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Aspects of economic costs and evaluation of health surveillance systems after a radiation accident with a focus on an ultrasound thyroid screening programme for children

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ABSTRACT

Health surveillance initiatives targeted at populations evacuated from, and residing in, areas affected by radiation contamination were implemented by international institutions as well as national and local governments after the nuclear accidents of Chernobyl and Fukushima Dai-ichi nuclear power plants. Most of these initiatives included a component of childhood thyroid cancer monitoring, with the more comprehensive schemes corresponding to national programmes of health monitoring for adults and children around general health and wellbeing. This article provides a short overview of available data on the costs and resources associated with surveillance responses to two recent nuclear accidents: Chernobyl and the Fukushima Dai-Ichi nuclear plant accidents. Moreover, because the balance of costs and benefits of health surveillance after a nuclear accident can influence decisions on implementation, we also present a brief overview of the principles of economic evaluation for collecting and presenting data on costs and outcomes of a surveillance programme after a nuclear accident. We apply these principles in a balance sheet analysis of a post-accident ultrasound thyroid screening programme for children.

1. Introduction

After the accidents at the Chernobyl and Fukushima Dai-ichi nuclear power plants, international institutions and local governments deployed a series of health surveillance initiatives targeted at populations evacuated from, and residing in, areas affected by radiation contamination. These initiatives typically included a component of childhood thyroid cancer monitoring along with monitoring of general health and wellbeing for adults and children. The outcomes of these surveillance programmes in terms of detected cancers, etc. have been extensively reported (Yamashita and Thomas, 2017); yet little has been reported about resources used and economic costs. This evidence gap is important given the substantial use of existing resources (e.g. medical infrastructure) and the new investments (e.g. specialist care centres, staff training) required for the implementation of surveillance programmes. At the same time, many of these health surveillance programmes have changed over time to curb unsustainable costs and to reflect new evidence on effectiveness. This illustrates the need for decision-makers to weigh the potential benefits of health surveillance against the associated costs in order to inform decisions on implementation.

An economic evaluation that considers the benefits, costs and potential harms of a health surveillance programme is only one of several dimensions decision-makers need to consider (Drummond et al., 1987). Nevertheless, an understanding of the costs alongside the benefits is an important step to establish what surveillance programme should be set up, and how that programme should evolve over time as circumstances change. An economic evaluation also sheds light on how benefits and costs are distributed across the population, i.e. who gains and loses out when resources are allocated to a surveillance programme (McIntosh et al., 1999). In this sense, the economic evaluation serves as an aid to decision-making (Public Health England, 2018; Hutubessy et al., 2003; Wilkinson et al., 2016), by presenting information that decision-makers can use in arriving to a decision. The economic evaluation of a health surveillance programme after a nuclear accident, however, presents challenges in assessing the outcomes and costs of health surveillance in the long-term, including determining the most appropriate economic measure of costs per outcome or outcomes of the surveillance programme.

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In light of these issues the objective of this study was twofold: first, to report available data on the economic costs and the resources used in providing the health surveillance systems adopted after the Chernobyl and Fukushima nuclear plant accidents; second, to illustrate the role of economic evaluation as an aid to decision-making, its advantages and the practicalities of economic evaluations of health surveillance programmes after a nuclear accident. We present a balance sheet analyses of a post-accident health surveillance programme. Given the importance given to childhood thyroid cancer screening in previous health surveillance initiatives, we focus on a routine ultrasound thyroid examinations surveillance system similar to that implemented in the Fukushima Health Management Survey (Yasumura, 2012). This work was part of the SHAMISEN (Nuclear Emergency Situations - Improvement of Medical And Health Surveillance) project under the Open Project for the European Radiation Research Area initiative (FP7 No. 604984) to develop comprehensive recommendations for health surveillance of populations affected by radiation accidents.

