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THE ALASKA BLIND CHILD DISCOVERY PROJECT:

RATIONALE, METHODS AND RESULTS OF 4000 SCREENINGS

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ABSTRACT

Background

Photoscreening allows lay persons to adapt the Enhanced Brückner Test to preschoolers in an attempt to identify refractive amblyopia. The Alaska Blind Child Discovery (ABCD) project is charitably funded and administered.

Methods

MTI® photoscreening was offered to children in rural and urban communities in southern Alaska from 1996 through June 1999. Parents answered questions concerning the child's health, family ocular history and whether the child had any eye "Warning Signs." The MTI® images were interpreted by two eye doctors using a modification in MTI® published guidelines.

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Financial Disclosure: MTI discounted photoscreeners 10% ($300) each to ABCD. None of the authors have any other financial association with MTI or other photoscreening techniques.

Results

Out of 4000 screenings performed on 3930 children, there was an overall "not normal" interpretation of 9% and an inconclusive rate of 1%. The mean S.D. age was 3.9 2 years. Only 6% had had a prior eye exam. The average number of Polaroid pictures per screening was 1.16. Follow-up data on "not normal" results was obtained on just over 50%. The positive predictive value during the first two years was 77% but improved to 92% from 1998-1999. Affirmative answers to the questions concerning previous eye exam, child's health, siblings eye health and positive "Warning Signs" were significantly associated with "not normal" interpretations but affirmative answers about eye health of mother, father and relatives were not. Community penetrance of photoscreening to the target age-group ranged from only 5% for Anchorage to almost 100% for the Bristol Bay public health nurses. Five percent of parents of "positive" results surveyed would not have recommended screening for their friends. Equipment functioned dependably even in remote Alaska.

Conclusion

Charitable volunteer Polaroid photoscreening detected amblyopia and significant pediatric eye disease in over 300 children during the first 3.5 years of ABCD.

INTRODUCTION

Amblyopia is a deficiency or interruption in the acquisition of vision by the brain during childhood which can result in lifetime cerebral visual loss(1).

It is the most common form of unilateral blindness in children and young adults. The three main types of amblyopia classified by anatomic factors are deprivational, strabismic and refractive(2). Deprivational amblyopia is due to the disruption of images by an infantile cataract, a corneal opacity or a malformed lid. Strabismic amblyopia results from misalignment of the two eyes due to constant esotropia, exotropia or hypertropia. Refractive amblyopia results from poorly focused images in one or both eyes due to high amounts of hyperopia or astigmatism, very high amounts of myopia or mild to moderate amounts of unequal hyperopic refractive error called anisometropia. Anisometropic amblyopia can also arise if one eye is very myopic. Anisometropic amblyopia is least likely to be noticed by parents and is a common component of the most severe cases of amblyopia.

The average child is moderately hyperopic with 90% of normal children having a cycloplegic spherical equivalent refractive error between +0.50 and +3.75 Diopters(3-5). These children see clearly by accommodating. During the teen years, a large portion of the population becomes myopic, requiring spectacles or contact lenses to clearly focus at distance.

There are critical periods in the development of a child's vision. The cellular architecture in the visual occipital cortex can be permanently impaired by school-age in severe forms of strabismic and refractive amblyopia or as early as four months of age with dense deprivational amblyopia. Amblyopia can be treated by combinations of surgery, spectacles or contact lenses, occlusion (patching) or blurring of the non-amblyopic eye. Early detection and persistent therapy can cure virtually all amblyopia(6).

Amblyopia meets World Health Organization (WHO) guidelines as a disease which merits screening in part because it is a significant public health problem which has a critical developmental period and effective treatment is available(7).

In 1996, the American Academy of Pediatrics (AAP) in association with the American Academy of Ophthalmology (AAO) and the American Association for Pediatric Ophthalmology and Strabismus (AAPOS) adopted guidelines for childhood vision screening(8). Age-specific screening is to be performed by pediatricians, family doctors, eye doctors and/or school professionals. The screening protocol involves observation of external anatomy and visual behavior with specific reference to pupillary 'red reflex" shortly after birth, fixation behavior and strabismus by 2-3 months, acuity by 3.5 years and during the first years of school. State-to-state adoption of policies for preschool vision screening have not been uniform(9).

An important and useful screening technique in children makes use of the red reflex and is called the Enhanced Brückner Test (E.B.T.)(10). Photoscreening records the image of the E.B.T. using on-axis or off-axis flash light source specifically designed to detect refractive errors in a range of concern(11-16). Howard Freedman, a pediatric ophthalmologist from Redmond, Washington developed an off-axis photoscreener using Polaroid technology which takes two orthogonal images. The actual machine is sturdy and portable; a 30.5 x 28 x 16.5 cm, 3 Kg camera, a manual and battery charger fit into a rugged 20.5 x 33 x 53 cm suitcase(17). Dr. Freedman's photoscreener is distributed by Medical Technology and Innovations, Lancaster, Pennsylvania (www.mtens.com) and is known as the MTI® Photoscreener (Figure 1,2).

Figure 1. An MTI photoscreener in use; orthoptist Diane Armitage screens a child. Lay screeners readily learn the process which rarely upsets the child.

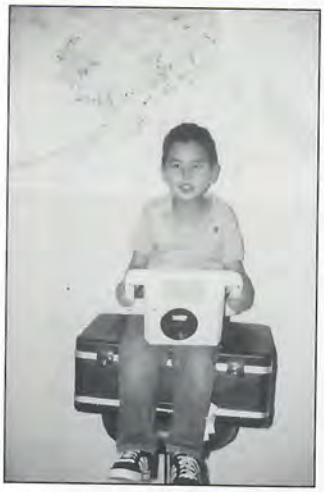


Figure 2. Anisometropic amblyopic rural Alaskan detected by ABCD. He holds the MTI camera and is perched upon the traveling suitcase.

The Alaska Blind Child Discovery (ABCD) is a cooperative charitable research project to photoscreen every Alaskan preschooler. Following is the report of our use of the MTI® photoscreener on the first 4000 young Alaskan screenings.

