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SUPPLEMENT

Evolution of multiple disease screening in Keelung: a model for community involvement in health interventions?

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Objective Screening for cancer and other chronic conditions tends to be conducted in independent programmes; that is, screening for one disease at a time. The Keelung community-based multiple disease screening programme, developed in Keelung city, Taiwan, is a notable exception. Here, we report on the Keelung programme's ethos and development within the community, focusing on equity of delivery of the service, and community involvement. In addition, we present some preliminary cost-effectiveness analyses of multiple disease screening.

Methods The Keelung programme offers screening for breast, colorectal and liver cancers, cervical and oral neoplasia, all of which have an evidence base for their efficacy, and for diabetes, hypertension, osteoporosis and hyperlipidaemia, which are of unknown efficacy. We assessed variability of coverage rates of the Keelung Community-based Integrated Screening (KCIS) programme with age and socioeconomic status, availability of facilities for referral of positive screenees, and numbers of community social workers, general practitioners and local hospitals involved in the programme. We also assessed in qualitative terms how the programme interacts with non-health agencies. Finally, we simulated activities and costs for a variety of single- and multiple-disease screening situations.

Results Between 1999 and 2003, coverage increased overall from 14.7 to 34.4%, and increased most dramatically in people aged 60–79 years (from around 30 to 60%) and in those of lower educational status (from around 40 to 70%). There was a significant growth in the involvement of social workers and volunteers in the programme, and an increase in the availability of local diagnostic and care facilities for those screened positive. In addition, there was substantial involvement of non-health agencies in publicizing the programme. In the health economic simulations, compared with no screening, the extra costs to gain an additional life year were estimated as US\$667, \$608, \$4227 and \$4789 for multiple screening with 100% attendance, multiple screening with 70% attendance, single disease screening with 30% attendance at each programme (i.e. 74% attendance for at least one out of four programmes), respectively.

Conclusions The innovative design and outreach procedures of the KCIS have led to a growth in delivery of screening services to groups sometimes overlooked (equity), community involvement in health care (participation) and the use of non-health organizations for publicity and health education (collaboration). Simulation studies indicate that multiple disease screening may be more cost-effective than single disease screening.

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INTRODUCTION

s many developing countries in Asia are faced with increasing incidence of cancers and other chronic diseases that are amenable to early detection by screening, population-based mass screening for early detection of these diseases has become an important issue. The implementation of organized population-based screening in western countries is largely guided by the concept of evidence-based medicine, dependent on the results of randomized trials, as in screening for breast or colorectal cancers. Programmes tend to be aimed at individual cancers. For example, screening for breast cancer and screening for cervical neoplasia are usually carried out independent of each other, in terms of timing, setting and facilities. The provision of multiple disease screening services for more than one disease is not generally practised.

The Keelung project is an innovative programme of multidisease screening, including several cancers and several non-malignant chronic diseases, in the city of Keelung, Taiwan. ¹ It was developed in line with the ethos of primary health care in the World Health Organization (WHO) Alma-Ata declaration. ² The latter statement emphasizes equity (delivery of service to all parts of the community), participation (having the community take responsibility by involvement of community members and volunteers in health-care delivery) and collaboration (involvement of non-medical agencies in health-care promotion or delivery). In this paper, we assess these features of the Keelung programme.

The first results of the Keelung programme have already been published. These published results do not include an economic analysis. From the cost-effectiveness viewpoint, several potential benefits may be derived from multiple screening services. There may be a beneficial effect on attendance rates. The multiple screening programme in Keelung, Taiwan, was accompanied by an increased attendance for cervical screening. Second, there may be economies of scale in administration and service provision, and additionally from the fact that some diseases are strongly associated with each other, for example, colorectal

cancer and hypertension. Third, because some chronic diseases are associated with certain cancers, multiple screening gives an opportunity to offer primary health education for reducing morbidities of chronic diseases, which in turn leads to morbidity or mortality reductions for specific cancers. The major disadvantage of multiple screening is the potentially high initial cost of early investment in a series of multiple activities with the benefit only being observed some years later. We therefore also report on a simulation study of costs and effects in multidisease and single disease screening in this paper, using a subgroup of the diseases screened for in the Keelung programme, as a first step towards a full economic analysis of the Keelung programme.

