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A population-level post-screening treatment cost framework to help inform vision screening choices for children under the age of seven

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ABSTRACT

Purpose/Background: Visual acuity (VA) screening in children primarily detects low VA and amblyopia between 3 and 6 years of age. Photoscreening is a low-cost, lower-expertise alternative which can be carried out on younger children and looks instead for refractive amblyopia risk factors so that early glasses may prevent or mitigate the conditions. The long-term benefits and costs of providing many children with glasses in an attempt to avoid development of amblyopia for some of them needs clarification. This paper presents a framework for modeling potential post-referral costs of different screening models once referred children reach specialist services.

Methods: The EUSCREEN Screening Cost-Effectiveness Model was used together with published literature to estimate referral rates and case mix of referrals from different screening modalities (photoscreening and VA screening at 2, 3–4 years and 4–5 years). UK 2019–20 published National Health Service (NHS) costings were used across all scenarios to model the comparative post-referral costs to the point of discharge from specialist services. Potential costs were compared between a) orthoptist, b) state funded ophthalmologist and c) private ophthalmologist care.

Results: Earlier VA screening and photoscreening yield higher numbers of referrals because of lower sensitivity and specificity for disease, and a different case mix, compared to later VA screening. Photoscreening referrals are a mixture of reduced VA caused by amblyopia and refractive error, and children with amblyopia risk factors, most of which are treated with glasses. Costs relate mainly to the secondary care providers and the number of visits per child. Treatment by an ophthalmologist of a referral at 2 years of age can be more than x10 more expensive than an orthoptist service receiving referrals at 5 years, but outcomes can still be good from referrals aged 5.

Conclusions: All children should be screened for amblyopia and low vision before the age of 6. Very early detection of amblyopia refractive risk factors may prevent or mitigate amblyopia for some affected children, but population-level outcomes from a single high-quality VA screening at 4–5 years can also be very good. Total patient-journey costs incurred by earlier detection and treatment are much higher than if screening is carried out later because younger children need more professional input before discharge, so early screening is less cost-effective in the long term. Population coverage, local healthcare models, local case-mix, public health awareness, training, data monitoring and audit are critical factors to consider when planning, evaluating, or changing any screening programme.

KEYWORDS
Child vision screening; cost-effectiveness; photoscreening; treatment costs

Introduction

Screening for amblyopia in childhood aims to reduce the long-term prevalence of amblyopia in the general population and fulfills many of the World Health Organisation's (WHO) screening justification criteria.[1] In particular, visual acuity (VA) screening is considered cost-effective in the long term,[2–4] but there is limited evidence that screening children before 3 years of age is justified.[5,6] Nevertheless, many countries recommend screening in early childhood even before children can reliably identify linear optotypes.[2–4]

It is possible to screen for photorefractive risk factors using automated devices (photoscreening) under 3 years of age. This has been widely promoted and adopted by some national, regional or local healthcare bodies[7] because if cases are detected earlier in the critical period, disease may be prevented, be less severe and treatment more effective.[8] Most automated screeners also detect

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large angle strabismus and media opacities, so it is an attractive option in settings without a skilled testing workforce. Automated screening has been shown to be more cost-effective than a comprehensive pre-school eye examination in a US rural setting, but photoscreening is less cost-effective than screening using an accurate good linear VA test,[9] especially if early testing is added to later VA screening.[10,11] Photoscreening is, however, easier within some current healthcare models.

The UK National Screening Committee recommended that, beyond the neonatal red reflex test, and the ability of health visitors and general practitioners (GPs) to refer any concerns from questions about vision at general health checks in the interim, children should receive their first formal VA test at 4/5 years of age on school entry, rather than VA testing under the age of 4.[12] This replaced previous screening by single letter VA testing, often including cover testing and motility assessment, a year earlier. Audits by the British & Irish Orthoptic Society[13,14] showed very significant advantages in population coverage, with little difference to final outcomes.

Treatment costs, however, are rarely included in cost-effectiveness calculations,[15–17] and if they are, they use very gross figures.[9,18] Often only short-term costs are reported, such as costs per screen,[19] or up to the point of diagnosis, e.g. cost per case detected.[16] Very few compare the total costs to the point of discharge from specialist services from different providers in similar populations.[9,10,19]

The EUSCREEN Screening Cost-Effectiveness Model[20] is an open-access interactive resource available to decision-makers and professionals which calculates the potential cost-effectiveness of different screening scenarios, taking local circumstances into account. However, even this resource only asks users to input broad figures for costs post-diagnosis, e.g. "what are the average costs of treating one child with amblyopia." This is not easy to estimate and does not account for the fact that although screening aims to identify individuals with amblyopia, it also results in children being referred to specialist secondary services without amblyopia. Children with false positive, untestable or equivocal results, or refractive errors without amblyopia, once referred, may still remain within a healthcare system. For instance, a child presenting at 2 years with hypermetropia may be monitored for several years. Different screening methods yield very different proportions of these groups.

Monitoring of screening and amblyopia treatment success on a community scale is difficult, especially, where different providers are involved, and data are rarely kept consistently even at local levels. Good long-term follow-up of outcomes in research projects and reports is rarely possible,[21–23] and the EUSCREEN project shows that data is very rarely publicly available.

This discussion paper presents hypothetical, but representative, examples of post-referral costs that may be incurred from different screening options, up to the point of discharge from specialist services. We look in detail at how changes to vision screening delivery and post-referral treatment patterns can make large differences to the costs of providing treatment. The UK is fortunate to have a highly skilled autonomous practitioner specialist workforce (orthoptists) who can supervise or deliver screening and can manage the bulk of referrals, with input from optometrists and ophthalmologists when necessary. This low-cost skilled option is not available in many countries around the world.

