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ORIGINAL ARTICLE

A general health economics review of the hidden costs involved in discharging coeliac patients from hospital-based specialty clinics to community-based management

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ABSTRACT

Aim: The aim of this work was to highlight the impact and hidden costs incurred by the NHS in supporting this management process.
Background: Coeliac disease (CD) is a common auto-immune condition which affects around 1% of the general population. In 2005 there was a drive by the government to discharge patients with CD from specialist hospital follow up to community-based management to improve cost efficiency.
Methods: A retrospective analysis of 1317 CD patients collected from a local coeliac database created between 2005 and 2016.
Results: During these 12 years, CD patients accounted for 1965 hospital admissions with a total 5716 days spent within the hospital setting. There were 33150 adult and paediatric OPAs attended equating to 25.17 per coeliac patient, or 2.29 per person per year. The cost to the CCG totalled £5,167,396. A total of 527 lower GI procedures were undertaken with findings of microscopic colitis, melanosis coli, inflammatory bowel disease and colon cancer. 420 (29%) of the coeliac cohort were found to have IDA with just 4% (17/420) receiving an intravenous (IV) iron infusion.
Conclusion: It would appear that the government's attempts to reduce the cost of CD care within the NHS was not particularly effective, from a financial, or patient care perspective. A hospital-based, specialist nurse led, virtual management system (with consultant over-view) may prove to be a more efficient compromise, to help reduce down waiting times and costs, whilst still providing coeliac patients with the specialist and holistic input they require and deserve.

Keywords: Adult, Coeliac disease, Patient discharge, Government.

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Introduction

Coeliac disease (CD) is a common auto-immune condition which affects around 1% of the general population and is associated with the development of other associated autoimmune conditions and malabsorption complications (1, 2). The classical presentation include symptoms of diarrhoea, steatorrhoea, weight loss or failure to thrive (3). It has been noted however that the rate of classical CD has decreased and more and more people with asymptomatic or non-classical disease has increased (4, 5).

Current BSG guidelines advise that a combination of serology and duodenal biopsies whilst on a gluten containing diet is required for diagnosis (6). There are subtle macroscopic features that can be seen at endoscopy, including features of villous atrophy, scalloping, a reduction of duodenal folds and nodularity. Definitive histology shows evidence of intra-epithelial lymphocytosis and villous atrophy. The histological presentation can be patchy, hence D1 and D2 biopsies are recommended (7). Milder cases can be confirmed where intraepithelial lymphocytes (IELs) and crypt hyperplasia is found in conjunction with positive coeliac serology. Seronegative CD is

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considered to be rare between 1-2% (8, 9). There are many histological mimickers of CD and these include other GI diseases like Crohn's disease and microscopic colitis, a number autoimmune diseases (e.g. Graves, SLE, T1DM) neoplasia and infections (H pylori, TB, HIV, post infectious diarrhea). Therefore in difficult cases genetic HLA associations with DQ2 and DQ8 can be used which has a significant negative predictive value. Approximately 30%–40% of Western populations carry HLA DQ2 or DQ8, while only 1% is affected by CD (10, 11). Conversely, the presence of HLA DQ2/DQ8 has been found to occur in 98.4% of patients with CD (12) thus, their absence makes a diagnosis unlikely.

Although CD is generally considered a relatively benign disease, patients can develop a number of complications. These include anaemia, reduced bone mineral density, liver disease, hyposplenism (leading to impaired immunity to encapsulated bacteria), lymphoma and an impact on mental health causing depression (6). Iron deficiency anaemia (IDA) is a common and manageable complication for patients with CD. 3-5% of patients with unexplained IDA are found to have CD (13). The current guidance for coeliac patients with IDA promotes the use of oral iron as first line treatment with consideration of intravenous (IV) iron replacement therapy (IRT) in coeliac patients at an 'early stage if oral IRT is judged to be ineffective or correction is particularly urgent' (14). The risk of developing other auto-immune conditions (e.g. diabetes, thyroid disorders, and adrenal problems) increases to 23.6% after 10 years of poorly controlled coeliac (15).

The mainstay of treatment is to encourage compliance with a gluten free diet (GFD), to avoid the precipitants that drive much of the allergic and auto-immune reaction (16), and to treat any complications that arise.