METHODS

The Keelung programme was originally designed to screen for oral (visual examination) and cervical (Pap smear) neoplasia, breast cancer (risk assessment followed by mammography if appropriate), colorectal cancer faecal occult blood test [FOBT], liver cancer (risk assessment followed by ultrasound if appropriate), Type 2 diabetes (fasting glucose), hypertension (blood pressure test) and hyperlipidaemia (total cholesterol, high-density lipoprotein [HDL] and low-density lipoprotein [LDL] testing). Recently, osteoporosis (heel ultrasound densitometry) was added to the screening remit. For the cancers and premalignancies screened for, there is an evidence base for efficacy.^{3–7} For the non-malignant chronic diseases, there is relatively little direct evidence of efficacy in preventing mortality or morbidity, but there is concern in Taiwan about their prevalence. Around one-third of the population had at least one of the non-malignant chronic diseases screened for. 1 Subjects in the population list were invited to attend for screening through invitation letter or telephone call. Each screening activity was carried out in cooperation with local health authorities. A series of stepwise multiple screening activities were conducted, including registration, fasting, blood-drawing, physiological measurement (including blood pressure, height, weight, waist-hip ratio), questionnaire administration, FOBT, physical examination of the breasts, Papanicolaou (pap) smear, oral examination and health education. The contents of the questionnaire consist of demographic characteristics, life-style variables, family history of cancer and chronic disease, personal history of disease and reproductive factors. The questionnaire responses were used to determine whether further referral to regular mammography is indicated. The responses also contributed to the decision process (along with a number of serological measures) as to whether liver ultrasound was required.

The target population consisted of residents aged over 30 years in Keelung, the most northern county of Taiwan. After excluding subjects registered in Keelung but residing outside Keelung, approximately 138,420 residents were listed as the target population. It was expected to take six years to achieve 100% invitation of the target population to multiple disease screening. We assessed whether coverage with screening (rather than invitation) was improving with time, particularly in lower socioeconomic groups and old people. We used educational level as our measure of socioeconomic status.

The KCIS programme also provides outreaching referral services such as the ambulatory sonographic service and organized referral systems for colonoscopic, mammographic

and coloposcopic examinations. The KCIS also aims to facilitate the evaluation of treatment of type 2 diabetes, particularly asymptomatic type 2 diabetes, in primary care. We therefore investigated the changes in patterns of referral and treatment over time.

We also considered the community aspect of participation: to what extent has KCIS activated people in the community to take part in the health promotion activity? In particular, we assessed the changes over time in participation of volunteers and non-health professionals, in particular, social workers and students. To disseminate information about the multiple screening programme, our project actively invited the press and media to get involved in the screening activity. This not only included the announcement of the screening time schedules but also critical comment on our activities. We also encouraged other major non-health agencies to contribute to publicizing the screening programme and to health promotion generally. We describe these activities in qualitative rather than quantitative terms below.

For our economic analysis, we included four cancers (colorectal, breast, cervical and oral cancer), for which there is a strong direct evidence base for screening. We also included two non-malignant chronic diseases, type 2 diabetes⁸ and hypertension, ^{9,10} for which there is at best some indirect evidence of a benefit of screening. We used Markov process models of disease progression with progression parameters estimated either from the Keelung project¹ or from the literature. 11-14 Parameters and costs included transition parameters for the disease, natural history for each disease, effects on these parameters of risk factors (e.g. betel quid chewing and progression of oral neoplasia), risk factor prevalence, survival, complication rates such as perforation in colonoscopy, associations between nonmalignant chronic diseases and cancers, screening costs, manpower costs, confirmation costs, treatment costs and average costs for terminal care.

Screening performance parameters such as sensitivity and specificity, and screening efficacy measures in terms of mortality, were derived from the literature review. ^{13–17} Full details of the parameters simulated are available from the authors (AMFY or THHC).

