) (*referential anomaly*), annotators next identified the grammatical type of the NP affected, according to the following NP sub-

classification scheme: pronominal/nominal ([p], [n]), definite/indefinite ([d], [i]), singular/plural ([s], [l]), animate/inanimate ([a], [t]), anomalous or not ([g], [b], as in the clausal case). In the pronominal case, it was also determined whether the pronouns were overt or covert ([o], [c]) and which grammatical person ([1], [2], [3]) they had. NPs were annotated even in truncated utterances or hanging topics (non-sentences), in which case no anomaly was annotated at the sentence (matrix) level (since there was no sentence). An exception to this rule was when this type of anomalous NP occurred in an embedded truncated clause, in which case the anomaly at the sentential level was annotated at the level of the superior clause (e.g. *En esta vida* [g] *pasa* [b] *que la la música,* lit. 'In this life it happens that music', where the [b] annotates the matrix clause anomaly, while the two NPs are unobjectionable). All instances of NPs that were not referential but predicational were disregarded, for example NPs in appositions, since they resume the same referent (e.g. the underlined NP in *Thor, the guy of the thunder*, …), in NPs forming predicates of copular sentences (e.g. *He was a policeman)* and secondary predications (e.g. *Me trajeron la última*, lit. 'They brought me the last one, meaning I was the last one they brought'). Crucially, referential NPs were *not* annotated as anomalous merely in virtue of reflecting rarified *beliefs* (e.g. of a religious nature, like having seen the virgin Mary, or simply unlikely to be true but not verifiable by annotators, like having talked to Mariano Rajoy in the Palacio de Congresos).

Turning to the remaining linguistic strata, annotations of lexis included anomalies that could be detected at the lexical level alone (without considering grammatical context), in the form of either neologisms, e.g. *espárramo*, *genitación*, clanging, lexical decompositions such as *arma de dedo* (lit. 'weapon of finger') for *pistola* ('pistol'), or anomalies relating to the use of light verbs e.g. *hacer convencimiento* (lit. 'make conviction') for *convencer* ('to convince'). Violations of argument structure were defined as local selectional relations between a verbal head and its thematic arguments; these could be anomalous if either the arguments were wrongly subcategorized at the level of grammatical category of the selected dependent, e.g. *hablé esto* ('I spoke this'), when *spoke* ('to speak') requires a prepositional complement; or else the selection was wrong semantically, e.g. *quisiera estar en la consideración y naturaleza de mi vida* (lit. 'I would hope to be in the consideration and the nature of my *live'*), where *estar* cannot subcategorize the following NP; or *Sí, están tramitando* (lit. 'Yes, they are processing'), where an object NP is grammatically obligatory but missing. Finally, errors in

morphosyntax comprised agreement and other errors compromising the formal-grammatical integrity of a phrase or clause, disregarding its meaning. In *Mi madre son muy monjas* (lit. 'My mother are very nuns').

Note that these four annotation levels (referentiality, argument structure, lexis, and morphosyntax) are not orthogonal to one another but can be at least partially ordered in a hierarchy. Thus, while morphosyntax is the most strictly formal level of grammatical organization, lexis involves lexical level semantic organization, while argument structure is grammatical but lexically projected, and reference in the present sense is a full-utterance level phenomenon. Referentially evaluable utterance-level propositional meaning presupposes a syntax matching this meaning, which in turn includes argument structure, which itself includes lexical meaning. In *Her grandmother broke a leg*, we need to understand the general lexical concepts *grandmother*, *break* or *leg*, before understanding which persons, objects, or event in the world the content words are used to refer to; and we need to grasp *what event it is*, conceptually, i.e. one of *a grandmother breaking a leg* (argument structure). Note that lexical items as such and as occurring in isolation (*grandmother*, *leg*, *break*) only have general conceptual meaning and cannot pick out particular persons, legs, or accidents; even complex NPs occurring in isolation, e.g. *her favourite grandmother*, do not as such give us any idea of who is being referred to. Both reference and truth are utterance level (root) phenomena, which require multiple grammatical elements to come together configurationally in the right way so to allow identifying a referent and event in context (Hinzen & Sheehan, 2015; Hinzen 2017).

Annotations were first made by three annotators (AT, CM, SS) working independently on sub-samples of 5 transcripts each, focused on the referential analysis only, which was the most complex. To ensure strict adherence to the same criteria, all three annotators then met to check all annotations in the entire sample, under the supervision of a senior rater who was not involved in the writing of this paper (Joana Rosselló, JR), so that all annotations were checked by four raters in total. All disagreements were resolved by consensus from all raters. Domains other than reference were pursued in the form of three bachelor theses completed a the Universitat de Barcelona in 2017 under the direction of JR, who verified every annotation made in these three domains.

2.3 Samples

We provide three extracts of the conversations to give the reader a sense of the type of speech investigated here and use of the annotation scheme. It needs to be kept in mind that in general, schizophrenic speech, particularly at this level of disorganization, is very difficult to translate, and errors in one language may not be errors in the best available English translation. Below, comments justifying codes are restricted to those codes ending with [b] (*bad*, i.e. anomalous by the above criteria). Superscripts identify such codes and are repeated in the comments. Clause-level annotations identifying whether a clause (specified as copular or non-copular) was *good* or *bad* are annotated directly behind the clause's finite verbs; NP annotations behind the relevant NPs. PAT: patient; INT: interviewer.

2.3.1 Sample 1

PAT1: (pcdxs1g) Estoy (ncopg) estupefacto.

INT: ¿Estupefacto?

PAT1: Estupefacto, sí. […] Bulle (ncopb)[1] a mi (podxs1g) alrededor (ndstb)[2] una distracción (nistb)[3] y un aliciente (nistb)[4] que seguramente debe ser (ncopb)[5] pero (pcds1g) no la (pods3tb)[6] noto (ncopb)[7] en mí (pods1b)[8], sino que (pcds1g) me doy cuenta (ncopg) que no (pcds1g) la (pods3tb)[9] tengo (ncopb)[10]. Un bulle (niqstb)[11] de un aliciente (niqstb)[12].

PAT1: ¿Un bulle?

FER: Un bulle (nistb)[13].

INT: Un bulle.

PAT1: Un bulle (nistb)[13] que debe (ncopb)[14] alrededor (nistb)[15] pero (pcds1g) no lo (pods3tb)[16] conexiono (ncopb)[17] conmigo (pods1g).

Literal translation: I am thunderstruck. - Thunderstruck? - Thunderstruck, yes. - A distraction boils around me and an incentive that it should probably be but I cannot notice it inside me, but I realize that I don't have it. - A boil? - A boil. - A boil. - A boil that should around but I cannot connect it with me.

Comments: [1]Deficient (non-copular) clause because of vague/unclear references in[2-4]. [5]Non-copular clause deficient for formal-grammatical reasons. [6]Reference of clitic unclear. [7]Deficit at clausal level inherited from referential deficits in its nominals. [8]*Una distracción y un aliciente* cannot be located 'in me'. [9]Reference of the clitic continues to be unclear. [10]Deficit at clausal level inherited from referential deficits. [11]The verb *bulle*, possibly a Catalanism, has transformed into a noun, which it cannot be in Catalan. [12]Inappropriate reference in this sentence context. [13]See previous comment (11). [14]Deficit inherited from referential and formal-grammatical problems inside the clause. [15]Adjective wrongly subcategorized. [16]Reference of clitic unclear. [17]Wrong verb form.

2.3.2 Sample 2

PAT2: Yo (pods1g) empecé (ncopg) a tomar Melleril (nistg) y eso (pods3tg) me (pods1g) producía (ncopg) desequilibrio (nistg), que es (copg) lo (pods3tg) que yo (pods1g) he sugerido (ncopg). He tenido (ncopb)[1] yo (pods1g) en mi vida (pods1g) desequilibrio (ndstg).

INT: ¿Qué quiere decir eso, 'desequilibrio'?

PAT2: Desequilibrio (nistg) es (copg) que no (pcis2g) estás equilibrada (ncopg). (pcis2g)Vas (ncopg) por un sitio (nistg) y te .., y por ejemplo, (pcil1g) vamos (ncopb)[2] a pasar por la pasera (ndstb)[3] y hay (ncopg) personas (nilag) que tienen (ncopb)[4] equilibrio que apartan (ncopb)[5] el equilibrio (nistg) y (pcil3ag) pueden (ncopg) pasar porque el cuerpo (nistg) lo tienen (ncopb)[6] bien, (pcil3ag) tienen (ncopb)[7] equilibrio. Sin embargo, otras personas (nilag) nos tambaleamos (ncopg), alguna (pois3ab)[8] que se descontrola (ncopb)[9]. Y entonces, pues eso, yo (pods1g) muchas veces (niltg) se lo (pods3tg) he dicho (ncopb)[10] las compañeras (ndflag) (…), que (pcds1g) me caeré (ncopb)[11] al arroyo (ndstb)[12] o quien dice una pendiente (nistg) (…), sitios (niltg) así de peligro (…) los sitios (niltg) así retirados. Eso (pods3tb)[13]es (copb)[14] equilibrio.

Literal translation: I started taking Melleril and it produced me imbalance, what I suggested, I have had in my life, imbalance. - What does this mean, 'imbalance'? - Imbalance is that you are not in balance. You go to a place and you … and for example, we go for a walk on the walkers [neologism] and there are people who have balance, who turn the equilibrium away and they can pass because their body is alright, they have balance. However, other people we stagger, someone gets out of control. And then, well, I have said it many times to the friends, that I will fall into the stream or who says a slope,

places of danger, the so distant places.

Comments: [1]Deficient grammatical selection of *desequilibrio* by *tener*. [2]Deficiency of the clause inherited from deficient NP: [3]*pasera* seems to be an instance of 'clanging', originating from *pasar* (*to pass*), with the intended meaning *places where you can go for a walk*, but the word exists in Spanish but only with a different meaning. [4]See comment (1) above. [5-7]Deficits at clausal level inherited from infelicitous selections inside with the NPs themselves in good shape. [8]Impossible reference after plural inclusive in previous clause. [9]Deficit at clausal level inherited from (8). [10]A missing preposition *a* introducing the object NP in *se lo he dicho las compañeras*: an argument structure violation. [11]Deficit at clausal level inherited from infelicitous definite reference to some stream not previously introduced (12). [13]Reference unclear. [14]Deficit inherited from previous example (13).

2.3.3 Sample 3

INT: ¿Cuánto tiempo estuvo usted en Barcelona ¿porque, usted, ¿dónde ha nacido usted?

PAT3: Yo (pods1g) nací (ncopb)[1] por aquí, por este mundo (nistb)[2], por el campo (nistb)[3].

INT: ¿Por el campo?

PAT3: Por el campo (nistb)[4]. Mi (pods1g) madre (ndsag) llevaba (ncopb)[5] una bata blanca (nistb)[6] e (pcds3ag) iba (ncopb)[7] por un barranco (nistg) y nací (ncopb)[8] yo (pods1b)[9].

INT: Su madre llevaba una bata blanca.

PAT3: Sí, (pcds3ab)[10] está (copb)[11] ahí, en la cocina (ndstb)[12].

INT: ¿Su madre?

PAT3: Sí.

INT: ¿Su madre vive?

PAT3: Sí, (pcds3ab)[13] está (copb)[14] aquí, en la cocina (ndstb)[15].

Translation: How long did you live in Barcelona? Where you were born? - I was born around here, in this world, in the countryside. - In the countryside? - In the countryside. My mother wore a white coat and she went to a ravine and I was born. - Your mother wore a white coat? - Yes, she is over there, in the kitchen. -Your mother? - Yes. - Is your mother alive? - Yes, she is here, in the kitchen.

Comments: [1]Deficiency at clausal level inherited from vague NP. [2-4]Vague NP reference. [5]Deficits at clausal level inherited from infelicitous reference in (6) to a white coat worn by the mother when giving birth, which the speaker presumably cannot know. [7]While there is nothing wrong per se in reference to *un barranco* (a ravine), the mother presumably did not go to a ravine when giving birth. [8]Deficit at clausal level inherited from anomalous reference in first person in (9), where the speaker misplaces herself as being born in the context outlined. [10-15]The covert subject is misplacing the mother as being in the kitchen.

2.4 Statistical analysis

Variables compared here are proportions of errors on a specific linguistic unit. For instance, the proportion of anomalous definites was calculated as the number of anomalous definite nouns or pronouns over the total number of definite nouns or pronouns produced. This was necessary to account for quantitative differences in the total number of words produced by patients. Paired-samples t-tests were applied within patient where normality as determined by a Shapiro-Wilk test and symmetry of the data allowed this. Wilcoxon signed-ranked tests were applied in cases of violation of normality only, and Sign tests if none of both conditions applied. Cohen's d for dependent samples was used to quantify effect size of differences that were significant. According to Cohen's (1988) suggested interpretation of this measure, almost all effect sizes reported are large (defined as > 0.80), only one being medium ($0.50 < d < 0.80$). All indicated p-values are two-tailed and the significance level is set at 0.05.

3. Results

3.1 Main comparisons

3.1.1 Proportion of anomalous definite vs. indefinite NPs

A paired-samples t-test revealed that, contrary to our predictions, the proportion of anomalous definites over the total of definites (M= 0.250, SD=0.137) was not significantly different from that of anomalous indefinites over all indefinites (M=0.314, SD=0.186), $t(14)= -1.41$, $p = .180$.

3.1.2 Proportion of anomalous nominals vs. pronouns

Wilcoxon signed ranked test showed, again contrary to our predictions, that the proportion of anomalous nominals out of all nominals (M = 0.339, SD = 0.176) was

significantly higher than that of anomalous pronominals out of all pronominals (in all grammatical Persons) (M = 0.222, SD = 0.118), V = 4, p < .001, Cohen's d = 0.85).

3.1.3 Anomalies across linguistic strata

Pairwise comparisons of anomalies divided by linguistic strata showed that language was affected over all strata distinguished here, and additionally that there was a linear progression between them in terms of mean proportion of anomalies. Specifically, starting from the most impaired, the pattern (with Means and SDs) was: NP (0.283 ± 0.134) >* Argument Structure (0.042 ± 0.034) >* Lexis (0.006 ± 0.006) > Morphosyntax (0.005 ± 0.004), where * indicates a statistically significant difference (see Figure 1 and Table 1).

Figure 1. Boxplots of anomalies in linguistic strata.