In 2005, due to the economic constraints placed on the NHS, there was a drive to discharge coeliac patients from hospital based specialist management to community-based primary care. It was perceived that the standard monitoring and management of these patients was relatively simple and that the outpatient waiting times and the cost of follow up for these patients would dramatically reduce by altering their care pathways. In 2006 the Primary Care Society for Gastroenterology released its guidelines to help educate GPs on how to manage these patients within the community, with recommendations on annual assessments, immunity screening and disease prevention (17). Areas of London had algorithms to help drive this process (18). However studies which assessed the outcomes of these alternative follow up strategies found a significant number of patients were either lost to follow up or their care needs weren't addressed appropriately. This raised concerns as to whether this would eventually lead to an increase in late complications (19).

In September 2015, new guidance by NICE & the BSG led to a reversal of practice. The updated guidelines stated "A healthcare professional with a specialist knowledge of CD" (20) should help manage the patient, listing a range of annual assessments, thus driving care back to the hospital setting.

The aim of this work was to highlight the impact and hidden costs incurred by the NHS in supporting the management of these coeliac patients during this 12 year period when they had been discharged from specialist hospital follow up to community-based management. The demand on inpatient admissions, endoscopy, outpatient burden and anaemia support were all reviewed within our local district general hospital.

Methods

The Luton & Dunstable University Hospital serves a catchment area of 330,000 people. A local coeliac database was created containing the records of 1317 patients with CD between 2005 and 2016. This was achieved by combining those patients deemed to have coeliac disease on the databases acquired from our local dietetic coeliac clinic reviews, the endoscopy reporting tool (HICSS Informatics – Hospital Integrated Clinical Support System), the local histology database (using the search terms "coeliac" and "gluten enteropathy") and the hospital coding system (for inpatient and outpatient, reviews, admissions and procedures). We combined those patients deemed to have coeliac disease within a single database and then validated the diagnoses by confirming a definitive histological diagnosis (on our hospital's histology reporting system on ICE – Sunquest's "Integrated Clinical Environment"), in combination with positive

coeliac serology results (also on our hospital's ICE clinical results system). Patients were removed if a definitive diagnosis was not confirmed.

Using this database for reference, the hospital coding system was then used to perform a 12 year retrospective study looking at the total number of inpatient and day-case admissions of coeliac patients between 2005 and 2016, within different admitting specialties. The coding system was also used to analyse the total number of outpatient appointments (OPAs) with total costs calculated using a range of different prices for different specialties and depending on whether the OPAs were new (£210-£265) or follow up (£83-£136) clinic appointments. New and follow-up clinic appointment prices were calculated using the local tariff provided by the regional CCG (Clinical Commissioning Group) at the time the study was performed. These were the prices paid by the local CCG to our District General Foundation Trust for the gastroenterology outpatient appointment service provided. From the 1st of July 2022, the UK introduced 42 new ICSs (Integrated Care Systems) to replace the old CCGs as new statutory bodies to co-ordinate local NHS commissioning, spending and workforce planning. The number, type and reason for endoscopic procedures were also reviewed and cross correlated in a similar method.

A 7 year retrospective analysis of iron deficiency anaemia (IDA) (defined as a haemoglobin of <12g/L for women and <13g/L for men with a ferritin of <50ng/ml) in this coeliac cohort was also performed using the database and coding records.

Absolute numbers and simple percentages were used, in the hope that this would make the data eminently transparent and allow simplification for future studies, should other units ever want to replicate this and make direct comparisons. The data was analysed using Microsoft Excel.

Results

We identified a coeliac patient cohort of 1317. Of these 467 patients were male and 850 were female. The