2. What is known about resource use and cost implications of long-term medical health surveillance programmes after a radiation accident

The economic burden of health surveillance systems varies with the depth and scope of the programmes implemented. The available evidence suggests that for the Fukushima and the Chernobyl accidents, the governmental and institutional health surveillance systems implemented as emergency and long-term responses to the catastrophes were very costly.

Systematic medical follow-up programmes and programmes providing regular health check-ups of affected or at risk populations after a nuclear accident are amongst the most costly health surveillance systems. For example, Belarus and Ukraine offered a wide range of health tests. They also offered additional compensation and benefit payments which were regulated by national laws, such as the 1991 “On legal regime of the territories affected by radioactive contamination as a result of the Chernobyl accident” in Belarus. A recent estimate of the cost of implementing this law in Belarus was BYR 474.5 billion (approximately €350 million) in 2010 and it was applicable to 1.14 million individuals in affected zones (National Report of the Republic of Belarus, 2011). Other reported costs include the BYR23.5 billion incurred between 2006 and 2010 (approximately €20 million) for the purchase of medical equipment required to monitor and treat the target population group and a further BYR22 billion (approximately €15.5 million in 2010 values) channelled from the republican budget for the purchase of medical equipment used in the national health surveillance programme (National Report of the Republic of Belarus, 2011).

In Fukushima, the Fukushima Health Management Survey (HMS) was implemented, which covered a range of health check-ups and tests for target population groups (children, pregnant women, evacuees), and in particular, follow-up ultrasound thyroid screening for children. The reported initial budget for the survey was 103.3 billion yen (€932.5 million), which included 78.2 billion yen (approximately €91.2 million) from the “Fukushima Resident Health Fund” with resources from the Ministry of Economy, Trade and Industry. Of this total budget, the costs for the first year of the survey were 10.1 billion yen (€91.2 million), with average annual cost of monitoring between 2012 and 2015 at 4.7 billion yen (€38.4 million) (Prefectural Oversight Committee, 2016).

The nature of these multi-component surveillance programmes following both the Fukushima and the Chernobyl accidents has resulted in substantial mobilization of physical resources. However, there is little information published on resource use for the different programmes. Some indication of the nature of resource use comes from the Ukrainian and Belarusian legislations, which dictate the types of tests and services that are made available to the different groups covered by the Chernobyl-specific legal regimes. Similar data also comes from the Fukushima Health Management Survey protocol. Table 1 below summarizes the type of resources used for health monitoring for children after the nuclear accidents of Chernobyl and Fukushima in the specific case of government-initiated and non-governmental but wide-ranging programmes.

3. Economic evaluation applied to health surveillance after a radiation accident: The case of paediatric ultrasound thyroid cancer screening

At low doses, the impact of radiation on thyroid cancer incidence is expected to be low and uncertain (Boice, 2012; Cardis, 2005). Outside of the context of post-nuclear accidents, active thyroid cancer surveillance for adults judged at low-risk has been reported as not being cost-effective (Wang, 2015). In the case of children, the prognosis of the disease is better than for adults, but the condition is more aggressive (Josefson and Zimmerman, 2008; Vaisman et al., 2011) and consequently screening might detect cases of disease at an earlier stage. This section presents an economic evaluation of a simple model of follow-up ultrasound thyroid cancer surveillance in children.

The economic evaluation described here is based upon a simple mathematical model. Models are widely used in economic evaluation of health care interventions, such as a health surveillance system after a nuclear accident. They are used to describe the cost and effects of a given surveillance system and compare these to an alternative surveillance system or no surveillance at all (Kaltenthaler, 2011; Manheim, 1998). The surveillance systems to be compared should ideally be informed by the opinion of experts and the preferences of public users of the surveillance system (Kaltenthaler, 2011; Scott, 2011). This is a fundamental principle in economic evaluations of health care interventions as it dictates the evidence demands for estimating costs and benefits, and ensures that the complexities of the system that is being evaluated are captured in the model (Kaltenthaler, 2011; Scott, 2011). At the same time, for both expert input and public opinion, caution is needed to prevent biases in the development of a model (Scott, 2011; Bowling and Ebrahim, 2001; Protheroe, 2012). More generally, the conceptualization of models for health resource allocation can be thought of as a two-step process: a first step is representation of the disease or health conditions, and a second step is the actual formalization of the model structure according to the specific purpose of the model and the level of detail needed (Roberts, 2012).