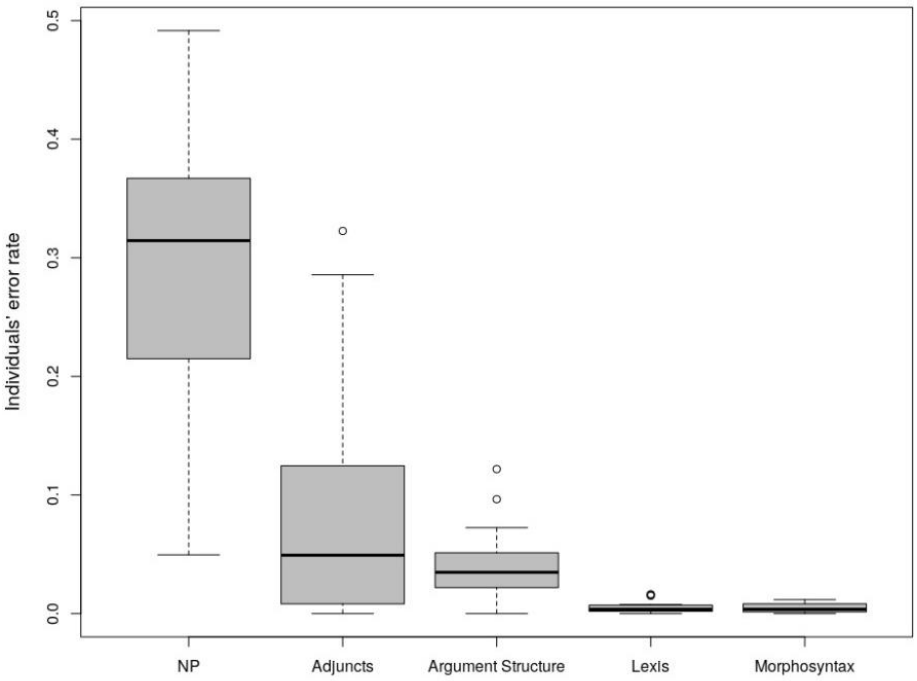


Table 1. Results on pairwise comparisons of linguistic strata.

Variable 1	Variable 2	Test	Statistic	P-value	Cohen's d
NP	Argument structure	Paired t-test	t(14) = 6.80	< .001	1.76
NP	Lexis	Paired t-test	t(14) = 8.17	< .001	2.11

NP	Morphosyntax	Paired t-test	t(14) = 8.13	< .001	2.09
Argument structure	Lexis	Sign test	13	.0023	1.13
Argument structure	Morphosyntax	Paired t-test	t(14) = -4.43	< .001	1.14
Lexis	Morphosyntax	Paired t-test	t(14) = 0.57	.578	

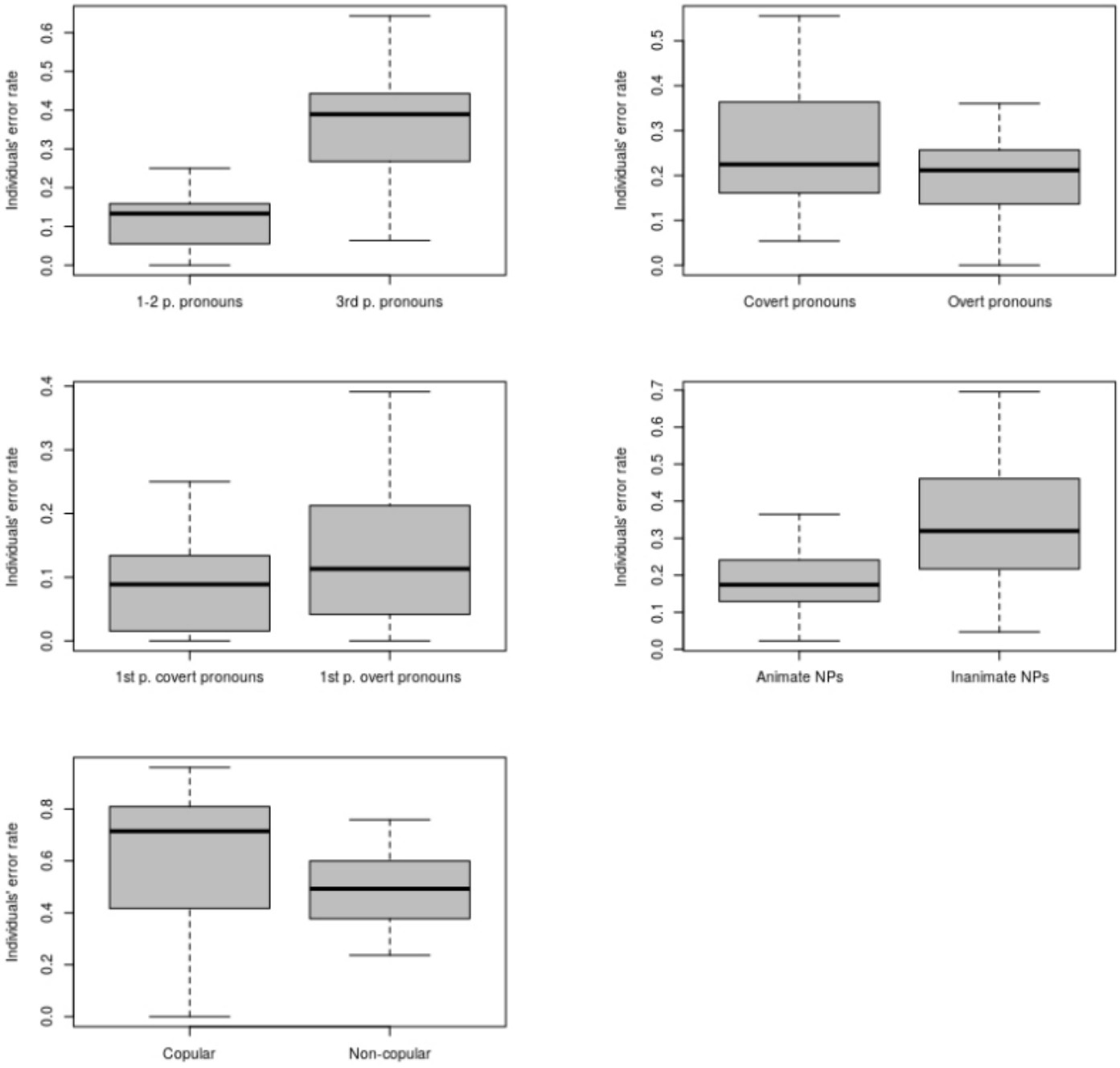
3.2 Finer-grained comparisons

Results from a series of paired-samples t-tests are summarized in Table 2; the corresponding boxplots can be found in Figure 2. There was a significant difference between the proportion of anomalous 1st and 2nd person pronouns out of all pronouns as compared with the proportion of anomalous 3rd person NPs, the latter being more affected. This in turn motivated restricting the comparison of the respective proportions of anomalous pronouns and lexical NPs to 3rd Person pronominals only, which eliminated the significant difference between anomalies in pronouns and lexical NPs found in the main comparisons. The difference between the proportion of anomalous covert pronouns out of all covert pronouns and that of anomalous overt pronouns out of all overt pronouns was also significant, with covert pronouns more affected than overt ones. When narrowing down this last comparison to 1st person pronouns only, on the other hand, the difference between covert and overt instances of 1st person pronouns trended in the opposite direction (p = .06). The proportion of anomalous animate NPs out of all animate NPs was significantly lower than that of anomalous inanimate NPs over all inanimate NPs. Finally, when comparing proportions of anomalous copular and non-copular clauses out of the total of copular and non-copular clauses, another trend (p=.06) emerged, with copular clauses more affected by anomalies than non-copular ones.

Table 2. Results of paired t-tests on fine-grained comparisons in the NP domain.

Variable 1	**Variable 2**	**$Mean_1 \pm SD_1$ $Mean_2 \pm SD_2$**	**Statistic**	**P-value**	**Cohen's d**
1st and 2nd person pronouns	3rd person NPs	0.111 ± 0.078 0.353 ± 0.175	t(14) = -6.36	< .001*	1.64
3rd person pronouns	Lexical NPs	0.352 ± 0.175 0.339 ± 0.176	t(14) = 1.22	0.244	
Covert pronouns	Overt pronouns	0.255 ± 0.193 0.193 ± 0.103	t(14) = 2.47	0.027*	0.64
1st person covert pronouns	1st person overt pronouns	0.091 ± 0.082 0.143 ± 0.117	t(14) = -2.12	0.053	
Animate NPs	Inanimate NPs	0.191 ± 0.100 0.335 ± 0.187	t(14) = -3.13	.007*	0.81
Copular	Non-copular	0.604 ± 0.310 0.489 ± 0.158	t(14) = -2.05	.060	

Figure 2. Boxplots of anomalies in the fine-grained comparisons.



Post hoc analyses of the sample with two participants removed due to their different profile of FTD as determined by Andreasen's (1986) TLC (see Section 2.1) by means of paired t-tests showed no differences in the pattern of results except in two cases where trends converted into significant results: covert (M = 0.098, SD = 0.084) vs. overt (M = 0.162, SD = 0.114) anomalous instances of 1st person pronouns (p = .035, t(12)=-2.37, Cohen's d=-0.66) with the latter more anomalous than the former; copular (M =

0.686, SD = 0.238) vs. non-copular (M = 0.524, SD = 0.137) anomalous clauses out of the total of copular and non-copular clauses (p=.010, t(12)=3.06, Cohen's d=0.85), with copular clauses more affected by anomalies than non-copular ones.

In order to ensure that results are not driven by possible outlier participants, we further searched for outliers in every comparison made using a common technique and, if one was found, the analysis was re-run omitting the outlier. Concretely, since our analyses are paired, for every comparison between variables V1 and V2 we looked for outliers on their paired difference (V1-V2). We calculated the Interquartile range (IQR) of this variable, which is the difference between the 75th and 25th percentiles. We then defined two cutoff points for outliers to be at a factor of k of the IQR above or below the 75th and 25th percentiles, respectively. If any patient lies beyond these points, it was considered an outlier. A common value for k is 1.5 (Tukey 1977).

A number of outliers were found with this method (2 in the Argument Structure - Lexis comparison, 2 in the Argument Structure - Morphosyntax comparison, 1 in the 3rd person pronouns - Lexical NPs comparison, and 2 in the covert - overt pronouns comparison). However, analyses excluding them resulted in very similar or smaller p-values to those of analyses using the full sample, and did not change their significance, showing that our results are not strongly driven by their influence (see plots in the Supplementary Materials showing the values of the compared error rates by patient).

4. Discussion

These results shed new light on language disintegration across different linguistic strata under conditions of clinical thought disintegration. Results partially supported and partially contradicted our main predictions. They did not support our expectations motivated by previous studies (Rochester & Martin1979; Wykes & Leff 1982; Harvey 1983; McKenna & Oh 2005), which had highlighted problems with pronouns and vague and unclear reference in spontaneous schizophrenic speech, while Cokal et al. (2018) and Sevilla et al. (2018) specifically highlighted problems with definiteness. Our results suggest that at least in severe FTD of the kind studied here and in a conversational task of this nature, the referencing problem seen in such patients is more general and reaches deeper into the organization of language, as opposed to primarily affecting pronominal or definite forms of reference, as we had predicted. Pronouns or definite NPs mediate specific discourse functions such as anaphoricity and (in the case of overt and covert

pronouns) aspects of information structure (Camacho2013; Sorace et al. 2009; Jiménez-Fernández 2016). The results suggest, therefore, that the referential problem is located at a more fundamental level, affecting the entire process of reference generation from the initial retrieval of a lexical content word to the final configuration of an act of reference via a full NP in a sentential context, without being restricted to anaphoric or discourse functions.

This failure to replicate results on definiteness in previous studies may be partially due to the fact that two of the studies mentioned above that have investigated this issue most directly (Sevilla et al. 2018 and Cokal et al. 2018), used narrative tasks, namely a fairytale retelling task and a retelling of a visually presented comic strip, respectively. These studies found that anomalies in definite NPs (Sevilla et al. 2018), and in the quantitative proportions of definite vs. indefinite NPs (Cokal et al. 2018), are linguistic identifiers of FTD as compared with controls and patients without FTD. But the tasks in question constrain the referential process more than the conversational task used here: a fairytale already provides a plot that is memorized, and in the case of the comic strip, the referents were visually present as and when the story was told. By contrast, in the present study, referencing was restricted only through the prompting questions of the interviewer, providing fewer constraints with regard to which lexical concepts are to be retrieved for reference.

Unlike in Sevilla et al. (2018), the proportion of anomalous lexical NPs turned out here to be significantly *higher* than that of pronouns. This could initially suggest that the problem *increases* when lexical content is involved, not when reference is not lexically mediated and, in this sense, more grammatically mediated, as in the case of pronouns. However, in the present study, when comparisons between anomalous pronouns and lexical NPs were restricted to 3rd Person pronouns as compared with lexical NPs (which are always 3rd or *non*-Person), significant differences in relative proportions of anomalies crucially disappeared (Table 2). In short, the initial *appearance* that lexical NPs are significantly more affected than pronominal ones is likely based on mixing in the other grammatical persons (1st and 2nd), which showed fewer anomalies in the domain of pronouns when compared with 3rd Person pronominals. Since personal pronouns are usually functioning deictically, this also suggests the conclusion that, *within* the domain of pronominals where lexical-descriptive content is absent, a specific difficulty with anaphora (referential dependencies) may indeed manifest itself: such a difficulty would

naturally affect 3rd Person pronouns in their most typical uses more than personal ones. Comparisons between the use of covert and overt pronouns reported above support this interpretation, since the former were more affected than overt ones (Table 2) and they tend to function anaphorically in Spanish. Interestingly, moreover, *within* the domain of 1st Person, this relation between anomalies in covert and overt 1st Person pronominals *reversed*, with overt 1st Person pronouns being more affected. This may be because there is no clear sense in which the 1st Person realized as a covert pronoun is anaphoric as opposed to deictic.

On the other hand, this interpretation of the pattern seen *within pronominals* should be qualified by the fact that no significant differences in respective proportions of anomalies between definite and indefinite NPs were found, even though the former tend to be anaphoric in their functions, unlike the latter. That is, if we include *all* NPs, whether lexical or not, the problem still does not appear to be a problem of one NP type (e.g. NPs with anaphoric functions) primarily: it affects definite NPs as much as indefinites.

Earlier studies have also supported the existence of formal syntactic anomalies in both FTD and schizophrenia at large, as compared with control subjects (Faber & Reichstein 1981; Hoffman & Sledge 1988; Oh et al. 2002; Moro et al. 2015, a.o.). Our data, on the other hand, suggest the *relative preservation* of morphosyntactic aspects of linguistic organization in even severe FTD. To put this insight in a different way, if all content words were replaced by pseudo-words in the speech of the patients studied here, particularly nouns, resulting in a radical version of Jabberwocky-style speech, very few anomalies would be noticeable.[1] We interpret this relative preservation of morphosyntactic aspects as showing that insofar as even severe FTD can exhibit relatively fluent discourse, the 'fluency' in question is largely *procedural* in nature – it reflects language at the level reflecting learned patterns in procedural memory of how phrases are built (Ullman et al. 1997). It is simply that, in terms of referential content, these phrases have become idle wheels, often effectively conveying no content at all. In short, what is surprisingly robust when our thought capacity is fundamentally lost, is morphosyntax in the sense of a *learned routine*, independent of the role that grammar plays in mediating a specific kind of content.