METHODS

Funding

The Alaska Blind Child Discovery has been charitably funded. The routine price for an MTI® photoscreener is $3000 and the price for each Polaroid film is between $1.10 and $1.50.

To date, no health policy recommends or requires photoscreening. The doctors at Ophthalmic Associates became aware of the potential for this technology to detect treatable childhood blindness(18). Rather than "participate" in the late detection of amblyopia, the doctors ordered 2 photoscreeners, film and paperwork. The local news media were contacted(19-23). Presentations were made to Alaskan pediatricians, ophthalmologists, optometrists, public health nurses and village health aids. Charitable organizations, specifically the Shriners, Masons, Lions Clubs and Kiwanis Clubs were contacted and presentations made.

Individuals and charitable organizations purchased 18 photoscreeners from Medical Technologies and Innovations for a special research price of $2699.

Alaskan Air Carriers donated space-available transport of photoscreeners in their protective suitcases to rural screening clinics.

An oil painting depicting children trying to peek at an Alaskan scene over an obstruction was donated by Alaskan artist Steve Gordon (Journal cover, figure 3). From this, posters were printed listing charitable sponsors. Individuals willing to donate $25 for a box of 20 Polaroid films were given a poster.

Charitable organizations donated to printing patient and clinic paperwork and to mailing packets of completed photoscreening material to the Coordinating Center. The interpretation of the images and the notification of families and clinics were donated by the doctors at Ophthalmic Associates.

Distribution

Donated photoscreeners were based in ABCD hubs in Anchorage, the Matanuska-Susitna Valley, Seward, Valdez, Dillingham, Kodiak, Bethel and Juneau. Data obtained from three cameras was not available for this report: two cameras functioned out of Fairbanks with Dr. Dan Karr and one camera was with the Nome public health nurses.

Charitable organizations organized community clinics in Anchorage, Palmer, Wasilla, Big Lake, Valdez, Bethel, Southeast and Kodiak. Low Vision professionals from Special Education Services Agency (SESA) conducted clinics with the Homer, Kenai, Eagle River, Anchorage and Kodiak Infant Learning Programs. Public health nurses from Dillingham(24), Seward, Anchorage, Delta Junction and Bethel incorporated photoscreening into their scheduled rural village visits. The Children's Miracle Network at Providence purchased a camera for free use in the outpatient sub-specialty clinic.

Figure 4 indicates the location of ABCD clinics and screening hubs.

Figure 3. Steve Gordon Painting (journal cover)

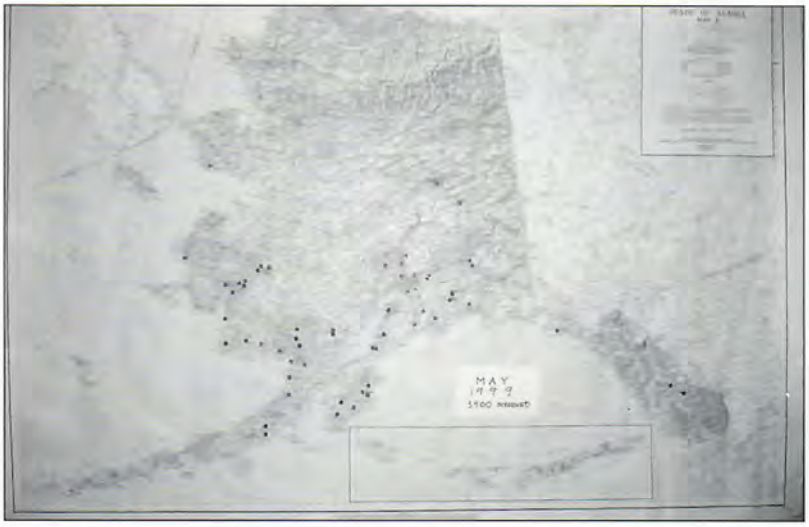


Figure 4. Screening clinics and hubs for Alaska Blind Child Discovery - February 1996-June 1999.

Data Collection

ABCD utilized three types of data. The first involved one or more MTI®. Polaroid photographs taped or stapled to an important data form (Figure 5). The second data came from a questionnaire on the data form concerning the child's health and the ophthalmic family history. The third type of data required reading a list of childhood vision "Warning Signs" (Figure 6) and signifying if the child exhibited any or not. Parents identified the name and birth date of the child, the return address of the parent in a special box corresponding to the return envelope window, the phone number and the child's pediatric caregiver.

Interpretation

All images were interpreted by Dr. Arnold or Dr. Coon with controversial photos reviewed by both doctors. The initial images were interpreted by the guidelines set forth in the manual that accompanies each new MTI® photoscreener. The MTI PowerPoint® demonstration was also carefully viewed. With increased experience and a reassessment of age-related detection goals, the ABCD interpretation guidelines were modified slightly as follows.

The two first Alaskan MTI® cameras have been based in each Ophthalmic Associates office and used frequently on patients of interest. From October 1995 through June 1996 approximately 1000 additional images from clinical practice were carefully reviewed. The camera is excellent for documentation of corneal size, pupil size and shape, media opacity (except <1 mm central opacities which hide behind the corneal light reflex), lid shape and height and Horner's syndrome (one image in dark and the other with more light). It can often identify subtle irregularities of refraction in a manner similar to streak retinoscopy near endpoint. MTI® recommends using 337 Polaroid black & white film due to fast film speed and reduced need for flash intensity. We have also used Polaroid 339 color film with good success, though the exposure is dark and the color process reverses the image so the right image is really the left eye, and vice-versa.

We tried to define the amblyogenic factors which we did NOT want to miss in Alaskan children aged 1-3 years under conditions of poor cooperation with photography. It is known that emmetropic small-pupil MTI® images taken off center can be interpreted as normal, especially with black and white film(25). We took multiple photos of known hyperopic anisometropes; none were able to normalize their MTI® photos even with off axis or out-of-focus images unless the pupils were very small (2.5 mm).

