The healthcare resource utilisation associated with managing Fragile X syndrome

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Background

- Fragile X Syndrome (FXS) is a genetic condition that is associated with cognitive impairment, a distinct physical phenotype and intellectual disability (1,2).
- Current treatments aim to manage co-occurring physical and mental health conditions such as epilepsy, anxiety and Attention Deficit Hyperactivity Disorder (ADHD).
- Psychopharmacological treatment and supportive therapy may be provided to improve quality of life and ease symptoms (3).
- From the UK perspective, there is a lack of data concerning resource use and costs associated with treating FXS. In this study we aim to address this data gap.

Methods

- Patients were selected from the Clinical Practice Research Datalink (CPRD) Aurum dataset linked to Hospital Episode Statistics (HES); a database derived from primary care that contains data for approximately 20% of the England population.
- Patients with FXS were selected by medcodes (Aurum) or ICD-10 codes (HES) (Table 1).
- The study period was 01/04/2007-31/12/2020. To capture an incident population, patients were required to be registered from birth, with a registration date within 3 months of their month of birth or within the same year of birth if month of birth was not available.
- Index date was defined as the date of first diagnosis of FXS.
- Patients with FXS were matched 1:1 to non-FXS control patients on age, gender and concurrent practice registration.
- Primary care contacts and costs, including prescriptions, were extracted from CPRD Aurum datasets. Inpatient, outpatient and accident and emergency (A&E) contacts and costs were extracted from HES.
- All healthcare categories were costed according to the relevant consultation and associated tariff (4-6).
- Healthcare contacts and associated costs (UK 2019/2020 prices) were calculated per person year (PPY) in the 1 year prior to index, 1 year post index and the subsequent follow up period.
- Generalised linear models were constructed to compare the incidence rate ratio (IRR; Poisson) and cost ratio (CR; Gamma) between patients with FXS and controls.
- This study received CPRD Research Data Governance approval (22_001814).

Table 1. The clinical codes used to select patients with Fragile X syndrome.

Description	Clinical code	Type
Fragile X Syndrome	2090010, 893501000006110, 940371000006118	Medcode
Fragile X Chromosome	315486012	Medcode
Cause of learning disability: Fragile X Syndrome	1009571000006115	Medcode
FRAXA – Fragile X Syndrome	2508311000006110	Medcode
Martin-Bell Syndrome	2508291000006111	Medcode
Fragile X Chromosome	Q99.2	ICD-10

Results

Baseline Characteristics

- 259 incident patients with FXS were selected and matched to 259 controls.
- The mean age at index date was 7.3 years, and the median age was 5.0 years.
- The majority of patients were male; 77.6% versus 22.4% female.

Healthcare Resource Use

- Patients with FXS had a significantly higher number of contacts per person year (PPY) in all healthcare sectors for each of the follow-up periods (Figures 1 and 2).
- Outpatients and inpatients were the healthcare sectors with the largest relative difference between patients with FXS and matched controls.