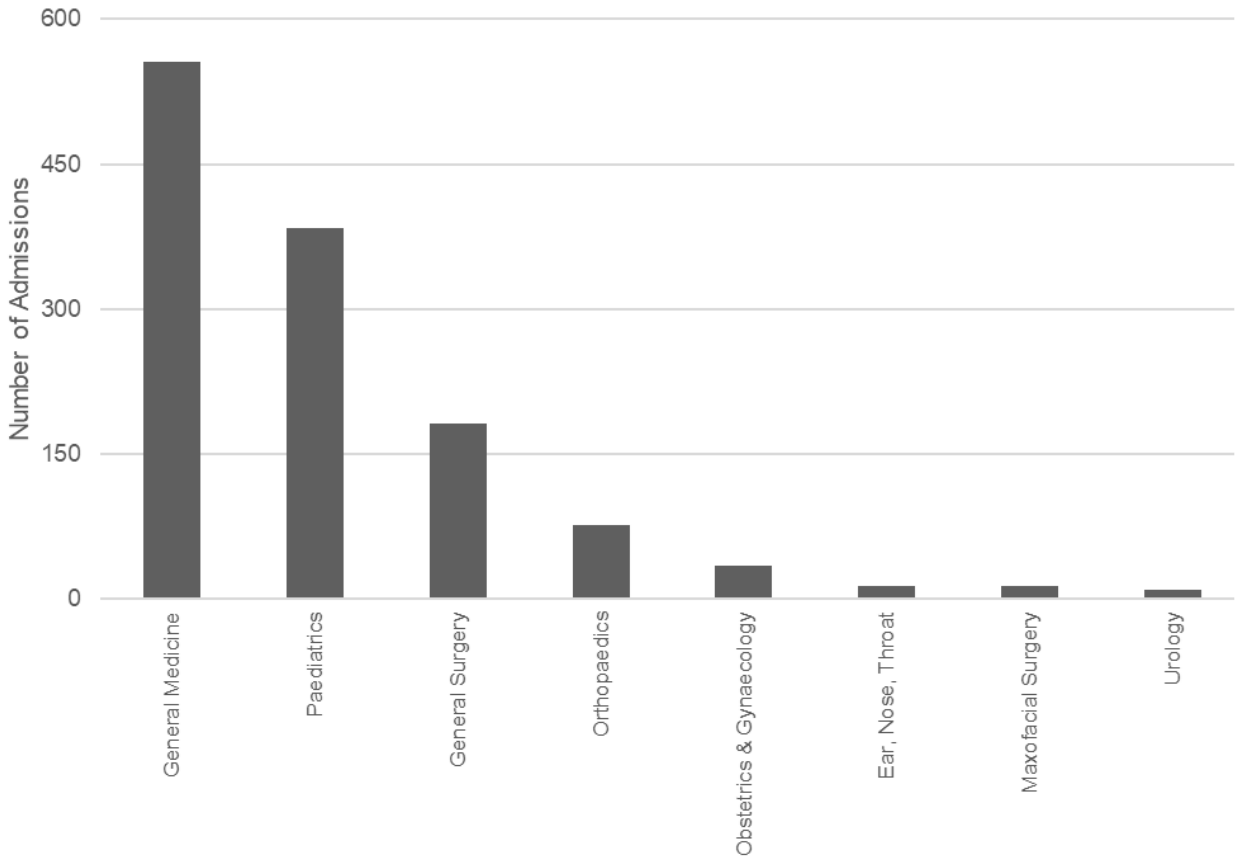

Figure 1. A graph to show the number of inpatient admissions for CD patients according to admitting specialty

age range was 1-102 years old with a mean age of 58. The paediatric population (age ≤ 16) accounted for 61 of these patients.

Hospital admissions

Patients with CD accounted for 1965 hospital admissions over 12 years (163.75 admissions/year), accounting for a total 5716 days spent within the hospital setting. Inpatient admissions accounted for 1266 of these whilst 698 day-case admissions accounted for the rest.

Out of 1266 inpatient admissions, the number of admissions per patient (all ages) ranged from 0-51 with an average of 1.49 per person. The length of stay ranged from 1-64 days, with a total 5018 days spent within hospital, at an average 3.96 days per inpatient stay. 30% of all inpatient stays were paediatric admissions. Figure 1 shows the number of patients admitted under each individual specialty.

Of the 698 day cases seen within this cohort, three specialties accounted for 43.4% of all the admissions. This included dermatology with 124 admissions, obstetrics and gynaecology who saw 91 admissions (with documentation of 24 miscarriages and 2 ectopic pregnancies), and ophthalmology who dealt with 88 admissions (of which, 74 were cataract operations, independent of age).

Outpatient appointments

33150 adult and paediatric OPAs were attended by patients with coeliac disease over the 12 year period. This equated to 25.17 per coeliac patient, or 2.29 per person per year. This cost the local CCG a total of £5,167,396, or £430,616 per year or £327 per coeliac patient per year.

