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Withdrawing gluten-free food from prescriptions in England: a mixed-methods study to examine the impact of policy changes on quality of life

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Keywords

coeliac disease, gluten-free diet, health services use, mixed methods, prescriptions, quality of life.

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Introduction

Currently, the only treatment for coeliac disease is a life-long gluten-free diet. Better dietary adherence leads to fewer symptoms ⁽¹⁾ and improved quality of life ⁽²⁾. Challenges with dietary adherence and the availability and cost of gluten-free food remain ^(3–6). The gluten-free diet is burdensome to many people with coeliac disease, even after several years of following it ⁽⁷⁾. Some countries support people with coeliac disease to adhere to the diet

Abstract

Background: Some local areas in England stopped have gluten-free prescriptions for coeliac disease. An explanatory mixed-methods study has investigated the impact of these changes.

Methods: A cross-sectional survey with 1697 participants was followed by 24 qualitative interviews. The survey included questions on the use of prescriptions and healthcare services, as well as the Coeliac Disease Assessment Questionnaire (CDAQ) to assess quality of life. The survey data were analysed by descriptive statistics, analysis of variance and regression analysis, and the interviews were analysed by thematic analysis. Findings from the interviews guided the survey analysis.

Results: Dietary burden was significantly different between prescribing and nonprescribing areas, with little impact on other aspects of quality of life. Survey participants in nonprescribing areas who felt more impacted by the prescription changes reported a lower quality of life. Satisfaction with and use of services was lower in nonprescribing areas. Interviews indicated that, after initial frustrations, most people adapted to the changed prescription policy. However, there was a clear preference for gluten-free prescriptions to be available, in particular for staple foods.

Conclusions: The main quality of life impact was on Dietary burden. It is encouraging that most participants in the present study maintained a good quality of life. However, issues of worse experiences of care, lower follow-up opportunities and inequity arose, and these should be taken into consideration in decisions on gluten-free food prescriptions. The new guidelines for the National Health Service in England have retained prescriptions for bread and flour mixes, which is more limited than the range of staple foods preferred in the present study.

through a variety of means. For example, gluten-free food is subsidised by the government in Italy, is tax deductible in the USA, and was provided on prescription by the English National Health Service (NHS) until 2015 according to National Prescription Guidelines ^(8,9).

As a result of the increasing financial strain on the NHS, providing gluten-free food on prescription has proven controversial in recent years and arguments both in favour of and against these prescriptions have been debated ⁽¹⁰⁾. The NHS aims to provide an equitable service, whilst allowing

Clinical Commissioning Groups (CCGs) to decide which specific services to commission locally⁽¹¹⁾. CCGs are clinically-led statutory NHS bodies that are responsible for planning and commissioning healthcare services for their local area (<https://www.nhs.uk/ccgs>). National Prescribing Guidelines⁽⁹⁾ set out recommendations on prescriptions for gluten-free foods and aim to offer equitable allowances for all UK patients with coeliac disease⁽⁸⁾. From 2015, some CCGs decided to no longer adhere to National Prescribing Guidance for gluten-free food⁽⁸⁾ and stopped or restricted prescriptions (in terms of providing fewer types of food or quantity). This has given rise to a concern that at least some people with coeliac disease may be negatively impacted by these changes. Evidence suggests that a quarter of people who do not receive gluten-free food prescriptions adhere less well to the diet⁽¹²⁾. Ensuring good outcomes and a positive experience of care remain a main focus of health policy in England⁽¹³⁾. The present study was conducted to assess the impact of stopping prescriptions for gluten-free food on adults with coeliac disease by comparing quality of life, finances and access to gluten-free food of people living in local areas where the CCG stopped prescriptions with people living in local areas where the CCG still provided prescriptions. Areas that restricted prescriptions were not included because there was no clarity of what 'restrictions' represented. Restrictions could be either restrictions of quantity or types of food, or a combination of both. This study was conducted prior to NHS England⁽¹³⁾ publishing guidance on the prescription of gluten free food in 2018, which recommends continuing with prescriptions of bread and flour mixes (see Discussion).

Materials and methods

A sequential explanatory mixed-methods study was conducted consisting of a cross-sectional survey (April to May 2017) followed by a qualitative study (August to November 2017). Mixed methods are valuable in health services research⁽¹⁴⁾ because they give insight into overlapping research questions by bringing together strengths from both qualitative and quantitative methodologies. Thus, mixed methods studies allow research questions to be addressed in greater depth than either method could on their own. Adults (≥ 18 years of age) living in England were eligible to participate if they confirmed receiving a diagnosis of coeliac disease by a medical professional. Ethics approval was obtained through the Central University Research Ethics Committee of the University of Oxford (Reference number R45890/RE001).

Survey

Participants were recruited for a postal survey via Coeliac UK, the leading charity for coeliac disease. The sample

size was determined by sensitivity to change data for the Coeliac Disease Assessment Questionnaire (CDAQ). To detect a difference at 90% power and 0.05 level of significance, the minimum sample size was 350 participants per group (i.e. people living in prescribing areas and people living in nonprescribing areas). With an estimated 40% response rate, a minimum of 2000 people needed to be invited into the study to achieve a sample of approximately 800 for the survey. A larger than necessary sample was recruited to allow for a wider geographical spread of local areas to be included.