Methods

To arrive at cost estimates, we needed representative examples of referral rates from screening schemes, which can vary widely. To identify screening programme types, referral rates and diagnostic outcomes we used data from a systematic literature search undertaken in 2019, when investigating the cost-effectiveness of photoscreening (for methodology see Horwood et al.).[18] This review is registered and reported on the PROSPERO International Database of Systematic Reviews,[24] which requires high methodological standards. All English language publications reporting vision screening in children were scrutinized and data extracted on screening method, referral rates, age group, referral criteria and outcomes while looking specifically for evidence of cost effectiveness. The search was repeated in 2022 for more recent literature. Three recently published datasets provided more

detailed data: VA screening at 4–5 years[25] (actual detailed data); VA screening in a similar cohort 6 months younger[26]; and photoscreening at 2 years.[27]

The treatment costs of each intervention were calculated for each of the programme examples using UK NHS 2019 published rates (in GBP £) as shown in Table 1. Private consultation costs at the time were mid-range typical costs for a general ophthalmologist in a large urban area in the UK outside London. These costs will vary widely between countries with different salary and healthcare structures, so every country would have to input typical local costs to make their own calculations.

The case mix from screening falls into seven main diagnostic categories:

(1) False-positive referrals, untestable children and equivocal results which trigger referral. These are more common when testing younger children and when screening programmes utilize nonspecialist personnel.[25] Once referred, some departmental protocols would require such children to be followed until they can demonstrate reliable linear equal VA within published normal ranges,[28] so after an initial diagnostic visit, they may still be followed.
(2) Simple refractive error e.g. myopia, astigmatism, anisometropia and hypermetropia without amblyopia. This group will include children who, if detected under 2 years of age, might be prevented from developing accommodative strabismus and amblyopia. Although emmetropisation is largely complete by the age of 2 years, some children with resolving hypermetropia just over the referral threshold might be referred at age 2, but their refractive error would not be considered significant a year later. Once referred, these children would also be observed by specialists at least until 3 years of age, and possibly longer.
(3) Refractive amblyopia: reduced VA on the return with the first glasses, but which resolves with glasses alone (after "refractive adaptation": the improvement of vision in the first months of wearing glasses).[29] This includes cases with myopia, hypermetropia, astigmatism and anisometropia. They may be discharged or observed once the best corrected VA is confirmed as normal, depending on whether there is provision in the community for them to be managed locally, for example by skilled local optometrists.
(4) Marginal or very mild amblyopia (0.2 LogMAR or better in one or both eyes). If the VA test screening referral threshold is "worse than 0.2 LogMAR," these children would not even be referred. If the threshold was more stringent and they were referred, they may be observed or not treated immediately. Such amblyopia seems to have few severe consequences.[30]
(5) Moderate amblyopia (0.4–0.6 LogMAR) requiring up to 3 months of occlusion or atropine, with more close supervision and longer follow-up compared to categories 2–4.
(6) Dense amblyopia (worse than 0.6 LogMAR) requiring intensive and longer treatment. Treatment and observation may continue until the child is older because crowding is more common[31] and VA may regress to unacceptable levels once treatment is stopped.[32] Children will therefore be discharged later.
(7) Eye pathology. In some cases, treatment will be necessary and costly at a very young age, e.g. a cataract, but in others, a child would be

Table 1. Published or estimated treatment costs in the UK setting based on 2019 rates.

Cost (rounded to nearest £10)	UK £
Cost of a glasses prescription per year (mean NHS voucher value including high/complex prescriptions).	50
Cost of one month's supply of patches/atropine	20
Orthoptist new case diagnostic visit	95
Orthoptist follow up visit	70
NHS ophthalmologist new case visit	110
NHS ophthalmologist follow up visit	100
Private ophthalmologist visit	150

discharged because the condition is stable and untreatable, e.g. a macular scar. They are rare (up to 1% of the referrals) and are excluded from further cost analysis. It is possible that older children may feign poor VA in order to get a pair of glasses, or as part of functional loss of vision, but this is rare in 4–5-year-olds, who are usually unaware of what a screening test might lead to.

Different methods of screening, and the age at which it is carried out, will result in different proportions of these categories within the total number of children referred. We made broad estimates of the probabilities of different case mixes from the literature. In the case of photoscreening, we used figures in the middle range of published values because data, methods, population prevalence and referral thresholds vary widely e.g.[33,34] We accept that these are approximate and often hypothetical figures, but we feel that using data from the systematic review makes them reasonable. They are only used as examples to illustrate how the framework can be used to compare post-referral costs.

We prepared an Excel spreadsheet that enables estimation of approximate costs of running a post-referral service for every 100 children referred (Figure 1) using case mix estimates and the costs listed in Table 1. There are input cells for case mix numbers and local costs. We assumed that most children given glasses would be discharged at 7 years of age. We also assumed that children referred as false positives or untestable would be observed until a linear VA is possible (at age 3–4 if tested by an experienced tester in a comprehensive eye test). We allowed for a private supplier profit margin for private glasses prescriptions or eyepatch provision if not eligible for state subsidy.

