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Bedside ROP screening and telemedicine interpretation integrated to a neonatal transport system: Economic aspects and return on investment analysis

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ABSTRACT

Background and aim: Peter Cerny Ambulance Service – Premature Eye Rescue Program (PCA-PERP) uses digital retinal imaging (DRI) with remote interpretation in bedside ROP screening, which has advantages over binocular indirect ophthalmoscopy (BIO) in screening of premature newborns. We aimed to demonstrate that PCA-PERP provides good value for the money and to model the cost ramifications of a similar newly launched system.
Methods: As DRI was demonstrated to have high diagnostic performance, only the costs of bedside DRI-based screening were compared to those of traditional transport and BIO-based screening (cost-minimization analysis). The total costs of investment and maintenance were analyzed with micro-costing method. A ten-year analysis time-horizon and service provider's perspective were applied.
Results: From the launch of PCA-PERP up to the end of 2014, 3722 bedside examinations were performed in the PCA covered central region of Hungary. From 2009 to 2014, PCA-PERP saved 92,248 km and 3633 staff working hours, with an annual nominal cost-savings ranging from 17,435 to 35,140 Euro. The net present value was 127,847 Euro at the end of 2014, with a payback period of 4.1 years and an internal rate of return of 20.8%. Our model presented the NPVs of different scenarios with different initial investments, annual number of transports and average transport distances.
Conclusions: PCA-PERP as bedside screening with remote interpretation, when compared to a transport-based screening with BIO, produced better cost-savings from the perspective of the service provider and provided a return on initial investment within five years after the project initiation.

1. Introduction

1.1. Screening for retinopathy of prematurity (ROP)

Retinal vessels develop relatively late during fetal development and the vascularization is completed by a gestational age of 36 to 40 weeks; the nasal retina develops earlier, and the temporal retina develops later [1,2]. Several factors, such as hypoxia, hyperoxia, variations in blood pressure, or acidosis can interrupt the normal process [2,3]. Due to the unique dynamics of retinal vascular development, ROP rarely emerges before 31 weeks of gestation and its progression stops after 44 weeks post-conception. ROP frequency essentially depends on the gestational age or birth weight: there is a 90% chance of ROP for weights below 500 to 750 g, a 78% chance between 750 g to 1000 g, and a 42% to 47% chance below 1000 or 1500 g. Its most serious forms develop in babies born below 1500 g or before 31 weeks [2,4]. Among Swedish babies with a gestational age < 32 weeks, the cumulative incidence of any ROP was 24.1% (including 8.5% of severe forms) in the screening period of the babies [5].

Most of the long-term consequences, such as retinal detachment and blindness, strabismus, refractive disorders, cataracts, glaucoma, loss of peripheral visual field and shrinkage of the eye [2,6,7], are irreversible and deteriorate the patient's health-related quality of life. Direct and indirect costs (such as costs of healthcare, productivity loss, loss of well-being, etc.) represent a substantial social economic burden in the first four decades of life [8].

Abbreviations: BIO, binocular indirect ophthalmoscopy; DRI, digital retinal imaging; km, kilometer; ROP, retinopathy of prematurity; NICU, neonatal intensive care unit; NPV, net present value; PCA, Peter Cerny Ambulance Service; PERP, Premature Eye Rescue Program.

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Recognizing and identifying the stage progression of ROP in time is essential to ensure the best possible outcomes: this is why adequate screening plays a central role in the ROP management guidelines [4,9–11]. However, these guidelines are slightly different in several ways: (i) the gestational age and/or birth weight limits that define the population to be screened; (ii) the postnatal age when the screening has to be initiated; and (iii) the follow-up interval between two screening examinations. The Hungarian Guideline [4] defines the population to be screened by <32 weeks of gestation or birth weight ≤ 1500 g; the screening initiation is at 4 weeks postnatal age but not earlier than 31 weeks post-conception, and the follow-up intervals range from one to four weeks.

The standard method of ROP screening is binocular indirect ophthalmoscopy (BIO). After instilling mydriatic eye drops to dilate pupils, the examiner visualizes the posterior pole and nasal and temporal peripheral area of the fundus. The new technology of digital retinal imaging (DRI) instrument uses a camera to take and a computer to record the images of the fundus. The retina cameras can be classified as narrow-angle or wide-angle cameras. DRI has several advantages over binocular indirect ophthalmoscopy (BIO), as DRI makes it possible: (i) for a nurse trained in retinal imaging to perform the bedside examination; (ii) for a remote ophthalmologist to interpret the images; and (iii) for images to be archived for further interpreting, documentation, or use in medical teaching, etc. Consequently, DRI with remote interpretation can decrease the need for transport (*neonatological benefit*), can decrease the workload of ophthalmologists so less ophthalmologists are able to meet the ROP screening requirements of a given population (*health system benefit*), and DRI can save images, which allows for the retrieval of images for quality control, patient follow-up, scientific analysis or even legal issues (*documentation benefit*). The guidelines referred to above [4,9–11] do not show a preference for either screening method, but the UK Guidelines acknowledge that DRI (RetCam) is useful and baby-friendly, and the USA Guidelines state that DRI has some benefit, e.g. in objective documentation. Based on a literature review, a recent joint technical report [12] concluded that telemedicine-based remote DRI did not supplant BIO, but evidence of moderate quality supported the use of the former in identifying patients with clinically significant or referral-warranted ROP [13]. It was shown [14] that DRI resulted in significantly lower stress-related heart-rate and respiratory rate responses than conventional BIO. The wide-angle cameras compared with narrow-angle cameras provide a greater view of the retina but are more expensive and less portable [15].