Coeliac UK sent a postal survey (including a participant information sheet, consent form, questionnaire and prepaid return envelope) to 4050 members. Completed questionnaires were returned to the research team. Eligible participants were invited from 13 local areas (or CCGs) that prescribed gluten-free food according to National Prescribing Guidance ($n = 2131$), as well as from 13 local areas that had stopped prescriptions ($n = 1919$). At the time when the sampling strategy was developed, approximately 33 local areas (out of 209) had stopped prescriptions for adults. The nonprescribing areas were selected first to give a geographical spread of urban and rural communities across England and these areas were matched for geographical location and level of deprivation with an equal number of prescribing areas. Coeliac UK selected members from their database in the selected geographical regions for the study. The aim was to invite 200 members per area; however, membership varied by area (range 10–624 members). In the 18 areas with fewer than 200 members, all members were invited. In the eight areas with more than 200 members, Coeliac UK randomly selected members to invite into the study.

In addition to the postal survey, an online advertisement was placed on social media sites Twitter and Facebook, with links to an electronic version of the survey (e-survey) and the participant information sheet. QUALTRICS, version April 2017 (Qualtrics, Provo, UT, USA) was used for the online survey.

All participants needed to sign a consent form. The consent form was provided and signed in paper form at the beginning of the postal survey (this page of the survey booklet was removed upon receipt of the questionnaire and stored separately) or in electronic format before the e-survey could be taken.

Questionnaire

The questionnaire comprised questions on the use of health services, availability and use of prescriptions (including a question on the impact of prescriptions being discontinued), and the availability and cost of gluten-free food. These questions were based on two previous

questionnaires on use of health services and prescriptions in Coeliac Disease^(15,16). The questionnaire, for use in this study, was reviewed, and where necessary amended, using input from people with coeliac disease and Coeliac UK. Additionally, the survey contained validated patient-reported quality of life measures [the EuroQol 5 Dimension 5 Level (EQ-5D-5L)⁽¹⁷⁾ and the Coeliac Disease Assessment Questionnaire (CDAQ)⁽¹⁸⁾] and a dietary adherence measure Coeliac Disease Adherence Test (CDAT)⁽¹⁹⁾. Data were also collected on demographics and health-related variables (e.g. time since diagnosis, overall impact of coeliac disease, comorbidities). This paper reports findings in relation to the Coeliac Disease Assessment Questionnaire (CDAQ)⁽¹⁸⁾ (EQ-5D-5L and CDAT data will be reported separately). The CDAQ has 32 items within five dimensions (Symptoms; Dietary burden; Social isolation; Stigma; and Worries and concerns). Items are rated on a five-point response scale from 'never' to 'always'. A single summary score (Overall index score) can be calculated from the individual dimensions. All CDAQ scores range from 0 to 100, with a higher score indicating a better quality of life.

Interviews

Survey participants were asked if they were willing to be invited for an interview and 872 consented and provided their contact details. Purposive sampling was employed to invite a diverse sample (in terms of coeliac history, prescription status, CDAQ scores and demographics) for a qualitative interview. The aim was to interview 20–25 participants. Potential participants were sent an invitation letter, consent form and participant information sheet. Anyone interested in participating was asked to return the signed consent form and they were contacted to arrange an interview. The interviews were conducted face-to-face (in the participant's home or at the University of Oxford) or via telephone at the participant's convenience. The interview topic guide, developed by all the researchers, focused on: (i) the impact of coeliac disease and the gluten-free diet; (ii) the use of prescriptions for gluten-free food; and (iii) the impact of changes in prescriptions.

Analysis

Survey

Participants were matched with information of their local area (i.e. local prescribing policy at the time when they completed the survey (March to April 2017) and Index of Multiple Deprivation (IMD) scores (<http://imd-by-postcode.opendatacommunities.org>). Because some participants reported not having prescriptions despite living in a

prescribing area (and a small number had prescriptions despite living in a nonprescribing area), a variable of 'self-reported prescription status' (with three categories: 'had prescriptions', 'did not have prescriptions' and 'had restricted prescriptions') was computed. A 'had restricted prescriptions' category was added as some people reported 'restricted prescriptions' (by which they meant they could have a smaller quantity or range of gluten-free food on prescription) despite living in a prescribing or nonprescribing area.

Descriptive statistics [means (SD)], chi-squared or analysis of variance (ANOVA) were used to investigate differences in demographics and quality of life in prescribing versus nonprescribing areas. Multivariate linear regression analyses were conducted where relevant (i.e. where univariate analyses showed a significant difference) with CDAQ dimensions and Overall index score as the outcome variables; and the local area prescribing rules or respondent self-reported impact of prescription changes on health as main explanatory variables. Additional variables were demographics, health-related variables and self-reported prescription status. Few data were missing for the CDAQ [less than 1% for all but one (i.e. pain was 1.2%) variables]. Dimension scores could not be calculated for 2% or less of the sample and the Overall index score for 5.3%. No data imputation was undertaken. $P < 0.05$ was considered statistically significant. Data were analysed in SPSS, version 25 (IBM Corp., Armonk, NY, USA).