Despite having been discharged from specialist outpatient review, 3235 of all OPAs were back with the gastroenterology team (836 new, 2399 follow up) accounting for 10.25% of total appointments and costing £462,858. There were also 1468 dietetic appointments (473 new, 995 follow up) costing £222,024, 949 dermatology appointments costing a total of £149,848, 1451 ophthalmology, and 906 audiology OPAs. This equates to a total of £3923.61 per person over the study time period.

Endoscopic procedures

During the 12 year review period, 1317 patients underwent a total of 527 lower GI procedures; 413 colonoscopies, 108 flexible sigmoidoscopies and 6 pouchoscopies (Table 1). These were performed for a number of primary indications including: 252 for change in bowel habit (CIBH) reflecting diarrhoea/loose stools; 53 for rectal bleeding; 54 for concomitant inflammatory bowel disease (IBD) assessment and 48 for anaemia. The breakdown of each individual indication is shown in Table 2.

Table 1. Breakdown of the total number of lower GI endoscopies performed in patients with coeliac according to procedure.

Endoscopic procedure	Number of procedures performed
Pouchoscopy	6
Flexible Sigmoidoscopy	108
Colonoscopy	413
Total	527

Table 2. The total number of lower gastrointestinal endoscopies performed for each indication.

Indication	Number of lower GI endoscopic procedures
CIBH	252
Rectal Bleeding	53
BCSP	52
Concomitant IBD assessment	54
Anaemia	48
Abnormal radiology	21
Post Cancer resection	15
Abdominal Pain	9
Other	23
Total	527

Microscopic colitis (MSC) was found in 4 patients presenting with CIBH (4/252, 1.6%). Number of new lower GI malignancies n=20 (indication for CIBH n=1). Twenty six new cases of IBD were identified (indication for CIBH n=16). Melanosis coli was confirmed histologically in 101 (indication for CIBH n=97, rectal bleeding n=3, anaemia n=1) (Figure 2).

Iron deficiency anaemia

A total 54977 blood tests were taken from 34395 patients over a 7 year period 2009-2016 reaching the criteria to confirm an iron deficiency anaemia. This equates to 10.4% of patients of the L&D catchment area. 46169 and 8807 were from females and males, respectively. The median haemoglobin was 121g/L (121g/L in women, 119g/L in men) with a median ferritin of 17.9ng/ml (17.5ng/ml women, 20ng/ml men). 20581 (37.4%) were repeated tests. 47103

(85.7%) tests were requested by GPs, 385 (0.7%) by community services, 6848 (12.4%) by hospital OP, 639 (1.2%) by laboratory and 1 private request.