Certain refractive states are much less likely to result in severe amblyopia than others, even though they may be detected by MTI® guidelines. Therefore, we specifically sought moderate (≥1 diopter) to high (>3 diopters) degrees of hyperopic anisometropia. Polaroid images demonstrating moderate amounts of astigmatism and myopia were interpreted as "normal" with a comment about astigmatism or nearsightedness. Abnormalities in pupil and corneal size potentially associated with neuro-ophthalmic disease or glaucoma were also specifically interpreted as "not normal." Symmetric, very light colored pupils can occur under normal mydriatic conditions in light-pigmented individuals and were eventually interpreted as "normal." If we were able to find a single image with reasonably focused pupils with passing refractive estimates, even if several other misaligned or out-of-focus images were also attached to the data sheet, a "normal" interpretation was given.

Notification

For the first year of ABCD, parents were mailed a postcard which they had pre-addressed. For clinics with multiple children screened, a "Clinic Report Sheet" was filled out by the screener identifying ten children per page with phone numbers listed (Figure 7). The postcard was marked with the photograph interpretation as either "normal", "abnormal" or "inconclusive." After 1997, the interpretation was clearly identified on the data sheet with the MTI® photo attached and a photocopy was mailed directly to the parents, and in many cases to the referring clinics. Accompanying the photocopied results was a color brochure describing the problem of undetected amblyopia, the potential benefits of early detection and the implications of "normal" or "not normal" interpretation. Parents of "abnormal" result children were instructed to seek a free repeat screening or obtain a complete eye exam from *the nearest or most convenient eye doctor*. If amblyopia was detected and the complexity of the problem challenged that doctor, the brochure suggests that the child may be referred to "one of Alaska's three pediatric eye specialists, Dr. Arnold or Dr. Robin Grendahl in Anchorage or Dr. Dan Karr in Fairbanks." Parents were requested to bring the photocopy of their MTI® Polaroid image to the eye doctor.

Figure 5.

We Serve

ALASKA BLIND CHILD DISCOVERY

A.B.C.D. is a cooperative, charitable research project to photoscreen every preschool Alaskan. Yukon-Kuskokwim

IDENTIFICATION (To be filled out by parent or Guardian)

Child's First Name:_____ Last Name_____ Child's Birth Date m_____/d_____/y_____

Parent's
Name: _____

Address:_____

Town:_____, Alaska zip:_____ (results mailed to this address!)

Phone: (907)_____-_____ Alternate Phone:_____-_____ Clinic or Doctor _____

HEALTH HISTORY and consent Filled out by parent or guardian: please sign:_____

Has the child ever had an eye exam? ___No ___Yes .....Results:_____

Child's Health Problem(s):___No ___Yes: _____

Eye problems in child's siblings (brothers or sisters):___No ___Yes: _____

Eye problems or strong glasses in mom? ___No ___Yes: _____

Eye problems or strong glasses in dad? ___No ___Yes: _____

Eye problems in relatives (grandparents, uncles, aunts) ___No ___Yes: _____

Does this child consistently have any "**WARNING SIGNS?**" (see back of this paper) ___No ___Yes
Do you object to having these pictures shown for medical education? ___No ___Yes

MTI® PHOTOSCREENING (filled out by technician) Tape or staple all MTI® photos here

Exam Date:_____/_____/_____
Cooperation (circle): Easy Moderate Challenge
Photographer's Name:_____

ABOUT PHOTOSCREENING WITH MTI®:

Good adult vision must be learned during the first ten years of childhood. Amblyopia is a potentially curable disease of disrupted vision learning due to blocked images (cataracts or corneal scars), poor or unequal focus (myopia, hyperopia, astigmatism or anisometropia) or misalignment (strabismus; cross-eye, wall-eye). Amblyopia treatment is most successful if started in pre-verbal children. Prior to photoscreening, this blinding condition has been difficult to detect.

The MTI® is one of the most portable and practical of a new group of instruments called photoscreeners. It has greater than 90% sensitivity and specificity in finding childhood amblyopia. This means most, but not all problems will be detected. The American Academy of Pediatrics recommends that children have frequent, thorough vision screening leading to complete eye exam by age 5. Photoscreening identifies which children should have their first complete eye exam early.

A.B.C.D. Gold '98 Coordinating Center:
542 West Second Avenue, Anchorage, Alaska 99501
(800)270-1617 or (907)276-1617 • fax 278-1705

RESULTS read by Drs. Robert Arnold or Lynn Coon

MTI® Reading Date:_____/_____/199____

NORMAL	NOT NORMAL	INCONCLUSIVE
Eye Exam by age 5	Eye exam SOON (take this to your eye exam)	Eye Exam or Rescreen in a year

Figure 6

WARNING SIGNS IN CHILDRENS' EYES

Guidelines for childhood eye exams:

The American Academy of Pediatrics and the American Association for Pediatric Ophthalmology and Strabismus agree that all children should have their eyes examined by the pediatric- or family doctor: 1) at birth, 2) at regular check-ups with vision testing using verbal charts before school. We feel that at least one thorough exam by an eye doctor including cycloplegic refraction and dilated retina check should be done by the age of five even in children who do not show signs of eye problems. Since brain visual development can be seriously and adversely affected before the age of 8-10 years ("Amblyopia") early and persistent intervention is critical for 5% of children. Urgent or more frequent eye exams are indicated if you observe one or more of the following **Warning Signs** in a preverbal child.