Interviews

Interviews were digitally audio-recorded and transcribed verbatim by a professional transcriber. The transcripts were not returned to participants for verification, although a research assistant (TL) checked all transcripts against the recordings for accuracy. Thematic analysis was undertaken, meaning that transcripts were read and re-read to identify the themes within the data. MP and TL coded three transcripts independently and developed individual coding frameworks. These two frameworks were compared and contrasted in a meeting and merged into one coding framework. A HC coded another three interviews to cross-check the merged coding framework and a final coding framework was agreed on by all of the investigators. This framework was systematically applied by the HC to all transcripts in NVIVO (QSR International, Melbourne, VIC, Australia), a software programme for the analysis of qualitative data.

Integration of findings

Qualitative findings were used to guide the survey analysis where relevant. The main research question was to compare quality of life between prescribing and

nonprescribing areas, although additional analyses were conducted to assess the relationship between quality of life and respondent reported impact of stopping

prescriptions and their 'self-reported prescription status'. For this publication, secondary analysis was conducted on the initial qualitative themes to be able to present the

Table 1 Demographics and health-related variables of survey participants

| Demographics | Local prescriptions | | No local prescriptions | | P |
|--|---------------------|----------|------------------------|----------|--------|
| | Mean | (SD) | Mean | (SD) | |
| Age (years) | 59.4 | (15.8) | 60.9 | (15.9) | 0.06 |
| | n | % | n | % | |
| Gender | | | | | |
| Female | 576 | (71.6) | 663 | (71.5) | 0.99 |
| Male | 229 | (28.4) | 252 | (28.5) | |
| Marital status | | | | | |
| Single | 140 | (17.5) | 127 | (14.4) | 0.15 |
| Married or civil partnership | 524 | (65.3) | 574 | (65.0) | |
| Separated, divorced or legally dissolved civil partnership | 73 | (9.1) | 83 | (9.4) | |
| Widowed or survivor of civil partnership | 65 | (8.1) | 99 | (11.2) | |
| Occupation (multiple responses are possible) | | | | | |
| Employment (full-time, part-time or self-employed) | 359 | (44.6) | 351 | (39.7) | 0.07 |
| Education (full- or part-time) | 19 | (2.4) | 15 | (1.7) | 0.33 |
| Unemployed | 12 | (1.5) | 9 | (1.0) | 0.38 |
| Permanently sick or disabled | 28 | (3.5) | 31 | (3.5) | 0.98 |
| Retired | 358 | (44.5) | 444 | (50.2) | 0.018 |
| Looking after the home | 120 | (14.9) | 134 | (15.2) | 0.89 |
| Other (e.g. volunteering/ being a carer) | 94 | (11.7) | 80 | (9.0) | 0.76 |
| Socioeconomic background | | | | | |
| Higher & intermediate managerial | 228 | (29.9) | 248 | (28.8) | 0.06 |
| Supervisory or clerical | 268 | (35.1) | 267 | (31.0) | |
| Skilled manual | 46 | (6.0) | 66 | (7.7) | |
| Semi and unskilled manual | 23 | (3.0) | 42 | (4.9) | |
| State pensioners or widows | 165 | (21.6) | 212 | (24.6) | |
| Casual or minimum wage earners | 33 | (4.3) | 27 | (3.1) | |
| Index of Multiple Deprivation quintiles | | | | | |
| 1 – most deprived | 98 | (11.4) | 67 | (7.7) | <0.001 |
| 2 | 140 | (18.0) | 205 | (23.4) | |
| 3 | 161 | (20.7) | 241 | (27.5) | |
| 4 | 184 | (23.6) | 199 | (22.7) | |
| 5 – least deprived | 205 | (26.3) | 163 | (18.6) | |
| Health-related variables | Mean | (SD) | Mean | (SD) | |
| Time since diagnosis of coeliac disease (years) | 13.9 | 13.0 | 13.9 | (12.4) | 0.95 |
| Comorbidities (n) | 1.93 | (1.55) | 1.85 | (1.58) | 0.31 |
| | n | % | n | % | |
| Overall impact of coeliac disease | | | | | |
| No impact | 148 | (18.4) | 158 | (18.1) | 0.79 |
| Mild impact | 297 | (37.0) | 325 | (37.1) | |
| Moderate impact | 250 | (31.1) | 286 | (32.7) | |
| Severe impact | 84 | (10.5) | 77 | (8.8) | |
| Very severe impact | 24 | (3.0) | 29 | (3.3) | |
| Impact of prescription changes on health* | | | | | |
| No impact | NA | NA | 282 | (53.9) | NA |
| Mild impact | NA | NA | 102 | (19.9) | |
| Moderate impact | NA | NA | 87 | (16.9) | |
| Severe impact | NA | NA | 28 | (5.3) | |
| Very severe impact | NA | NA | 21 | (3.9) | |

NA, not applicable.

*532 participants whose prescriptions stopped < 2 years ago.

qualitative findings under the same dimensions as those of the CDAQ.

Results

Participants

In total, 1653 postal survey responses (40.8%) were received, with 1615 (39.9%) valid for inclusion. The e-survey received 234 responses, with 82 valid for inclusion. Reasons for exclusion were being below the age limit; not living in England; not having a medically confirmed diagnosis of coeliac disease; living in an area that restricted prescriptions; or if the majority (or all) of the answers were missing. The 1697 respondents included in the analysis all confirmed receiving a diagnosis of coeliac disease by a medical professional. Survey participants' demographics and disease-related characteristics are presented in Table 1. In the nonprescribing areas, there was a significantly higher proportion of retired people ($P = 0.018$) and a higher likelihood of living in a more deprived area ($P < 0.001$). Of 54 people (31 women and 23 men) invited for an interview, 24 (13 women and 11 men) participated from nine local areas, three of which prescribed and six of which did not prescribe gluten-free food (Table 2; see also Supporting information, Table S1).