Coeliac patients accounted for 1.7% (967/54977) of

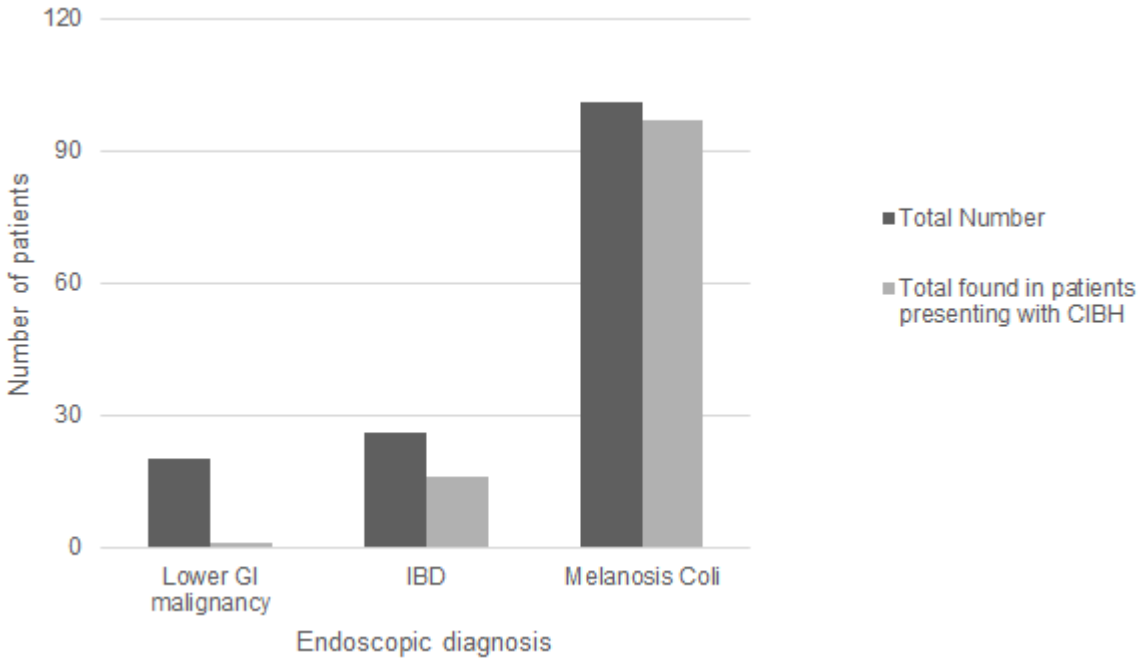

Figure 2. A graph to show the number of endoscopic diagnoses of lower GI malignancy, IBD, and melanosis coli and the proportion of those who presented with a CIBH.

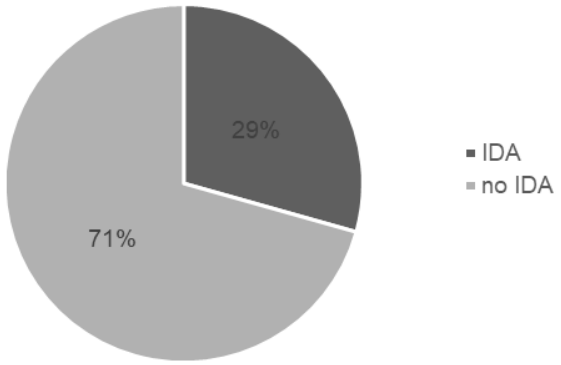

Figure 3. A graph to show the proportion of the coeliac cohort diagnosed with IDA

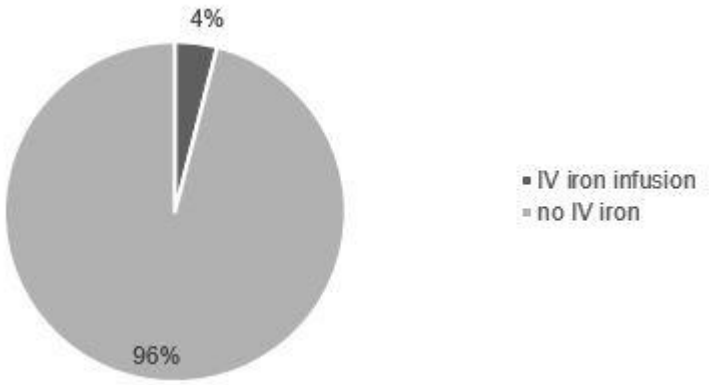

Figure 4. A graph to show the proportion of the coeliac cohort with IDA who received IV iron

all blood tests with IDA. 420 (29%) of the coeliac cohort were found to have IDA (Figure 3). There was an average of 2.5 blood tests per patient with evidence of IDA. Despite tiredness and lethargy being amongst the most commonly reported symptoms impacting the daily lives of CD patients, just 4% (17/420) went on to receive an intravenous (IV) iron infusion (Figure 4).

Discussion

It is clear from our results that there was evidence of an ongoing financial cost for the Luton and Dunstable hospital to care for our coeliac patient cohort during this 12 year period. These results add further weight to the previous studies that have suggested that coeliac disease is not optimally managed within the community.

Whilst the emphasis was on discharging patients to primary care between the years of 2005 to 2016 our results demonstrate a significant number of OPAs were attended during this time totaling 33150 adult and paediatric OPAs costing an estimated total of £4.7millon. Over 15% of these OPAs were unsurprisingly gastroenterology and dietetic related however the majority were follow up appointments (1399 gastroenterology, 995 dietetics) suggesting there was an ongoing need for continued specialist tertiary input for these patients. The results also suggest that a wide range of specialist services were required for our CD cohort including dermatology and ophthalmology. Interestingly these specialties only account for 24% of the total number of OPAs made. Whilst this could be a result of coding error it may also suggest that what was perceived to be a relatively benign and easily managed cohort of patients, may actually be a more complex group than previously appreciated.

There was also a surprisingly high number of audiology OPAs suggesting a possible greater association between ontological extra intestinal manifestations of CD than previously thought (21). Another interesting finding was the significant proportion of cataract operations performed in this cohort. Previous studies have suggested a causal relationship between cataracts and CD (22) with the suggestion that nutritional deficiencies predispose these patients to ophthalmic complications, however more research is required to further define this relationship. 24 miscarriages and 2 ectopic pregnancies were also documented suggesting that good coeliac control may play a more important role in fertility and fecundity than previously thought (23, 24).