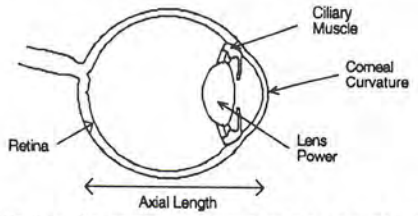
- ❑ Lack of fixation: After a few weeks of age, a normal baby should be able to look at your face and follow your eyes as you move from side to side. Even before that, a normal baby will quickly close both eyes when exposed to bright light.
- ❑ Jerking Eye Movements: As a baby begins to fix his/her eyes, they should rest steadily without jerking side-to side or up-and-down. Such perisistent or intermittent eye movements called "nystagmus" can indicate brain dysfunction or subnormal visual potential.
- ❑ White Pupil: The pupil is the hole in the iris through which light enters tha back of the eye and the retina. Under nomal conditions, the pupil is black or it may appear reddish-orange in photographs. A white or discolored pupil can indicate a cataract or a life-threatening tumor in the eye.
- ❑ Slow or unequal pupils: The pupils should be round, roughly equal in size and each should get larger in the dark and smaller in bright light. Irregular pupils can indicate serious eye disease or abnormal development. Unequal or slowly reacting pupils may indicate retinal or brain disease.
- ❑ Excess Sensitivity to light: Called "photophobia," this can be caused by harmful inflammation in or on the eye or by an abnormally functioning retina.
- ❑ Redness: Inflammation and infection in or on the eye will cause the tiny blood vessels overlying the white sclera to dilate causing an injected, red appearence. A broken blood vessel on the eye ball may make a bright red blood blister which is usually not as serious unless caused by trauma.
- ❑ Drooping Lid: Abnormalities of the brain or tissue around the eye ball may cause one or both lids to droop (ptosis) or retract. Other children have a drooping lid at birth which may cause vision loss secondary to astigmatism.
- ❑ Misalignment: Days to weeks after birth, a baby's eyes should be aligned (most of the time) on interesting objects, near and far, left and right, and up and down. Any persistent misalignment called "strabismus" will usually cause vision loss (amblyopia) and may be due to nerve or brain problems.
- ❑ Head Tilt: When a baby's eyes are better aligned in one direction than another, a head tilt or head turn may result.
- ❑ Swelling around the eyelids: Lumps, changes in color or swelling around the eyes and lids can be caused by tumors or life-threatening infections.
- ❑ Pain or headache: Inflammation or high pressure in an eye can cause pain ranging from a dull ache to excruciating and radiating back to the rest of the head. In addition, the eyes may be involved in the cause or diagnosis of some other kinds of headache.
- ❑ Excess tearing: Blocked tear ducts are not the only cause of excess tearing. Serious inflammations, blurry vision and nerve problems are also possible reasons.
- ❑ Squinting or frequent blinking: Partially closed eyelids may produce temporary improvement in some types of blurry or double vision. Frequent blinking may occur with eye inflammation or allergies or with neurologic disorders.
- ❑ Moving close to see: A baby's eyes can focus much closer than an adult's. However, children who persistently sit close to the TV, or who hold objects close to their eyes may have significant visual impairment.
- ❑ Large Eyes: Vision-robbing congenital glaucoma may cause very large eyes.

Child's Name:_____

❑ Please Check (√) any Warning Signs that apply to your child.

When children begin to talk, the health of their eyes becomes less of a mystery. Even before that, your pediatric care giver should be able to rapidly screen even the most wiggly of infants for most of these serious problems*. Preverbal children can also be Photoscreened. If parents or primary doctors remain concerned, or if the Photoscreen is abnormal, children should then see (visit) the eye doctor.

REFRACTIVE ERRORS (glasses problems): **Farsightedness** and **Nearsightedness** depend on the length of the eye, the power of the lens, the curvature of the cornea and the pull of the cilliary muscles distorting the young flexible lens. **Astigmatism** is usually due to irregular curvature of the cornea; the front of the eye is more like the side of a football than the shape of a basketball (no astigmatism). **Anisometropia** is unequal power of the eyes. Children with large amounts of farsightedness (hyperopia), astigmatism and anisometropia are at high risk of refractive amblyopia...if left untreated potential brain blindness in one or both eyes!



The infant eye: the cornea and lens should be capable of focusing light from near and far objects through the pupil allowing the retina to send an images from both eyes to the brain.

Robert W. Arnold, M.D., Lynn J. Coon, O.D. and M. Diane Armitage, orthoptist

* **Arnold RW:** Vision screening in Alaska: Experience with Enhanced Brückner Test. Alaska Medicine 35(2): 212-215, 1993

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Figure 7.

A.B.C.D. SCREENING REPORT

CLINIC:______________________________ Date: ___/___/___
ADDRESS:______________________________________
PHONE: (907) ______-________ • FACSIMILE: (907)______-__________
Photographer(S)______________________
Camera (yellow plaque):______________

NORMAL
(Eye exam on or before age 5)

NOT NORMAL
(Eye Exam Soon)

Patient:______________________ Result: ☐ ☐
Phone: _________-_____________

Patient:______________________ Result: ☐ ☐
Phone: _________-_____________

Patient:______________________ Result: ☐ ☐
Phone: _________-_____________

Patient:______________________ Result: ☐ ☐
Phone: _________-_____________

Patient:______________________ Result: ☐ ☐
Phone: _________-_____________

Patient:______________________ Result: ☐ ☐
Phone: _________-_____________

Patient:______________________ Result: ☐ ☐
Phone: _________-_____________

Patient:______________________ Result: ☐ ☐
Phone: _________-_____________

Patient:______________________ Result: ☐ ☐
Phone: _________-_____________

Patient:______________________ Result: ☐ ☐
Phone: _________-_____________

Read by: ______________________________ Date:______________

ALASKA BLIND CHILD DISCOVERY

A cooperative charitable research project to photoscreen every rural pre-school Alaskan
A.B.C.D. Coordinating Center: 542 West Second Avenue, Anchorage 99501 -(907)276-1617
(800)270-1617 • fax (907)278-1705 • eyedoc@alaska.net

Mailed:

Follow-up

Alaska's ophthalmologists and optometrists were informed in 1996 of the ABCD study and that they were likely to see some children who had been notified of "abnormal" or "inconclusive" results. We implored them to notify the Coordinating Center of the outcome of their complete exam on any ABCD referred children.