[1]We owe this point to conversations with Joana Rosselló.

The type of content that is lacking concerns meaning that arises when lexical-level content is turned into referential expressions via grammar, which is in line with earlier suggestions of an anomalously lexically-driven speech generation process, contravening a proper 'balance' between such lexical and grammatical processes of encoding meaning (Ditman et al. 2011). In accordance with this interpretation, lexis as such (disregarding its referential use in context) was as comparatively unaffected in the present study as morphosyntax was. Put differently, from the viewpoint of our comparative results across different variables and linguistic layers, it is difficult to detect FTD, even at this level of severity, by looking at a lexical level only (neologisms, clanging, etc.) while abstracting from the normal referential function of lexical items retrieved in language use. The problem does not lie so much in lexical content *per se* as in the *grammatical meaning* that arises when grammar accesses the lexicon so as to produce referential and propositional meaning on an occasion of language use. Such meaning is inherently *contextual* insofar as it locates given abstract and general concepts (*man*, *birth*, *village*, etc.) in specific objects or events existing in space and time as identified relative to the time and space of speech.

Although personal (1st or 2nd Person) pronouns were proportionally less affected than 3rd Person ones, it is worth noting that, at a qualitative level, remarkable anomalies showed in the former as well, which precisely relate to contextual embedding. An example is from the participant of sample 3 in Section 2.3, who insists that her mother wore a white coat (*bata blanca*) during her birth, upon which the interviewer asks how she could know this, given that she had just been born. The patient answers: *Porque yo nací por el campo y me dijo: "Estate aquí que yo ahora vengo"* ('Because I was born around the countryside and she told me: "Stay here as I come now"'). We interpret these as misinterpretations of when a speaker is an addressee, i.e. 2nd Person, and hence as a mis-localization of herself as a 1st Person. Problems with felicitous uses of personal pronouns thus deserve further study, in line with theoretical approaches stressing the importance of disturbances of deixis to the psychopathology of schizophrenia (Crow 2010; Hinzen & Rosselló 2015; Hinzen et al. 2017). Deictic disturbances clearly extend beyond personal pronouns, reflecting remarkable problems of these patients in locating events or themselves as event participants in space and time, e.g. a patient saying *Yo nací por aquí, por este mundo* ('I was born around here, in this world'), another specifying a time incomprehensibly as *la hora de víctimas*, a third commenting: *Llevo aquí un mes o*

bien han adelantado el calendario? Yo llevaba la cuenta de los días y la he perdido ('I am here for a month, or have they advanced the calendar? I kept track of the days but I have lost track now').

We speculate that the special role of referentially anomalous NPs in the linguistic profile of FTD may also explain the interesting and novel result that copular clauses were proportionally more often anomalous than non-copular ones (see Table 2 and post hoc results with two outliers removed). Copular clauses lack a lexical verb, hence they necessarily rely on NPs for their lexical structure more than any other part of speech (e.g. 'I was my husband', 'This is equilibrium'). As a consequence of that, they also have a restricted range of possible meanings, which particularly includes statements of identity, as just illustrated. This is what might make copular clauses more anomalous as compared with non-copular clauses, which have lexical support in their verbs and in this sense depend on less on the lexical content of NPs only. Investigating clause structure is an important task in future work. A completely unexpected post-hoc result (Table 2), on the other hand, was that reference to animate entities was proportionally more impaired than reference to animates. We do not know how to interpret this result. Very speculatively, reference to persons will often be deictic and rooted in the 1st and 2nd Person (e.g. reference to speaker and hearer, or persons directly relating to them, e.g. *my sister*), which were less impaired. While referencing is unstable in this population across all forms of reference including deixis, reference to non-personal objects without anchoring in the immediate speech context may become particularly unstable.

Although the lexical level showed a low proportion of anomalies comparatively to the other strata distinguished here, two phenomena transpired in the course of these annotations that have to our knowledge not been noted before and bear brief mentioning here to motivate future research. Firstly, a recurring phenomenon in this subsample were lexical decompositions of nouns or verbs into their conceptual ingredients, e.g. *hace convencimiento* (*lit.* 'make conviction') in the place of *convencer* ('to convince'), *son de credo* (are of faith) in the place of *creyentes* ('believers'), *artistas de hielo* (artists of ice) in the place of *patinadores* ('ice skaters'), *arma de dedo* ('finger weapon') for *pistola*, *corrida de la vida* ('course of life') for *prostitute* (prostitute), *general del tráfico* (traffic general) for *policía de tráfico* ('traffic police'), *nervio de hombre* ('man's nerve') for *pene* ('penis'). A second noteworthy phenomenon was the pervasive existence of lexical NP repetitions or stacked NPs (see e.g. the end of sample 2 above).

Sandwiched in between lexis and referential and deictic meaning lies argument structure, as an early layer of grammatical complexity encoding basic thematic structure: participants organized around an event. In line with the above interpretation, statistically significant differences in the proportion of anomalies seen with respect to both lexis and morphosyntax appeared at this layer already, though by no means as severe as in referential, utterance-level meaning. Reference in this latter sense is where language and thought connect: language cannot be used except referentially, i.e. with words used so as to pick out objects, persons or events, which the thoughts expressed in the sentences are about. Though abstract poetry takes this idea to its limits, language never functions in the way that music, say, does. It does allow us to talk about fictions, yet only if these fictions are distinguished from reality and appropriately placed in relation to existing objects and a shared deictic frame relative to which fictions are recognized as such. Again, the absence of such anchoring in a shared space of reference would be a sign of pathology, as in delusional speech.

In line with this, reference in the normal (i.e. a declarative) sense has long been linked to language, given its essential absence in non-human primates (Butterworth 2003; Tomasello 2006; Tomasello & Call 2018), its close association with language development even in its nonverbal forms in humans (Iverson & Goldin-Meadow 2005; Colonnesi et al. 2010), and given its severe reduction or absence in non- or minimally verbal children with autism spectrum disorders (Maljaars et al. 2011; Slusna et al. 2018). Since, in turn, thought that was not expressible in language as used normally would not be thought of the same kind (but might be emotion, imagery, music, or pathological thought), it is arguable that language, thought, and reference are inseparable in humans, forming an integrated, single species-specific scheme, in which they are all co-dependent (Davidson 2004; Hinzen & Sheehan 2015; Hinzen 2017). From this point of view, it makes sense that language in FTD is seen to disproportionally disintegrate at this referential end the level of grammatical complexity where thought becomes referentially anchored in speech. Referential language is unthinkable without thought; as is thought without reference.

Overall, then, we conclude that the present results suggest that, in formal thought disorder, language and thought disintegrate together: the language disintegration seen cannot be made sense of independently of the thought that language inherently conveys, nor can the thought disturbance be separated from specific linguistic dimensions and

parameters in which it is manifest. *Language* and *thought* in this sense imply a conceptual distinction that ceases to be empirically meaningful. To be sure, data reported here and elsewhere (Cokal et al. 2018; Sevilla et al. 2018) do not *rule out* that referential anomalies seen in FTD might be due to some language-independent, currently unknown cognitive mechanism, in which case a language-thought dissociation would be re-vindicated. However, it seems unnatural to split the referential function of language off from language, when referentiality is intrinsic to all linguistic functioning and grammar is systematically sensitive to referentiality. Language cannot be used other than referentially, and it never resembles a system like music, where referential meaning of the same type is not seen. Moreover, reference as investigated here concerns a specific type of meaning arising configurationally, i.e. from an NP in a grammatically referential position within a structured utterance; and significant differences in the use of specific NP types are seen in the present results, as they were in other studies (Rochester & Martin 1979; Docherty et al. 1996; Docherty et al. 2003; Seville et al. 2018; Cokal et al. 2018). This suggests that language dysfunction in FTD should be studied at a linguistic level, though it is also true that language functioning is always integrated with other domain-general cognitive functions such as attention, executive functioning, or working memory. Docherty (2012) in particular found significant correlations between 'communication failures', which often relate to reference in the present sense, and measures of attention, working memory, and conceptual sequencing. Nonetheless, whether such mechanisms can illuminate the specific and differentiated linguistic pattern seen here, is unclear. Current studies on pronoun resolution specifically find correlations between reference skills and executive functions (Hendriks et al. 2014; Ladányi et al. 2017; Sorace 2016), yet as noted, pronouns were not specifically more impaired than lexical nominals in the present study. A primary linguistic deficit in how grammar configures reference clearly remains an option to be considered in the neuropsychology of FTD. This would be consistent with current evidence from meta-analyses of neuroimaging studies about the neural correlates of FTD, which center on core language territory in the brain (Wensing et al. 2017; Cavelti et el. 2018), though interconnected with other cognitive functions.

A limitation of this study is that it lacks comparable data from a neurotypical control group using the same measures. The study focused on relative differences between error rates to profile a particular dataset of clinical speech. We therefore cannot assert to what extent the same types of errors would also be found in controls, nor whether a similar

progression from the levels of Lexis and Morphosyntax to Argument structure and Referential errors might be seen there. Regarding absolute proportions of errors, some previous studies have found no differences in the proportion of syntactic errors between schizophrenia and control groups (Sevilla et al. 2018), while others have (e.g. Cokal et al. 2018). By contrast, both of these studies and many others have documented significant increases in referential errors in schizophrenia groups vs. controls, particularly in FTD. It is nonetheless empirically possible that, in controls, a significant difference between syntactic and referential errors could be found, too, though we are not aware of data on this. When comparing formal syntactic with referential errors specifically, a reason for a gradient of increased error proportions towards referential errors might be a greater cognitive demand imposed when language is put to a referential use, as opposed to merely being produced in a formally correct manner. Cokal et al. (2018) (Supplementary Materials) reported means of ratios of referential errors divided by total utterances to be .35 in a group with FTD and .11 in neurotypical controls. By contrast, means of ratios of syntactic errors were .11 in both schizophrenia groups and .07 in the control group, suggesting a much smaller gap. In the present study, the means were .28 for ratios of referential errors and about .05 for syntactic errors (including both argument structure and morphosyntactic errors). However, criteria of annotation were partially different in Cokal et al. (2018), the task was a picture description rather than free conversation, and referential errors certainly had a distinctively different quality and scale in the present group. Nonetheless, data from that study and the present one certainly suggest remarkably low rates of syntactic errors in both schizophrenia and control groups, despite of the severity of FTD involved in the present study; and only slightly more elevated mean rates in referential errors in the controls of Cokal et al. (2018) relative to syntactic errors. In a recent study of an elderly Spanish-speaking neurotypical control group (Martínez-Ferreiro et al. 2017), participants (N=15) produced only 2.3% of ungrammatical utterances; though different from a ratio of syntactic errors, this figure again suggests such errors to be relatively rare in neurotypical speech. Overall, it seems reasonable to conclude that compared to potential differences in (low) rates of syntactic errors between people with schizophrenia (whether with or without FTD) and controls, which may or may not exist, a wide gap opens at the other end of our spectrum of linguistic strata, i.e. in referential errors, with a steep slope of the gradient from syntactic to referential errors, particularly at the levels of severity of FTD studied here.

In sum, this study has revealed, for a rare corpus of severely thought disordered speech and a conversational task, that the disorganization of thought in question affects the organization of language differently at different levels: proportionally the least anomalies are seen at the morphosyntactic and lexical levels, while proportions increase the moment that meaning is involved at a structural level (argument structure), which is still at least partially lexically driven by the meaning of the selecting verbal head. Anomalies peak when lexical meaning and argument structures are put to a referential use at the level of sentences and utterances, in a way that affects NPs in their referential uses in general, though within pronouns, distinctive patterns of differential impairment can also be seen. This result informs theories of the language-thought interface by showing that and how, as thought disintegrates, language is affected. Future work needs to confirm and fine-grain the referential anomalies seen in such speech, and determine the neural basis of the gradient of decline across the four strata distinguished here.

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Author contributions: WH, AT, and WSSN conceptualized the study. AT, WSSN and CMM annotated the corpus. AG designed and performed the statistical analysis. WH, AT, and AG wrote the first draft of the paper.

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Supplementary Materials

Sample annotation

(1) Original version (Spanish)

T: Trabajaba como criada.
You-worked as maid

„You worked as a maid.“

P: Sí, y me dijo la señora: “Ya más tiempo de criada no.”Yes and to-me said the master already more time of maid no

“Yes, and the master said to me: “You have worked long enough as a maid.”

P: “Estate aquí que nosotros nos vamos al pueblo te dejamos el piso.”
Stay here that we us go to.the village to-you we-leave the apartment

“Stay here as we are going to the village, we leave you the apartment.“

P: Y yo me fui de estar con mi señora.And I me went of to-be with my master

“And I ended my relationship with my master.“

T: ¿Y quién?
and who

“And who?“

P: Y vinieron a mi casa y usted estaba.
and they-came to my house and you were-there

“And they came to my house and you were there.'

T:¿Yo estaba en su casa?
I was in your house?

“I was in your house?“

P: Sí.
Yes.

"Yes."

T:¿Cómo es eso?
How is this.

"How can that be?"

P:Estaba usted en mi casa cuando llegaron mis señores.

Were you in my house when came my master.

"You were in my house when my masters came."

(2) Codified version

P: "Sí	y	**me**	**dijo**	**la**	**señora**	más	**tiempo**	de	criada no.
		✓ Pronoun	✓ Clause		✓ Noun		✓ Noun		
		1 Person			Animate		Inanimated		
		Definite			Definite		Indefinite		

P: ***pro***	**Estate**	aquí que	**nosotros**	nos	**vamos**	al	**pueblo**,	
✓ Pronoun	✓ Clause		✓ Pronoun		✓ Clause		✓ Noun	
2nd person			1st person				Inanimate	
Definite			Definite				Definite	

pro	**te**	**dejamos**	el	**piso**."
✓ Pronoun	✓ Pronoun	✓ Clause		✓ Noun
1st person	2nd person			Inanimate
Definite	Definite			Definite

P: Y	**yo**	me	**fui**	de	estar	con	**mi**	**señora.**
	✓ Pronoun		X^1 Clause				✓ Pronoun	✓ Noun
	1st person						1st person	Animated
	Definite						Definite	Definite

Y	***pro***	**vinieron**	a	**mi**	**casa**	y	**usted**	**estaba.**
	X^2 Pronoun 3rd person Definite	X^3 Clause		✓ Pronoun 1st person Definite	✓ Noun Inanimate Definite		X^4 Pronoun 2nd person Definite	X^5 Clause

P: **Usted**	**estaba**	allí	en	**mi**	**casa.**
X^6 Pronoun 2nd person Definite	X^7 Clause			✓ Pronoun 1st person Definite	✓ Noun Inanimate Definite

P: **Sí.**
X^8 Clause

P: **Estaba**	**usted**	en	**mi**	**casa**	cuando	**llegaron**	**mis**	**señores.**
X^9 Clause	X^{10} Pronoun 2nd person Definite		✓ Pronoun 1st person Definite	✓ Noun Inanimate Definite		✓ Clause	✓ Pronoun 1st person Definite	✓ Noun Animated Definite

Explanations:

✓ Ok

X = Anomaly

X^1: Morphosyntactic anomaly. X2: Vague reference.