On review of our anaemia data, our results showed IDA was found in 29% of our coeliac cohort. An average of 2.5 blood tests were taken per patient confirming IDA, however only 4% of patients received IV iron replacement. These results may suggest poor compliance of a GFD, undertreatment of IDA and ultimately a potential failure of monitoring and managing coeliac patients with IDA in the community.

Altered bowel habit is a commonly reported symptom in coeliac patients. Out of a total of 527 lower GI endoscopic procedures, 48% were performed for CIBH. Whilst microscopic colitis is a key differential to exclude (25), it was only found in 1.5% of these patients suggesting a weak association. Melanosis coli on the other hand was histologically identified in 38.5% of procedures performed for CIBH. We know constipation is a relatively common presenting symptom in CD2 and melanosis coli is often attributed to use of laxatives (26). A direct pathological link between CD and melanosis coli cannot be excluded. Whilst a focus on constipation and use of laxatives is important, the role of calprotectin and endoscopy still remain crucial, given the number of new cases of colorectal cancer (20, 4%) and IBD (26, 5%) identified in our CD cohort. This shows the importance of an overall understanding of bowel related conditions in order to identify those requiring further evaluation.

Following a decade of the community trial, in September 2015, NICE & BSG released new guidelines to help support the management of Coeliac patients quoting “A healthcare professional with a specialist knowledge of CD” (20) should help manage the patient and listed a range of annual assessments. Whilst this does not specify that patients cannot be managed in primary care, the likelihood of finding a coeliac specialist within the community care system is low.

It goes without saying that we should continue to be aware of the ways in which the government try to save money and streamline services. In March 2017 the Department of Health and Social Care (DHSC) underwent a consultation exercise to review the NHS expenditure on gluten free products. In 2016 1.1 billion prescriptions had been made dispensed in primary care at a cost of £9.2 billion (27). In an attempt to make

savings and "release funds elsewhere in the NHS, DHSC decided to restrict GF prescriptions to certain bread and mixes." Their guidance was published to try and avoid major variations in prescribing practice throughout the UK (28). The long term outcome from these decisions are yet to be fully evaluated.

It would appear that the government's attempts to reduce the cost of CD care in the NHS, between the years of 2005 and 2016 was not particularly effective, from a financial or patient care perspective. A hospital-based, specialist nurse led, virtual management system (with consultant over-view) for coeliac patients may prove to be a more efficient compromise to help reduce down waiting times and costs, whilst still providing coeliac patients with the specialist and holistic input they require and deserve.

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Conflict of interests

The authors declare no conflict of interests.

References

1. Mustalahti K, Catassi C, Reunanen A, Fabiani E, Heier M, McMillan S, et al. The prevalence of celiac disease in Europe: results of a centralized, international mass screening project. Ann Med 2010;42:587-595.

2. Johnson MW, Ellis HJ, Asante MA, Ciclitira PJ. Celiac disease in the elderly. Nat Clin Pract Gastroenterol Hepatol 2008;5:697-706.

3. Ludvigsson JF, Leffler DA, Bai JC, Biagi F, Fasano A, Green P et al. The Oslo definitions for coeliac disease and related terms. GUT 2013;62:43-52.

4. Rampertab SD, Pooran N, Brar P, Singh P, Green P. Trends in the presentation of coeliac disease. Am J Med 2006;119:355.

5. Bottaro G, Cataldo F, Rotolo N, Spina M, Corazza GR. The clinical pattern of subclinical/silent celiac disease: an analysis on 1026 consecutive cases. Am J Gastroenterol 1999;94:691–696.

6. Ludvigsson JF, Bai J, Biagi F, Card T, Ciacci C, Ciclitira P et al. Diagnosis and management of adult coeliac disease: guidelines from the British Society of Gastroenterology. GUT 2014;0:1-20.

7. Hopper AD, Cross SS, Sanders DS. Patchy villous atrophy in adult patients with suspected gluten-sensitive enteropathy: is a multiple duodenal biopsy strategy appropriate? Endoscopy 2008;40:219-224 .

8. Schiepatti A, Sanders DS, Biagi F. Seronegative coeliac disease: clearing the diagnostic dilemma. Curr Opin Gastroenterol 2018;34:154-158.

