Bibliographic reference	Nordyke (2011): Mass screening for celiac disease from the perspective pf newly diagnosed adolescents and their parents: A mixed method study
Study type and aim	Nested case-referent study
Study quality	Was there a clear statement of the aims of the research? Yes – aim is clear Is a qualitative methodology appropriate? Yes – appropriate methodology for this type of research question Was the research design appropriate to address the aims of the research? Yes – design was appropriate Was the recruitment strategy appropriate to the aims of the research? Yes – all screening participants included Was the data collected in a way that addressed the research issue? Yes – standardised HRQOL Eq5D used

Bibliographic reference	Nordyke (2011): Mass screening for celiac disease from the perspective pf newly diagnosed adolescents and their parents: A mixed method study
	Has the relationship between researcher and participants been adequately considered? Not clear Have ethical issues been taken into consideration? Yes – study approved by ethical board Was the data analysis sufficiently rigorous? Yes – all EQ5D data analysed Is there a clear statement of findings? Yes How valuable is the research? Valuable
Number of patients	N=103 CD and 483 non-CD
location	Sweden
Patient characteristics	**Inclusion criteria:** 10041 children invited and 7567 consented to participate. 6th graders from 5 regions in Sweden when they were 12 years old. 145 had screening detected CD and 61 reported CD prior to screening. 4 refernts per CD child were randomly chosen to match age and gender **Exclusion criteria**: 2 participants with CD were found not to have CD (61 diagnosed prior and 144 screening-detected CD cases) **Mean age at diagnosis**: 13.4 **Mean age at follow-up:** 14.6
Intervention	Mass screening for CD
Investigations	Questionnaire: EQ5D Swedish child-friendly pilot version Baseline questionnaires were filled out before results fed back to participants Questionnaires mailed out to participants one year at follow-up Responses were included for the screening-detected cases and respondents when they answered all 5 dimensions Cases = 103 Referents = 483 VAS thermometer also filled out where fill in health today from worst to best imaginable (0 - 100)

Bibliographic reference	Nordyke (2011): Mass screening for celiac disease from the perspective pf newly diagnosed adolescents and their parents: A mixed method study
	Blood sample and biopsy Serological testing done. Biopsy confirmed CD. No further information on type of serological testing No further information on who was given biopsy (i.e. all seropositive?) or biopsy histological criteria for diagnosis
Length of follow up	1 year
Outcome	Change in EQ5D and VAS scores between cases and referents at baseline and at follow-up
Results	Eq5D and VAS Few participants reported severe symptoms, so collapsed into 'no problems' vs. 'problems'. HRQOL similar between cases and referents both at baseline and at follow-up Only dimension where difference was pain, where fewer cases reported problems than referents: OR = 0.50 (95% CI: 0.27 – 0.97) This only significantly different in boys at follow-up In anxiety dimension both cases and referents had small increase between baseline and follow-up (not significant) No significant change in VAS score between baseline and follow-up in either group
Source of funding	Study was supported by grants from the following: Swedish research council: Swedish research council for environment, agricultural sciences, and spatial planning; Swedish council for working life and social research grant, European union supported project
Comments	

Bibliographic reference	Cederborg (2011): Living with children who have Coeliac disease: a parental persepective
Study type and aim	Qualitative cross-sectional study: uses interpretative phenomenological approach to enhance the understanding of how

Bibliographic reference	Cederborg (2011): Living with children who have Coeliac disease: a parental persepective
	to support family adjustment to a GFD
Study quality	CASP QUALITATIVE TOOL: Was there a clear statement of the aims of the research? Yes - aim to understand impact on family of child with CD Is a qualitative methodology appropriate? Yes - no other method applicable Was the research design appropriate to address the aims of the research question? Yes - structured interview Was the recruitment strategy appropriate to the aims of the research? NO - unclear recruitment. No mention of how participants were found or approached Was the data collected in a way that addressed the research issue? Yes - thematic analyses of key interview themes undertaken Has the relationship between researcher and participants been adequately considered? NO - unclear relationship between researcher and participant, and who analysed data Has ethical issues been taken into consideration? Not applicable Was the data analysis sufficiently rigorous? Yes - key themes thoroughly explored Is there a clear statement of findings? Yes - thematic analyses and supportive quotes supplied in text How valuable is the research? Valuable - limited information available to date on impact on family of having a child with CD.
Number of patients	20 parents of 14 children interviewed
location	Sweden