Prescriptions

Eight-hundred and nine (47.7%) survey participants lived in a prescribing area and 888 (52.3%) lived in a nonprescribing area. In terms of 'self-reported prescription status', 247 (33.5%) participants living in prescribing areas reported not using prescriptions and 178 (24.2%) reported access to restricted prescriptions. In areas that stopped prescriptions, 14 (1.6%) reported still having prescriptions and 32 (3.8%) reported restricted prescriptions. A mean (SD, range) of 9.4 (5.7, 0–20) months had passed since prescriptions had stopped in nonprescribing areas. Of the 532 participants in nonprescribing areas whose prescriptions had stopped in the 2 years prior to the survey, half reported no impact of prescriptions stopping ($n = 287$; 53.9%), 106 (19.9%) reported mild impact, 90 (16.9%) reported moderate impact and 49 (9.2%) reported severe or very severe impact.

Of the 970 (57.2%) participants who had used NHS services (such as for consultations, prescriptions or tests) for coeliac disease in the 12 months prior to the survey, significantly more respondents from nonprescribing areas rated the NHS as 'fair' or 'poor/very poor' ($n = 187$; 19.3%) compared to those from prescribing areas ($n = 156$; 16.1%) ($P = 0.016$). Participants from nonprescribing areas reported a significantly lower mean (SD) number of GP consultations [$n = 0.58$ (1.46)] compared

to those from prescribing areas [$n = 0.77$ (2.10)], as well as dietitian consultations [$n = 0.27$ (0.66) versus $n = 0.34$ (0.77)] ($P = 0.03$).

Table 2 Summary demographics of the 24 qualitative interview participants (full demographic details are provided in the Supporting information, Table S1)

| Variable | <i>n</i> * |
|--|-------------------------------|
| Gender | |
| Male | 11 |
| Female | 13 |
| Age (years) | |
| 18–29 | 3 |
| 30–39 | 0 |
| 40–49 | 3 |
| 50–59 | 3 |
| 60–69 | 6 |
| 70+ | 5 |
| Marital status | |
| Single | 11 |
| Married or civil partnership | 9 |
| Separated, divorced or legally dissolved civil partnership | 1 |
| Widowed or survivor of civil partnership | 3 |
| Occupation (multiple responses are possible) | |
| Employment (full-time, part-time or self-employed) | 11 |
| Education (full- or part-time) | 2 |
| Unemployed | 1 |
| Permanently sick or disabled | 3 |
| Retired | 10 |
| Other (e.g. volunteering/being a carer) | 1 |
| CDAQ Overall index score (range 0–100, with higher scores indicating a better quality of life) | |
| <20 | 1 |
| 20–39 | 8 |
| 40–59 | 3 |
| 60–79 | 8 |
| >80 | 2 |
| Index of Multiple Deprivation quintiles | |
| 1 – most deprived | 3 |
| 2 | 9 |
| 3 | 6 |
| 4 | 2 |
| 5 – least deprived | 2 |
| Time since diagnosis (years) | |
| Up to one year | 6 |
| 2–5 | 3 |
| 6–10 | 2 |
| 10–20 | 9 |
| 20 or more | 2 |
| Local prescriptions | |
| Yes | 6 (2 not using prescriptions) |
| No | 18 |

CDAQ, Coeliac Disease Assessment Questionnaire.

*Totals do not always add up to 24 as a result of missing data.

Interview participants (Table 3) talked about three main aspects of prescriptions: (i) frustrations about the changes in prescriptions; (ii) coping and adapting to the changes; and (iii) suggestions for the future of prescriptions. Participants valued prescriptions and expressed a strong sense of frustration and irritation with the decision to stop them. The frustration was especially strong if there was no local consultation before the changes were introduced. The issue of inequity of the health service was raised because some questioned why their particular health problem or their local area should not get support. A few participants speculated that people from lower income groups may not be able to adhere to the diet as a result of the high cost of gluten-free food. There was little evidence of problems with dietary adherence, but there was evidence of higher dietary burden for more vulnerable participants such as Participant 4 who found the withdrawal of prescriptions particularly difficult as she had additional morbidities, was a wheelchair user and lived on her own in a rural community.

Despite negative views and feelings, the majority of interview participants reported adapting to the changes, continuing to adhere to the diet, and finding ways to cope, even if these could be burdensome. This involved buying more gluten-free foods out-of-pocket, going to multiple shops that stock the preferred gluten-free options, restricting the amount of gluten-free food bought and eaten, and stocking up on food to not run out. Stocking up on gluten-free food meant alternatives needed to be found for food storage, such as buying a larger freezer, or storing foods differently. This adaptation and coping may explain why little impact on quality of life, apart from Dietary burden, was found in the survey (see below).