9. Volta U, Caio G, Boschetti E, Giancola F, Rhoden K, Ruggeri E et al. Seronegative celiac disease: Shedding light on an obscure clinical entity. Dig Liver Dis 2016;48:1018–1022.

10. Husby S, Koletzko S, Korponay-Szabo R, Mearin ML, Phillips A, Shamir R et al. European society for pediatric gastroenterology, hepatology, and nutrition guidelines for the diagnosis of coeliac disease. J Pediatr Gastroenterol Nutr 2012;54:136–160.

11. Ciclitira PJ, Johnson MW, Dewar DH, Ellis HJ et al. The pathogenesis of coeliac disease. Mol Aspects Med 2005;26:421-458.

12. Cecilio LA, Bonatto MW. The prevalence of HLA DQ2 and DQ8 in patients with celiac disease, in family and in general population. Braz Arch Dig Surg 2015;28:183–185.

13. Mahadev S, Laszkowska M, Sundstrom J, Bjorkholm M, Lebwohl B, Green P et al. Prevalence of celiac disease in patients with iron deficiency anemia - a systematic review with meta-analysis. Gastroenterology 2018;155:374–382.

14. Snook J, Bhala N, Beales I, Cannings D, Kightley C, Logan P et al. British society of gastroenterology guidelines for the management of iron deficiency anaemia in adults. GUT 2021;0:1–22.

15. Ventura A, Maguzzu G, Greco L. Duration of exposure to gluten and risk for autoimmune disorders with celiac disease. SIGEP study group for autimmune disorders in celiac disease. Gastroenterology 1999;117:297-303.

16. Leffler DA, Edwards-George J, Dennis M, Schuppan D, Cook F, Franko DL et al. Factors that influence adherence to a gluten-free diet in adults with celiac disease. Dig Dis Sci 2008;53:1573-1581.

17. Coeliac UK document: Primary Care Society for Gastroenterology (2006). Available from: https://www.coeliac.org.uk/document-library/1464-primary-care-society-for-gastroenterology-2006.

18. Palmer R. City and Hackney CCG Pathway for Coeliac Disease in Adults. Available from: https://gp-website-cdn-prod.s3.amazonaws.com/pathway-downloads/1518527625-bfe9ad178137ab437060fd0f739563bc.pdf.

19. Pritchard L, Waters C, Murray I, Bebb J, Lewis S. Comparing alternative follow-up strategies for patients

with stable coeliac disease. Frontline Gastroenterol 2019;11:93-97.

20. NICE. Coeliac disease: recognition, assessment and management; NICE guidelines. Published Sept 2015. Avaiable from: https://www.nice.org.uk/guidance/ng20/chapter/recommendations.

21. Volta U, Ferri G, De Giorgio R, Fabbri A, Parisi C, Sciajno L et al. Sensorineural hearing loss and celiac disease. A coincidental finding. Can J Gastroenterol 2009;23:531–535.

22. Mollazadegan K, Kugelberg M, Lindblad BE, Ludvigsson J. Increased risk of cataract among 28000 patients with celiac disease. Am J Epidemiol 2011;174:195-202.

23. Castano M, Gomez-Gordo R, Cuevas D, Nunez C. Systematic review and meta-analysis of prevalence of coeliac disease in women with infertility. Nutrients 2019;11:1950.

24. Butler M, Kenny L, McCarthy F. Coeliac disease and pregnancy outcomes. Obstet Med 2011;4:95-98.

25. Munch A, Aust D, Bohr J, Bonderup O, Fernandez Banares F, Hjortswang H et al. Microscopic colitis: current status, present and future challenges: statements of the European microscopic colitis group. J Crohns Colitis 2012;6:932-945.

26. Yang N, Ruan M, Jin S. Melanosis coli: A comprehensive review. Gastroenterol Hepatol 2020;43:266-272

27. NHS Digital Prescription Cost Analysis, 2017. Available from: https://digital.nhs.uk/data-and-information/publications/statistical/prescription-cost-analysis/prescription-cost-analysis-england-2017.

28. Prescribing Gluten-Free Foods in Primary Care: Guidance for CCGs. Available from: https://www.england.nhs.uk/wp-content/uploads/2018/11/prescribing-gluten-free-foods-primary-care-guidance-for-ccgs.pdf.