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Cerebral visual impairment (CVI) should be considered as a verifiable visual dysfunction not attributable to disorders of anterior visual pathways or ocular disorder. Manifestations reported to be part of CVI include reduced acuity (of non-ocular cause), visual field defects, oculomotor disorders, abnormal crowding ratio (difference between visual acuity tested with single and crowded optotypes), impaired motion detection, and many visuo-cognitive or visuoperceptual impairments. Some authors regard reduced visual acuity as essential for the diagnosis of CVI, while others do not.

Cerebral visual impairment has been described after preterm birth or in neurodevelopmental disorders including cerebral palsy, autism, or genetic or chromosomal conditions. Academic underachievement is also associated with CVI. Children’s behaviour may indicate CVI, while CVI-related behaviours may be misinterpreted as manifestations of autistic spectrum or attention-deficit/hyperactivity disorders. There is an unmet visual need, including due to CVI, in children who attend special schools, but little is known about the unmet visual need in mainstream schools even though half of children with Education, Health and Care plans in the UK attend these.

The diagnosis of CVI usually involves a multidisciplinary team, inputs from parents, and multiple assessments. Therefore, we aimed to estimate the prevalence of CVI-related vision problems in a one-off survey of a sample of mainstream primary schools in England. A secondary aim was to investigate indicators (red flags) that could identify children as being at risk of having these problems. We investigated having extra educational help, a neurodevelopmental disorder, and responses to behavioural or CVI-specific questionnaires as potentially useful red flags.

**METHOD**
This was a cross-sectional survey, using questionnaires and school information to identify children potentially at risk of CVI who were then assessed to look for specific, non-ocular, CVI-related vision problems.
Sampling and recruitment
Our sampling was opportunistic: we invited all mainstream primary schools local to the collaborating ophthalmologists. The head teachers of 11 mainstream schools and one special school volunteered to participate (the latter only if parents/caregivers could accompany their children). The information sheets for mainstream school parents contained an ‘opt out’ slip to return if they wanted their child not to be included.

Data collection
The Strengths and Difficulties Questionnaire (SDQ) has good predictive value for behavioural and mental health problems in children. The short Impact supplement is almost as predictive of behavioural problems as the full SDQ. Teachers were asked to complete the full SDQ including the Impact supplement and parents were asked to complete the Impact supplement only. Parents were also sent a short set of five questions suggested as screeners for CVI-related vision problems (Gordon Dutton’s five questions). Schools shared information about extra educational help and children’s medical diagnoses. We obtained information on the proportion of children in each school who had free school meals, which we used as a proxy for local area deprivation, from the local authority websites.

Nested case-finding examinations
We invited children to a vision examination if they were in any of the following groups: potentially at risk for CVI or comparison groups.

Potentially at-risk for CVI children were defined as follows: having extra educational help (My Plan, My Plan+, Education, Health and Care plan, or equivalent); teacher-reported SDQ Impact score of +2; parent-reported SDQ Impact score of +2; 3 out of 5 positive CVI screening questions (Gordon Dutton’s five questions). These questionnaires are shown in Appendix S1 (online supporting information).

The comparison groups consisted of a random 5% sample of mainstream-educated children (we used Microsoft Excel [Microsoft Corporation, Redmond, WA, USA] to generate random numbers based on the study IDs, ordered them, took the top 5%, and removed children with extra educational help since they were already invited) and children attending the special school (as long as an adult could accompany them to the assessment).

Children in the random 5% and in the special school were included to provide comparison groups expected to have fewer or more children with CVI-related problems respectively, than the at-risk mainstream-educated children. We wanted to avoid stigmatizing invited children, so made it clear in the information leaflets that we were inviting a random sample of typically developing children as well as children at risk of CVI.

Parents of invited children were sent a 51-item CVI-related questionnaire (from which the Gordon Dutton’s five questions are taken), questions about the child’s medical and ophthalmic history, information sheets for the child and parent, a consent form, and a child assent form.

Vision examinations
Children who returned a signed consent form to their school were examined by a study orthoptist on school premises. The vision assessments lasted 45 to 60 minutes and followed a set protocol comprising a range of tests.