"Routine follow-up intervals" and prescribing patterns are known to vary within professional communities, both when prescribing glasses[35–37] during occlusion therapy, and when checking for stability once occlusion is stopped. Typical UK intervals were used, using the pre-COVID departmental protocols from the Horwood et al. example[25]: an annual refraction and prescription, 8-week follow-up intervals during occlusion, with younger children and those being occluded for the first time followed at 6 weeks. On stopping occlusion, follow-up was at 3 months, and then every 6 months until discharge at 7 years of age. We are aware that it is not clear at what point it is safe to discontinue occlusion or full-time glasses in childhood beyond 7 years of age, but we have used common practice in the UK. Another source of variation could be whether occlusion is stopped only when maximum VA is achieved, or at an agreed criterion, e.g. 0.1 logMAR. For the purposes of this exercise, we assumed occlusion was stopped once 0.1 LogMAR was reached in the amblyopic eye.

	A	B	C	D	E	F	G	H
1								
2	Age at referral (mean screening age in years)							
3		False +ves	Refractive error without amblyopia	Refractive error with amblyopia responding to glasses alone	Mild amblyopia (0.2 logMAR or better)	Moderate amblyopia (0.225 - 0.7 logMAR)	Dense amblymyopia (worse than 0.7 logMAR)	
4	Numbers per 100 children referred							=SUM(B4:G4)
5	Age at discharge or leaving costed service (years)							
6	Years between screen and discharge	=B5-B2	=C5-B2	=D5-B2	=E5-B2	=F5-B2	=G5-B2	
7	Typical number of visits between referral and discharge							Total number visits per 100 children referred
8	Total number of visits of all children with this diagnosis	=B7*B4	=C7*C4	=D7*D4	=E7*E4	=F7*F4	=G7*G4	=SUM(B8:G8)
9	Mean duration of occlusion in months	n/a	n/a	n/a				
10	Typical Annual Frequency of refraction							
11	Cost of pair of glasses (e.g €50)							
12	Total cost of glasses per child to discharge		=B11*B10*C6	=B11*B10*D6	=B11*B10*E6	=B11*B10*F6	=B11*B10*G6	Total cost of glasses per 100 children referred
13	Total cost of glasses		=C12*C4	=D12*D4	=E12*E4	=F12*F4	=G12*G4	=SUM(B13:G13)
14	Cost of a month's supply of patches or atropine(€)							Total cost of occlusion
15	Cost of patches/atropine per treated child				=E9*B14	=F9*B14	=B14*G9	per 100 children referred
16	Total cost of patches/atropine				=E15*E4	=F15*F4	=G15*G4	=SUM(E16:G16)
17	Salary and overhead costs per visit of the treating professionals e.g. billed cost for time (average per visit)							Total salary /consultations
18	Salary & overhead costs per child to discharge	=B17*B7	=B17*C7	=B17*D7	=B17*E7	=B17*F7	=B17*G7	=SUM(B18:G18)
19	Total cost per child to discharge	=B18+B12	=C18+C12	=D18+D12	=E18+E15+E12	=F18+F15+F12	=G18+G15+G12	Total cost of treatment per 100 children referred
20	Total cost to service of treatment	=B19*B4	=C19*C4	=D19*D4	=E19*E4	=F19*F4	=G19*G4	=SUM(B20:G20)
21								

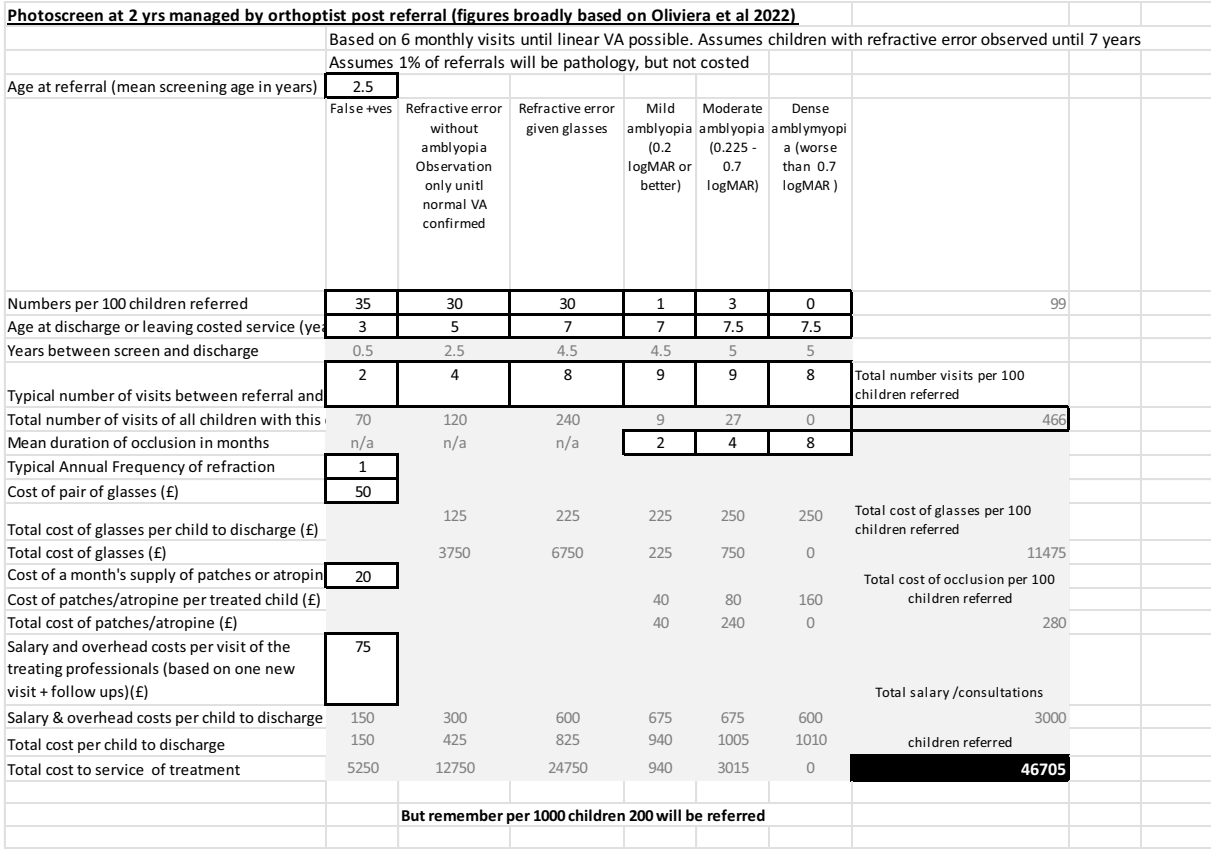
Figure 1. Spreadsheet used to calculate treatment costs.