Most interview participants thought that prescriptions for gluten-free food should be available. This is in conflict with the survey findings indicating that the impact of prescriptions stopping mostly had no or a mild impact (see above). It is likely that participants were biased towards the view that prescriptions for gluten-free foods

Table 3 Interview participants' views on prescriptions for gluten-free food

| Theme | Quote(s) |
|---|---|
| Frustrations in relation to changes in prescriptions policy | '... it's made things a bit more difficult, but I suppose ... now it's been like two months or so, I've got into the ... but at first, because it was such that sudden change, it was like, ooh, there was a bit of a panic at first ...' (Participant 2, 18 year old woman) '... I'm sure there are other diseases and illnesses that people have, and they get the necessary medication for on prescription ... So, why can't we have what we need? It's difficult to ... I guess the government think it's a luxury. I don't think it's a luxury; I think it's a necessity ... I understand the need to cut costs, although I'm not sure that they're cutting in the right direction. I think there's other ways possibly that they could save money ... without penalising a group of people ...' (Participant 14, 61 year old man) '... I don't cost the NHS very much because I don't have anything else done, apart from [coeliac disease] ... I've had things done to my eyes, but I don't cost the NHS very much ... I paid my dues [laughs], always paid my taxes and always paid all the things I'm asked for, and now they're taking this away. No, I was angry; I was angry ...' (Participant 16, 82 year old woman) |
| Adapting and coping | '... I've still been able to stick to the diet but it's like just more of an inconvenience really ...' (Participant 2; 18-year-old woman) '... I'm retired, financially I'm still able to ... you know I would still arguably, I would have to find the money to purchase it [gluten free food] because as far as I'm concerned, my health comes before anything else ...' (Participant 9; 54 year old man) '... since I've been buying all the stuff, it's quite difficult because it's now been, I think eighteen months at least, that I've been doing this, and over that period the availability has improved. But it can be a disaster because I go every week to two or three supermarkets, and you can get there intending to buy certain products ... only to find that they've got no stock, and you're forced then to buy something else which you may not like. Occasionally, you go back the following day or something, and find that the items are in stock. The whole process is totally chaotic really ...' (Participant 21; 85-year-old man) |
| Suggestions for the future of prescriptions | '... I think they ought to have the staples. So, the basic mix and bread and pasta ought to still be available. I've never been ... never been in agreement with having sweet biscuits or cake mixes, but then most GPs certainly, while I've been diagnosed, have never allowed those anyway ...' (Participant 5; 65-year-old man) '... I would value going back to having two or three basics that you need ... I mean if I could have the oats I would be ... it would change my life completely; honestly ...' (Participant 4, 69-year-old woman) '... people on low incomes really do need something. So, if the NHS could find a way of means testing that would be, for me, the best alternative ...' (Participant 6; 59-year-old man) '... [prescriptions] should be an option because there are people that can't afford it, you know and they're affecting their health by eating food with gluten in it. So, I think it should be an option there definitely for people, especially families with young kids ...' (Participant 11; 55-year-old man) |

are important. Interview participants thought prescriptions were particularly important for vulnerable people or large families. They understood that the NHS is facing financial challenges and made suggestions for improving the system, such as only allowing the prescription of staple foods or basics (including bread, flour, flour mixes, pasta and cereal), the introduction of a voucher system, or means testing prescriptions. Essentially, participants suggested that it would be better if prescriptions were restricted rather than stopped completely.

Quality of life

Of the five dimensions of the CDAQ, ANOVA showed that Dietary burden was the only quality of life dimension significantly different for survey participants living in prescribing versus nonprescribing areas ($P = 0.005$) (Table 4). Stigma, Dietary burden and the Overall index score were significantly different for self-reported prescription status (Table 4). Interestingly, those who self-reported restricted prescriptions had the lowest quality of life scores on all dimensions, whereas those self-reporting receiving prescriptions had the highest quality of life scores on all dimensions. It is worth noting that the study included only participants who lived in prescribing or nonprescribing areas and those reporting 'restricted' prescriptions lived predominantly in prescribing areas. ANOVA analyses showed that lower quality of life (on all CDAQ dimensions and the Overall Index Score) was associated with a lower satisfaction with the NHS (all $P < 0.001$) both in prescribing and nonprescribing areas. Participants in nonprescribing areas who reported higher impact of the prescription changes on their health reported significantly lower quality of life (Table 5).

In the interviews, participants described quality of life impacts and challenges of living with coeliac disease, which for some included prescriptions being stopped (see also Supporting information, Table S2). Quality of life issues included stigma [e.g. other people not understanding that the gluten-free diet is the only treatment for coeliac disease (as opposed to a lifestyle choice)]; symptoms (such as experiencing adverse consequences of eating gluten); or the impact on social life (e.g. difficulties eating out). Challenges with the diet were also discussed and included issues such as sourcing gluten-free food (which can be particularly challenging when travelling or eating away from home), avoiding cross-contamination, spending time checking food labels, buying and cooking gluten-free food, and the cost of gluten-free food. Participants struggled to make links between the prescriptions policy and their quality of life, potentially because they adapted to life without prescriptions after