Cerebral visual impairment-related vision tests, all binocular (included to answer the primary research question) included the following: crowding ratio at distance; crowding ratio at near; horizontal and vertical supranuclear eye movements (pursuits and saccades); peripheral visual field awareness to confrontation; contour integration cards; simple visuo-cognitive tests (LEA Mailbox, LEA Rectangles); alternate items from the Motor-Free Visual Perceptual Test.

Routine visual function and oculomotor tests (included to investigate the overlap with the CVI-related vision problems) included the following: ocular alignment (cover and alternate cover tests); near point of accommodation and convergence; stereopsis; colour vision.

Additional tests (included to provide preliminary data for future research) were: low contrast acuity (LEA 10M shapes); gratating orientation acuity; eye movements and peripheral visual field awareness with auditory distractor; numbers, shapes, and letters near acuity test (LEA 3-in-1 test).

All tests were carried out with the child’s own glasses if available. Data were entered online into a secure REDCap database (v7 and v8) held at the University of Bristol.

Feedback of test results
Each child’s results were discussed with the school staff by CW. A brief summary letter for each parent was provided and another letter for onward referral to an optometrist or the local eye unit was given if habitual binocular distance or near acuity was 0.2 logMAR or worse, or if there was strabismus or field loss that was not mentioned in the parent questionnaires.

Data analysis
Vision test results are given as proportions of children with impairments using clinical criteria or local or published norms (except for some of the additional tests). We
describe the prevalence of CVI-related vision problems by age, sex, and by whether the children were at risk or in the random 5% sample. We assessed associations with established risk factors: preterm birth,12 previous admission to a special care baby unit; deprivation; and ethnicity.27 More details of the tests and analysis are given in Appendix S1.

Likelihood ratios were used to estimate the usefulness of the red flags as indicators that a child was at increased risk of having CVI-related vision problems. Positive likelihood ratios indicate a higher prevalence of the target condition than baseline if the flag is present: 15% higher if the positive likelihood ratio is equal to 2; 30% higher if the positive likelihood ratio is equal to 5; and 45% higher if the positive likelihood ratio is equal to 10. The converse applies for negative likelihood ratios: 15% lower if the negative likelihood ratio equals 0.5; 30% lower if the negative likelihood ratio equals 0.2; and 45% lower if the negative likelihood ratio equals 0.1.28

Ethical permission
Research ethics approval was granted by the University of Bristol Faculty of Health Sciences Ethics (ref. 39801), including the parental opt out and consent for examination arrangements.

RESULTS

Participants
Of the 2372 children in the mainstream schools, 74 (3.1%) were opted out by their parents, leaving 2298 (96.9%) plus 14 children from the special school; thus, 2312 children took part. A flow chart of data collection (carried out from 2017 to 2019) is shown in Figure S1 (online supporting information). Fewer opted out children attended the schools with at least a quarter of children eligible for free school meals (12% compared with 21%; p=0.093) but similar proportions had extra educational help (14.9% compared with 19.4%; p=0.328) or a diagnosed neurodevelopmental condition (5.4% compared with 3.9%; p=0.496).

Questionnaire data returns
We received 2217 out of 2298 (96.5%) teacher-reported and 714 out of 2298 (31.0%) parent-reported questionnaires for the mainstream-educated children and 10 out of 14 (71.4%) teacher-reported and 5 out of 14 (35.7%) parent-reported questionnaires for the special school-educated children.

Invitations and consent for testing
A total of 547 out of 2298 (23.8%) mainstream-educated children were invited to the vision assessment (Table 1). We invited an additional 16 mainstream-educated children at the request of their teachers.

Table 1: Indications for invitation to vision assessments

<table>
<thead>
<tr>
<th>Invited</th>
<th>Attended</th>
</tr>
</thead>
<tbody>
<tr>
<td>Extra educational help only</td>
<td>143</td>
</tr>
<tr>
<td>Extra educational help + teacher report</td>
<td>125</td>
</tr>
<tr>
<td>Extra educational help + parent report</td>
<td>20</td>
</tr>
<tr>
<td>Teacher + parent report</td>
<td>7</td>
</tr>
<tr>
<td>Extra educational help + teacher report + parent report</td>
<td>24</td>
</tr>
<tr>
<td>Teacher report only</td>
<td>102</td>
</tr>
<tr>
<td>Parent report only</td>
<td>19</td>
</tr>
<tr>
<td>Total</td>
<td>440</td>
</tr>
</tbody>
</table>

*After removing 12 due to having extra educational help. **After removing 14 due to having extra educational help.

were the 14 children in the special school who had a parent/caregiver who could accompany them.