Table 2. Illustrative examples of different case mix (and numbers) yielded from every 1000 children screened *Actual data from Horwood et al. 2021.[25.]

	1. Single photoscreen @ 2 years	2. Repeat photoscreen @ 2 years & 3 years. Refer only most severe cases after 1st screen	3. VA screen @ 3–4 years	4. VA screen @ 4–5* years	5. Photoscreen + VA screen @ 4–5 years
Referral rate	18%	15%	15%	5%	8%
Children referred per 1000 screened	(180)	(150)	(150)	(50)	(80)
False +ve, discharged or observed without treatment	60% (108)	40% (60)	15% (22)	5% (3)	2% (2)
Simple Refractive error (if corrected)	May be corrected or observed	May be corrected or observed	May be corrected or observed	35% (18)	40% (32)
Optical treatment only	31% (56)	50% (75)	65% (98)	35% (18)	33% (26)
Marginal amblyopia (0.0–0.2logMAR)	Undetectable	Undetectable	4% (6)	5% (3)	5% (4)
Mild (0.3–0.5 logMAR)	5% (9)	5% (8)	10% (15)	10% (5)	10% (8)
Dense amblyopia (>0.5 logMAR)	1% (2)	1% (2)	5% (8)	9% (5)	8% (7)
Eye pathology (not considered further)	1% (2)	1% (1)	1% (1)	1% (1)	1% (1)

Results

Table 2 illustrates five different common examples of screening programmes used 1) a single photoscreen at 2 years; 2) repeating the photoscreen 1 year later, with only the most severe cases referred after the first screen[38]; 3) a VA screen at 3–4 yrs where a linear test is not used or is not possible on less able children (a commonly used UK model before the 2013 UK National Screening Committee recommendation[12]; 4) a single test episode using a linear VA test at 4–5 years[25] and 5) adding a photoscreen to the VA test at the same



Photoscreen at 2 yrs managed by orthoptist post referral (figures broadly based on Oliviera et al 2022)							
	Based on 6 monthly visits until linear VA possible. Assumes children with refractive error observed until 7 years						
	Assumes 1% of referrals will be pathology, but not costed						
Age at referral (mean screening age in years)	2.5						
	False +ves	Refractive error without amblyopia Observation only unitl normal VA confirmed	Refractive error given glasses	Mild amblyopia (0.2 logMAR or better)	Moderate amblyopia (0.225 - 0.7 logMAR)	Dense amblymyopia (worse than 0.7 logMAR)	
Numbers per 100 children referred	35	30	30	1	3	0	99
Age at discharge or leaving costed service (ye	3	5	7	7	7.5	7.5	
Years between screen and discharge	0.5	2.5	4.5	4.5	5	5	
Typical number of visits between referral and	2	4	8	9	9	8	Total number visits per 100 children referred
Total number of visits of all children with this	70	120	240	9	27	0	466
Mean duration of occlusion in months	n/a	n/a	n/a	2	4	8	
Typical Annual Frequency of refraction	1						
Cost of pair of glasses (£)	50						
Total cost of glasses per child to discharge (£)		125	225	225	250	250	Total cost of glasses per 100 children referred
Total cost of glasses (£)		3750	6750	225	750	0	11475
Cost of a month's supply of patches or atropin	20						
Cost of patches/atropine per treated child (£)				40	80	160	Total cost of occlusion per 100 children referred
Total cost of patches/atropine (£)				40	240	0	280
Salary and overhead costs per visit of the treating professionals (based on one new visit + follow ups)(£)	75						Total salary /consultations
Salary & overhead costs per child to discharge	150	300	600	675	675	600	3000
Total cost per child to discharge	150	425	825	940	1005	1010	children referred
Total cost to service of treatment	5250	12750	24750	940	3015	0	46705
		But remember per 1000 children 200 will be referred					

Figure 2. Scenario A: Photoscreen age 2 years, orthoptist treated.

age. It uses reasonable predictions to show that different screening methods have the potential to yield very different patterns of referrals and actual numbers (in brackets) of children per 1000 children screened.