After 3.5 years, volunteer research students compiled data on the patient report sheets. A list of individuals with "abnormal" or inconclusive" results was generated and a mail merge letter sent to the parents. This letter reminded parents that a photoscreen was done on a given date and that the results were not normal. We queried: had an exam been done?. by whom?, was the exam finding normal?, what vision problem was diagnosed? In addition, the parents were asked to select an adjective for their feelings on learning that the photoscreen was not normal and whether they would recommend ABCD to a friend.

Letters were sent to the eye doctors in ABCD screening hubs of Dillingham, Kodiak and Bethel as well as the Alaska Native Medical Center (ANMC) in Anchorage. Accompanying these confidential letters was a list of children who had yielded "abnormal" photograph interpretations.

RESULTS

From February 1996 (Fur Rendezvous) through June 1999, 4000 MTI® photoscreens were offered to Alaskan children as a part of the ABCD study (Figure 8). One parent declined to have his child photographed after completing the paperwork. There were 76 initial repeat photographs and 3 second repeat photographs leaving 3930 children who had MTI® photographs recorded. Parents successfully completed parts of the health and family history questionnaire on the ABCD datasheet. The question about the child's health generated the least answers (2940) while the most complete answers concerned strong glasses or eye problems in the mother (3498). The presence or absence of "Warning Signs" was reported by 3211 respondents. Partial or incomplete answers accounted for the remainder. Prior eye exam were reported by 5.9% of respondents.

The mean ± standard deviation age for 3666 children screened (birthdate correctly given) was 3.89±2.08 years with a range of 0.08 to 14.9 years. The distribution of age at screening by the various screening hubs is given in Figure 9. The influence of age on the MTI® interpretation is given in Table 1. By unpaired t-test, the "inconclusive" results were younger than the "positive" results ($t = 2.5$, $p=.01$) and were particularly younger than the "negative"