The proportions of children having extra educational help were similar in those who did and those who did not bring a consent form (58.7% vs 58.1%, \( \chi^2 = 0.02, p = 0.884 \)); however, children with a signed consent form were more likely to have a neurodevelopmental disorder than those without (18.2% and 7.3% respectively, \( \chi^2 = 15.6, p = 0.001 \)).

Characteristics of participating children
Among all 2298 participating mainstream-educated children, 1144 (49.8%) were male, 1073 (46.7%) were female, and data were missing for 81 (3.5%); 1874 (81.5%) were white British, 193 (8.4%) were of a different origin, and data were missing for 231 (10.1%). Four-hundred and fifty (19.6%) were at schools where a quarter or more children were eligible for free school meals. The mean age was 8 years 4 months (SD 1y 9mo; range 5y 10mo–11y 3mo). Overall, 435 (18.9%) were having extra educational help and 77 (3.4%) had a diagnosed neurodevelopmental disorder (autism, attention-deficit/hyperactivity disorder, cerebral palsy, developmental coordination disorder, epilepsy, learning difficulties or developmental delay, chromosomal or genetic syndrome). Teachers in mainstream schools reported high Impact scores for 261 out of 2217 (11.8%) of children. Parents reported high Impact scores for 70 out of 714 (9.8%) and high Gordon Dutton’s five questions scores for 12 out of 682 (1.8%) children.

Among the 14 participating children from the special school, 12 (85.7%) were male and two (14.3%) were female; 12 (85.7%) were white British and two (14.2%) were of other ethnicities. The mean age was 7 years 11 months (SD 1y 5mo; range 5y 10mo–11y 3mo) and 43.5% of the children at the school were eligible for free school meals.

The results for the full SDQ completed by the teachers and the full vision questionnaire completed by the parents of invited children will be presented elsewhere.

Table S1 (online supporting information) shows the characteristics of the 248 children who were examined.
There was the intended excess of children having extra educational help (58%), a neurodevelopmental disorder (18%), or high Impact score reported by teachers (43%) or parents (47%). The Gordon Dutton’s five questions indicated risk for CVI in 10% of the children with parent-completed questionnaires. One in six children were born preterm and one in eight had been on a special care infant unit.

Prevalence of CVI-related vision problems

As shown in Table 2, almost a third of mainstream-educated children tested (78 out of 248, 31.5%) had at least one, and 26 out of 248 (10.5%) had multiple CVI-related vision problems. The results for males (33.5%) and females (31.1%) and for children in different age bands were all similar (33.3%, 38.5%, and 25.3% for ages 5–6y, 7–8y, and 9–11y respectively, having any CVI-related vision problems).

Sixty per cent (47 out of 78) of mainstream-educated children with any CVI-related vision problems had been identified by only one test: abnormal crowding ratio at near (n=8) or distance (n=3); abnormal pursuits (n=5) or saccades (n=8); impaired peripheral field awareness (n=2); contour integration at or below the 5th centile (n=1); estimated in the lowest 5th centile of the Motor-Free Visual Perceptual Test (n=6); problems with the LEA Mailbox (n=10) or LEA Rectangles (n=4).

In the mainstream schools, 60 out of 144 (41.7%) of children having extra educational help and 23 out of 45 (51.1%) of children with a neurodevelopmental disorder (NDD) (equivalent to ≥0.3 LogMAR difference between acuity with single vs crowded optotypes) had had at least one CVI-related vision problem and 22 out of 131 (16.8%) and 12 out of 37 (32.4%) respectively had more than one CVI-related vision problem. More children had CVI-related vision problems in the at-risk group (73 out of 207, 35.3%) than in the random 5% sample (5 out of 41, 12.2%; χ²=8.4, p=0.004). The child in the random 5% sample with three CVI-related vision problems was born preterm and had a parent-reported high Impact score. The prevalence of CVI-related problems in the at-risk mainstream-educated children was lower than in special school-educated children (73 out of 207, 35.3% and 8 out of 14, 57.1% respectively; χ²=2.7, p=0.100).