In general, the younger the children, the higher the referral rate and the lower the precision in detection of both amblyopia or refractive amblyopia risk factors, and the more false positives and untestable children. A few more severe amblyopes will be detected if screening is carried out later. Photoscreening refers children with refractive error as well as those with low VA and amblyopia, so many more younger children will be given glasses.[27]

Figures 2 and 3 use outputs of the spreadsheet illustrated in Figure 1 for two common alternative screening methods (further worked examples are in the Supplement). Both figures itemize the costs per 100 children referred. Scenario A is a single minimally skilled photoscreen at age 2 years with the treatment service managed by orthoptists/hospital optometrists (Figure 2). Scenario B is an orthoptist VA screening at 4–5 years, also delivered and treated by orthoptists/optometrists. Costs are 36% higher per 100 referrals in Scenario A, and it is also important to note that 18% of the screened population will be referred by Scenario A and only 5% in B (using examples of recently published data).[25,27]

Table 3 shows calculated total costs (mean cost per child × estimated number of referrals) from eight different types of screening/treatment in a community with an annual birth cohort of 6000 children (chosen because it was the cohort size of the most detailed of our datasets).[25] Costs can vary by a factor of 16 or more (e.g. orthoptist treated from age 5 vs private ophthalmologist treated from age 2). Even if referrals from a photoscreening at age 2 was at the low end of published figures, at 10%, e.g. Goodman et al.[39] instead of the 18% used in Scenario A, the age at referral, which is the main driver of costs, would still be approximately the same. Even the best photoscreening refers more children than later VA screening, and they usually need longer follow-up. The higher sensitivity and specificity of screening for detection of

Orthoptist VA screen at 5 years, orthoptist managed (actual data from Horwood et al 2021)							
Age at referral (mean screening age in years)	5						
	False +ves	Refractive error without amblyopia	Refractive error with amblyopia responding to glasses alone	Mild amblyopia (0.2 logMAR or better)	Moderate amblyopia (0.225 - 0.7 logMAR)	Dense amblymyopia (worse than 0.7 logMAR)	
Numbers per 100 children referred	5	57	24	*	10	3	99
Age at discharge or leaving costed service (ye	5.5	6	6.5	6.8	7	7.5	
Years between screen and discharge	0.5	1	1.5	1.8	2	2.5	
Typical number of visits between referral and	1	3	4	5	6	8	Total number visits per 100 children referred
Total number of visits of all children with this	5	171	96	0	60	24	356
Mean duration of occlusion in months	n/a	n/a	n/a	2	5	8	
Typical Annual Frequency of refraction	1						
Cost of pair of glasses (£)	50						
Total cost of glasses per child to discharge (£)		50	75	90	100	125	Total cost of glasses per 100 children referred
Total cost of glasses (£)		2850	1800	0	1000	375	6025
Cost of a month's supply of patches or atropin	20						Total cost of occlusion per 100 children referred
Cost of patches/atropine per treated child (£)				40	100	160	
Total cost of patches/atropine (£)				0	1000	480	1480
Salary and overhead costs per visit of the treating professionals (based on one new visit + follow ups)(£)	75						Total salary /consultations
Salary & overhead costs per child to discharge	75	225	300	375	450	600	2025
Total cost per child to discharge	75	275	375	505	650	885	Total cost of treatment per 100 children referred
Total cost to service of treatment	375	15675	9000	0	6500	2655	**34205**
		* would pass screening unless other refractive error					

Figure 3. Scenario B: VA screen age 5, orthoptist treated.

Table 3. Comparative treatment costs for a birth cohort of 6000 per annum compared to the lowest cost option in row 8 (see supplement for worked examples 1–7 & 9).

	Cost per child referred (cost per 100 referred/ 100)	Estimated referrals per 6000 children (from Table 2 x 6)	Post-treatment cost for service (£)	Multiplication factor compared to example in row 8 below
1. Single photoscreen aged 2yrs orthoptist managed	467.05	1080	504,900,	x7.4
2. Single photoscreen at 2 with state funded ophthalmologist care	606.85	1080	655,398	x9.6
3. Photoscreen at 2 with private ophthalmologist care	833.15	1080	899,802	x13.2
4. Photoscreen at 2 with private ophthalmologist care, one more visit due to later discharge	1011.65	1080	1,092,582	x16.5
5. Photoscreen with confirmatory repeat before referral	As above	900	420,345	x6.17
6. Photoscreen aged 3.5 yrs orthoptist managed	489.95	900	440,955	x6.47
7. VA screen aged 3.5 yrs orthoptist managed	603.10	900	441,000	x6.47
8. Orthoptist screen at 5 and treat with repeat screen for borderline fails *actual data	342.05	199	68,067	Reference
9. School nurse screen at 5. * actual data ** probably more children missed	298.05	215	64,080	x.94

poor VA at age 5 years means that the treatment costs are much lower due to lower referral numbers and fewer visits before discharge.

Discussion

This paper models the rarely considered whole-patient-journey cost implications of earlier versus later amblyopia screening. This aspect of costs may have escaped attention because patient care is rarely fully coordinated or audited from screen to discharge. Nevertheless, the costs are still incurred by states and parents, and these are important considerations when making health policy decisions. The simple model structure applied may help decision-makers plan and modify services to best suit local resources and circumstances.

It does *not* suggest withdrawing or modifying existing screening programmes without careful audit and piloting of any changes. The importance of the use of data to evaluate services was one of the major findings of the EUscreen study.[21]

The question is not "should screening be a priority," because it is. But rather "what form of screening carries the least burden and optimum population outcome," for children, parents and countries. Most of the arguments revolve around whether the benefits of early detection and treatment of amblyopia refractive risk factors outweigh a more precisely targeted approach incorporating VA measurement slightly later in the critical period, which reaches all children and is cheaper, but which does risk slightly worse outcomes for a few children.