The present study used a decision-tree with rollback model structure (Brennan et al., 2006). The surveillance strategy modelled was informed by published literature, reports from Fukushima Prefecture on the thyroid ultrasound examination rounds (Prefectural Oversight Committee, 2016), and the protocol of the Fukushima Health Management Survey (Public Health England, 2018). The model was extended beyond the surveillance protocols mentioned above to include both screening/surveillance activities and response/treatment activities (Drummond et al., 1987; Herida et al., 2016; World Health Organization, 2005). The comparator for the analyses was a “no thyroid screening” scenario, which we also referred to as an ‘opportunistic case finding’ alternative, i.e. detection of cases through primary care presentations. The model for this latter alternative was based upon guidelines for paediatric thyroid management (Mazzaferri and Sipos, 2008; Pacini and De Groot, 2014). The difference between the ultrasound screening alternative and the primary care case finding alternative is whether cancers are detected and the stage at which cases are detected (stage/severity). A graphical representation and more detail on the model structure is available in the online supplementary material. The model assumed that a follow-up ultrasound surveillance was offered to a cohort of children who were aged from 0 to 18 years at the time of the accident. As in the Fukushima context, participation was modelled as voluntary but strongly encouraged and assumed to reach 70% of the target population. The target population was assumed to be for a low dose scenario, with an average thyroid dose exposure assumed to range between 10 mGy and 60 mGy similar to the range of exposures for evacuees and inhabitants of the

Table 1
Summary of the type of resources used in child thyroid cancer surveillance and medical follow-up in Belarus and Ukraine, and Fukushima.