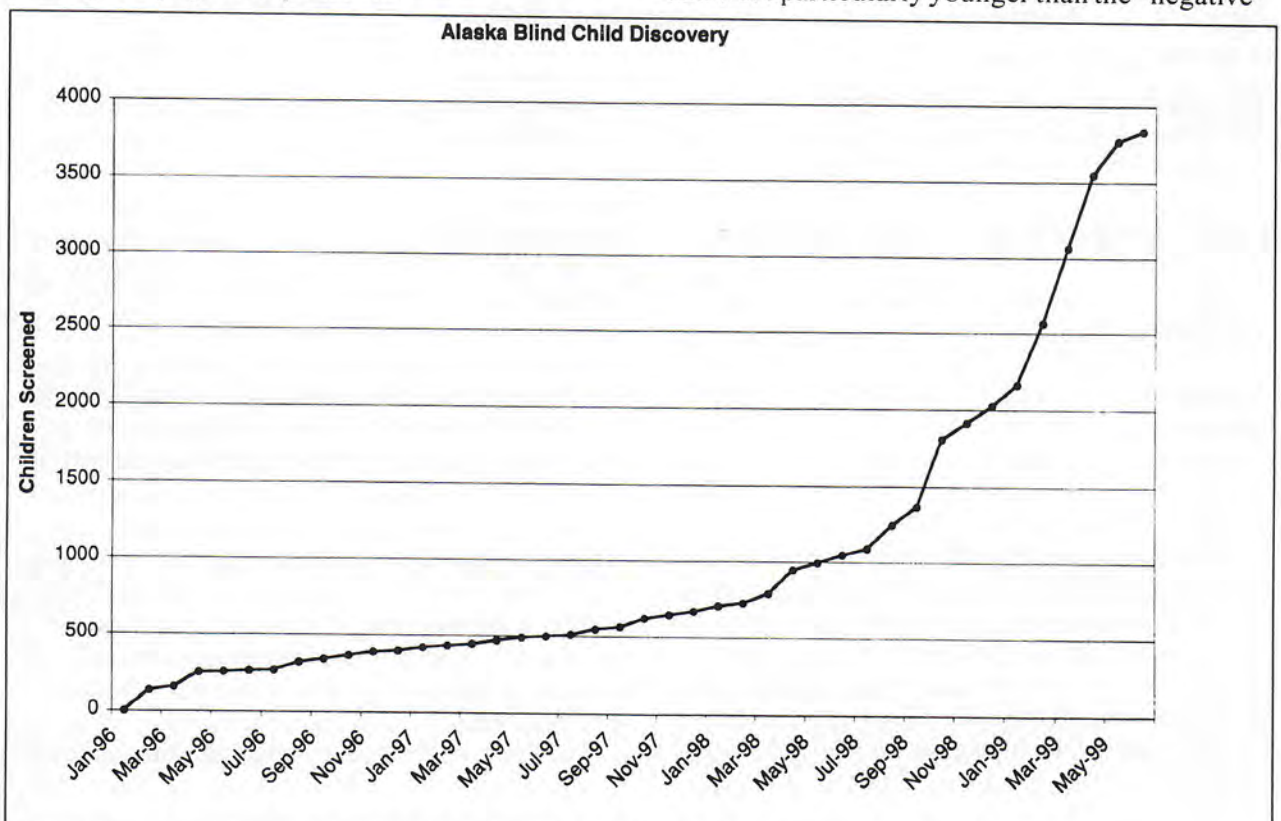

Figure 8. Accumulation of Cases by Alaska Blind Child Discovery 3.5 years

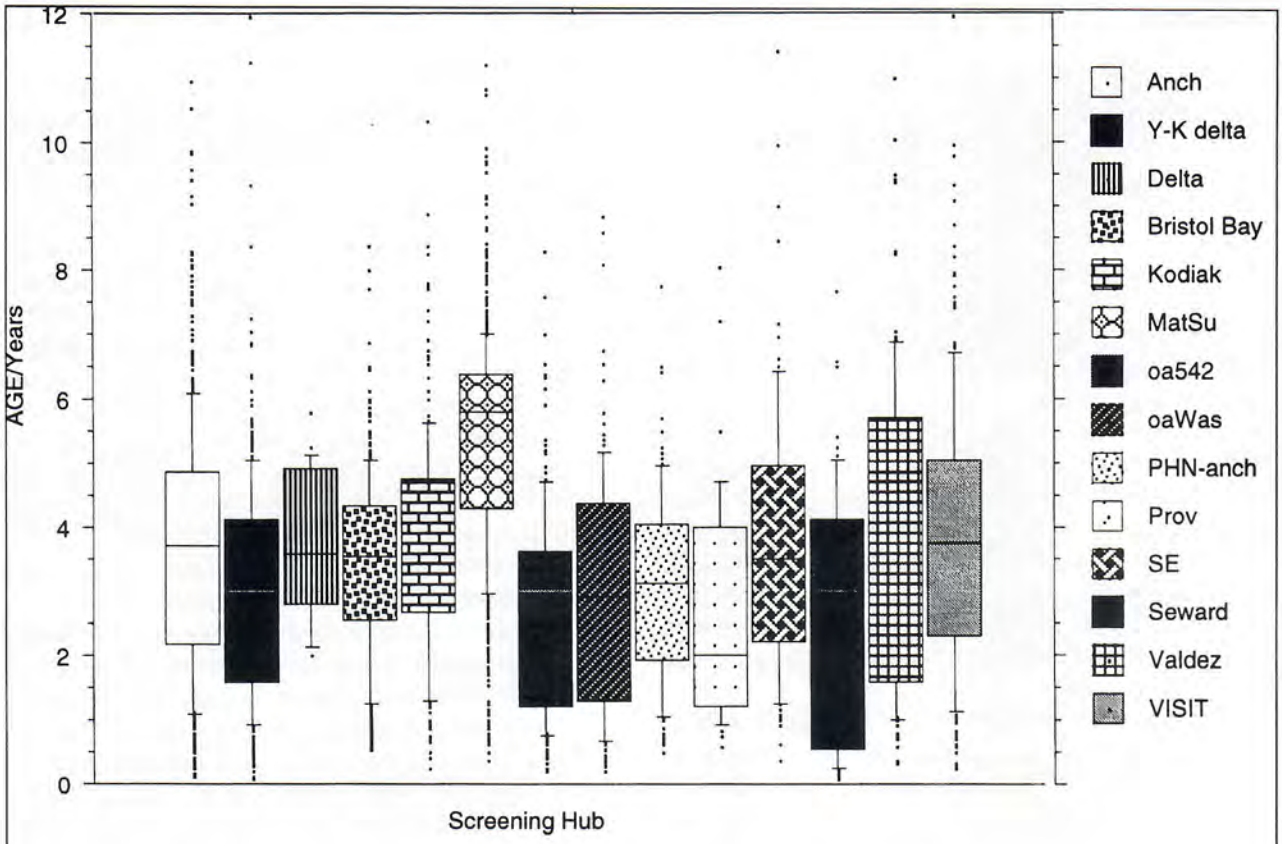

Figure 9. Distribution of age of children screened by ABCD screening hub.

results (t = 3.5, p=.0005). The "positive" results were younger than the "negative" results (t = 1.9, p=.055).

Table 1. Influence of Age on ABCD Interpretation

MTI interpretation	Mean age/years	Standard deviation	Count
"normal"	3.92	2.05	3289
"positive"	3.70	2.26	344
"inconclusive"	2.68	1.97	33
Total	3.89	2.08	3666

Table 2 gives chi Square results for the ABCD health/family questionnaire section comparing positive (+) and negative (-) questionnaire results with MTI® photograph interpretations (Y=abnormal, I= inconclusive and N= normal). Totals do not equal 4000 due to incomplete reporting by parents. Those questions with answers correlating with MTI interpretation were Prior Eye Exam, Child's health problem, Strong Glasses or Lazy eye in sibling, and the presence or absence of "Warning Signs."

The percentage of MTI interpretation and the total number screened for various ABCD hub screening centers is given in Table 3. Seward and Anchorage contributed the earliest screenings for which the interpreters were in their "learning curve." Numbers do not total 4000 due to incomplete reporting of screening site.

Follow-up data on "abnormal" results: Table 4 shows results of mailed surveys, phone calls, and actual exams on ABCD screenings which resulted in "Abnormal" results. Complete cycloplegic eye exams were considered abnormal if the following conditions were detected: amblyopia (acuity decrease of $\geq$2 lines), hyperopic anisometropia $\geq$1 diopter, astigmatism $\geq$2 diopters, tropia, hyperopia $\geq$4 diopters, cataract >1mm, anisocoria $\geq$1.5 mm, myopic anisometropia >4 diopters, ptosis, abnormalities of pupil shape, buphthalmos, myopia >8 diopters. Spherical equivalent (sphere + 0.5x cylinder) was used.

The Positive Predictive Value was extrapolated from a calculation by adding the survey, phone and exam results to determine actual Eye Problem / Abnormal Result in Table 5.

Table 2. The influence of affirmative answer to ABCD health, family ocular history and "Warning Signs" on photoscreening image interpretation.

Question	N/+	1/+	Y/+	N/-	1/-	Y/-	Chi Square p
Eye Exam	475	3	56	2501	12	133	<.0001
Health Problem	304	2	47	2395	17	175	<.0001
Siblings	446	4	49	2569	14	158	.0021
Mother	1140	5	86	2115	15	137	.36
Father	820	4	58	2411	15	164	.88
Relatives	1590	7	119	1580	12	94	.13
Warning Signs	130	0	30	2760	32	259	<.0001

Table 3. MTI® photoscreen interprtation percentages by screening hub.