We demonstrate that the age at which screening occurs, the case mix of the referrals, who treats the children, and how long children are under the care of specialist health services greatly influence treatment costs. We used typical treatment costs from the UK NHS, and because orthoptists are not available worldwide, we also included an example using local ophthalmologist reimbursement rates, so that different screening methods could be compared within a common infrastructure. Costs are likely to differ even more widely internationally, and adding a profit motive for healthcare providers may also be a significant multiplier to costs.

There is no doubt that early detection and treatment of amblyopia leads to better individual outcomes and shorter periods of active amblyopia treatment for affected children.[40–45] Some cases of amblyopia might be prevented by early detection, and dense amblyopia may not become so entrenched.[46–48] Children with severe amblyopia detected very early may benefit individually, but there are hidden long-term costs even for these children, and modest outcome benefits need to be evaluated in comparison to the same amblyopia treated, still within the critical period, but later. Whatever age treatment starts, most children need monitoring until at least age 7. Treatment starting at 4–5 years can be very effective,[25,49,50] but the monitoring period is shorter than if referred aged 2–3.

Childhood vision screening programmes target amblyopia and treatable eye pathology, but photoscreening, in particular, also identifies children *at risk* for developing amblyopia[51] or children with mild problems that are not amblyogenic, such as mild myopia, while missing others.[52] Some normal children may also be referred due to poor testability, equipment limitations and lower test specificity.[18] For the many children outlined in this paper with no, or mild problems, also referred from screening, the lifetime benefit of early referral is more questionable, and can be costly.

Countries with limited resources may need to make hard choices about when best to screen and start treatment, and currently there are few sources of objective comparative information. Many countries have patchy health surveillance, wide healthcare provider choice and minimal national data collection, and it is almost impossible to compare population-level benefits from different approaches. More published data are the key.

Paediatric ophthalmologists and orthoptists are busy and scarce. Early photoscreening is generally not as sensitive or specific for detecting amblyopia risk factors as later VA screening is for detecting amblyopia. False or trivial referrals are unpopular with parents and ophthalmologists.[53] Donahue suggests screening test specificity of 97% would be ideal,[54] but this is rare in most programmes, despite great efforts to maximize sensitivity and specificity over the years, so false referrals are more common. Amblyopia that develops before age 3 is mostly caused by strabismus alone or strabismus in combination with refractive error, and most of these cases enter treatment after self-referral anyway.[55] Trust in screening is important to make it acceptable[56] and to ensure treatment uptake, especially in regions where new services are introduced, as was found in the EUSCREEN study of implementation of vision screening in Romania.[57] It is important that the right children are referred *and* that they can, and will, access treatment.

A single VA screen aged 4–5 years carried out by an expert can be highly effective in detecting children with visually significant refractive errors and amblyopia not spotted by their parents, with a very low false-positive rate.[25] When a linear VA is accurately measured by someone experienced, adding photoscreening could be considered superfluous if VA is normal. Photoscreening *without* VA testing at age 4 will still miss some amblyopes, some significant hypermetropes and some pathology if there is no significant refractive error.[52] Regular eye checks should still be advised throughout the school years, but it is not clear how significant mild refractive errors are to very young children as long as they can achieve good vision later.

Solebo et al. argue that the age of a single screen should be 4–5 years.[49] In 2013, the UK National Screening Committee recommended moving the screening age from 3 to 4 years in the community to 4–5 years in school. Two large national audits before and after the change did not find poorer outcomes, but coverage improved dramatically because parents did not have to bring their child to be screened.[13,14] However, it is important that the single, later, screening test is highly accurate, so high-quality training, feedback and audit are vital to maintain standards.

Population coverage is critical. Williams et al. emphasize the point that on a population level, coverage at screening makes the difference to finding positive effects of early detection.[47] Horwood et al.[25] demonstrated that the overall outcomes of screening of children aged 5 years were not very much different from those of Oliveira et al. who screened at 2 years of age,[27] but Horwood et al. reported 97% coverage, compared with Oliveira et al. who reported a coverage of 54.3%. The EUSCREEN Screening Cost-Effectiveness Model[20] clearly shows that cost-effectiveness increases where there is good coverage of the eligible population of children. Coverage of pre-school children can be very good if attached to a well-established early child surveillance episode, e.g. approaching 70% in Flanders, Belgium (personal communication with the "Kind en Gezin" service) but is commonly much lower if parents need to make a specific visit for the screening. Low coverage means that many children will be missed or only detected later. These are also more likely to be from socially and/or economically disadvantaged groups, so poor coverage usually means a lack of equity in the first stage of access to healthcare. By leaving the screen until compulsory education age, as in the UK, or a compulsory pre-school vaccination, means that many more are screened, and

lower false-positive rates mean that only the right children are referred. A few, however, could have developed denser amblyopia or an accommodative esotropia that could have been prevented by earlier detection, and treatment may be marginally more difficult.

In countries such as the UK, where the state funds all child health services via central taxation, there is a strong motivation to make the most cost-effective choices and make strong recommendations for commissioning bodies.[49] Amblyopia is well treated by orthoptists, often in lower-cost facilities such as community clinics, and does not necessarily need a much more expensive ophthalmologist or specialist visit. In the UK, ophthalmologists rarely manage non-complex amblyopia personally, but we have included an example here because they do in countries with few or no autonomous-practitioner orthoptists. This paper illustrates how any well-trained *and audited* specialist workforce can increase the cost-effectiveness of screening dramatically. An important question for decision makers is whether funding high-quality training for locally available workforces and better public health awareness, particularly for parents of children under 4 years of age, is more cost-effective than "cheaper" early automated screening that feeds more children into very expensive and scarce secondary services.