Programme	Resources
Belarus (2001–2010) Medical surveillance according to population groups and risk. As of 2008, children less than 1 year of age were seen twice a year by specialist, and annual check-ups provided for children of school age from contaminated areas. At the beginning of 2010, there were 1,137,253 adults and 279,271 children and adolescents receiving periodic medical examinations (National Report of the Republic of Belarus, 2011).	Periodic health check-ups (National Report of the Republic of Belarus, 2011) • Therapist or paediatrician examination. • Endocrinologist examination. • Erythrocyte sedimentation rate (ESR), leucocytes and haemoglobin tests. • Dosimetry control examination (Cs-1370 exposure dose using whole-body counters) • Ultrasound scan of thyroid gland. • Puncture biopsy for those with nodular pathology less than 1 cm and those with abnormalities of the ultrasound scan with additional lymphadenopathy. • Thyroxin and thyrotrophic hormones (pre-natal and up to 3-years), or those with 100 Gy per gland.
Fukushima follow-up ultrasound thyroid examinations for children The preliminary/first-round of the thyroid examination survey (baseline) began on 9 October 2011 and was completed at the end of March 2014. The first full-scale survey began in April 2014 and finished at the end of March 2016. The second full-scale survey was programmed for May 2016 to April 2017. Between 2016 and 2017, the screening programme surveyed 336,609 children between the age of 0 and 18 years (Prefectural Oversight Committee, 2016; Suzuki, 2016).Responsibility for screening was with Fukushima Prefecture, whereas treatment plans were the responsibility of hospitals (Prefectural Oversight Committee, 2016).	Ultrasound screening (Suzuki, 2016) • Ultrasound equipment. GE 7.75-MHz probes (12L-RS linear array transducer) and LOGIQ Expert ultrasound (GE) with a 12RS linear probe (10–12 MHz) and the Noblus (Hitachi-Aloka co.) with a 12 MHz linear probe. • Trained ultrasonographer. • Specialist employed in retrospectively reviewing and categorizing the ultrasound images (endocrine surgeons, thyroid surgeons, ultrasonic medicine professionals). Suspicious images retrospectively reviewed twice by specialist. Confirmatory screening (Suzuki, 2016) • Precision ultrasound (18-MHz or higher frequency device) • Specialist physician consultation. • Fine needle aspiration biopsy (FNAB) of suspicious lesions. • Blood tests (fT3, fT4, TSH, and thyroglobulin concentrations, as well as TgAb and TPOAb) (Suzuki, 2015). • Urine sample (Suzuki, 2015).
Chernobyl Sasakawa Health And Medical Cooperation Project Five year program to examine children aged 10 years or less at the time of the accident in Kiev and Zhitomir (Ukraine), Gomel and Mogilev (Belarus) and Bryansk (Russian Federation). A total of 160,000 children were surveyed (Shigematsu, 2002).	Mobile case detection (Shigematsu, 2002) • 10 × 5 Mobile examination units (equipped with ultrasound equipment, hemoanalyzer, whole-body counter, etc.). • 10 × 5 passenger busses for the transport of children. Screening infrastructure (Shigematsu, 2002) • 5 fixed examination centres (equipped with ultrasound equipment, hemoanalyzer, whole-body counter, etc.). Thyroid and dosimetry examinations (Foundation, 1994) • FNAB of suspicious lesions. • Blood tests (serum free thyroxine T4, thyroid-stimulating hormone TSH, titers of anti-microsome antibody AMC, antiglobulin antibody ATG). • Dosimetry control examination (Cs-137 exposure dose using whole-body counters). • Ultrasound scan of thyroid gland. Staff training and community education (Shigematsu, 2002) • 310 experts, over 90 visits to project centres. • Training for examination centre staff (130 staff). • Production of educational materials for residents of the affected area.
BelAm (1997–2008) and UkrAm cohorts (1998–2007) Cohort studies of thyroid cancer and other thyroid diseases. Four biannual screening rounds covering around 13,000 and 11,000 children in Ukraine and Belarus, respectively, who had had the radioactivity of the thyroid gland measured in 1986 shortly after the accident (Stezhko, 2004). Screening organized into mobile screening teams and fixed screening sites (two locations in Belarus and two locations in Ukraine). Focus of the investigations was not exclusively thyroid cancer but thyroid function more generally, the protocol for the study included tests for thyroid function (Stezhko, 2004).	Screening (Stezhko, 2004) • Phlebotomist appointment. • Blood test for thyroid-stimulating hormone (TSH) levels.* • Free-thyroxine assay performed when TSH abnormal.* • Serum antithyroid peroxidase (antiTPO), antithyroglobulin (antiTG) and thyroglobulin (TG).* • Serum-ionized calcium (parathyroid disfunction). • Spot urine sample.* • Ultrasound scan of thyroid gland. • FNAB of suspicious lesions.

*These tests were excluded from the protocol in the last round of the Ukraine-American (UkrAm) cohort follow-up.

most affected areas after the Fukushima Dai-ichi nuclear plant accident. The activities included in the model and the associated costs followed a service-provider perspective (i.e. a healthcare perspective), which is concerned mainly with costs of running the service as opposed to a national government perspective, where societal benefits and costs might also be considered (World Health Organization, 2005).

More generally, irrespective of the perspective adopted, the reliability and usefulness of an economic evaluation requires a technically rigorous costing exercise that allows for a credible comparison of costs alongside the benefits of a health service (McIntosh et al., 1999). From an economic point of view, costs can include direct costs (medical or non-medical), indirect costs such as lost wages due to illness, and intangible costs or harms which do not have an easily estimable monetary value. These intangible costs can, for example, be those associated with the pain and discomfort caused by treatment (Kielhorn, 2000) and the broader impacts of anxiety and reasurrance to populations undergoing health surveillance. For costs that can be monetized, only those areas of resource use where change occurs as the result of the health surveillance programme should be included in the economic evaluation (McIntosh et al., 1999).