Bibliographic reference	Cederborg (2011): Living with children who have Coeliac disease: a parental persepective
Patient characteristics	**Inclusion criteria:** families of which children who had a definite diagnosis of CD and had been livin with the disease and a GFD for at least 2 years. Among those that met inclusion criteria consecutively chose 15 families with a child diagnosed with CD. All but one of the representatives consented to being interviewed. Interviewed in 3 groups: First group: parents whose children performed their first small intestine biopsy (SIB) before 2 years of age (7 children, 13 parents) at time of interview children between 3 and 5 years Second group parents whose children were >23 years when went through first SIB (3 parents and 3 children) Third group: parents whose children had performed first SIB before 2 years of age but were older than first group at time of interview (16 years old) Exclusion criteria: None listed **Mean age**: group 1: 4.3 years; group 2: 16.3 years, group 3: 16 years Mean age at diagnosis: NA Mean years since diagnosis: NA
Signs and symptoms	NA
Investigations	Interview: Interview took place in home Recorded all interviews and used semi-structured interview guide that includes open ended questions about how parents experienced their children's disease Depending on parents answers, asked follow-up questions to obtain a deeper understanding of their experiences

Bibliographic reference	Cederborg (2011): Living with children who have Coeliac disease: a parental persepective
	Transcribed verbatim and exhaustively examined for references to similarities and differences Then identified sections of the text that illustrate how parents experience their children's disease before and after diagnosis and how manage to adopt a GFD Then chose among the examples to find those that most obviously captured participants' thoughts and beliefs
Length of follow up	
Outcome	Resolution of symptoms Patient experience Complications of cd Adherence Health related quality of life Impact on carers
Results	Organized results into 2 categories with subthemes: 1) struggle to understand child's disease before the diagnosis; 2) process of transforming to a GFD Struggle to understand disease Mother of a 5 year old boy suspected something was wrong with her son when she tried to give him ordinary food – " when we gave him ordinary food hejust cried…he bawled through meals" 5 year old lost weight dramatically – " she lost more than a kilo so she was really weak. It was terrible" One parent did not suspect. Her child was coincidentally tested with no symptoms – " she never showed any symptoms, she had never been sick"

Bibliographic reference	Cederborg (2011): Living with children who have Coeliac disease: a parental persepective
	Parents described process of gaining understanding among HC professionals before the diagnosis as a ‘struggle’ and concerns not taken seriously Mother 4 year old, 5 months to diagnosis. Staff at well-baby clinic told her not to worry – “ I felt everything was not as it should be. They went against me many months before the diagnosis was made. Now looking back, I regret I did not stand my ground more than I did or go to a private doctor”. Most of parents said they were relieved when they knew what was wrong with their child Mother 4 year old girl – “it was wonderful to get the diagnosis. It was a relief” Getting diagnosis meant parents knew how they could help their child to reduce symptoms Transforming to a GFD Most parents reported rapid normalization process to a GFD. One mother of 2 year of said was confused for about 2 months after diagnosis – “I panicked about everything…the first 2 months were a mess. Parents express appreciation of child’s response to GFD – “as she gets older she is more aware of this” Mother 17 year old who got diagnosis as teen said harder for her child – “it might be different if she got sick as soon as she ate gluten food. Theyn you know you cannot eat this because you will get sick and not feel well afterwards” Parents whose children were diagnosed when young have had opportunity to socialize their children into a GFD. These children usually haven’t experienced taste of gluten food and were not aware of what they are missing. Mother of 5 year old could not stop worrying about what woud happen if her daughter tasted something she should not eat – 2 it is always ther that she could get access to crumbs”