It could be argued that the example of photoscreening referral rates for 2-year-olds used here (18%) is high. It rests somewhere in the mid-range of reported rates, and many large studies include a much wider age range.[58] Even using a lower (10%) referral rate, e.g. Goodman et al.[39] it is still a more expensive option. Repeating the screen before referral may improve sensitivity and specificity somewhat[38] but the optimal interval between screenings is unclear. Children at risk may, or may not, go on to develop amblyopia, or their vision may not be significantly reduced without glasses, but all are still referred into healthcare systems. Some may be given glasses of marginal benefit and can be monitored for months or years. Experience of UK practice suggests that most ophthalmologists or orthoptists would monitor young children until good equal vision can be confirmed, so, in particular, photoscreening of infants means that many would enter, and stay in, expensive secondary services at least until old enough to test VA accurately.

In the end, the public are rarely given a vote on what screening services are offered – but always pay for treatment costs. In some countries, parents may be unable to afford early, longer, more costly treatment for marginal benefit. Every parent and professional wants the best for every child, but public health decision-makers may have to make the harder choice – "is earlier detection worth it?." High income, highly health-aware, countries may decide their population is prepared to pay for earlier detection and arguably slightly better individual outcomes, but on a population level, and especially where healthcare is paid for wholly or partly from taxation, cost-effectiveness may take precedence. The literature is so sparse in this area that decision-makers, or their advisors, have little evidence upon which to make public policy decisions, and may unwittingly choose more expensive options.

Table 3 suggests that the cheapest option is apparently a school nurse screen/orthoptist treated service, but this is somewhat misleading. The cohort screened by less well-trained and -audited school nurses used for this study[25] had a very high number of false positives (42%) compared to its neighboring district with orthoptist delivered screening (9%), despite very similar referral rates and populations. If referral numbers were the same, this implies that some children with poor VA may have been missed by the school nurses (false negatives). These children may remain undetected or only identified elsewhere in later childhood. Later presenting or missed children also incur a cost and negatively influence the later population-prevalence of amblyopia. The school nurses were rarely given feedback about the accuracy of their referrals, or the outcome of the treatment, so this could probably be managed by better communication, better training and emphasis on completing the audit cycle. The orthoptist-led and administered service reported in 2021,[25] which has a close audit from screen to final outcome, is highly efficient and late-presenting amblyopia is exceptionally rare.

It is notable that both VA screening[26] and photoscreening at 3–4 years of age can produce fairly similar referral rates and costs[59,60] (although

different prevalence rates may pertain to these studies from different countries), but costs are still higher than if the screening waited a year until 4–5 years.

Our study has some limitations, and many aspects were not specifically considered. Firstly, generalizability is limited. We have used approximate figures for the scenarios presented, as there is a huge variation in the reported data, even among studies that seem superficially similar. We use three actual published examples because they were particularly comprehensively reported, but other scenarios presented here are more speculative. Our examples should only be considered as a basis to start analyzing how differing screening scenarios can influence costs. It is important to realize that national and regional populations, healthcare provision, ethnicity (which may influence population prevalence of conditions), public health awareness and healthcare-seeking behavior differ widely. It must be emphasized that this paper only serves as a framework for decision-makers to approach planning, piloting and audit based on using their own actual, not hypothetical, data.

It would have been preferable to perform a meta-analysis of different published reports, but despite the efforts of researchers in the field,[23,61] data are patchy, without agreed reporting benchmarks.[21] A major finding of the EUSCREEN study was that data collection, a core dataset, ongoing monitoring, and a full audit cycle of outcomes should be embedded in all screening services.

There are multiple possible scenarios and factors which we did not address:-

(a) False negatives that are very difficult to capture from reports of community screening. These are likely to be higher where non-automated tests involving expertise, such as acuity and cover tests, are administered to very young children by less skilled personnel.[62]
(b) Referral criteria will vary and may be adjusted as schemes progress to optimize outcomes.[33,51,63] These criteria will determine how many children are referred. The referral criteria for different photoscreeners at different ages are a large research topic and are under constant scrutiny. We used one of the many published datasets, because it was recent and quite comprehensive. This paper highlights how the length of follow-up for referrals acts as a significant multiplier to costs, so getting referral criteria right is even more important.
(c) Orthoptic tests at the age of 6–24 months[44] and VA testing at the age of 36 months have been shown to be of limited value, so have not been considered.
(d) We did not calculate the cost of diagnostic nonattendance ("no-shows") or loss to follow up, either due to poor parental awareness or inability to access care. These costs can be high[33,58,64] and are known to impact upon the efficiency of healthcare services. High attrition rates are probably best approached by increasing public health awareness.
(e) We did not attempt to calculate other costs to parents (e.g. travel expenses or lost income due to time off work), but as with the other costs, these obviously increase when more visits are necessary.
(f) Referral rates may also differ if VA is tested to a threshold or just to a pass/fail criterion (e.g. a mild amblyopia of two lines difference (R 0.175, L −0.1 logMAR) would pass the screen if screening stopped at a 0.2 pass/fail criterion).
(g) Costs may differ if there is a community workforce, e.g. optometrists, suitably qualified and expert enough to treat and monitor pre-literate children in the community
(h) Although the examples presented here did not estimate the costs of eye pathology, earlier detection may be important for countries with a high prevalence of potentially blinding conditions such as trachoma.
(i) Strabismus and minor media opacities are often considered target conditions for screening but do not in themselves fulfill as many of the WHO criteria for screenable conditions. VA screening without other tests will pick up these conditions if they are visually significant, and most are referred via other routes (via parental concern or red reflex testing in infancy).