X^3: When a linguistic element is anomalous at a phrase level, the whole clause turns anomalous at the clausal level.

X^4: Referent misplaced.

X^5: Anomalous relationship established between the predicate and the misplaced referent.

X^6: Same referent misplaced.

X^7: Same anomalous relationship established between the predicate and the misplaced referent.

X^8: Fragments are wrong as well when they refer to anomalous clauses.

X^9: Same anomalous relationship established between the predicate and the misplaced referent.

X^{10}: Same referent misplaced.

Supplementary Table S1: Total and incorrect frequencies across linguistic variables

Patient ID	Definite NPs		Indefinite NPs		Pronouns		Nouns	
	Total	Incorrect	Total	Incorrect	Total	Incorrect	Total	Incorrect
1	137	27	67	15	109	21	95	21
2	88	37	33	22	63	26	58	33
3	192	50	149	97	181	34	160	113
4	118	29	226	80	154	32	190	77
5	189	13	54	4	147	8	96	9
6	261	48	84	27	208	34	137	41
7	141	32	112	32	138	35	115	29
8	118	32	41	17	93	22	66	27
9	235	20	67	8	158	11	144	17
10	129	19	87	42	128	23	88	38
11	309	125	70	23	258	100	121	48
12	150	6	74	3	145	4	79	5
13	06	50	42	12	65	23	83	39
14	174	77	115	18	160	54	129	41
15	125	35	93	29	136	37	82	27

	1st and 2nd person pronouns		3rd person NPs		Covert pronouns		Overt pronouns	
	Total	Incorrect	Total	Incorrect	Total	Incorrect	Total	Incorrect
1	60	11	144	31	53	7	56	14
2	40	10	81	49	27	15	36	11
3	142	19	199	128	100	19	81	15
4	77	5	267	104	82	21	72	11
5	77	4	166	13	63	4	84	4
6	122	0	223	75	109	22	99	12
7	103	16	150	48	89	20	49	15
8	52	7	107	42	41	11	52	11
9	103	6	199	22	59	4	99	7
10	77	6	139	55	49	11	79	12
11	139	32	240	116	147	60	111	40
12	83	0	141	9	74	4	71	0
13	30	4	118	58	42	18	23	5
14	58	2	231	93	69	30	91	24
15	74	12	144	52	47	15	89	22

	1st person covert pronouns		1st person overt pronouns		Animate NPs		Inanimate NPs	
	Total	Incorrect	Total	Incorrect	Total	Incorrect	Total	Incorrect
1	21	3	32	8	132	23	73	19
2	7	0	24	4	103	31	50	28
3	82	13	54	6	179	28	171	119
4	32	1	13	1	174	24	187	85
5	16	2	47	2	184	14	64	3
6	48	0	69	0	233	28	149	47
7	53	3	32	10	171	41	92	23
8	19	2	22	4	118	28	48	21
9	45	4	49	2	185	12	143	16
10	22	0	53	6	186	31	64	30

	1st person overt pronouns		Animate NPs	
	Total	Incorrect	Total	Incorrect
1	32	8	132	23
2	24	4	103	31
3	54	6	179	28
4	13	1	174	24
5	47	2	184	14
6	69	0	233	28
7	32	10	171	41
8	22	4	118	28
9	49	2	185	12
10	53	6	186	31
11	46	18	299	109
12	50	0	134	3
13	10	2	55	12
14	27	1	136	33
15	40	9	139	48

	Morphosyntax		Lexis	
	Total(words)	Incorrect	Total(words)	Incorrect
1	796	1	796	843
2	407	10	407	445
3	1407	6	1407	1474
4	819	5	819	885
5	599	15	599	645
6	598	10	598	642
7	1193	14	1193	1255
8	594	8	594	629
9	564	10	564	603
10	1904	13	1904	1978
11	980	14	980	1038
12	704	12	704	728
13	924	16	924	983
14	1044	14	1044	1099
15	796	20	796	854

Figure S1: Error rates on linguistic strata across patients

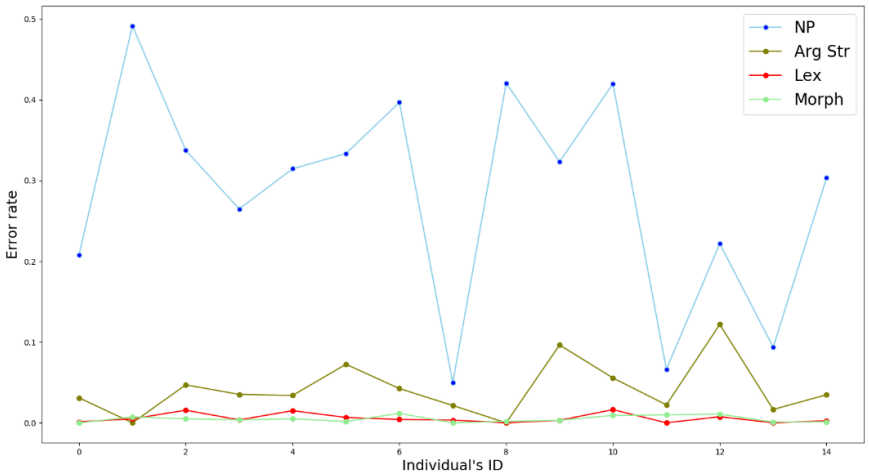


Figure S2: Error rates on covert and overt pronouns across patients

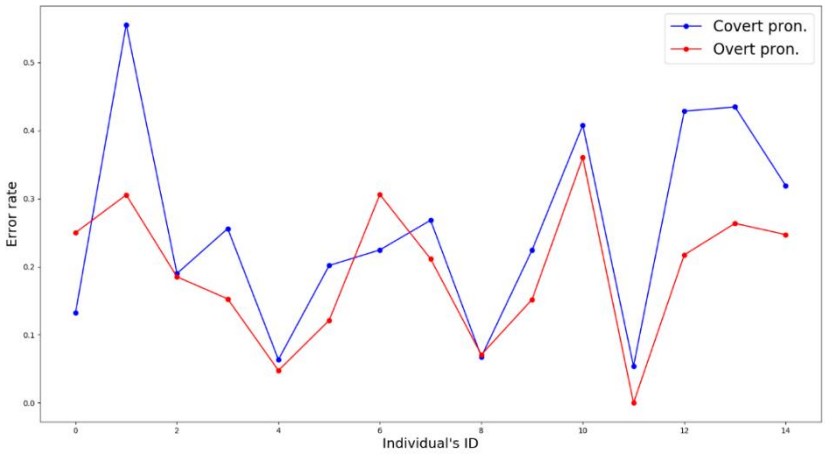


Figure S3: Error rates on 1st person covert and overt pronouns across patients

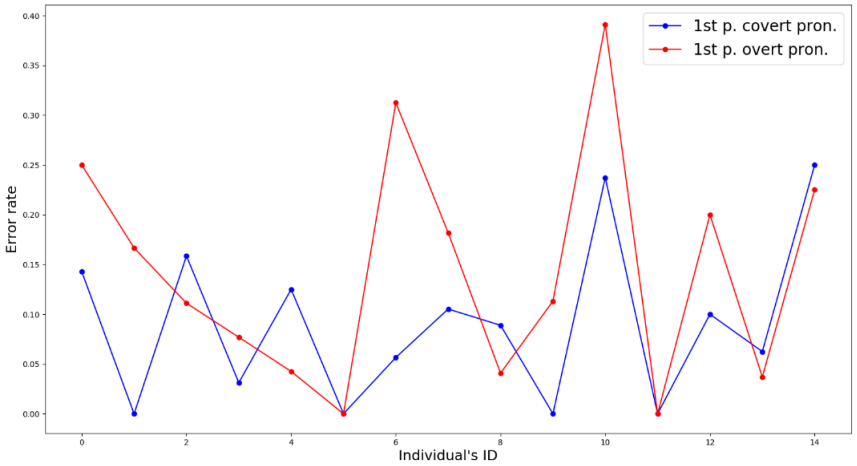


Figure S4: Error rates on 1st and 2nd person pronouns and 3rd person NPs across patients

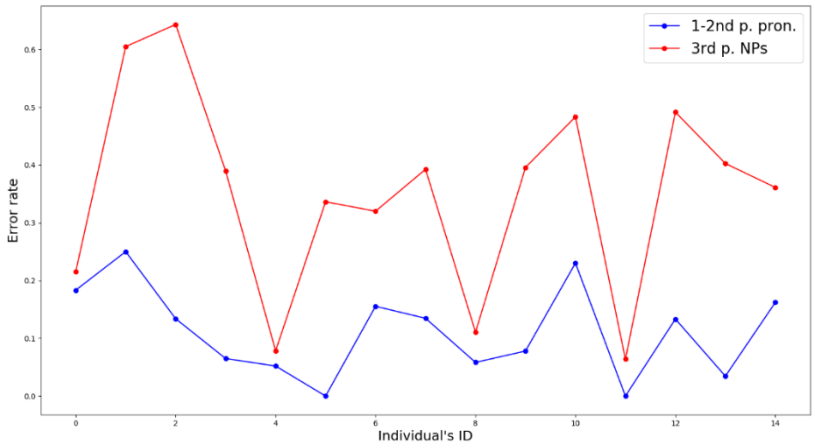
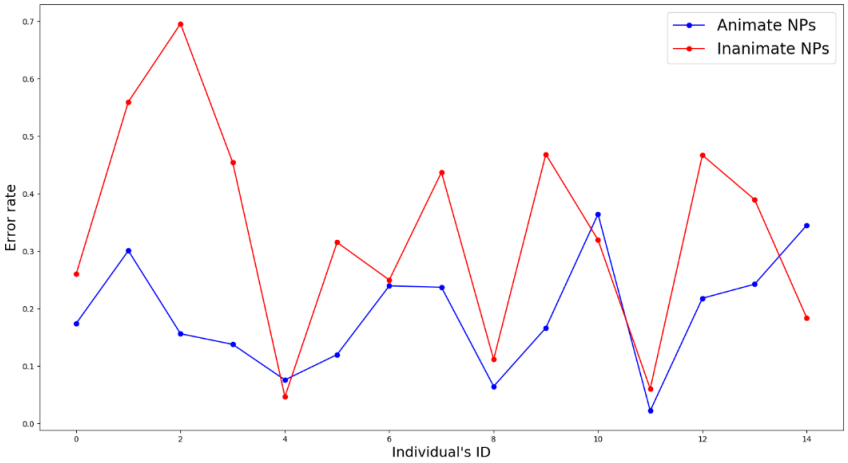


Figure S5: Error rates on animate and inanimate NPs across patients



7. STUDY 5: THE LINGUISTIC SIGNATURE OF HALLUCINATED VOICE TALK IN SCHIZOPHRENIA

Abstract. Very few studies have investigated the formal linguistic aspects of auditory verbal hallucinations (AVHs), though speech is a defining aspect of AVHs. Hallucinated speech heard by 19 patients with schizophrenia and highly frequent voices was obtained online, as and when they spoke, and annotated for pre-selected linguistic variables. Results showed that, consistently across the sample, (i) the grammatical first Person was significantly less represented than both second and third person, and often absent altogether; (ii) overwhelmingly, isolated clauses with no grammatical connectivity (parataxis) were produced, as compared with subordinations, coordinations, and adjunctions; (iii) in all participants except one, virtually no noun phrases (NPs) were anaphoric ones, back-referring to previous NPs, illustrating again a lack of connectivity across utterances. (vi) Sentence-level con- tent was largely personal rather than impersonal, and in impersonal utterances, it was generally vague. (v) Formal syntactic errors were consistently nearly absent, as were semantic level errors such as paraphasias. Voice talk was not generally stereotyped. These results indicate that, despite a certain amount of individual variation, there is a distinctive linguistic profile to voice speech, which constrains theories of AVHs and their neurocognitive basis.

1. Introduction

Hallucinations are one of the core symptoms of schizophrenia and by far the most common form they take is hearing voices (auditory verbal hallucinations, AVH) (Andreasen and Flaum, 1991; Bleuler, 1914; Slade and Bentall, 1998). In some ways, a considerable amount is known about the clinical features of AVH: in different patients (and sometimes in the same patient), the voices can be single or multiple, heard inside or outside the head; they may range in complexity from single words to sentences to conversations, refer to the patient in both the 2nd or 3rd person, and they have themes that are often but not al- ways being derogatory (Jones, 2010; McCarthy-Jones et al., 2014; Nayani and David, 1996). In one important way, however, knowledge about AVH is limited. Namely, while language is a defining feature of the notion of AVH, little is known about the linguistic features they show. One study (Stephane et al., 2003) used hierarchical clustering and multi-dimensional scaling to show that linguistic complexity (voices heard talking in words, sentences, or discourses), as assessed through semi-structured interviews, is one dimension along which the voice hearing experience can

differ. One other study (de Boer et al., 2016) compared the linguistic complexity of voice talk in psychotic and non-psychotic voice hearers as based on direct transcriptions, and found that the former group had a lower mean length of utterance and verb complexity. More systematic linguistic profiling of voice talk based on more fine-grained linguistic measures could contribute important constraints on neurocognitive models of AVHs. Inner speech models, in particular, regard the voice-hearing experience as arising from a failure of properly self-monitoring one's own inner speech (Frith, 1992; Frith and Done, 1989; Jones and Fernyhough, 2007). Leudar et al. (1997) already argued that the pragmatics of voice talk is consistent with this model. However, the degree of phenomenological fit between inner speech and AVH has remained a complex and open issue (Jones, 2010; Rosen et al., 2018). Insights at other levels of linguistic organization can provide further constraints to evaluate such models.