Screening Center	% normal	% inconclusive	% not normal	Total number
Anchorage	84.07%	0.59%	15.34%	678
Y-K Delta	94.06%	0.48%	5.46%	421
Delta/Tok	95.46%	0%	4.54%	22
Bristol Bay	92.90%	0.59%	6.51%	338
Kodiak	94.05%	0.74%	5.20%	269
MatSu	92.36%	1.18%	6.46%	929
OA542	89.50%	0.55%	9.95%	181
OAwas	78.68%	0%	21.32%	136
PHN-Anch	92.55%	1.06%	6.38%	94
Prov	94.29%	0%	5.71%	35
South East	96.30%	0%	3.70%	108
Seward	76.00%	6.67%	17.33%	75
Valdez	90.27%	1.77%	7.97%	113
VISIT	92.13%	0%	7.87%	216
TOTAL	90.26%	0.83%	8.91%	3615

Table 4. Follow-up surveys, phone calls and complete eye examinations on ABCD "positive" results.

Survey	Bad address	17
Survey	No result given	12
Survey	Normal eyes	7
Survey	Eye problem	20
Phone	Normal Eyes	1
Phone	Eye Problem	1
Eye Exam	Normal Eyes	14
Eye Exam	Eye Problem	102
Other (incomplete info)	Not counted	10
Total Contacted		184

Table 5. The positive predictive value P.P.V. (extrapolated) for ABCD in the first two years, in the subsequent 18 month and overall.

Total P.P.V.	123/145	84.8%
1996-97 P.P.V.	53/69	77%
1998-99 P.P.V.	70/76	92%

Two other doctors sent notice of their findings in ABCD "abnormal" patients. To date, no information has been received from the eye doctors serving the ABCD remote screening hubs. The majority of "positive" result patients for whom we do not know the results of complete exam reside in the vicinity of the remote screening hubs. *Unfortunately, we do not know if the patients are not seeking follow-up, or if the doctors are not reporting.*

Parental survey results: We received survey results from parents of 64 "positive" screenings. The results for three queries are given in Table 6. The parents impressions of their children's eye exams agreed with our findings in all but one case. Most parents were initially worried or shocked at "positive" test results but were relieved to have detected a treatable condition after complete exam. One parent reported that she was "mad," and despite the initial and subsequent survey notifications, indicated that she did NOT intend to have the child examined.

CONCLUSION

Successes: MTI® photoscreening is a durable and portable technique which can be used by relatively inexperienced screeners to reliably detect amblyogenic refractive errors in toddlers. There is a long learning curve for MTI® interpretation, but a positive predictive value above 90% can be achieved. The ABCD program, using charitably purchased cameras, film, paperwork, and transport achieved a high penetrance in rural communities, especially when adopted by public health nurses and Infant Learning programs. Another large scale, Lions Club sponsored study is taking place in Tennessee with MTI® image interpretation performed by three specifically trained readers at Vanderbilt University(26).

Shortcomings: Despite general acceptance by pediatricians, public notification by the media, health fairs and free clinics, only about 5% of eligible preschool children in urban Anchorage were screened. ABCD had good penetrance in Dillingham, Prince William Sound, Kodiak and Bethel but has yet to make a significant dent in the Interior, Norton Sound, the North Slope and South East. The Polaroid film was relatively temperature and time sensitive, yielding partial exposures if the emulsion was cold, or half images if the batteries were old. ABCD paperwork, clinic-mailing, interpretation and then result-mailing often had a lag of a month or more between screening clinic and initiating a referral for the follow-up eye exam. Inconclusive results, often from lack of fixation or incomplete accommodation, was more common in very young toddlers or infants. Some screeners had a high percent of children directing their fixation on one or both of the images high above the camera. In November 1998, a letter was mailed to the screening hubs with a suggestion on how to hold the camera to reduce high fixation (Figure 11).

Implications for false negatives: Since we do not have data on complete eye exams of every child, we are at a loss to calculate false negative data. However, families living near the rural ABCD hubs are less likely to relocate outside Alaska or even their immediate borough. If a child whose ABCD photoscreen was interpreted as "normal" eventually develops fairly severe amblyopia, these children will eventually be detected in school acuity checks in kindergarten, first and second grades. Therefore, a careful survey of elementary schools in regions with prior high penetrance ABCD photoscreening will allow an approximation of false negative data and careful review of the Polaroid images associated with eventual vision loss.

Costs: The total cost to the children and parents of the 4000 Alaska Blind Child Discovery screenings and interpretations was $0.00. Follow-up on "not normal" or inconclusive" results motivated many parents to comply with the recommended complete eye exams. During 1996-1997, an estimated 37 children (23%) were false positives, and therefore were charged for eye exams with normal

Table 6. ABCD survey results: presence of eye disease, emotional reaction and notification and whether recommended to a friend.

Eye Disease? Survey/Exam	Number	Notification reaction	Number	Recommend for friend?	Number
+/+	16	Worry	21	Yes	53
-/+	1	Shock	13	Maybe	3
+/na	17	Mad	1	No	3
na/+	3	Concern	2	Blank	5
-/-	1	Sad	3		
-/na	7	Surprise	1		
blank/na	11	Expected	2		
		Relief	9		
		Joy	1		

Figure 11. Suggested photoscreener handling to reduce high-fixation or off-center fixation.

High Fixation

Correct Fixation

Table 7. Clinical tests of photoscreeners: sensitivites, specificities, PPV, inconclusive rate and prescreening probabilities.

Ref.	Technique	Population (prescreen prob)	Sensitivity	Specificity	PPV	Inconclusive
17	MTI	202 Children, 63% disease	87%	89%	93%	
18	MTI	1003, aged 6-59 mo, 20% disease	82%	91%	69%	3%
34	MTI	100, <3yrs, 74% disease	37%-88%	40%-88%		
35	MTI	100, 4 mo-12 yr, 46% disease	86 % (80 - 91%)	52% (20 - 67%)		3% (miosis)
33	MTI	45 mentally delayed	77%	70%		0
36	MTI	112 ?high risk, aged 6-48 months	83%	62%		1
37	EyeDx	206, peds, 0.7-16 years, 66% disease	89%	83%	91%	9
38	Vision Research	14,591 school-age	?	?	?	3.5%
39	Otago	1245 Kindergarten, 8% (3% est) disease	81%	98%	77%	

results. As the false positive rate falls with more accurate interpretation, follow-up exam costs are reduced accordingly.