Bibliographic reference	Cederborg (2011): Living with children who have Coeliac disease: a parental persepective
	Most parents reported seldom visited restaurants for reason such as not trusting staff's description of ingredients or lack fo food for child One parent spoke of restricted leaisure activities for her 16 year old son – " he cannot spontaneously be with his oeers, everything has to be checked and questioned if he eats with them, I think he fears his peers will think he is a bother to be with. I think the disease hinders him socially" Parents said travelling could be demanding because of difficulties getting acces to propoer food Visiting houses can be difficult. One parent always called house before to check food and make soue would be GF food available Expressed struggle to get staff at daycare and school to understand their childrens GFD Daycare staff not sufficiently educated Negative attitudes from staff at school's dining hall Parents actively and constantly try to find out as much as possible about the disease and how to meet childs GFD needs. Aprents of a 3 year olf search for knowledge through people who know about the disease, on the internet, and through the CD association Most parents have regular contact with a dietician Parents have concerns for children's future. Mother of 5 year olf worries about how child will cope when living alone Parents put hope into new treatments based on scientific breakthroughs
Source of funding	Swedish society for coeliacs, FORSS and the Swedish research council

Bibliographic reference	Cederborg (2011): Living with children who have Coeliac disease: a parental persepective
Comments	

Bibliographic reference	Rosen (2011): Mass screening for celiac disease from the perspective of newly diagnosed adolescents and their parents: a mixed method study
Study type and aim	Mixed-method using both qualitative and quantitative study designs, which aimed to explore adolescent' and parent' experiences having the adolescent' CD detected through mass screening and their attitudes towards possible future screening
Study quality	Was there a clear statement of the aims of the research? Yes – aim is clear Is a qualitative methodology appropriate? Yes – appropriate methodology for this type of research question Was the research design appropriate to address the aims of the research? Yes – design was appropriate Was the recruitment strategy appropriate to the aims of the research? Yes – all screening participants included Was the data collected in a way that addressed the research issue? Yes – standardised focus groups structured and questionnaires were validated in previous study Has the relationship between researcher and participants been adequately considered? Not clear Have ethical issues been taken into consideration? Yes – study approved by ethical board Was the data analysis sufficiently rigorous? Yes – all data sufficiently rigorously analysed Is there a clear statement of findings? Yes How valuable is the research? Valuable
Number of patients	N=145

Bibliographic reference	Rosen (2011): Mass screening for celiac disease from the perspective of newly diagnosed adolescents and their parents: a mixed method study
location	Sweden
Patient characteristics	**Inclusion criteria:** Same pool of participants described in Nordyke (2013). All 145 screening-detected and biopsy-verified CD cases and their parents were contacted for this study. 31 adolescents and 43 parents participated in focus group discussions, 91 adolescents and 105 parents submitted written narrative, and 114 parents filled in questionnaires **Exclusion criteria**: **Mean age of adolescents:** 14.6 years **Mean age since diagnosis:** 15.9 months
Intervention	N/A
Investigations	Focus group discussion: Families in four of the five study sites invited to participate 14 focus groups held involving 31 adolescents and 43 parents Main reason non-participation was lack of time, but a few adolescents expressed reluctance to discuss their disease – parents of the latter did participate Adolescents and parents attended different groups Flexible topic guide and hypothetical case stories used to stimulate discussions and infomants encouraged to discuss issues most important to them Topic guide focussed around informants reasoning when deciding to take part in a screening, and their attitudes towards CD mass screening All interviews digitally recorded Recordings transcribed and later cross-checked to ensure accuracy Transcribed texts entered into Open code Follow-up questionnaires Short reflective narratives Adolescents and their parents asked to write narratives Encouraged to reflect on their overall experience of CD screening and specifically to elaborate on both how they felt about receiving diagnosis and on their recommendation about possible future CD screening Length = one or two hand written pages