Examination of the linguistic features of voice talk is of interest also in the wider context of schizophrenia, where linguistic abnormalities have been documented to also occur in the patients' expressed speech and comprehension, particularly in patients with formal thought disorder (FTD, speech that is difficult to follow, sometimes to the point of complete incomprehensibility) (Chaika, 1974; Covington et al., 2005; Kuperberg, 2010; McKenna and Oh, 2005). A further well-replicated finding in thought-disordered schizophrenic speech is presence of un- clear reference, particularly in the poorly specified use of personal pro- nouns such as he, she, they, etc., to refer listeners back to previous aspects of discourse (Barch and Berenbaum, 1996; Docherty et al., 2003; Rochester and Martin, 1979). There are certainly hints of some such linguistic abnormalities in one of the very few verbatim accounts of AVHs, from a patient reported by Kraepelin (1913). His voices consisted of a succession of short sentences, probably from different speakers, some of which were nonsensical (‘We inhale you’; ‘Because we have to fear your brain grease’). Linguistic abnormality has also been documented in spontaneous speech of patients with schizophrenia without FTD, where it takes the form of syntactic simplification and errors (Hoffman and Sledge, 1988; Morice and Ingram, 1982; Oh et al., 2002; Thomas et al., 1990).

The aim of the present study was to provide a more systematic linguistic profiling of voice speech in patients with a diagnosis of schizophrenia. Our primary research question was which linguistic variables would characterize this profile and which would not. We also attempted to corroborate the existence of some of the features of AVH

suggested by the existing clinical and research literature. Specifically, given that voices are commonly heard so as to speak in the 2nd (2P, e.g. ‘you’ or 2P agreement on the verb), and 3rd Person (3P, e.g. ‘he/it/the God/a man’), we examined the distribution of grammatical person. Since voices are also commonly reported to take the form of brief phrases and this was a feature in the example of Kraepelin as well as the study of de Boer et al. (2016), we also examined grammatical connectivity between clauses. This specifically included the proportion of clauses that were embedded within other clauses, and how they were embedded, namely as adjuncts (e.g. You are bad because you killed your mother), as clausal arguments (e.g. I think you are bad), as coordinated (You are bad and you killed your mother), or without any grammatical connectivity at all (parataxis). The different types of noun phrases (NPs) produced, i.e. configurations like the devil, your body, she, etc. can also reflect degrees of connectivity between subsequent utterances: e.g., correct use of anaphoric NPs such as the man or he de- pends on the referent of these NPs having been mentioned previously, thus reflecting a narrative connection. Voices are also usually reported as being personal, i.e. directed at the patient, often in a negative way, so we also examined the frequency with which they were personal as opposed to being impersonal in content, in the sense that they reflect states of the world that obtain irrespective of the speaker's relation to them and his or her mental states.

Our specific hypotheses based on clinical impression and previous phenomenological reports were that (i) use of the 1st Person (1P) sin- gular and plural (e.g. Spanish equivalents of ‘I’, ‘my’, or 1P agreement on the verb) would be uncommon, (ii) that clauses would exhibit little grammatical connectivity, with a preponderance of parataxis, (iii) that the proportion of anaphoric NPs would be low, and (iv) that sentence-level content would be largely personal.

Finally, in a more exploratory fashion, we investigated the pattern of formal syntactic errors in voice speech, since normal expressive language in schizophrenia has been found to feature an increased number of such errors in production (Marini et al., 2008; Morice and Ingram, 1982; Morice and McNicol, 1986; Tavano et al., 2008; Thomas et al., 1990), while in language perception, patients with schizophrenia show less sensitivity to syntactic errors as compared with neurotypical controls (Moro et al., 2015). Moreover, since single-sentence semantic- level anomalies characterized the language of formal thought disorder in a previous study (Oh et al., 2002), we explored whether such

anomalies, along with other features of thought disorder such as paraphasias and neologisms, would also be found in voice speech.

2. Methods

2.1 Participants

The patient sample consisted of 19 patients with a diagnosis of schizophrenia or schizoaffective disorder, recruited from five psychiat- ric hospitals in Barcelona (Benito Menni CASM, Hospital Sagrat Cor de Martorell, Hospital Sant Rafael, Parc de Salut Mar) and Zaragoza (Centro Neuropsiquiátrico N.S. del Carmen), Spain. Results are reported from 18 patients, since inspection of one transcript revealed that only 9 words were produced by this patient's voice, grouped into three sentences separated by long pauses to which our analysis scheme could not be ap- plied, after which the patient was removed prior to analysis. All participants met DSM-IV-TR criteria for schizophrenia or schizoaffective disorder, based on review of their clinical history by the patient's psychiatrist and a member of the research team. They were excluded if they (a) were younger than 18 or older than 65, (b) had a history of alcohol or substance abuse/dependence in the last year, (c) had a history of head injury, neurological disorder or medical disorders affecting cognition, (d) had hearing loss, and (f) had had treatment with electroconvulsive therapy in the last six months. All patients were also required to have a current IQ in the normal range (i.e. N70). Current IQ was prorated from 4 subtests from the Wechsler Adult Intelligence Scale (WAIS-III; Wechsler, 1997): Vocabulary, Similarities, Matrix reasoning and Block design. The patients were all on antipsychotic treatment (typical [n = 1], atypical [n = 7], both kinds [n = 10], missing data for one patient). All participants gave written informed consent. All the study procedures were approved by the local research ethics committee and adhered to the Declaration of Helsinki.

2.2 Clinical assessment

The patients were administered the PANSS (Kay et al., 1987) to as- sess positive and negative psychotic symptoms. AVHs were rated with the Psychotic Symptom Rating Scales (PSYRATS, Haddock et al., 1999), auditory hallucinations subscale (AHS). This subscale consists of a semi-structured interview with 11 items referring to frequency, duration, controllability, loudness, location; severity and intensity of dis- tress; amount and degree of negative content; beliefs about the origin of voices; and disruption caused

by the AVHs (see Supplementary Table 1 for a list of the qualitative aspects of AVH in the present sample as obtained from the PSYRATS). To be included in the study, patients were required to score at least ‘once an hour’ in the ‘frequency’ item of the PSYRATS-AHS. This was done to ensure that the patients would experience voice hearing during the time of the assessment. Voice frequency was further examined by asking the patients to signal (tap on the table) each time they experienced an instance of AVHs during a period of 5 min in a quiet environment.

2.3 Procedure

Patients were placed in a quiet environment and asked to repeat verbatim everything their voices said over a 5 to 25-minute period (mean 11 min and 45 s, one patient reported to stop hearing voices after 2 min and therefore was only recorded for that period). Patients were recorded with the built-in microphone in a Dell laptop using in-house developed software written in TCL.

2.4 Transcriptions and annotation

Transcriptions were strictly literal and hence included all repetitions, unintelligible speech (which was marked as such), and indications of speech pauses, which were indicated without time specifications as these had no impact in the linguistic variables. Uncertain words and other non-linguistic sounds and screams were also reported, but not analyzed. Relevant non-verbal aspects of communication were specified, such as tone. Other significant data were reported, such as when the patient started to sing or laughed. Linguistic variables were manually an- notated in CLAN (MacWhinney, 2000) by a linguist rater (AT), who had previously not been involved in the study, including in the transcriptions (performed by PF-C). In the linguistic analysis we annotated, for every single NP, which grammatical Person (1P, 2P or 3P, subdivided into singular and plural) was grammatically specified. In the domain of syntactic complexity, four modes of grammatically connecting clauses were distinguished: subordination (embedding), coordination, adjuncts and parataxis, where the last of these reflects the absence of a grammatical connection. NPs were further annotated for whether they were anaphoric or non- anaphoric, where anaphoricity means that the NPs refer to an object previously mentioned by another NP. In terms of the sentence-level content of utterances, we distinguished between personal and impersonal content, where personal means that speech participants or other objects of the immediate context are the subject of the

utterance, while impersonal content concerns facts about the world relatively independent of the speech context. Formal syntactic errors, i.e. violations of grammatical well-formedness conditions, were also counted. Finally, semantic-level anomalies were annotated according to four variables, capturing paraphasias, violations of semantic selectional restrictions, neologisms and clanging. Definitions and examples (in English translation) are presented in Table 1.

Table 1. Linguistic variables, definitions, and examples.

Variable	Definition	Example
1st Person NP	NP in grammatical 1st Person	*I* don't know.
2nd Person NP	NP in grammatical 2nd Person	*You* will fail.
3rd Person NP	NP in grammatical 3rd Person	*He* didn't say *that*.
Subordination	One clause is embedded in another and dependent on it	You want to hurt people *that are around you*.
Coordination	Two clauses are coordinated when both are at same level.	Why are you talking in Spanish *and* why are you saying this?
Adjuncts	Dependent clauses specifying further information that is not grammatically required.	This has happened *because you are a junkie*.
Parataxis	Utterances not connected grammatically to others.	Ceremony. Don't be afraid.
Anaphoric NP	NPs picking up the referent of a previous NP.	*He* has not repeated *this*.
Personal content	The content of the utterances is directly referring to speakers/hearers or objects/events present in the speech context.	*I* love *you*.
Impersonal content	Sentences stating facts of the world without reference to speaker/hearers.	Spain is a democracy.
Vagueness of content (only annotated in impersonal contents)	The fact referenced lacks specificity or is overly generic.	It is a mortal sin. Respiration is the method.
Impersonal content non vague	A specific fact of the world is referenced informatively.	Here one can speak.
Formal grammatical error	Violations of grammatical well-formedness conditions.	And you pay her in this way [laísmo]. (Y la pagaste de esa manera.)

Variable	Definition	Example
Paraphasia	Word choice for a given referent where another word is expected.	Red Riding Hood walked into the *park* [instead of forest].
Violations of semantic selectional restrictions	Conceptually impossible combination of lexical features	The *pond fell* in the front doorway.
Neologisms	Creation of a new word	*Noises* [non-existent word in Spanish in a context where Moses, the biblical figure, appears to have been intended].
Clanging	Association of words based on similar sounds or inside the same lexical field	Tú eres un culturista. El niño no tiene cultura [lit.: You are a bodybuilder. The child has no culture]

2.5 Statistical analysis

All absolute counts of grammatical Persons in singular and plural were normalized by the total quantity of noun phrases (NPs) produced. This was done both for total occurrences of each of the three grammatical Persons, and separately for their respective occurrences (i) in isolation and (ii) as occurring jointly with other grammatical Persons within the same sentence (e.g., 'I love you', where 1P co-occurs with 2P, or 'I like him', where 1P co-occurs with 3P). We proceeded in the analogous way for the four different modes of clausal connectivity, which we normalized relative to the total utterances produced; and with the relative proportions of anaphoric vs. non-anaphoric NPs, of impersonal vs. personal sentence-level content, of utterances with formal syntactic errors, and with semantic-level anomalies. Results are stated descriptively as averaged percentages. In addition, and wherever appropriate, related-samples analyses of variance (Friedman's related samples two-way analysis of variance by ranks) were carried out to test for significant deviance from equal distributions of instances of the same variable under different conditions, specifically the three grammatical Persons in the case of grammatical Person, and the four modes of clausal connectivity. The α-level was set at 0.05. Multiple comparisons were corrected for by multiplying the observed p-value from the significance tests by the number of tests, k. Then if any k P is <0.05, the test is significant at the 0.05 level. Only significant comparisons are reported.

3. Results

3.1 Demographics and clinical information

Demographic and clinical information for the participants is shown in Table 2. Subjective AVH frequency, as indicated by asking the patients to tap each time they experienced a voice during a 5-minute period, ranged from 1 to 360. The mean was 51.27 (SD = 98.39), median = 13.50 (Interquartile range, IQR = 33.5).

Table 2. Demographic, clinical and neuropsychological variables.

	Mean (SD)
Sex (M/F)	15/4
Age (years)	43.89 (7.71)
Illness duration (years)	19.58 (9.81)
PANSS-total	77.39 (16.09)
PANSS-positive	20.00 (5.13)
PANSS-negative	22.22 (6.72)
PANSS-general	35.33 (10.41)
PSYRATS-AHS	27.83 (8.04)
Estimated IQ	93.59 (11.50)
Current antipsychotic dose (CPZ equivalent, mg)	751.99 (526.35)

3.2 Linguistic variables

3.2.1 Differences in the use of grammatical persons

Use of 1P in isolation, in both singular (S) and plural (P), was rare across the group and completely absent in 12 (singular) and 13 (plural) out of 18 cases (Table 3). A Friedman test (related samples two-way analysis of variance by ranks) confirmed statistically significant differences between total uses of the three Persons ($\chi2 = 19.681$, $p < .001$). Post hoc pairwise comparisons corrected for multiple comparisons showed significant differences between total use of 1P and both 2P ($p < .001$), and 3P ($p = .001$). The same test confirmed that distributions in the combinations of different Persons was not equal either ($\chi2 = 11.019$, $p < .004$). Pairwise comparisons revealed significant differences between combinations of 1w2 (1P with 2P) and of 2w3 (2P with 3P) ($\chi2 = 11.019$, $p < .014$), with the latter significantly more frequent than the former (all significance values Bonferroni corrected). For examples see Supplementary materials.

Table 3. Percentages (Medians) of grammatical Persons (1P/2P/3P: totals; 1/2/3 PS: 1/2/3 Person Singular in isolation; 1/2/3PP: 1/2/3 Person Plural in isolation; w: one Person in combination with another in the same utterance).

Person	**1P**	**2P**	**3P**	**1PS**	**2PS**	**3PS**	**1PP**	**2PP**	**3PP**	**1w2**	**1w3**	**2w3**
%	12,6	48,4	44,3	0,0	19,4	8,8	0,0	0,0	0,5	0,0	5,9	16,1
Total	110	381	391	16	174	107	8	2	31	19	67	186
Median	2	13	12	0	7	3	0	0	0	0	1	5
Mean	5,79	20,05	20,58	0,84	9,16	5,63	0,42	0,11	1,63	1,00	3,53	9,79
IQR	5	23,5	20	1	13,5	4,5	0,5	0	1,5	1	3	11
Min.	0	2	2	0	0	0	0	0	0	0	0	0
Max.	45	72	99	10	30	24	4	2	13	5	31	48
% subjects with 0 instances	36,84	0	0	68,42	5,26	15,79	73,68	94,74	52,63	57,89	47,37	15,79

3.2.2 Modes of clausal connectivity

Median percentages of uses of grammatical connectivity types summarized in Table 4 show that parataxis was by far the most frequent connection type (for examples see Supplementary material). A Friedman test confirmed significant differences between distributions of different types of connectivity ($\chi 2 = 38.92$, $p < .001$). Post hoc pairwise comparisons revealed significant differences in the comparison of ratios of parataxis with coordination ($p < .03$), with subordination ($p < .000$), and with adjuncts ($p < .001$).