Table 4 ANOVA analysis on quality of life, as assessed by the Coeliac Disease Assessment Questionnaire (CDAQ) (score range 0–100 with higher scores indicating a better quality life) in relation to local area prescription policy and self-reported prescription status

| CDAQ dimensions | Local prescriptions policy | | | | | | Self-reported prescriptions | | | | | | | | | |
|----------------------|----------------------------|-------|---------|------------------|-------|---------|-----------------------------|-------|---------|-----------------------|-------|---------|--------------------------|-------|---------|----------|
| | Prescriptions | | | No prescriptions | | | No prescriptions | | | Current prescriptions | | | Restricted prescriptions | | | |
| | <i>n</i> | Mean | (SD) | <i>n</i> | Mean | (SD) | <i>n</i> | Mean | (SD) | <i>n</i> | Mean | (SD) | <i>n</i> | Mean | (SD) | <i>P</i> |
| Stigma | 790 | 53.85 | (23.76) | 873 | 53.61 | (23.22) | 1029 | 53.42 | (23.41) | 319 | 57.03 | (23.80) | 207 | 50.85 | (22.83) | 0.008 |
| Dietary burden | 791 | 45.99 | (19.96) | 877 | 43.31 | (18.57) | 1032 | 44.37 | (19.08) | 321 | 46.92 | (20.42) | 206 | 41.78 | (18.65) | 0.010 |
| Symptoms | 792 | 67.29 | (21.73) | 871 | 67.47 | (20.67) | 1032 | 67.13 | (20.76) | 319 | 69.50 | (21.16) | 203 | 65.54 | (21.97) | 0.08 |
| Social isolation | 798 | 70.57 | (23.82) | 887 | 69.83 | (22.30) | 1035 | 70.24 | (22.68) | 322 | 72.17 | (23.16) | 207 | 67.97 | (23.63) | 0.12 |
| Worries and concerns | 795 | 53.94 | (23.32) | 875 | 53.57 | (22.62) | 1032 | 53.46 | (22.87) | 321 | 55.18 | (23.78) | 207 | 51.63 | (22.15) | 0.22 |
| Overall index score | 760 | 58.66 | (19.11) | 847 | 57.58 | (17.93) | 1003 | 57.81 | (18.17) | 307 | 60.29 | (19.41) | 195 | 55.86 | (18.48) | 0.024 |

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Table 5 ANOVA analysis on the impact of prescription changes on quality of life, as assessed by the Coeliac Disease Assessment Questionnaire (CDAQ) (score range 0–100 with higher scores indicating a better quality life) for survey participants living in areas that stopped prescriptions

| Impact CDAQ | No impact | | | Mild impact | | | Moderate impact | | | Severe impact | | | Very severe impact | | | P |
|----------------------|-----------|-------|---------|-------------|-------|---------|-----------------|-------|---------|---------------|-------|---------|--------------------|-------|---------|--------|
| | n | Mean | (SD) | n | Mean | (SD) | n | Mean | (SD) | n | Mean | (SD) | n | Mean | (SD) | |
| Stigma | 286 | 58.01 | (21.65) | 104 | 54.24 | (22.00) | 87 | 46.88 | (20.76) | 27 | 41.67 | (24.77) | 20 | 38.75 | (19.75) | <0.001 |
| Dietary burden | 286 | 48.10 | (17.11) | 105 | 40.65 | (15.31) | 89 | 37.18 | (13.79) | 27 | 27.55 | (15.65) | 20 | 25.31 | (16.84) | <0.001 |
| Symptoms | 282 | 73.85 | (16.83) | 103 | 67.33 | (18.51) | 88 | 60.28 | (19.15) | 28 | 53.04 | (26.89) | 21 | 49.29 | (26.09) | <0.001 |
| Social isolation | 283 | 76.64 | (19.95) | 106 | 67.22 | (21.50) | 90 | 62.50 | (22.97) | 27 | 53.70 | (20.87) | 21 | 51.19 | (27.88) | <0.001 |
| Worries and concerns | 284 | 58.33 | (20.74) | 103 | 53.76 | (18.76) | 90 | 46.48 | (21.46) | 27 | 38.27 | (19.41) | 21 | 29.37 | (20.00) | <0.001 |
| Overall index score | 280 | 62.87 | (15.75) | 98 | 56.64 | (15.36) | 85 | 51.06 | (16.10) | 27 | 42.98 | (17.75) | 19 | 39.57 | (19.08) | <0.001 |

The impacts were similar between participants with and without prescriptions.

Regression analysis

Dietary burden, which was the only quality of life dimension significantly related to prescription change in the preliminary ANOVA analysis, was examined further. The regression model was significant ($P < 0.001$, adjusted $r^2 = 0.23$) (see also Supporting information, Table S3). Dietary burden was significantly higher in the following groups: if prescriptions had stopped ($P = 0.007$); by employment status (unemployed ($P = 0.006$), permanently sick/disabled participants ($P = 0.002$), 'other' employment ($P = 0.018$) with employed as reference group); socio-economic group (skilled manual workers' ($P = 0.017$), semi-skilled manual workers ($P = 0.001$) and pensioners/widow(er)s ($P = 0.006$) with higher and intermediate managerial workers as reference category); and a higher number of comorbidities ($P < 0.001$). Dietary burden was significantly lower with increased satisfaction with the NHS ($P < 0.001$); increasing age ($P < 0.001$); and longer time since diagnosis ($P < 0.001$).

For participants living in nonprescribing areas, regression analyses were performed with all CDAQ dimensions and the Overall index score as outcome variables. All models were significant ($P < 0.001$) and showed that higher perceived impact of stopping prescriptions was significantly related to lower quality of life (Table 6; for details, see also Supporting information, Tables S4–S9).