Resource use in the model developed here included an appointment with a specialist consultant, ultrasound scans (to detect abnormalities), precision ultrasound (if an abnormality is detected), blood tests, fine needle aspiration biopsy (FNAB) (to confirm cases), and surgical excision of a tumour, postoperative follow-up and treatments for recurrence and progression (all of which are labelled as treatment activities). Other activities associated with surveillance case registration, reporting, data analysis and feedback, training and supervision were not costed because of lack of available data or because they would be a relatively small component of overall costs. Nevertheless, they are important considerations when the infrastructure needs to be created as part of the surveillance strategy (McNabb, 2002). The model excluded non-medical direct costs (e.g. costs to children and carers of attending follow-up appointments and treatment consultations) and indirect cost (e.g. lost wages due to illness). These costs were excluded as the model took the service-provider perspective (see above) and these costs are not borne by the service-provider. Potential harms from the ultrasound thyroid examinations surveillance for which it is difficult to assign a monetary value (see below) were included in the model qualitatively, which is an advantage of the balance sheet approach (Kielhorn, 2000).

Unit costs for the resources used in the activities of the surveillance programme (screening, confirmatory screening, surgery, recurrence and follow-up) are summarized in the online supplementary material. Cost for specialist consultations, ultrasound scans, FNAB, chest x-ray, urine test, cytopathology and histology were sourced from 2014 to 2015 UK National Health Service National Schedule of Reference Costs. The cost of Levothyroxine (thyroid hormone treatment) was taken from the Medicines Complete pharmaceuticals database of the Royal Pharmaceutical Society, based on a 1.6 μg/kg dose (Mistry, 2011) taken over the duration of the individual's life under the screening programme (55 years, annual discount rate 3.5%). Other costs were sourced from the literature. Monetary costs were expressed as 2016 Euros using purchasing power parity conversion rates to account for different currencies used in different studies, (http://eppi.ioe.ac.uk/costconversion/default.aspx, online supplementary material).

In addition to estimating the level of resource use and their cost, information on the short and long-term health consequences associated to a nuclear accident, and the likelihood of these consequences, is a crucial part of an economic evaluation of surveillance programmes. Benefits from a post-accident health surveillance programme can include measures of the effectiveness of the programme for affecting expected health outcomes, such as the number of disease cases detected and deaths averted. Table 2 presents a basic list of data pertaining to the effectiveness of health surveillance systems that apply to post-nuclear accident situations. In this sense, `diagnostic specificity' and `diagnostic sensitivity' of the surveillance methods used are included to capture how well these methods provide reassurance that cases that need to be identified and further investigated can be detected as early as possible. In many instances, these data can be sourced from epidemiological studies and randomized controlled trials, as well as grey literature reporting on previous accidents and on similar accident situations (World Health Organization, 2005). Importantly, the final measures of effectiveness and benefit of health surveillance programmes included in an evaluation are chosen in line with the principle of developing informed models for economic evaluation. As such, typical measures of the effects of health surveillance systems such as deaths avoided as a result of surveillance (Herida et al., 2016) were not applicable to this model as childhood thyroid cancer has very low disease-specific mortality rates (Hay, 2010; Ji, 2008; Tuttle et al., 2011). Measures of effectiveness therefore included cases detected and cancer recurrence rates.

Table 2
Example of variables describing the effectiveness of a health surveillance programme used to inform an economic evaluation.

Epidemiological studies and public health data	
	Disease incidence
	Disease specific mortality risk
	All-cause mortality rate
	Screening uptake rates
	Detection rates
Natural history of disease	
	Probability of progression to different disease states
Clinical outcomes	
	Measures of sensitivity and specificity of diagnostic tests
	Probability of recurrence after treatment
	Probability of complications from treatment and adverse events