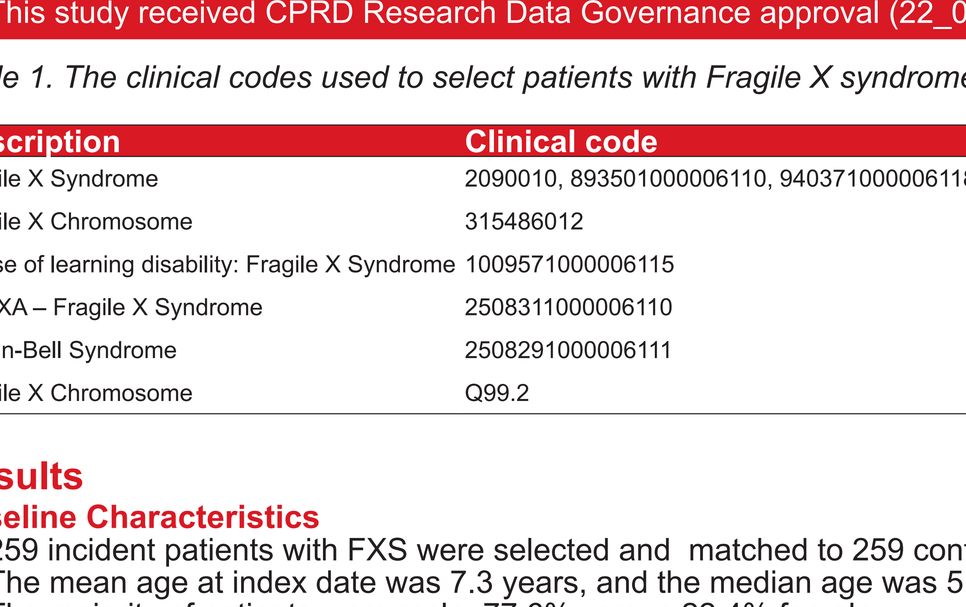

Figure 1. Rate per person year of healthcare contacts for patients with Fragile X syndrome and matched controls from the Clinical Practice Research Datalink and Hospital Episode Statistics 2019.

This study was funded by Shionogi. This study is based in part on data from the Clinical Practice Research Datalink obtained under licence from the UK Medicines and Healthcare products Regulatory Agency. The data is provided by patients and collected by the NHS as part of their care and support. HES data (Copyright © 2023), re-used with the permission of The Health & Social Care Information Centre. All rights reserved.

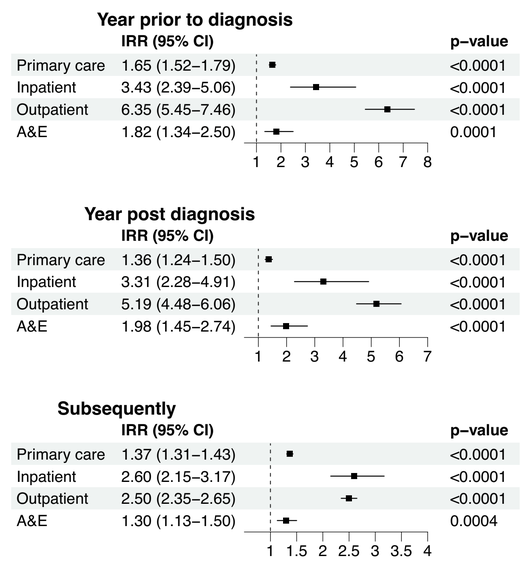

Figure 2. Generalised linear model for healthcare contacts for patients with Fragile X syndrome and matched controls in the year prior to Fragile X diagnosis, year post Fragile X diagnosis and subsequently.

- The cost of healthcare resource use was also higher PPY for cases compared to controls for primary care, inpatients and outpatients in both the 12 months prior and 12 months post index date (Figures 3 and 4).
- In the period beyond one year after index date there was no significant difference in costs except for outpatients and primary care prescription.