Table 4. Median percentages of uses four grammatical connectivity types of clauses.

Clausal connectivity	**Subordination**	**Adjuncts**	**Coordination**	**Parataxis**
%	4,0	5,5	3,9	86,7
Total	47	57	37	566
Median	1	1	1	18

Clausal connectivity	Subordination	Adjuncts	Coordination	Parataxis
Mean	2,47	3,00	1,95	29,79
IQR	3	3	2,5	25,5
Min.	0	0	0	0
Max.	12	15	12	108
% subjects with 0 instances	36,84	36,84	42,11	5,26

3.2.3 Ratio of anaphoric vs. non-anaphoric NPs

Inspections of raw counts of anaphoric NPs revealed one outlier who produced 26 out of a total of 32 instances of anaphoric NPs occurring in this corpus. Of the remaining 17 participants, the voices of 13 had no anaphoric NPs, 2 participants had 1, and 2 had 2. The median percentage of anaphoric NPs of our total NPs was 0% (mean 0,6%).

3.2.4 Impersonal vs. personal content

Median percentages of Personal and Impersonal content out of total utterances were 92,3% and 7,7%, respectively. A Wilcoxon signed-rank test showed a statistically significant difference between utterances with personal vs. non-personal utterance content, $z = 3.44$, $p < .001$. Impersonal content turned out to be often vague upon inspection (e.g. *la respiración es el método* [the respiration is the method]), and only 5 utterances in the corpus (out of a total 72 impersonal utterances) were classed as impersonal-non-vague (see Supplementary materials).

3.2.5 Formal syntactic errors

An extremely low number of utterances with formal syntactic errors was found, corresponding to a raw total count of 6 in the entire corpus.

3.2.6 Single-sentence semantic-level anomalies

Very few instances of such anomalies were found: 0 cases of paraphasias, 1 semantic selectional restrictions (*este niño ni lleva la cultura* [this child does not even carry the culture]), 1 neologism (*Moises*) and 1 possible instance of clanging (*sabes sabueso*).

4. Discussion

Results confirmed our hypothesis that 1P would be significantly under-represented in comparison to the other two grammatical persons, which between

themselves did not significantly differ. This difference is seen (i) when total uses of the three Persons are compared, (ii) in the case of combinations of 1P and 2P, which were significantly less frequent than combinations of 2P and 3P, and (iii) in the case of 1P singular and plural as used in isolation, which were absent altogether in a majority of participants. In such isolated uses, the NPs in question are used in simple sentences in which only this single NP occurs, which allows assessing uses of grammatical Persons controlling for the effects of the co-presence of other Persons. This is also relevant since use of 1P in combination, where it occurred, often did so in such a way that it was still directly related to the voice hearer as addressed in 2P, as e.g. in *te quiero* (I love you), without much additional personal information provided about the 1P referent, i.e. the voice itself. In short, 1P can be 'carried along' by a discourse largely centred around 2P and could in this sense be in part a by-product of this fact and the use of a verb with two nominal arguments.

On the other hand, 2P plural and 3P plural in isolation were also very rare or absent, raising the question of why the absence of 1P in isolation should be special or be of any theoretical interest. In the case of 2P plural, a natural explanation is the nature of the speech situation itself: there is only *one voice* hearer to whom the voice speech is directed, and while, in theory, 2P plural could also be used to address several other voices, this apparently simply never happened in sentences figuring a single NP. Such a natural explanation is not available, on the other hand, for the case of the absence of 1P in isolation, since there always is at least one voice talking. Why then should it be that this voice, which is normally heard as personified, almost never says 'I' in simple sentences with only one NP, and does so much more rarely also when NPs occur in combination with others?

The answer would be immediate if voice talk was largely the impersonal one of a news reader on TV, say, who will barely if ever use 1P except in direct quotations. The observation is startling in the present case, however, where speech content precisely is largely personal. Voice speech is not merely largely personal, moreover, but often takes place in a quasi-conversational setting, in which the voice hearer is, for the most part, directly addressed in 2P by the voice, or else is the 3P topic of a conversation of one voice with another. There seems to be no reason at all, in such a setting, why 2P and 3P should be nearly four times as frequent in sentences with two nominal arguments and be used near-exclusively in simple sentences. A tendency to under-use 1P by a speaker in a

personal conversational setting would mean that this speaker does not tend to talk about himself (engages in self-reference): he is not present in the conversation as a first-person self. Insofar as the voice hearer also does not reply, there is thus little or no 1P reference in this conversational setting at all. This is even more remarkable in light of the fact that numerous elements of such speech, such as swearing, are considered 'expressive' and expressives are inherently related to a speaker's self (Potts, 2007). What transpires, then, is that selfhood at this deeper, emotional level dissociates in AVH from the kind of 'conversational' or thinking self that is referenced in English with the 1P pronoun.

As for the rarity of 3P plural in isolation, this suggests that in sentences featuring a single NP, no pluralities of things or persons were ever referred to: the voices stuck to single, individual referents. While this may reflect a form of 'concretism' in voice speech that is worth noting, our finding about 1P again does not have such a straightforward answer. As noted, the absence or rarity of 1P in a personal, conversational setting could not simply be an accident, and a form of concretism would certainly not predict it either. One could further speculate that this finding might be due to the tendency of the voices to often engage in insults, threats, or praise (which itself, in order to be explanatory as a hypothesis, would then have to have some other explanation, unrelated to the poverty of the language and the distribution of grammatical persons used). However, while voice talk is indeed frequently marked by such speech act types, it is by no means the case that this type of speech necessarily entails the absence of 1P: *I hate you, I will kill you, Don't piss me off, I love you, Listen to me, Don't talk to me, You are worse than my dog, I am the Master, I am fed up*, etc., are all sentences in the insulting/praising/commanding mode, and they all contain 1P. Moreover, while this argument might make us expect more 2P use than 1P use, it would not explain why 1P in total was underused compared with both 2P and 3P, and that this also applied to combinations of 1P with 2P relative to combinations of 2P and 3P.

While we thus have no convincing explanation to offer for our finding, it is worth highlighting it as an explanandum for any theory of AVH, which we would characterize descriptively as follows. Person distinctions are deictic distinctions: they regulate whether a person referenced as taking part in some event is identical with a speech participant or not: with the speaker in the case of 1P, and with the hearer in the case of 2P, or with neither of them in the case of 3P. The normal conversational speech situation is that there is a speaker identifying himself in 1P, i.e. the 'center' or 'origo' (Buehler, 1934) of the

deictic space in which speech takes place. This person speaks to a second person (a 'you', the 2P) about the world (the 'it/he/she', i.e. the 3P). Voice talk in this sense thus effectively lacks a deictic center – it is deictically de-centered. Theories of AVHs need to explain this deictic shift, which is in line with theoretical approaches pointing to a disruption of deictic anchoring of thought in schizophrenia potentially not specific to AVH (Crow, 2010; Hinzen et al., 2016). The result is also significant with regards to studies of voice hearing in non-clinical populations. It is noteworthy that all examples given in de Boer et al. (2016) of voice speech in which 1P occurred, were from the non-clinical sample.

Results on grammatical connectivity of clauses also confirmed our predictions. Clauses are the minimal structural configurations that are meaningful at a propositional level. Connections between clauses in normal speech are reflected in the kind of grammatical devices we distinguished here: subordinations, coordinations, and adjuncts, all of which are crucial to narrative, dialogue, and discourse. A restricted kind of lack of connectivity occurs in specific rhetorical contexts (e.g. *he came, he saw, he won*), or when a thematic connection makes connectors superfluous (e.g. *Inspector Clouseau arrived in the evening. The house was dark. He rang the bell*.). But cases annotated as 'parataxis' in our study were not of these kinds: they either exhibit no grammatical connectivity at all or else exhibit only semantic connectivity in the sense of being broadly subordinated under a given broad lexical-thematic field (e.g. *killing*, see example 5). Effectively, then, voice speech in our sample is language with a strong tendency to be reduced to the single-sentence level, lacking connectivity and embedding. This was further confirmed by the virtual absence of anaphoric NPs – and ipso facto the absence of referential connections across utterances that this entails.

In line with results from phenomenological studies, linguistic annotations based on analysing referencing revealed that a median 92,3% of utterances were personal-level utterances, and that the vast majority of the remaining non-personal utterances were defective in the sense that their content was not clearly identifiable. This shows that where voice speech deviated from its pattern of operating at the personal level, it was rarely the case that language strayed away from the speech context and lived up to its normal function and potential, namely capturing informative content about the world in objective, impersonal terms. It is inherent to language in its normal use in mental health that it can convey thoughts which, while expressing the mental states (beliefs, desires) of the

speaker, capture states of the world that hold independently of that speaker (e.g. *Gold is yellow, John is married*).

Finally, with regards to syntactic and semantic errors, our hypothesis concerning syntactic errors was not confirmed. Voice speech was virtually error-free in either the syntactic or the semantic terms we distinguished, demonstrating that, within schizophrenia symptomatology, it is its own register that has a distinctive profile. We speculate that the low grammatical complexity of such speech may act as a kind of protector of its syntax, since if speech lacks complexity, syntactic errors are less likely to occur. As for semantic-level distinctions, lexical and phrasal selection are clearly not what makes such speech deviant.

Summarizing, what emerged in this study as distinctive of voice speech are: (i) the shift in the deictic space in which speech takes place, away from 1P to non-1P, while at the time being largely personal; (ii) the effective lack of grammatical connectivity, with speech reducing to the single-clause level without embedding, and (iii) reduction to the personal level language which does not reach informative levels depending on the presence of objective (impersonal) meaning.

These results constrain theories of AVHs. In particular, on the still widely maintained 'inner speech' theory of AVHs going back to Frith (1992), voice talk is inner speech misattributed to an external source (Jones and Fernyhough, 2007). As noted, the degree of 'fit' of this model with phenomenological features of AVHs has remained complex and contentious (Jones, 2010; Rosen et al., 2018). But it seems difficult to fit this model to almost any of the linguistic features identified here. It is important in this regard to distinguish inner speech from a different linguistic genre, namely self-talk (Holmberg, 2011), which tends to involve a person referring to herself in 2P (e.g. *You idiot!, Now pull yourself together!, You messed this up again!*). By contrast, inner speech or ruminations will often, if not mostly, be 1P (*I really don't like this, I still need to do the shopping, Damn, my secretary is still waiting, Why does he stare at me like this,* etc.), though they can also be impersonal (*They'll never repair this bridge, Joe has really gotten old,* etc.). Voice speech in our sample did not have the former feature, nor were impersonal utterances qualitatively of the same kind. The 'perceptual' theory (Mørch-Johnsen et al., 2017), according to which AVHs arise from abnormal activations of temporal cortex involved in language perception, predicts the specificity of the patterns we have found even less.

These failures could be rescued by more specific such models. Thus, one particular conception of inner speech regards it as based on internalized interpersonal dialogue as a raw material (Fernyhough, 2004). This would still not predict the low proportions of grammatical 1P, whether referring to the voice hearer or to other persons or voices featuring in such dialogue. This conception also proposes that external dialogical speech, when internalized, undergoes important transformations leading to syntactic and semantic changes, particularly a form of 'syntactic abbreviation' involving the dropping of subjects in favor of predicates, and to fragments of verbal images rather than fully formed utterances, until inner speech ultimately 'loses most of the acoustic and structural qualities of external speech' (Fernyhough, 2004: p.55). Yet again, none of these features were observed in the AVH studied here, where utterances were mostly fully formed, very little ungrammaticality was found, and virtually no anaphoric NPs were present (which dropped subjects in normal speech would typically be, corresponding to 'old' information); moreover, despite voices present identified as people talking to or about the voice hearer continuously, the dialogical or conversational involvement of the voice hearer was very rare in our sample; and it is doubtful whether a voice's running commentary on what the voice hearer is thinking, or a series of instructions or commands that the voice hearer is following, can count as 'dialogical'. Ultimately, the question of whether inner speech exhibits a similar linguistic profile as hallucinated voice talk, whether with regard to grammatical Person or grammatical connectivity, could only be answered by a direct comparison using the same method of annotation in order to obtain such a profile. But this is hardly possible since, by definition, inner speech is not hallucinated voice talk, i.e. AVH, and hence is not presented and transcribable in the same way, though other methods are available (Alderson-Day and Fernyhough, 2015; Hurlburt et al., 2017).

This illustrates that and how linguistic analysis of AVH can provide an objective basis against which theories of AVHs can be evaluated and neurolinguistic correlates of such distorted speech can be explored. Specific linguistic features of voice talk documented here may also provide important clues for their neural correlates. This in particular applies to the processing of deictic distinctions (grammatical Persons), which has been explored with fMRI in autism spectrum conditions as compared with neurotypical individuals (Mizuno et al., 2011), but has not yet been a focus of research in AVH.

Our results suggest further studies integrating language as a dimension of AVH that is not captured by traditional phenomenological descriptions and methods, and exploring links between linguistic analysis and phenomenological data as obtained from interviews. de Boer et al. (2016) already explored these links but did not find a correlation between a sum score of syntactic complexity in AVH and the amount of negative emotional content. Nor is it clear conceptually why such a link should obtain, i.e. which implications such content would have for structural linguistic complexity; or why it should obtain between other features of voices, such as whether they are heard inside or outside of the head, whether their loudness is like that of the own voice, or what beliefs are held about them. In line with this, post hoc inspection of qualitative aspects of voices as captured by the PSYRATS (Supplementary Table 1), did not suggest any tendencies or correlations, though small numbers prevent formal correlational analyses. This may indicate that the linguistics of the voices is a dimension of AVH at least partially independent of their non-linguistic phenomenological dimensions.

5. Conclusion

This study reveals that AVHs have a less explored linguistic dimension that is subject to formal analysis and shows a distinctive profile, which may illuminate the nature of voice hearing as such and informs neurocognitive models. Specifically, while voice talk was generally neither stereotyped nor ungrammatical, grammatical complexity was seen to virtually reduce to clauses and NPs with no grammatical connectivity to other clauses or NPs, respectively. The use of grammatical Person showed a striking pattern, in which the grammatical first Person was significantly less represented than both the second and third persons and often absent altogether, whether referring to the voice hearer or another voice heard. As Person distinctions are deictic distinctions, this may suggest a disruption in the deictic anchoring that is a necessary requirement whenever thought or speech is generated, linking thought content to a person thinking it.

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Conflict of interest

All authors declare that they have no conflicts of interest.

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Supplementary Materials

Voice speech samples

1. **Differences in the use of grammatical Persons**

Typical examples of voice talk with 1P are the following (for examples without 1P see examples 3, 4, 6). Examples are presented with literal word-word translations as well as English glosses and represent utterances following each other in sequence:

Examples:

(1) A este tío lo vamos a matar.
To this guy him we will to kill
'We are going to kill this guy'

Pacifícate joder.
Calm down fuck
'Calm down, damn it.'