Implications for early detection: Photoscreening in general and the ABCD program in particular must be evaluated in light of other childhood vision screening paradigms (Table 8). Congenital cataracts are best detected by careful Brückner or red reflex testing by experienced pediatricians. Large-angle strabismus is usually first noticed by parents, and then confirmed by pediatricians. Symptoms for several types of pediatric refractive error can be noticed by parents. Highly hyperopic children often present with symptoms and signs of progressive, intermittent esotropia if accommodation is adequate(27). Highly astigmatic children may avoid bright light or squint. Children

Table 8. Mechanisms of pediatric eye disease detection: symptomatic and vision screening techniques compared.

Vision Screen Technique	Age to be screened	Target disease
Parent recognition of signs and symptoms	Any	*Large angle strabismus*, myopia. *inflammation*
Red Reflex	Newborn	*Congenital cataract*
Fixation and cover test	3-4 months	Esotropia
Acuity	4 years	*Amblyopia*, myopia
Acuity/cover test	Kindergarten, 1 st grade	Refractive error, intermittent strabismus, amblyopia
Photoscreening	1-3 years	*Anisometropia*, high refractive error

with high myopia will move very close to view things. Anisometropic hyperopic children are often not symptomatic for parents and are not easily detected by pediatricians until a [non-cheating] acuity is eventually obtained. Isolated, small angle, constant strabismus is a relatively rare eye disease which is best detected by the Enhanced Brückner Test(10) and later with a careful acuity check.

Photoscreening has an excellent potential to detect almost all of the high spherical, astigmatic, and particularly the anisometropic refractive errors in children aged 6 months to 4 years. Thorough ABCD photoscreening will probably result in these children obtaining spectacle and/or patching therapy 1-4 years earlier than with symptomatic or school detection. It is difficult, and/or unethical (except in England (28,29)) to do a natural history study of early detected and then untreated high anisometropic hyperopia. Therefore, it is not known whether early treatment may influence emmetropization (a gradual refractive error trend toward plano). High anisometropia is associated with dense amblyopia, especially in the presence of small-angle strabismus(30) and aniseikonia(31).

Implications for early notification of "normal" ABCD: The text of ABCD paperwork and brochures emphasize that the preschool photoscreening is only part of a multi-pronged vision screening. The results of ABCD are photocopied on paper, the reverse side of which is printed with "Warning Signs" of children's eye disorders. We continue to urge school-aged acuity screening and/or complete eye exams. It is possible, however, for a parent to presume that the child with "normal" photoscreen results will develop normal vision and fail to pursue future eye exams or care.

In an environment of provided complete annual pediatric eye exams (a "Perk" of several managed care plans, Alaska Medicaid), a photoscreen "normal" result could be used to identify those children who do NOT need an eye exam that year. In a similar manner, photoscreening can be used to identify those for whom it is prudent to postpone eye exams in high risk and developmentally delayed populations(32,33).

Various techniques for objective pre-school vision screening have been recently reviewed(40). Some of the potentially portable devices are compared in Table 7 including the recent addition "EyeDx" (www.eyedx.com) which employs digital flash photography and on-site, synchronous computerized interpretation(37). Unfortunately, there is a lack of agreement about exactly which amblyogenic refractive errors, and/or anatomical factors qualify for an abnormal pediatric "gold standard" complete exam failure. Therefore, in addition to different prescreening probabilities in these studies, the sensitivities, specificities and PPV cannot be directly compared. To address these issues, a group of interested scientists and clinicians have formed the PESSG (Pediatric Eye Screening Study Group). PESSG aims to perform standardized evaluations of existing and new technologies with the aid of Pediatric Research in an Office Setting (PROS). The results from these studies will then allow health care policy makers to adjust implementation of wide-spread photoscreening. More studies on the natural history of pre-school amblyogenic factors, and the treatment success for each amblyogenic factor in current socio-economic climates will be required to know the exact cost-benefit of screening plus treatment.

ABCD recommendations for a preschool vision screening ideal: The intent of vision screening is

early detection allowing successful treatment/elimination of reversible childhood blindness(41). The most successful amblyopia reduction programs to date have utilized careful acuity checks(6) in cooperative children aged 4-5 years. Therefore, we emphasize the importance of visual acuity measurement at age 4, in kindergarten and first grades. Inventive children with a poor eye will often cheat on an acuity exam; the tester must have a high index of suspicion even after having completed 30 normal tests in a row. *One technique which enhances detection is to apply an occlusion patch over the non-tested eye before presenting the optotypes. Then the patch is placed over the other eye. Observation of the behavior of an amblyope with the better eye patched is often pathognomonic.* Pediatricians are to be encouraged to persist with careful red reflex testing of newborns, and fixation testing with cover test in young infants. We suspect that babies from home births and children obtaining non-allopathic care are at higher risk for non-detected or late detected congenital cataracts, essential infantile esotropia, congenital glaucoma and retinoblastoma. Parents are often able to detect strabismus and ocular inflammation before health professionals. Unequal pupils in bright or dim illumination are also noted by parents; anisocoria of $\geq$1.00 mm was detected in 6.6% of Ottar's screenings(18). The better educated the parents, the earlier and more appropriate the detection. It is for this reason that ABCD circulates pediatric eye "Warning Signs" twice for each screening.

Table 8 lists mechanisms by which pediatric eye disease may prompt appropriate care. The ideal screening paradigm would detect severe amblyogenic factors and serious eye disease objectively in very young children; newborn red reflex, experienced pediatric cover test at age 3-4 months and photoscreening between age 9-24 months. As soon as sensory testing is practical (age 3.5-4 years), acuity testing should be very carefully and systematically performed. Acuity checks should be repeated through age 8 (kindergarten, first and second grade). Parents should be educated about age-appropriate"Warning Signs."

We hope the efforts of ABCD allow early detection of treatable blindness in young Alaskans and provides data on practical application of this technology in some of the least accessible communities in North America. Additional efforts are aimed at calculating the cost of delivering such screenings to urban and remote communities.

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