Most (68 out of 86, 79.1%) children with at least one CVI-related vision problem were getting extra educational help or had a diagnosed neurodevelopmental disorder. An additional 9 out of 86 (10.5%) had a high Impact score reported by either teacher or parent.

Had we found all children with CVI-related vision problems, then the prevalence in the mainstream schools would be 78 out of 2298 (3.4%, 95% confidence interval [CI] 2.5–4.0) with any and 26 out of 2298 (1.1%, 95% CI 0.7–1.7) with multiple CVI-related vision problems.

Associations between CVI-related vision problems and known risk factors

Children who had been admitted to a special care infant unit were more likely to have CVI-related vision problems than children who were not: 16 out of 31 (51.6%) compared with 51 out of 175 (28.5%; χ²=6.5, p=0.011). However, we found similar rates in children born before 37 weeks gestation as in those born at 37 weeks gestation or more: 15 out of 39 (38.5%) and 52 out of 172 (30.2%) respectively (χ²=1.0, p=0.319). There was a trend towards a higher prevalence in children who attended schools with greater free school meal eligibility: 31 out of 75 (41.3%)...
compared with 55 out of 187 (29.4%; $\chi^2=3.5$, $p=0.063$), but no association with ethnicity (33.2% for white children compared with 40.1% non-white; $\chi^2=0.3$, $p=0.589$).

**Prevalence of routinely tested visual and oculomotor problems**
In Table S1, approximately one-third of children tested had at least one instance of strabismus, reduced distance acuity, an extended nearpoint of convergence, or worse than average accommodation. These routinely tested for impairments were more frequently present in children with CVI-related vision problems than in children without: 40 out of 78 (51.3%) and 46 out of 170 (27.1%) respectively ($\chi^2=13.9$, $p<0.001$). Only 12 out of 78 (15.4%) children with CVI-related problems had binocular distance acuity worse than 0.3 logMAR. Seventeen children had colour vision abnormalities: 12 had a red-green defect (all males); four had red-green and blue-yellow defects (one female); and one male had a blue-yellow defect. Half of tested children in the mainstream-educated group had worse than foveal stereoacuity.

**Additional visual impairments**
As shown in Table S1, testing eye movements and peripheral visual awareness in the presence of an auditory distraction resulted in more children with abnormal pursuits or impaired visual field awareness in the at-risk group than in the random 5% sample (Fisher’s exact test, $p=0.035$ and $p=0.031$ respectively), but similar numbers in each group had abnormal distracted saccades. Children in the special school-educated group were more likely to have poor low contrast acuity than children in the mainstream-educated group (Fisher’s exact test, $p<0.001$).

More children with at least one CVI-related vision problem had additional impairments than children without CVI-related vision problems: impaired distracted saccades (27.1% vs 4.0%; $\chi^2=30.1$, $p<0.001$); impaired distracted pursuits (30.6% vs 10.2%; $\chi^2=16.9$, $p<0.001$); impaired distracted fields (22.4% vs 4.0%, $\chi^2=21.6$, $p<0.001$); poor low contrast acuity (46.5% vs 31.9%; $\chi^2=5.4$, $p=0.02$); and letters greater than 0.2 logMAR worse than numbers or shapes (4.0 vs 0.6; $\chi^2=3.9$, $p=0.083$).

**Usefulness of potential red flags**
Table 3 shows the sensitivity, specificity, and positive and negative likelihood ratios of the potential red flags (unless the likelihood ratio 95% CIs included unity, so they were not significantly predictive).

If the target condition was any CVI-related vision problem, the most sensitive red flag was having extra educational help, but this was poorly specific with a positive likelihood ratio of 1.5. Conversely, the most specific flags were having a neurodevelopmental disorder (which had a positive likelihood ratio of 2.3) and the Gordon Dutton’s five questions, both of which had low sensitivity.

We hypothesized that the number of CVI-related vision problems may reflect the severity of disturbance to the visual system and compared the results for when the target was one or multiple CVI-related vision problems (Table 3). None of the red flags had positive likelihood ratios different from 1 when predicting children with only one CVI-related vision problem; however, having a neurodevelopmental disorder and/or a parent-reported high Impact score were predictive for multiple CVI-related vision problems, with positive likelihood ratios of 3.8 and 2.7 respectively. The negative likelihood ratio for parent-reported Impact scores indicated a prevalence of 30% below baseline if this flag was confirmed to be absent.