Estate tranquilo.
Be quiet
'Be quiet.'

Qué pesado que eres.
How heavy that you are
'You are so annoying.'

Mañana le cuento la verdad mañana.
Tomorrow him/her I-tell the truth tomorrow
'Tomorrow I am going to tell him/her the truth tomorrow.'

Qué joder idiota.
What fucking idiot
'What a fucking idiot'

Qué esquizofrenia ni qué pollas.
What schizophrenia not that cocks
'Don't talk to me about fucking schizophrenia.'

Quiero ir a la guardia civil.
I want go to the guard civil
'I want to go to the Civil Guard'

¿Te crees muy importante no?
You you-believe very important no
'You think you are very important, don't you?'

(2) De verdad ¿tú qué piensas?
Of truth you what you-think
'You, what do you really think?'

No estás loco.
Not you-are crazy
'You are not crazy.'

Te *quiero*.
You I-love
'I love you'

Vas a cambiar el mundo.
You-will to change the world
'You are going to change the world.'

De verdad no me crees.
Of truth not me you-believe
'You do not really believe me.'

Es raro pero es así.
It-is weird but it-is so
'It is weird, but that is how it is.'

Te *quiero* desde siempre.
You I-love since always
'I have loved you always.'

Que no te dé vergüenza.
That not you give shame
'Do not be ashamed.'

Los tienes flipando a todos.
Them you-have freaking to all
'Everyone is crazy for you.'

2. Modes of clausal connectivity

Two typical examples for a stretch of voice talk without grammatical connectivity is given in examples 3 to 5:

Examples:

(3) Eres un cobarde.
You-are a coward
'You are a coward.'

Ya vienen.
Already they-come
'They are coming.'

Ya están aquí.
Already they-are here
'They are already here.'

Por qué no fuiste al cementerio.
For what not you-were at-the cementary
'Why did you not go to the cemetery'

(4) Mata a otra gente.
Kill to other people
'Kill other people.'

Haz una matanza.
Make a slaughter
'Create a bloodshed.'

Joan haz una matanza de gente.
Joan make a slaugher of people
'Joan, slaughter people.'

Suicídate después suicídate.
Kill-yourself after kill-yourself
'Kill yourself, afterwards kill yourself.'

(5) Está cobrando por la puta cara.
He-is charging for the fucking face
'He is charging money for the fucking face.'

Pilla colillas.
Catch cigarettes
'Get cigarettes.'

Hay que robar la cartera.
There-is that steal the wallet
'You have to steal the wallet.'

El Corte Inglés no le puede ni ver.
The Corte Inglés not him can not see
Colloquial expression for 'they hate him'

Su paga tiene que quedar para el estado.
His salary must that leave for the state
'His salary has to be given to the state.'

Me cago en los muertos de la asistenta social y del rey de España.
I shit on the dead of the assistant social and of-the king of Spain.
'I shit on the dead of the social worker and the King of Spain.'

Hijo puta este.
Son bitch this-one
'This son of a bitch.'

Estamos hartos ya de este.
We-are tired already of this-one
'We are already tired of him.'

Pilla colillas para porros.
Catch cigarettes for joints
'Get cigarettes for joints.'

Hostia puta.
'The fuck.'

Hijo puta.
'Son of a bitch.'

Por qué no lo dejamos ciego?
For what not him we-leave blind

'Why don't we make him blind?'
Este tiene mucha cara.
This has much face
'This guy is shameless.'

3. Impersonal vs. personal content

The full list of impersonal-non-vague utterances (coming from the voices of different participants) is given here:

Examples:

(6) Ya son las cuatro de la tarde. [The statement was accurate]
Already are the four of the evening
'It's already four in the afternoon.'

Es raro pero es así. [Referencing a previous statement.]
'It is weird but it's like that.'

Cuánto tiempo falta para terminar?
How-much time lacks for finish
'How long until we finish?'

Aquí se puede hablar.
Here one can talk.
'Here one can speak.'

Això no ho ha repetit. [Talking about a different voice.]
That not that has repeated
'He did not repeat that.'

Supplementary Table 1. Qualitative aspects of AVH in the present sample: frequency table showing the amount of patients that scored in each level of the PSYRATS scale items (missing data for one patient).

Item	Levels	Number of patients
Frequency	Voices not present or present less than once a week	0
	Voices occur for at least once a week	0
	Voices occur at least once a day	1
	Voices occur at least once an hour	9
	Voices occur continuously or almost continuously	8
Duration	Voices not present	0
	Voices last for a few seconds, fleeting voices	6
	Voices last for several minutes	3
	Voices last for at least once an hour	4
	Voices last for hours at a time	5
Location	Voices not present	0
	Louder than own voice	1
	Extremely loud, shouting	3
Beliefs about origin of voices	Voices not present	0
	Believes voices to be solely internally generated and related to self	4
	Holds < 50% conviction that voices originate from external causes	3
	Holds > 50% conviction (but < 100%) that voices originate from external causes	5
	Believes voices are solely due to external causes (100% conviction)	6
Amount of negative content	No unpleasant content	3
	Occasional unpleasant content (< 10%)	1
	Minority of voice content is unpleasant or negative (< 50%)	3
	Majority of voice content is unpleasant or negative (> 50%)	7
	All of voice content is unpleasant or negative	4
Degree of negative content	Not unpleasant or negative	2
	Some degree of negative content, but not personal comments relating to self or family, e.g. swear words or comments not directed to self	1
	Personal verbal abuse, comments on behavior, e.g. 'shouldn't do that or say that'	3
	Personal verbal abuse relating to self-concept, e.g. 'you're lazy, ugly, mad, perverted'	5
	Personal threats to self, e.g. threats to harm self or family, extreme instructions or commands to harm self or others	7
Amount of distress	Voices not distressing at all	3
	Voices occasionally distressing, majority not distressing (< 10%)	4
	Minority of voices distressing (< 50%)	3
	Majority of voices distressing, minority not distressing (> 50%)	1
	Voices always distressing	7

Intensity of distress	Voices not distressing at all	3
	Voices slightly distressing	4
	Voices are distressing to a moderate degree	2
	Voices are very distressing, although subject could feel worse	5
	Voices are extremely distressing, feel the worst he/she could possibly feel	4
Disruption	No disruption to life, able to maintain social and family relationships (if present).	2
	Voices cause minimal amount of disruption to life e.g., interfere with concentration although able to maintain daytime activity and social and family relationships and be able to maintain independent living without support.	4
	Voices cause moderate amount of disruption to life causing some disturbance to daytime activity and or family or social activities. The patient is not in hospital although may live in supported accommodation or receive additional help with daily living skills.	3
	Voices cause severe disruption to life so that hospitalisation is usually necessary. The patient is able to maintain some daily activities, self-care and relationships while in hospital. The patient may also be in supported accommodation but experiencing severe disruption of life in terms of activities, daily living skills and/or relationships.	4
	Voices cause complete disruption of daily life requiring hospitalisation. The patient is unable to maintain any daily activities and social relationships. Self-care is also severely disrupted.	5
Controllability	Subject believes they can have control over the voices and can always bring on or dismiss them at will.	0
	Subject believes they can have some control over the voices on the majority of occasions.	3
	Subject believes they can have some control over their voices approximately half of the time.	1
	Subject believes they can have some control over their voices but only occasionally. The majority of the time the subject experiences voices which are uncontrollable.	2
	Subject has no control over when the voices occur and cannot dismiss or bring them on at all.	12

8. GENERAL DISCUSSION AND CONCLUSIONS

8.1. Summary of the five studies: the importance of language decline

In general terms, the five studies forming this doctoral thesis corroborate the importance of language in the clinical context. The starting point of this thesis was the idea that so-called 'cognitive' or 'mental' disorders also harbour important linguistic diversity. As mentioned in the introduction, my main objective was to delve deeper into language decline in both disorders, refining the relevant variables for the analysis of language in the clinical setting. More specifically, we have provided significant results on referential dysfunction in both HD and SZ. These contributions will be expanded in the following pages.

This section, as a preamble to what will be presented throughout chapter 8, is a brief summary of the results obtained in each study. Section 8.2 discusses the most relevant results of each study. Section 8.2.1 presents a comprehensive linguistic profile of the HD population in the conversational context. Section 8.2.2 on the processing of illicit syntactic movement, and section 8.2.3 focuses on the referential domain. Regarding language in SZ, section 8.2.4 lists the referential abnormalities, and section 8.2.5 presents the linguistic profile of AVHs. This is followed by the most important contributions of the research forming this thesis (8.3), the limitations of each study (8.4) and future lines of research (8.5).

In the first study *(chapter 3*), a linguistic profile of HD patients was developed by analysing spontaneous speech samples from 20 patients (compared to 20 NT subjects). In this research, a total of 56 variables related to grammar organisation were validated, and drawn from a previous study analysing narrative discourse in HD (Hinzen et al., 2018). Results confirmed that there is a set of features characterising the linguistic profile of this disorder. Pre-symptomatic and symptomatic patients exhibited differing but complementary language disturbances in certain grammatical domains. In the domain of fluency, prodromal subjects showed patterns marked by prolongations, full pauses and repetitions, whereas symptomatic patients employed more empty pauses, truncations and reformulations. In the domain of sentence connectivity, the discourse of HD subjects was characterised by poor grammatical connection, with a predominance of isolated clauses. The domain of reference also reflects this lack of grammatical connection through changes of topic, vague or truncated topics, and inclusion of unknown referents.

The second *(chapter 4*) and third study *(chapter 5*) arose from the same corpus of subjects: 31 NTs and 31 patients in pre-symptomatic, early and advanced stages. Since the general linguistic profile of HD has already been extracted in the previous study, here we delved into two linguistic mechanisms that have proved problematic for this population: reference and syntactic movement. Thus, in the second study, a grammaticality judgement task was devised to show that HD involves a loss of cognitive control over the manipulation of complex sentences in terms of structural hierarchy and syntactic movement. On the other hand, in the third study, through a picture-sentence matching task, it is shown that HD patients are less sensitive to syntactic locality constraints, particularly affecting co-referential structures.

As for linguistic anomalies in subjects with SZ, two different corpora were analysed. On the one hand, in the fourth study *(chapter 6*) a linguistic profile of AVHs was created by analysing the transcriptions of 19 patients with AVHs. AVHs are characterised by the dominance of parataxis (isolated clauses with no grammatical connection), due to the majority use of non-anaphoric nominal syntagms (no connection with previous phrases) and the absence of the first person, grammatical errors and semantic errors. In the second study *(chapter 7*) a corpus of spontaneous conversations of 38 FTD patients was re-analysed. In this study, the use of nominal phrases was investigated through anomalies related to different grammatical domains: reference, argument structure, lexicon and morphosyntax. As the results show, the referential domain was the most affected compared to the other categories. Morphosyntax and lexicon were less affected compared to the number of errors, whereas argument structure was positioned in the middle of both domains.

In summary, the results of the five studies show that it is possible to create specific profiles of language decline for both HD and SZ. In certain cases, this impairment is related to the clinical symptomatology, characteristic of the disorder, such as the distorted thought, characteristic of SZ. In other cases, the study of linguistic abnormalities can detect the cognitive change that standard neuropsychological batteries fail to track, as in the case of HD.

8.2 Discussion of the results

Thus, the main aim of this thesis is to detect anomalous linguistic patterns both in HD and SZ patients. Given that these diseases have different symptomatology, language

disturbance will also manifest show characteristics for each of them. In both disorders, the importance of language as part of the generalised cognitive deficit has been underestimated. The clinical literature defines HD as a disorder of motor origin with progressive cognitive impairment starting in pre-symptomatic stages. In the case of SZ, it is considered as a disease that primarily affects thought and behaviour and is accompanied by a number of cognitive deficits.

Therefore, standard contemporary cognitive models of both diseases do not give language a central status: neither theoretically nor clinically. In our view, some of the symptoms associated with both HD and SZ may be partially language-dependent, and language disturbance may even precede the onset of other symptoms considered more relevant in the clinical profile of the two diseases. In the following section, the most relevant results of each research are discussed with the intention of analysing the role of language in the cognitive decline of HD and SZ.

8.2.1 Expanding the language profile of HD

While it is true that Hinzen et al. (2017) produced a comprehensive profile of the linguistic change witnessed in HD, the analysed discourse sample belongs to the narrative genre. Study 1 *(Chapter 3*) replicated their methodology in a more natural linguistic setting: unguided conversation simulates normal social language use and allows for the detection of errors associated with the spontaneity of context. The extracted results provided a more accurate linguistic profile of this disease, maintaining language degeneration as a marker of HD progression even in pre-symptomatic stages.

Among the most relevant results, we may remark the pattern of fluency that distinguishes the pre-symptomatic population from the group with overt HD. While the speech of pre-symptomatic subjects was characterised by prolongations, full pauses and repetitions, patients exhibited a linguistic profile marked by empty (non-filled) pauses, rephrasings and truncations. This suggests that manifest HD population has ‘gaps’ in their speech (pauses and truncations) and that pre-symptomatic subjects are aware of the disruption to the flow of speech and attempt to fill these gaps by using prolongations and repetitions. Following this pattern, the same tendency of discursive repair was found in the domain of reference: the variable called ‘definiteness repair’ captures processes of self-correction of anomalously introduced referents, and has only shown significant

results in the pre-symptomatic group. Therefore, there is a certain degree of perception of communication failures that loses impact as the disease progresses (Illes, 1989).

It is important to note that these patterns of fluency break arise in positions related to clause boundaries, suggesting the importance of indexing syntactic positions where patterns of disfluencies originate. Following this line of research, results in the connectivity domain confirmed the importance of syntactic organisation in HD. As this disease progresses, a reduction in syntactic complexity was observed, and monoclausal or grammatically independent structures stand out.

However, the discourse of pre-symptomatic subjects was characterised by the anomalous use of subordinate and co-ordinate structures. In total numbers, they used these structures to the same extent as NTs, the difference being that they used them incorrectly. As neurodegeneration progresses, the use of more complex hierarchical structures is drastically reduced and unconnected clauses appeared instead. These results followed the same line of thought as previous research indicating a reduction of hierarchical complexity in this population, and using different linguistic analysis variables (Gordon, & Illes, 1987; Murray 2000, Murray & Lenz, 2001; Hinzen et al., 2018).

This lack of grammatical connection appeared not only in the way clauses were combined and in the resulting loss of syntactic complexity, but also in the domain of reference. The discourse of manifest HD subjects was riddled with topic changes, anomalous truncations, vagueness and ambivalence of referents.