A total of 66,791 participants with the demographic characteristics in Keelung were simulated. Each participant in no screening (NS) followed the transition parameters from the disease natural history without interruption by early detection and treatment. For multiple screening (MS) and single screening (CS), each participant was simulated for a six-year screening programme with different interscreening intervals for different diseases. Details of the regimes are given by Chen *et al.* We simulated follow-up for 20 years or until 99 years of age. Costs incurred in US dollars and life years gained were estimated and the costs were discounted at 5% per year.

RESULTS

Table 1 shows the screening coverage rate increasing with the time after the introduction of the KCIS programme. There was some health screening before programme, but it reached only 6% of the population. By 2003, 34% of the target population had been screened. In absolute terms, the coverage increased most for those aged 50–79 years, and for those of low educational status. In these groups, the majority of the population is now covered.

The proportion of persons reimbursed for hospital health checks who also used the outreaching services increased

S56 Wang et al.

Table 1 Evolution of the equity indicators in the KCIS

Indicators	Before KCIS	1999	2003
1. Coverage rate (%)			
(A) Age 20–29		. –	
20–29	0.0	3.7	9.9
30–39	0.0	11.5	26.6
40–49	8.9	16.4	38.3
50–59	13.0	22.7	49.1
60-69 70-79	15.0 16.7	28.9 26.7	63.3
80+	11.8	17.3	66.3 19.8
Overall	6.0	14.7	34.4
Overdii	0.0	14./	34.4
(B) Educational levels	17.0	0.5.4	47.0
Illiterate	17.8	35.6	67.2
Literate	14.8	48.1	88.9
Elementary School	12.5	19.5	48.0 30.8
Junior high School	4.6 2.9	11.9 11.3	28. <i>7</i>
High School College	2.9	10.9	25. <i>4</i>
Post Graduate	1.6	14.7	35.2
1 Osi Ordabale	1.0	14.7	33.2
2. Proportion using outreaching (%)	5.9	15.5	20.8
3. Referral and care patterns			
(A) Diabetic Care	•	•	. _
Number of GPs providing diabetic care	0	26	47
(B) Sonography		0.10/ / 07/1010\	20 20/ /1574 /412 4)
Patients referred to local hospital services	_	2.1% (27/1312)	38.2% (1576/4124)
(C) Referral to colonoscopy Patients referred to local hospital services	_	7.8% (57/737)	100.0% (308/308)
·	V 11 11 %	, , ,	•
4. Financial Support	NHI*	NHI+subsidy from local government	NHI+subsidy from local government+ community-based education funding from
5. Clinics or hospitals involved	0	0	central government 7

^{*}National health insurance

Table 2 Manpower involved in the fieldwork of screening of 1999 and 2003

District	Social worker A	Professional staff B	Total manpower A+B	Professional/ social ratio B/A	Total screened C	Average served attendants per manpower C/(A+B)
1999						
Chung-chen	27	152	1 <i>7</i> 9	5.6	525	2.9
Chi-du	56	1 <i>7</i> 4	230	3.1	426	1.9
Nuannuan	34	13 <i>7</i>	1 <i>7</i> 1	4.0	412	2.4
Jen-ai	141	307	448	2.2	769	1 <i>.7</i>
Chungshan	47	182	229	3.9	867	3.8
Anula	78	162	240	2.1	563	2.3
Hsin-l	88	270	358	3.1	547	1.5
Total	471	1384	1855	2.9	4109	2.2
2003						
Chung-chen	188	490	678	2.6	3347	4.9
Chi-du	249	462	711	1.9	3250	4.6
Nuannuan	212	644	856	3.0	3654	4.3
Jen-ai	237	476	<i>7</i> 13	2.0	2906	4.1
Chungshan	396	539	935	1.4	4703	5.0
Anula	276	556	832	2.0	3489	4.2
Hsin-l	191	530	<i>7</i> 21	2.8	2468	3.4
Total	1 <i>74</i> 9	3697	5446	2.1	23817	4.4

during the years since the programme was introduced. Table 1 also shows how patterns of care in Keelung have changed since the programme was introduced. The number of general practitioners (rather than hospital departments) directly involved in diabetic care increased, and there was a shift in provision of sonography services to hospital rather than having the examination on the screening site. There was also a complete shift of colonoscopy referrals so that they are

now referred to local services in Keelung and not to central services in Taipei. Table 1 also shows the expansion of sources of financial support for the KCIS programme over time.