Retinal imaging and remote interpretation can be classified by several aspects [16] such as (i) what angle of view the camera uses (wide or narrow), (ii) who performs the examinations (a qualified nurse or an ophthalmologist), (ii) who interprets the images (ophthalmologist or pediatric ophthalmologist), (iv) what patients are covered by the screening program (all infants or ROP cases only), and (v) how many examinations are performed per patient (single or repeated).

The diagnostic performance, i.e., the sensitivity and specificity of DRI, found in some published studies are summarized in Table 1. The results show that DRI has good or even excellent diagnostic accuracy with high sensitivity, especially in those screening outputs that are used in the PCA-PERP screening program (see later).

1.2. Premature eye rescue program of the Peter Cerny Ambulance Service for Curing Sick Babies

Peter Cerny Ambulance (PCA) Service for Curing Sick Babies was founded in 1988 with the primary aim to ensure a special neonatal inter-hospital transport facility for premature or sick newborn babies and infants between referral hospitals and level III neonatal intensive care units (NICU III). PCA working as a neonatal emergency and transport service similar to a "mobile NICU III" covers the central region of Hungary (within a 120 km vicinity of Budapest) [17]. In addition, PCA performs inter-facility transport from NICUs to special examinations or interventions. Since it was launched in 1988, PCA has transported > 61.000 babies.

The *Premature Eye Rescue Program of the Peter Cerny Ambulance Service (PCA-PERP)* was established in 2008, and this program was based on the PCA's facilities, their logistic system, their highly qualified and trained staff (Neonatal Nurse Practitioners), and their skills and experiences accumulated over 25 years. Before the launch of PCA-PERP, bedside ophthalmologic screening of premature babies in the central region of Hungary could be ensured only in NICUs by local ophthalmologists. When local ophthalmologists asked for consultation, the premature baby had to be transported to a university ophthalmologic department by PCA. Moreover, some of these babies required more than one screening examination. Hence, the obvious drawbacks of this "transport-based" screening system were the significant burden on the babies with a potential risk of deterioration in cardiac, respiratory or neurologic status, and the significant burden on PCA as well, considering that the ambulance vans almost always ran "unoccupied" before and after transporting the babies.

The objectives of establishing the PCA-PERP have been to reduce the burden on these extremely vulnerable babies by decreasing their transport needs and to optimize PCA daily transport services by decreasing "empty" transport vehicle running time. The bedside retinal imaging is

Table 1
Some characteristics of studies investigating the diagnostic performance of digital retinal imaging (DRI) in ROP (ND: no data).

Ref	Number Of infants screened	Gestational age	Outcome Of ROP screening with DRI	Sensitivity	Specificity
[13]	36	Range: 23–33 weeks	Referral-warranted ROP (ROP in zone 1, plus disease or any stage 3 ROP)	100%	96%
[19]	27	Range: 28–36 weeks	Any ROP	85.71%	91.66%
[20]	64	Range: 23–32 weeks	Any ROP	81.6%–86.4%* (*ranges of the values of three image readers)	49.3–95.5%* (*ranges of the values of three image readers)
			Treatment-requiring ROP	85.0–90.0%* (*ranges of the values of three image readers)	95.3%–97.3%* (*ranges of the values of three image readers)
[21]	67	Range: 23–33 weeks	Mild or worse ROP	72.9%–93.8%	89.3%–97%
			Type 2 pre-threshold or worse ROP	71.4%–85.7%	92.8%–96.9%
			Treatment-requiring ROP	ND	93.8%–100%
[22]	51	Median: 26.9 weeks (interquartile range: 2.43 weeks)	Clinically significant ROP (that required on-site examination by an ophthalmologist)	92%	37.2%
[23]	43	Range: 23–33 weeks	Pre-threshold and threshold ROP	100%	97.5%
[24,25]	After 18 months: 97 After 36 months: 230	Range: 25–35 weeks	Referral-warranted ROP (ETROP type 2 or greater, threshold disease, any plus disease, and any stage 4 or higher disease)	After 97 infants screened: 100% After 230 infants screened: 100%	after 97 infants screened: 98.9% after 230 infants screened: 99.5%
[27]	1257	Mean: 27 weeks (standard deviation 2.2)	Referral-warranted ROP (considering both eyes)	90%	87%