Bibliographic reference	Rosen (2011): Mass screening for celiac disease from the perspective of newly diagnosed adolescents and their parents: a mixed method study
	All narratives entered into Open Code software Questions on future screening Parental questionnaire included 2 questions that were utilised in this study: I) whether a CD screening should be implemented (Y/N) At what age screening should be conducted (open answer)
Length of follow up	1 year post diagnosis
Outcome	Adolescents and parents' reported experiences of process of CD diagnosis and consequences of this
Results	**Immediate Reaction to the diagnosis:** '**like a bolt of lightning' – changed life**: adolescents – 75% parents 70% emphasized that more specific information about the consequences of the screening]and having CD] should be given before the test researhers informed parents over the phone and parents were messengers to their children adolescents described this as awkward because neither they nor their parents knew what it really meant this lack of knowledge fostered anxiety among both parents and adolescents "[when receiving the results] I wasn't totally sure either, but I had a little hop[e that maybe it wasn't so, but what was it then? Something even worse… I was scared about that and searched the internet and got nightmares that it was something even worse" –mother Some adolescents felt betrayed by the information given before the test, as they thought it had not sufficiently prepared them for the consequences of participating in the screening. Described being disappointed by their parents having decided on their behalf for them to participate worlds like "getting caught" or getting stuck frequently used to describe receiving the diagnosis **Suddenly everything made sense – adolescents 5% parents 18%** Some described how the diagnosis came as a relief as they had had unexplained symptoms "We'd been to paediatric clinics earlier for different diffuse problems, so when we found out about this, it was as if it suddenly dawned on me" – father

Bibliographic reference	Rosen (2011): Mass screening for celiac disease from the perspective of newly diagnosed adolescents and their parents: a mixed method study
	<u>Looking back at screening</u> **Feeling grateful for being made aware – 38% adolescents, 72% parents** Knowledge of previously undetected diagnosis was perceived as important and both expressed gratitude Reasons differed depending on adolescents perceived health before the screening If had symptoms becoming aware of diagnosis gave them a means to feel better Adolescents who had no symptoms expressed screening even more important to them as they would not have known about the disease “You’re happy when it’s detected. Since she wasn’t sick, its even better that we found out now. It could have gone on forever.” Both concerned about future complications – these different based on which centre diagnosed at Some sites greatest concern was developing diabetes, others it was cancer risk “ I think knowing is positive. I think It would be worse not knowing and risk of developing all those complications” – father “I think you’re more motivated to eat gluten-free food than to not start smoking because smoking is still your own choice” girl “If you get the recommendation to eat gluten-free food, then it’s more personal.” Boy “it sort of feels more important” girl **Ambivalent feelings towards personal benefit 10% adolescents, 8% parents** Some were ambivalent – this was associated with not perceiving any health improvement and being ambivalent about whether heath complications would really occur This also related to social consequences of having to adhere “I got very annoyed when the doctor called and said that I was gluten intolerant, not because I was gluten-intolerant, but because I had no symptoms.”
Source of funding	
Comments	
Bibliographic reference	**Hogberg (2003): Better dietary compliance in patients with coeliac disease diagnosed in early childhood**

Bibliographic reference	Rosen (2011): Mass screening for celiac disease from the perspective of newly diagnosed adolescents and their parents: a mixed method study
Study type and aim	Cross sectional study to assess whether young adults diagnosed with CD before the age of 4 have better dietary compliance than those diagnosed later in life
Study quality	Was there a clear statement of the aims of the research? Yes – aim is clear Is a qualitative methodology appropriate? Yes – appropriate methodology for this type of research question Was the research design appropriate to address the aims of the research? Yes – design was appropriate Was the recruitment strategy appropriate to the aims of the research? Yes – all screening participants included Was the data collected in a way that addressed the research issue? Yes – standardised protocol for serological testing used. Specifics of questionnaire used not listed Has the relationship between researcher and participants been adequately considered? Not clear Have ethical issues been taken into consideration? Yes – study approved by ethical board Was the data analysis sufficiently rigorous? Yes – serological and questionnaire data analysed sufficiently Is there a clear statement of findings? Yes How valuable is the research? Valuable
Number of patients	29 adults with CD diagnosed at childhood
location	Sweden
Patient characteristics	**Inclusion criteria:** consecutively recruited for the study. Patients were consecutively diagnosed before 18 years of age in one clinic between 1975 and 1981. 9 men and 20 women. Group 1: n=15, aged 4 or younger at diagnosis. Group 2: n= 14, older than 4 years at diagnosis Diagnosis confirmed by biopsy in all according to ESPGHAN criteria. **Exclusion criteria**: noen listed **Mean age at diagnosis**: 5.8 (1.6 – 15.1) **Mean age at follow-up:** 26 (19 – 34)
Intervention	NA
Investigations	Questionnaire Sent by mail to participants