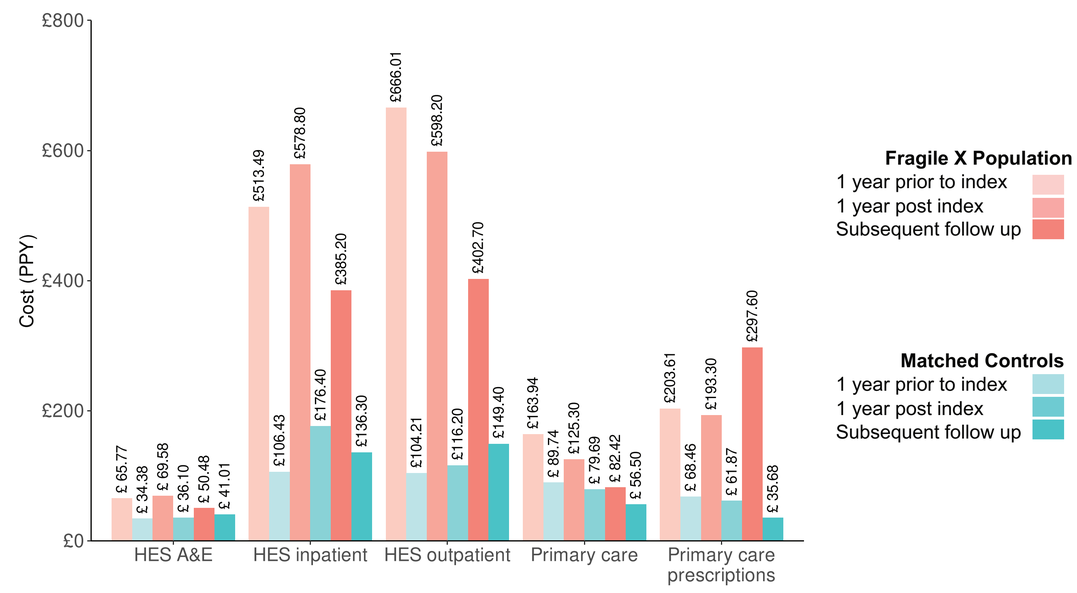

Figure 3. Healthcare costs (per person year) for patients with Fragile X syndrome and matched controls from the Clinical Practice Research Datalink and Hospital Episode Statistics 2019.

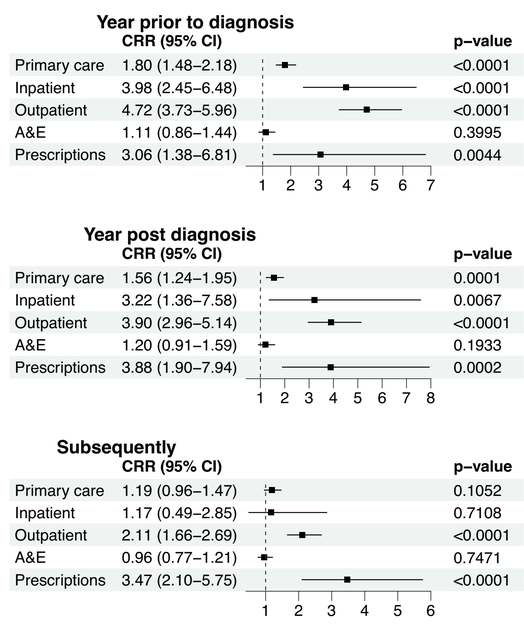

Figure 4. Generalised linear model for healthcare costs for patients with Fragile X syndrome and matched controls in the year prior to Fragile X diagnosis, year post Fragile X diagnosis and subsequently.

Conclusion

- This study demonstrated that patients with FXS had significantly higher healthcare contacts than controls in the primary care, inpatient, outpatient and A&E sectors.
- In both the 12 months before and after diagnosis, this was mirrored in significantly increased costs in all sectors with the exception of A&E.
- Beyond the 12 months after diagnosis, the significant increase in costs was only observed in outpatients and primary care prescriptions.
- However, this study only considered direct medical costs. A large proportion of costs associated with FXS may be indirect costs such as informal care, social care and education and these are likely to remain constant.

References

1) Fragile X Society. About Fragile X syndrome [Internet]. 2021. Available from: https://www.fragilex.org.uk/syndrome, 2) National Fragile X Foundation. Fragile X 101 [Internet]. Available from: https://fragilex.org/understanding-fragile-x/fragile-x-101/, 3) Nazareth T, Li N, Marynchenko M, Zhou Z, Chopra P, Signorovitch J, et al. Burden of illness among patients with Fragile X syndrome (FXS): A Medicaid perspective. Current Medical Research and Opinion. 2016 Mar;32:405-16. 4) Personal Social Services Research Unit. Unit costs of health and social care [Internet]. 2020. Available from: https://www.pssru.ac.uk/project-pages/unit-costs/, 5) NHS Business Services Authority. Prescription cost analysis - England 2019 [Internet]. 2020. Available from: https://www.nhsbsa.nhs.uk/statistical-collections/prescription-cost-analysis-england/prescription-cost-analysis-england-2019, 6) NHS England. National tariff 2019/20: Documents and policies [Internet]. 2020. Available from: https://www.england.nhs.uk/publication/past-national-tariffs-documents-and-policies/