Keelung also developed a training programme for volunteer social workers to provide aspects of the service including questionnaire administration, blood pressure measurement, dental hygiene training, checking for periodontal disease and health education. Tables 2 and 3 show the

GP, general practitioner.

Table 3 Balance of volunteer sources over time – social workers and students

District	1999			2003			
	Social worker volunteers	Student volunteers	Total	Social worker volunteers	Student volunteers	Total	
Chung-chen	34 (100.0%)	0 (0.0%)	27	289 (86.8%)	34 (13.8%)	333	
Chi-du	27 (63.0%)	10 (37.0%)	56	212 (86.2%)	28 (11.1%)	246	
Nuannuan	88 (78.4%)	19 (21.6%)	34	187 (76.0%)	44 (13.2%)	246	
Jen-ai	141 (85.1%)	21 (14.9%)	141	230 (73.0%)	85 (27.0%)	315	
Chungshan	56 (100.0%)	0 (0.0%)	47	225 (88.9%)	41 (20.3%)	253	
Anula	47 (93.6%)	3 (6.4%)	<i>7</i> 8	161 (79.7%)	43 (16.1%)	202	
Hsin-l	78 (94.9%)	4 (5.1%)	88	224 (83.9%)	59 (24.0%)	267	
Total	471 (87.9%)	57 (12.1%)	471	1528 (82.1%)	334 (17.9%)	1862	

Table 4 Simulated results in terms of mortality by different screening regimes

	MS, 100%	MS, 70%	CS, 100%	CS, 30%	NS
Cancer death Number Relative risk	377.49 0.53 (0.47-0.60)	475.10 0.66 (0.59–0.75)	382.82 0.54 (0.47-0.61)	609.62 0.85 (0.77-0.95)	715.07
Chronic disease Number Relative risk	related or cancer death 5643.08 0.91 (0.87–0.94)	5837.64 0.94 (0.90-0.97)	5941.62 0.95 (0.92–0.99)	6134.01 0.98 (0.95–1.02)	6233.08

Table 5 Simulated results of incremental cost-effectiveness analyses (ICE) by different screening regimes

Mode	Cost	Effectiveness (life years)	ICE, by reference group			
			NS	MS, 70%	CS, 100%	CS, 30%
NS MS, 100% MS, 70% CS, 100% CS, 30%	151,290,562 154,139,608 153,177,299 163,882,998 156,177,986	849,871.29 854,143.59 852,975.20 852,850.39 850,891.85	 666.86 607.86 4226.93 4788.96	823.62 — Dominated Dominated	Dominate Dominate — Dominated	Dominate Dominate 3934.06

growth of community social workers and other volunteers. This suggested that the programme offers an opportunity to enable the community to be involved in health care. This had in turn enabled the social workers to develop health promotion models such as a lipid-reducing project, antiobesity programmes, anti-smoking programmes and promotion of healthy diet and exercise.

The programme also invited students in the Technological College of Jing-Gou, the only nursing school in Keelung to participate in the outreaching screening service. Student volunteers comprised 18% of the volunteer workers in 2003 (Table 3).

Since the programme was an outreaching service, it offered an opportunity to interact with community leaders and non-health agencies. Examples included the publicity of the programme on public buses in Keeling and on the local cable TV system. The latter not only provided the time schedule of the KCIS programme but also issued health education messages and critical comment on the activities of the programme.