Bibliographic reference	Rosen (2011): Mass screening for celiac disease from the perspective of newly diagnosed adolescents and their parents: a mixed method study
	Sked how after had gluten in diet: never, once a year, once a month, once a week, or always Gluten intake > once a month considered non-compliance
Length of follow up	N/A
Outcome	Self-reported compliance and serological maker of compliance
Results	Questionnaire Dietary compliance significantly differed between the 2 groups from questionnaire measure Serology** 11/29 had elevated EMA 10/28 elevated TGA 80% patients in group 1 vs 46% in group 2 kept a GFD according to serology
Source of funding	Odd Fellow foundation, Sweden
Comments	
** Sera were collected 3 years before questionnaire was filled out!!	

Bibliographic reference	Kurppa (2014): Benefits of Gluten-free diet for asymptomatic patients with Serologic markers of Coeliac disease
Study type and aim	Study investigated whether screen-detected and apparently asymptomatic adults with positive EMA benefit from a glutenfree diet
Study quality	Was there a clear statement of the aims of the research? YES Is a qualitative methodology appropriate? YES Was the research design appropriate to address the aims of the research? NO: Particpants are EMA positive only, so there

Bibliographic reference	Kurppa (2014): Benefits of Gluten-free diet for asymptomatic patients with Serologic markers of Coeliac disease
	is no way to verify how many of this population actually have CD. Optimal research design would have confirmed CD diagnosis histologically. Was the recruitment strategy appropriate to the aims of the research? YES Was the data collected in a way that addressed the research issue? YES Has the relationship between researcher and participants been adequately considered? UNSURE Have ethical issues been taken into consideration? MAYBE; EMA positive individuals were randomised to either GFD or Gluten containing diet for one year. It is possible that the QoL of those with positive EMA who were randomised to gluten containing diet would have significantly benefited from a GFD and their diagnosis of CD was delayed by at least a year. However, if patients exhibited significant symptoms they were withdrawn from the study for further investigation. The authors justify their methodology with the statement that if they had not been part of this study these individuals would never had had testing for EMA anyway and therefore would have continued on their normal gluten containing diet Was the data analysis sufficiently rigorous? YES Is there a clear statement of findings? YES How valuable is the research? Highly valuable - no other studies exist which address this issue. Overall risk of bias = Low
Number of patients	40
location	Finland
Patient characteristics	**Inclusion criteria:** Positive EMA antibodies; aged between 18 - 75; absence of clinical symptoms; **Exclusion criteria**: <18 or >75; symptomatic of CD; Any concommittent conditions; pregnancy; **Mean age at diagnosis**:NA **Mean age at follow-up:** NA
Investigations	3031 individuals who were relatives of coeliac patients (deemed higher risk than the general population) screened for EMA. Of these, 108 were positive and of those, 40 met inclusion criteria. The following investigations were carried out: Serology and HLA genetics Gastrointestinal and heat-related quality of life - GSRS and VAS Laboratory parameters: haemoglobin; iron, folate, albumin

Bibliographic reference	Kurppa (2014): Benefits of Gluten-free diet for asymptomatic patients with Serologic markers of Coeliac disease
	Bone mineral density using X-ray Gastrointestinal endoscopy Questioned on dietary adherence and willingness to continue diet in the future
Length of follow up	2 year
Outcome	GSRS; VAS
Results	All study groups comparable in age sex medical history and associated medical conditions All subjects had HLA DQ2 or DQ8 status Baseline score GSRS = 1.8 (0.6) in GFD and 1.7 (0.6) in gluten group After intervention total GSRS significantly reduced in GFD group (p=0.49) Anxiety alleviated in GFD group in PGWB score (p=0.25) Mean change in SF-36 not significantly different between groups in any dimension Perception of current health as evaluated by VAS improved in the GFD group (p=0.17)
Source of funding	None listed
Comments	
Serological histological and bone mineral density not reported on here as not listed as relevant outcomes in the review protocol.	