Discussion

This sequential explanatory mixed-methods study aimed to investigate the impact of policy changes in prescriptions for gluten-free food on adults with coeliac disease. The survey showed that Dietary burden was significantly

increased by stopping prescriptions. Quality of life issues described in the interviews support the findings reported in other studies^(3,20). Self-reported prescription status was associated with a wider range of quality of life aspects, with participants reporting restricted prescriptions also reporting a worse quality of life compared to those not reporting restrictions. The number of participants self-reporting restricted prescriptions (either lower quantity of food or more limited types of food) was surprisingly high because local areas that had implemented restrictions were excluded from the study. With self-reported prescription status impacting more strongly on quality of life than actual prescription policies, it is clear that people with coeliac disease need to be informed correctly about their local prescriptions policy. This finding may indicate that some general practitioners make exceptions when following prescription rules set by their local area. The qualitative data supported the findings that the impact on quality of life was predominantly an issue of dietary burden. Although initially frustrated or annoyed by the changes, most participants adapted to the withdrawal of prescriptions because they thought it was essential to adhere to the gluten-free diet.

The survey findings showed that participants living in nonprescribing areas who reported a higher impact from the change in prescription policy also had significantly lower quality of life in all domains compared to participants in nonprescribing areas who reported no or low impact from the policy change. Qualitative findings indicated that more vulnerable people, such as those less able to afford gluten-free food, had or were thought to experience greater impact by the prescription changes. Gluten-free food alternatives can be two to four times the cost of gluten-containing equivalent foods^(21,22). Findings on financial impact and availability of gluten-free food will be reported separately.

Table 6 Significant variables in regression models for Coeliac Disease Assessment Questionnaire (CDAQ) dimensions and Overall index score (with perceived health impact, satisfaction with NHS, demographic and disease-related factors as explanatory variables) for participants living in nonprescribing areas

| Outcome variables CDAQ | Model | | Explanatory variables |
|---------------------------|-------------|----------|--|
| | r^2_{adj} | <i>P</i> | |
| Stigma | 0.20 | <0.001 | Impact on health (<i>P</i> = 0.006) Satisfaction with NHS (<i>P</i> < 0.001) Age (<i>P</i> < 0.001) Socio-demographic 'Semi-skilled/unskilled manual' (<i>P</i> = 0.03) Employment 'Looking after the home' (<i>P</i> = 0.005) Time since diagnosis (<i>P</i> = 0.033) Number of comorbidities (<i>P</i> < 0.001) |
| Dietary burden | 0.26 | <0.001 | Impact on health (<i>P</i> < 0.001) Satisfaction with NHS (<i>P</i> < 0.001) Age (<i>P</i> = -0.005) Socio-demographic 'Semi-skilled/unskilled manual' (<i>P</i> = 0.007) Socio-demographic 'Pensioners/Widow(ers)' (<i>P</i> = 0.021) Time since diagnosis (<i>P</i> < 0.001) Number of comorbidities (<i>P</i> = 0.024) |
| Symptoms | 0.22 | <0.001 | Impact on health (<i>P</i> < 0.001) Satisfaction with NHS (<i>P</i> = 0.001) Age (<i>P</i> = -0.001) Number of comorbidities (<i>P</i> < 0.001) |
| Social isolation | 0.25 | <0.001 | Impact on health (<i>P</i> < 0.001) Satisfaction with NHS (<i>P</i> < 0.001) Employment 'Permanently sick/disabled' (<i>P</i> = 0.001) Employment 'Supervisory or clerical' (<i>P</i> = 0.019) Time since diagnosis (<i>P</i> = 0.001) Number of comorbidities (<i>P</i> < 0.001) |
| Worries and concerns | 0.21 | <0.001 | Impact on health (<i>P</i> = 0.004) Satisfaction with NHS (<i>P</i> < 0.001) Age (<i>P</i> = 0.031) Socio-demographic 'Semi-skilled/unskilled manual' (<i>P</i> = 0.014) Employment 'Permanently sick/disabled' (<i>P</i> = 0.016) Time since diagnosis (<i>P</i> = 0.001) Number of comorbidities (<i>P</i> < 0.001) |
| Overall index score | 0.29 | <0.001 | Impact on health (<i>P</i> < 0.001) Satisfaction with NHS (<i>P</i> < 0.001) Age (<i>P</i> = -0.001) Socio-demographic 'Supervisor/clerical' (<i>P</i> = 0.023) Socio-demographic 'Semi-skilled/unskilled manual' (<i>P</i> = 0.007) Time since diagnosis (<i>P</i> < 0.001) Number of comorbidities (<i>P</i> < 0.001) |

NHS, National Health Service.

Participants rated experiences with the NHS less favourably in areas where prescriptions stopped. Another survey in areas that had restricted or stopped prescriptions found that participants felt less supported in the management of their coeliac disease as a result of changes in prescriptions⁽²³⁾. Ensuring a positive experience of care is part of national English health policy⁽²⁴⁾ and the withdrawal of prescriptions was met with frustrations and feelings of inequity. Despite a stated commitment of the NHS to equity of access, the CCGs ability to commission

services locally to fit local population needs may lead to inequities⁽¹¹⁾. Also, participants in nonprescribing areas consulted the GP and dietitian less often, which means that there are fewer follow-up opportunities for people living in these areas.