Table 4 shows the results of the simulations with respect to mortality. For cancer deaths, both MS and CS with 100% attendance rate were estimated to confer a 45–50% mortality reduction from cancer. MS with 70% attendance was estimated to reduce deaths from cancer by 34% compared with NS. CS with 30% attendance independently for each of the four regimes (i.e. 76% attendance for at least one programme and 65% attending at least two) was estimated to confer a 15% reduction. The absolute numbers of cancer deaths estimated to be prevented were 240 for

MS with 70% attendance rate and 105 for CS with 30% for each screening regime. The corresponding numbers needed to invite were 279 for MS and 638 for CS. In multiple screening with 70% attendance, an additional 145 deaths are estimated to be prevented from causes related to diabetes and hypertension; however, these must be regarded as tentative estimates in view of the lesser evidence base for the efficacy of the non-malignant disease screening.

Compared with no screening, we estimated that the extra costs to gain an additional life year were US\$667, \$608, \$4227 and \$4789 for MS with 100% attendance rate, MS with 70% attendance rate, CS with 100% attendance rate and CS with 30% attendance rate, respectively (Table 5). Table 5 also suggests that in cost-effectiveness terms, MS dominated over CS in general.

DISCUSSION

The Keelung programme has demonstrated the development of certain important secondary preventive aspects of primary health care in terms of equity (delivery, participation and collaboration.) A key feature of these is integration of administration: different administrative institutions, not necessarily health-care institutions, cooperated in a unified effort for prevention of morbidity and mortality. There is internal integration between different divisions of the health authority, including, control and prevention of disease, medical and drug administration, health promotion,

S58 Wang et al.

nutrition and laboratory services. The external cooperation included the integration of health check-ups for women and elderly people conducted by social welfare organizations.

A major feature of the impact of the programme has been the delivery of services to elderly and deprived groups. Also, the programme provides medical and preventive services for two remote areas in Keelung, Ma-Lin and You-Rui, where there was previously a lack of any medical service.

A consequence of the programme is the community development of self-sufficiency for the implementation of secondary prevention and downstream health education and promotion activities. Within the secondary prevention programme, health promotion and education activities are also carried out to contribute to the primary prevention of cancer and chronic diseases. In addition, there is a possibility of improving both professional standards and public knowledge of the importance of high-quality tertiary prevention in rehabilitation and recovery of health. One example is the formation of a patient club for colorectal neoplasm patients. It is likely that this has contributed to a recent rise in compliance with referral to colonoscopy from 30 to 65%.

The economic analyses suggest that multiple disease screening can be more cost-effective than individual programmes. These results are to be regarded as suggestive rather than conclusive, since some of the non-malignant disease screening activities do not have proven efficacy. This does not mean that all the screening activities are necessary or that there is no room for further improving the programme. Although this is a community-based and service-oriented screening project, it also creates a study base for research. This includes epidemiological, health policy, medical management, occupational and environmental health research. There have also been experimental intervention studies and decision analyses. The current studies integrated into the Keelung programme include a population-based randomized trial for the evaluation of efficacy of toluidine blue staining for oral screening, the comparison of the efficacy between direct culdoscope and Pap smear screening, and prospective studies on epilepsy and Parkinson's disease. A particular point of interest for the future is whether screening for some of the non-malignant conditions can be replaced by a population intervention. For example, blood pressure is generally high in Taiwan, 1 and it is likely that almost all the population members would benefit from a reduction.18

There may be a role for legislation and regulation to facilitate the screening activities and their research sequelae. Legislation enables health personnel to conduct screening activities in the community without any barrier. The formation of a serum bank in conjunction with the screening would facilitate the development of biotechnology, but this probably requires a strong legislative framework to deal with ethical aspects and maintain the confidence of the public. Finally, social equity for high-risk groups with respect to cancer and chronic diseases may be facilitated through legislation to provide statutory services.

The programme activity may be criticized as duplicating the current policy of health check-ups for adults who have been reimbursed by national health insurance. However, the programme has successfully integrated the current strategy of health check-ups for adults into the model and demonstrated economic implementation in the community,

making full use of the community volunteers and infrastructure already present.

Conclusions

The above-mentioned data suggest that the Keelung model can achieve major goals of primary health care at an acceptable cost, and exemplifies the principles of equity, participation and collaboration.

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