In summary, our results reinforce the idea that neurodegeneration in HD affects central domains of language processing since prodromal stage. However, it is not possible to detect this language decline with traditional clinical tests. Although the manifest HD group differs from the other two groups during language tests designed for aphasic patients (BDAE), performance on these tests remains high and does not correlate with the linguistic variables introduced in our study. Moreover, other standardised neuropsychological tests do not reveal any cognitive decline in the pre-symptomatic population, except for MMSE. But even in manifest HD, MMSE scores do not indicate severe cognitive impairment and do not correlate with any language domain in the investigation.

Language impairment is therefore not predicted by non-linguistic cognitive impairment. In line with the study by Hinzen et al. (2018), these results corroborate that linguistic change is evidence of cognitive decline in HD prior to the onset of motor symptomatology, and it is not possible to detect this impairment through standard neuropsychological tests. Thus, language plays a key role in tracing the evolution of HD since prodromal stages.

8.2.2 Syntactic movement processing in HD

As mentioned in previous literature, the HD population exhibits difficulties in processing non-canonical sentences, such as passive constructions (Teichmann et al., 2015; Szalisznyo et al., 2017). Furthermore, it has been observed that the linguistic production of these patients is characterised by a reduction in syntax complexity (Gordon, & Illes, 1987; Murray 2000, Murray & Lenz, 2001; Hinzen et al., 2017; Tovar et al., 2020). As discussed in section 8.2.1, the last two studies noted an increase in the use of clausal coordination over subordination from prodromal stages, indicating a loss of specific forms of hierarchical complexity.

Given these results, in Study 2 *(chapter 4*), using a violation detection paradigm, we investigated whether HD impacts on locality constraints in syntactic movement. These linguistic rules govern whether or not it is possible to move a constituent out of a local phrasal context. Broadly speaking, results showed that the ability to detect illicit syntactic movements in the HD population is profoundly affected.

It is not possible to attribute these results to a generalised linguistic processing problem in judging complex stimuli, as patients are able to judge correct complex sentences. Moreover, a specific pattern has been observed when judging incorrect sentences: sentences with semantic errors are judged significantly more accurately than those with syntactic anomalies. Thus, there is a problem in detecting illicit syntactic movement: when sentences are manipulated to violate a syntactic restriction on movement, HD subjects do not interpret them as violations.

Despite this apparent specificity, it would be wrong to characterise this deficit as exclusive to the syntactic domain. Semantic processing, although significantly less affected, is also impaired in this population. Significant results, however, are only obtained in subjects with initial HD. As discussed in the introduction, there is a semantic deficit in HD that was related to the processing of complex lexical-semantic structures.

In our view, these results do not indicate the existence of a semantic deficit *per se*, but rather that the problem originates in the step of combining concepts using syntactic structures. Therefore, we conclude that neurodegeneration of cortico-striatal circuits in HD has an impact on the integration of linguistic information: both in terms of hierarchical manipulation through syntactic movement, and when integrating lexical concepts into a proposition.

In summary, these results indicate the importance of the striatum in processing the illicit movement of constituents. Moreover, neuropsychological predictors did not reveal relevant results, which could lead to the existence of a specifically linguistic deficit in HD.

8.2.3 Processing of referential structures in HD

Previous studies showed that the subcortical damage in HD has a direct impact on the reference domain (Hinzen et al. 2018, Tovar et al. 2020), and more specifically, difficulties were detected in applying the syntactic rules governing co-referentiality (Sambin et al., 2012). For example, in sentences like *He entered when Paul was getting dressed*, Principle C blocks the co-reference between the pronoun *he* and the NP *Paul*. The results obtained in Sambin et al (2012), after controlling for working memory limitations, showed that HD subjects are able to correctly establish referential dependencies between NPs over long distances, but fail when they must avoid co-reference as dictated by Principle C.

Taking this previous research into consideration, Study 3 *(Chapter 5*) tested the sensitivity of the HD population to the three principles of BT. Principle A requires establishing the co-reference of reflexive and reciprocal pronouns in a local domain. In *Mary combs herself,* the grammatically plausible reading is that Mary does her own hair, not that of a different referent. In contrast, Principle B states that in sentences such as *She tucks Mary in*, the referent of the pronoun *she* must be placed outside the local context and cannot be coreferential to *Mary.*

Therefore, in this study we researched the impact of striatal neurodegeneration on the comprehension of referential dependencies in the three contexts established in BT, both in simple and complex sentences. Results showed that both early HD subjects and advanced HD patients performed significantly worse than NTs. Although pre-symptomatic patients were included, no significant results were obtained for this stage.

Contrary to predictions, no differences were found between the three BT conditions or between simple and complex sentence contexts.

Overall, these new results confirm sensitivity loss to syntactic locality restrictions in HD patients in both Principle C and the other BT principles. A generalised problem of comprehension and referential processing is therefore evident, in accordance with the results obtained by Sambin et al. (2012). In conclusion, the striatum seems to play a relevant role in the processing of referential structures.

8.2.4 Examining referential deficits in SZ

As discussed in the introduction, although SZ has traditionally been considered a thought and behavioural disorder, certain authors suggested that it is possible to reinterpret the three main positive symptoms of SZ as a result of language collapse (Hinzen & Rosselló 2015). Thus, they conceptualised SZ as a language disorder that has an impact on thought structures mediated by the linguistic system. Therefore, considering that it is not so simple to differentiate between language and thought, SZ is the perfect model of pathological change to investigate the relationship between thought and linguistic systems.

Study 4 *(chapter 6*) investigated referential function in SZ and +FTD patients. In particular, a specific type of referentiality (configurationally originated) was analysed: NPs may take up referential positions within utterances. In general, significant differences were observed regarding the use of different types of NPs, as suggested by previous studies (Rochester & Martin, 1979; Docherty et al., 2003, McKenna and Oh, 2005).

More specifically, Study 4 looked for linguistic disintegration patterns across different hierarchical layers of linguistic organisation, including: the referential domain, the argument structure, the lexical domain and the morphosyntactic domain. Results showed that referential anomalies are the most relevant within the FTD linguistic profile. In contrast, both morphosyntax and lexicon appeared comparatively better preserved. It seems that, although at a superficial level the language shows some integrity, in reality we are faced with a profound deterioration in referential functioning. Previous studies already pointed out that referential anomalies play a key role in the linguistic profile of SZ, and particularly the +FTD population (Sevilla et al., 2018; Cokal et al., 2018, 2022).

Overall, these results showed how language and thought disintegrate together in FTD. Thought is referentially anchored in language and language serves to convey thought. Therefore, it is not possible to separate the referential function from language: referentiality is the basis of the linguistic system and linguistic parameters are sensitive to referential content. Thought disturbances cannot be separated from the concrete linguistic mechanisms that externalise them. These results also provided evidence that linguistic disintegration and thought disorder progressively affect different hierarchical layers of linguistic organisation.

8.2.5 Linguistic profile creation of AVHs

Very few studies have explored the formal linguistic aspects of AVHs. As study 6 *(chapter 7*) revealed, despite some individual variation, there is a distinctive linguistic profile that characterises the language of the reported voices. In general, structural complexity was found to be reduced to clauses and NPs with no grammatical connection to other linguistic units. These data confirmed the conclusion of Boer et al. (2016), stating that reduced syntactic complexity is a distinctive feature of the psychosis population. This lack of grammatical connection was also observed at the NP level. According to results, 84% of the NPs were non-anaphoric, i.e., they have no connection with the NPs previously introduced in the discourse.

In terms of content at a sentence level, results revealed a mostly personal discourse with no objective content. Impersonal statements were also uninformative, as they were vague and inaccurate. Despite the abundance of personal content, the use of the first person was significantly reduced compared to the second and third person, and sometimes disappeared completely from the discourse. This suggests that the voices do not tend to talk about themselves, as is usually the case in normal conversation.

These results limit interpretations of the voices reported in psychosis episodes. Some authors analysed AVHs as abnormal experiences of inner speech (Stephane et al., 2003) or inner speech misattributed to an external source (Jones & Fernyhough, 2007). These theories do not explain the transformation of the deictic space into the second and third grammatical person, both in the case of the listener and in the case of the voices participating in the dialogue. Authors also suggested that discourse, when internalised, is syntactically and semantically distorted. However, our results show that formal syntactic errors are almost non-existent, as are errors in the semantic domain, such as paraphasia.

To sum up, this study illustrates that, although AVHs have a less explored linguistic dimension, it is possible to extract a distinctive linguistic profile. Simultaneously, research on the language of AVHs allows us to evaluate theories about the origin of this phenomenon and to explore the neurolinguistic correlates of such distorted speech (Fuentes-Claramonte et al. 2022). Both objectives are interrelated: discovering the main features of linguistic impairment leads to the development of linguistic paradigms that are verified in neuroimaging studies. Ultimately, these studies make it possible to detect the brain regions involved in language processing.

8.3 Main contributions of this research

Our results are direct evidence of the relevance of linguistic impairment in the clinical profiles of HD and SZ. Previous research already showed that it is possible to construct a specific profile of language change as a diagnostic measure in many disorders, most notably Alzheimer's disease (Ahmed et al. 2013, Chapin et al., 2022), Parkinson's disease (Liu et al., 2015; García et al., 2017), autism (Eyler et al., 2012; Lombardo et al., 2015, Sterponi et al. 2015) and Down syndrome (Ypsilanti & Grouios, 2008; Carvalho et al., 2018).

In the case of HD, we contributed to the broadening of the previous linguistic profile of this disease (Hinzen et al. 2018), including prodromal stages where non-linguistic cognitive change is (apparently) not yet visible. After an exhaustive analysis of the most relevant linguistic features of conversational discourse, the extent and implications of this primary language decline have been qualified by investigating two specific grammatical phenomena: referential dependencies and syntactic movement. As results showed, the processing of both linguistic mechanisms is profoundly impaired in HD. Taking into account that both, the displacement of elements within the syntactic hierarchy and the referential function, are phenomena that cross and connect more than one grammatical domain, we consider that it would be necessary to investigate other linguistic contexts in order to know to what extent these two mechanisms disintegrate in HD.

As for language in SZ, we have contributed to the ratification of previous studies pointing to a problem with referential definiteness in SZ, and in FTD in particular (Sevilla et al., 2018, Cokal et al., 2018, Cokal et al., 2022). These results encouraged us to propose that SZ should be characterised as a disorder that affects both linguistic system and

thought structures equally, as it is not possible to dissociate one domain from the other. In the specific case of AVHs, few studies have tracked the linguistic anomalies inherent to this central symptom in the diagnosis of SZ. Thus, our study is one of the first approaches to the creation of a specific linguistic profile for AVHs, ratifying and extending previous proposals (de Boer et al., 2016).

8.4 Final thoughts: implications, limitations and future research

Language is a complex system that can deteriorate in different ways and at different levels: language impairment in mental disorders rarely follows the same pattern, even in different clinical groups of the same disease. It is therefore important to design clinical tests that are sufficiently accurate to detect and assess the different patterns of linguistic disintegration. These linguistic-based clinical tests can also serve as part of the neuropsychological batteries. Indeed, recent studies confirmed that automated speech graph analysis in psychotic disorders such as bipolar and SZ serves to quantify discursive disorganisation (Palaniyappan et al., 2019), with the intention of detecting specific patterns of dysfunctional thought flow, such as divergence and recurrence (Mota et al., 2012).

Furthermore, as our research showed, standard tests have not revealed the cognitive change present in prodromal stages of HD. Therefore, the in-depth study of language ability makes it possible to locate cognitive anomalies that go unnoticed by traditional neuropsychological tests. In Tovar et al. (2020), the cognitive batteries did not reveal any sort of decline in the premanifest stage, except for the MMSE. Even in manifest HD, the scores on most clinical tests do not show a pressing cognitive impairment. In the future, it would be interesting to investigate to what extent or in what way linguistic decline is related to cognitive changes, or what its origin really is. As a first limitation, our methodology does not allow us to confirm whether we are dealing with a language deficit *per se*.

However, while Study 1 outlined the grammatical abnormalities typical of the pre-symptomatic population, studies 2 and 3 failed to qualify the impact of neurodegeneration at this early stage. In this sense, more precise research is needed in order to detect the subtle cognitive change that precedes motor symptomatology in HD. Longitudinal studies are also very useful in this population, as they allow for a progressive comparison of

cognitive dysfunction, both linguistic and non-linguistic, throughout the different stages of the disease.

Another limitation of this research is the lack of interrelations with neuroimaging studies. Data on neural atrophy were not available in our studies, but we would like to relate patterns of linguistic impairment to patterns of neuronal degeneration in the future. Hinzen et al. (2018) failed to identify neural correlates for the linguistic abnormalities they researched (except in the Quantitative domain), so the relation between language decline and striatal neurodegeneration in HD remains to be discovered.

By constructing specific linguistic paradigms and testing them in neuroimaging studies, it is therefore possible to detect which brain regions play a relevant role in language processing. Given that HD primarily affects the striatal nucleus, research into the neural correlates of HD may shed light on the role of the striatum in language (Jacquemot & Bachoud-Levi, 2021).

On the other hand, although it was possible to use a NTs group in HD studies, the research on SZ lacked neurotypical data. In future work it would be interesting to compare the language patterns extracted in the patient sample with NTs subjects performing the same linguistic tasks, as in De Boer et al. (2016). They conducted a comparison of AVHs reported in patients with a psychotic disorder and in non-psychotic individuals, with the intention of determining the specific linguistic features of the clinical population.

Finally, in future research, it would be useful to replicate our linguistic paradigms in other languages, both Romance and non-Romance. Our studies are mainly on European Spanish, but, as de Freitas (2022) points out, for example, investigating the pronominal system of SZ patients speaking colloquial Portuguese from Brazil is especially interesting, since is a partially pro-drop language, and the use of certain forms is more restricted.

With future projects in mind, we conclude this dissertation with the anecdote about how George Huntington's interest in HD began. In this way, we hope to keep that the same research spirit that makes us constantly ask ourselves, 'what could it mean?':

> 'Over 50 years ago, riding with my father on his professional rounds, I saw my first case of 'that disorder,' which was the way in which the natives always refereed to

the dreaded diseases. It made a most enduring impression upon my boyish mind, an impression every detail of which I recall to-day, an impression which was the very first impulse to my choosing chorea as my virgin contribution to medical lore. We suddenly came up two women, mother and daughter, both tall, thin, almost cadaverous, both bowing, twisting, grimacing. I stared in wonderment, almost in fear. What could it mean? My father passed to speak with them and we passed on. Then my Gamaliel-like instruction began; my medical institution had its inception. From this point on my interest in the disease has never wholly ceased.' (Lecture to NY Neurological Society, 1909)

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