New guidelines recommend retaining prescriptions across England but restricting them to bread and flour mixes⁽¹³⁾. The re-introduction of prescriptions in areas that have withdrawn them will negate at least some of the cost savings to the NHS that were made as a result

of their withdrawal⁽²⁵⁾; however, more restricted prescriptions in areas that were still prescribing may lead to cost savings. Interview participants understood that changes may be necessary but wished that 'staple foods' would remain available on prescription. The range of foods labelled as 'staple foods' by interview participants, however, included items such as cereal and pasta. Hence, although the new guidance is positive in terms of prescriptions still being available, the choice of food items on prescription within the new guidance may be too limited. Although this study has focused on the NHS in England, other countries have policies for supporting people with coeliac disease with their gluten-free diet; for example, via tax deductions in Canada, prescriptions in New Zealand or direct food provision in Spain⁽²⁶⁾. These countries may be influenced by the NHS England approach of limiting support; however, the specific outcomes may vary across countries as a result of cultural and socio-economic characteristics, as well as differences in national healthcare systems and policies.

Some limitations need to be acknowledged. The response rate to the postal survey was 40.8%, and it is possible that the quality of life of nonparticipants may be different. The response rate to the e-survey cannot be known. Furthermore, many of the e-survey participants did not continue to fully complete the survey once they had clicked on the link. The postal survey response rate is not unusual for surveys posted by Coeliac UK^(18,27), although it does mean that the results need to be interpreted with caution. The diagnosis of coeliac disease was self-reported; however, all participants were asked to confirm that the diagnosis had been given by a medical professional. Finally, the study mainly included members of Coeliac UK who may have been more aware of prescribing changes, and the way they have coped with the changes may be different to non-members. The e-survey did not achieve a sufficiently high participation rate to explore any differences in members versus non-members.

Conclusions

In conclusion, the present study has found that, in terms of quality of life, it is mostly dietary burden that is impacted by the changes in prescription policy. However, some people who were not able to have prescriptions, reported significantly higher impact on quality of life. Overall, it is encouraging that most participants in the present study maintained a good quality of life, which suggests that they are adhering to the gluten-free diet despite the policy changes on prescriptions. However, issues of worse experiences of care, lower follow-up

opportunities and inequity arose as a result of differences in prescriptions across local areas, and these should be taken into consideration in decisions of whether to prescribe or not prescribe. Also, because this was a cross-sectional survey, the potential longer-term impact on quality of life of not prescribing gluten-free food remains unknown. The clear message from the qualitative interviews was that people with coeliac disease want staple foods to remain available on prescription. The new guidelines for the NHS in England have retained prescriptions for bread and flour mixes, although this is more limited than the range of staple foods suggested by interview participants in the present study.

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Conflict of interests, source of funding and authorship

MP, HC and CJ are developers and copyright holders of the Coeliac Disease Assessment Questionnaire (CDAQ), which is licensed through Oxford University Innovation.

The study was funded by Coeliac UK.

MP, CJ and MV conceived the study and raised the funding. All authors were involved in the development of the new questions used in the survey and the interview topic guide. HC and MP collected and processed the survey data. Survey data entry and collection of the qualitative data were undertaken by a TL under the supervision of MP. MP led on the survey analysis, with the support of MV and CJ. HC led on the qualitative data analysis with the support of MP. MP led on integrating the quantitative and qualitative data, and the drafting of this manuscript, with HC, CJ and MV providing input throughout. All authors have approved the final version of the manuscript submitted for publication.

Transparency declaration

The lead author affirms that this manuscript is an honest, accurate and transparent account of the study being reported. The reporting of this work is compliant with STROBE guidelines (for the survey part of the study). Overall the work is compliant with best practice for reporting mixed methods studies. The lead author affirms that no important aspects of the study have been omitted. There were no discrepancies from the study as planned.

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Supporting information

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

Table S1. Demographics of the interview participants.

Table S2. Interview participants' accounts of quality of life issues.

Table S3. Linear regression for Dietary burden (outcome variable) and local prescription policy, self-reported prescriptions, demographics and health-related variables for all survey participants ($r^2_{\text{adj}} = 0.23$, $P < 0.001$).

Table S4. Linear regression analysis for CDAQ Stigma (outcome variable) and impact of change of prescription policy, demographics and health-related variables for participants living in areas where prescriptions had stopped ($r^2_{\text{adj}} = 0.20$, $P < 0.001$).

Table S5. Linear regression analysis for CDAQ Dietary burden (outcome variable) and impact of change of prescriptions policy, demographics and health-related variables for participants living in areas where prescriptions had stopped ($r^2_{\text{adj}} = 0.26$, $P < 0.001$).

Table S6. Linear regression analysis for CDAQ Symptoms (outcome variable) and impact of changes of prescription policy, demographics and health-related variables for participants living in areas where prescriptions had stopped ($r^2_{\text{adj}} = 0.22$, $P < 0.001$).

Table S7. Linear regression analysis for CDAQ Social isolation (outcome variable) and impact of changes of prescriptions policy, demographics and health-related variables for participants living in areas where prescriptions had stopped ($r^2_{\text{adj}} = 0.25$, $P < 0.001$).

Table S8. Linear regression analysis for CDAQ Worries and concerns (outcome variable) and impact of changes of prescriptions policy changes, demographics and health-related variables for participants living in areas where prescriptions had stopped ($r^2_{\text{adj}} = 0.21$, $P < 0.001$).

Table S9. Linear regression analysis for CDAQ Overall index score (outcome variable) and impact of changes of prescription policy, demographics and health-related variables for participants living in areas where prescriptions had stopped ($r^2_{\text{adj}} = 0.29$, $P < 0.001$).