(j) We also did not address the effects on other patients being treated by stretched and scarce pediatric ophthalmology services. Many mild or equivocal cases referred from screening may lead to extended waiting times for these children, who often have more serious diseases.

National opinions differ widely as to what the target conditions are, but this paper refers to amblyopia and corrected and uncorrected low vision only. Choices, diagnostic thresholds, "success" definitions, prescribing patterns, follow-up intervals and costs vary widely around the world and in the literature. We could not address them all here, but by choosing standardized, fairly representative definitions and costs across the different scenarios we were able to illustrate the main drivers to high patient-journey costs. Local piloting, feedback and audit are the only way to make more accurate cost predictions and efficiencies.

Further directions

There is strong evidence that screening at 4–5 years is warranted,[65] but it is weaker for screening earlier in childhood. Current arguments for pre-school correction of modest refractive error are weak, but stronger ones may yet emerge. A recent review by Evans et al.[5] highlights that even the best evidence around thresholds for glasses prescription is still based on professional consensus, rather than empirical evidence of need or benefit. So if detected, how early, and which, refractive errors need correction? Another pertinent question is "when is it safe to discontinue occlusion, monitoring or glasses"? Every extra visit increases costs, so how many children benefit from longer observation before discharge, and do any suffer significant lifetime harm if mild residual amblyopic VA drops a little beyond the age of 7?

There is ongoing debate as to what we are actually screening *for*.[66] If correction of refractive error in non-amblyopic pre-school children was proven to help their general development, or make a significant difference to final acuity, the argument for earlier detection of refractive error would be much stronger, and needs more research. How reduced does VA have to be to need correction, and does the type of refractive error matter? Currently, early correction is a costly option, with weak evidence of need. Whilst there is some evidence that children over 4 years benefit from correction of hyperopia,[67,68] very young children can function well with modest blur because their visual requirements are generally not for fine detail, and they can often accommodate to overcome blur. Reported VA outcomes from later screening and correction are still good.[13,14,30] Research is, however, needed to investigate the effect of optical correction on both cognition, generalized attention and literacy, as well as purely visual outcomes, and such studies are under way.[67–69]

There is also growing interest in lifetime consequences of amblyopia and its treatment, with evidence that amblyopia is associated with poorer quality of life (QoL) in some studies[70,70–75] (but not in others).[30,76,77] It is not always clear, however, whether earlier detection and treatment would change anything. Lower QoL could be due to self-perception of disadvantage, such as of "being a patient" or "having a physical difference,"[78] or stigma associated with glasses or patches,[6] which would remain after any amblyopia is treated, whatever the age. Amblyopia is associated with defective binocularity and fine depth perception, and some subtle defects in the non-amblyopic eye[79] which, while sometimes improved[80] are rarely fully restored with conventional occlusion (although recent dichoptic treatments improve these outcomes).[81] It is unclear how much treatment improves daily function in terms of fixation instability,[82] fine motor deficits and slower reading.[83] If subtle neurological impairment or primary poor binocularity caused the initial amblyopia, they will persist. Few papers consider possible adverse effects of treating amblyopia,[6] and amblyopia treatment itself carries risks to QoL.[84] There are small benefits from early treatment of amblyopia,[85] especially for the most severe amblyopia, but the significance of one or two more lines of mild residual amblyopia in health economics terms (e.g. QALYs) needs further research.

Mildly myopic children have good near vision and no amblyopia so appear to have fewer educational problems and indeed may do better educationally,[86] but now that treatment is available to mitigate the development of high myopia, the

point at which early myopia should be identified is another important emerging issue. It is possible that different types of screening become a preferable option (e.g. looking for longitudinal refractive trends, or repeating VA screening later into childhood). Our approach, advising consideration of post-screening costs and benefits could still be applied, but this is beyond the scope of this paper.

Early screening of children under 4 years of age is inefficient on many levels, so it would be interesting to conduct a trial of outcomes comparing more intensive education for parents about how to spot strabismus or monocular or bilateral poor vision in their pre-school children, with a comparative cohort screened early. It may be better or more cost-effective to inform parents of how to detect severe refractive error and dense amblyopia with simple measures at home, e.g. objection to occlusion, inability to identify near or distance detail, and leave the more subtle defects to be screened at a later time.

Conclusions

We do NOT suggest that existing screening services are changed, and certainly not withdrawn, without proper audit and piloting of proposed improvements. Countries make very different screening decisions,[7] but these are usually based on local factors and expert opinion informed by a literature heavily weighted toward what tests, which thresholds and at which age – not what happens to the children *after* the screening, or to the children who are not screened. The EUSCREEN implementation study of vision screening[57] taught us the importance of good groundwork with communities. Preparing the population to be willing and able to access screening and aftercare; making sure that local services can cope efficiently with referrals; collecting data at all points in the patient's journey from screen to discharge; making data available to others, and using training, feedback and audit to make incremental improvements are all vital.

Where funding is limited, where there is poor public awareness of the importance of early detection and treatment, or where there are few people with the skills to treat patients, then it is important to target the most important cases as efficiently as possible.

Cost-effectiveness is not about how much it costs, but is it worth it, in terms of reduction of population prevalence of disease or disadvantage. In the end, societies always have to pay for treatment, either personally, or via taxation or insurance, and only some countries and populations may be prepared to pay more. This paper presents a framework for considering post-screening costs when making screening decisions.

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