**DISCUSSION**
Our primary aim was to estimate how many children in mainstream primary schools had CVI-related vision problems. Our results suggest that on average, there would be at least one affected child in every class of 30 children. It is important to recognize that not all children we identified with CVI-related vision problems would be diagnosed as having a CVI if they presented to a clinical setting. However, most had risk factors or difficulties associated with CVI.

We found that most of the affected children (79%) were already recognized as being at risk due to having extra educational help or a known neurodevelopmental disorder. Impact scores from the parents were more informative than the equivalent teacher-reported scores and the parent-reported Gordon Dutton’s five questions was the most specific of the flags but had low sensitivity (12%). However, our results for parent-reported data are limited by the low numbers and low statistical power.

One advantage of using likelihood ratios is that they are not generally affected by the baseline prevalence of the target condition and can be used in high- and low-risk settings, unlike sensitivity and specificity, which vary with the underlying prevalence. Across all participating mainstream-educated children, the estimated baseline prevalence of children with any CVI-related vision problems was 3.4%; it was 1.1% for children with multiple CVI-related vision problems. Therefore, increases in prevalence of between 15% and 30% above baseline for children with red flags whose positive likelihood ratio is between 2 and 4 (as in this study) would still result in most children with the red flag not having the condition.

Given these results, providing assessments to identify or exclude CVI for children who need intervention for educational or behavioural problems may be preferable to attempting to screen for CVI. Also, paediatricians and teachers need to be aware that children who are managing at school but have emotional or behavioural problems at home can have CVI-related visual problems (and possibly CVI) as seen in our random 5% sample.

Although many of the affected children we identified were receiving extra educational help, they were not getting vision-based support, perhaps due to lack of awareness or to other competing problems. However, teachers may find information about vision problems useful when supporting children.
vision problems affected children so that opportunities to help them by associated. Pathways to assessment and support are needed for be more prevalent in primary schools than has been appre-
at-risk children and further research would be useful.
an auditory distractor may unmask some abnormalities in of our protocol. The additional tests suggested that using sample than in at-risk children, supporting the specificity of CVI-related problems (using the number of tests failed) may be inaccurate since one major impairment may be more disabling than two or three minor ones. Using the full Motor-Free Visual Perceptual Test or other formal neuropsychology assessments may have given different results.

The strengths of this study include the range of validated assessments, examiners being masked to the child’s risk factors, and a community rather than clinical setting. We identified fewer affected children in the random 5% sample than in at-risk children, supporting the specificity of our protocol. The additional tests suggested that using an auditory distractor may unmask some abnormalities in at-risk children and further research would be useful.

These results suggest CVI-related vision problems may be more prevalent in primary schools than has been appreciated. Pathways to assessment and support are needed for affected children so that opportunities to help them by meeting their visual needs are not missed. Studies that are powered to assess different approaches to identifying affected children are needed, as is research into the benefits of doing this for individual children and their families and for the school and health care systems that support them.

ACKNOWLEDGEMENTS
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This study has several limitations. Our schools were self-selected, more disadvantaged, and less ethnically diverse than schools in England overall. We were unable to examine half of the children we invited so we would have underestimated the true prevalence of CVI-related vision problems. We did not check for refractive errors and so may have underestimated visual acuity, stereopsis, and/or ability to accommodate. Peripheral field awareness testing was by confrontation and may have missed some visual field defects. Our estimate of severity of CVI-related problems (using the number of tests failed) may be inaccurate since one major impairment may be more disabling than two or three minor ones. Using the full Motor-Free Visual Perceptual Test or other formal neuropsychology assessments may have given different results.

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DATA AVAILABILITY STATEMENT
The data that support the findings of this study are available from the corresponding author upon reasonable request.

SUPPORTING INFORMATION
The following additional material may be found online:

REFERENCES

Figure S1: Flow chart of data collection.
Table S1: Summary of child characteristics, routinely tested vision problems and additional visual impairments identified
Appendix S1: Questionnaires used to identify children as at risk of CVI-related vision problems, details of vision tests used, reliability of test procedures, and analysis.
Appendix S2: Participating schools.