The number of cases detected are routinely reported as part of monitoring in surveillance systems, but these must be interpreted with caution given the possibility of screening or surveillance bias. This sort of bias relates to a seemingly observed rise in the outcome of interest solely because detection efforts are more intense and all that is detected are otherwise occult cases (Hautt and Pronovost, 2011). Efforts to detect thyroid cancer cases through screening may also reveal other thyroid conditions in the population. In Japan, cases of ectopic thymus, diffuse goiter, ultimobranchial body, lymph node swelling and thyroid agenesis were identified as part of examinations for thyroid cancer (Hayashida, 2013). These conditions were not believed to be caused by radiation exposure and were incidental findings, often with unclear clinical significance. Thus, as with thyroid cancer incidence, the potential for over diagnosis (with the attendant anxiety this causes) and overtreatment (with the attendant harms and adverse effects of treatment) is another effect included qualitatively in the model of ultrasound surveillance screening.

Although not captured in the current model, in addition to clinical measures of effectiveness, other effects from health surveillance after a nuclear accident are the psychological effects from having access to a system for monitoring disease, both in terms of reassurance from being followed-up and testing negative to the presence of disease, and anxiety from undergoing tests and from testing positive to a condition (Hiroko and Miyamoto, 2017). Considering these effects is part of principles of economic evaluation, which should aim to include all costs and all benefits no matter on whom they may fall. This would include any unintended consequences from surveillance (Craig et al., 2006) as well as satisfaction with the programme for those receiving follow-up. It is likely that for many decision-makers quality of life will also be an important measure of effectiveness. Measuring and valuing the psychological consequences of health monitoring is, however, difficult and these impacts have only been reported qualitatively (World Health Organization, 2005; OECD/NEA Expert Group, 2015; Radiation Protection Division, 2014).

Within an economic evaluation the outcomes and costs of a health surveillance programme can be compared through the frameworks of a cost-effectiveness, cost-utility and cost-benefit analyses. The choice between these frameworks should reflect the question that the decision-maker seeks to answer and the availability of good quality data to construct the measures of benefit and costs. To date, the majority of previous studies of economic evaluations of public health surveillance systems have used cost-effectiveness analysis (Wang, 2015; Herida et al., 2016), where costs are equated to a single clinical measure like cases detected or lives saved. Very few studies have undertaken a cost-utility or a cost-benefit analyses, partly because of difficulties in accessing appropriate data on benefits (e.g. cost-utility analyses requires data on both quality and quantity of life and reports measures such as quality adjusted life years, or disability adjusted life years (World Health Organization, 2005; Drummond et al., 1987). Cost-benefit analyses aim to directly compare costs and benefits by monetizing outcomes of an intervention). Among other reasons, the difficulties in assigning monetary values to outcomes of health programmes has led to the development of a more flexible form of economic evaluation known as the balance sheet approach. This approach seeks to identify and compare 'pros' (in terms of gains in health or wellbeing, and cost saving) and 'cons' (the harms and costs) of the surveillance system. It seeks to specify

Table 3
Balance sheet summary of outcomes and costs associated to paediatric ultrasound screening after a radiation accident.

'Pros': Outcomes or Savings Favour Screening	'Cons': Outcome or Costs Against Screening
80% of those with detectable disease^ are detected by ultrasound and between 80 and 85% with FNAB. Some of this disease will be clinically relevant. Early detection is relevant in terms of complications that come from later diagnosis (surgical and medical complications, adverse effects, remission, etc.). Reassurance provided to the 75–90% identified correctly as having no disease* using ultrasound, and 85% with FNAB This provides reassurance even when the risk of clinically relevant disease is lower than detection rates.	Higher expected cost per child of detecting thyroid cancer with universal ultrasound screening than by opportunistic case detection, estimated at €194 to €350 (70% participation in screening) to €32 to €24 (30% participation in screening). Potential for overtreatment from screening effect. Some estimates suggest that detection rates can be increased by as much as 30 times the baseline prevalence rate. ֍ False reassurance for those with disease (a smaller number of which might have clinically relevant disease) who are incorrectly identified as negative on screening. Anxiety and overtreatment for those children who are incorrectly identified as having disease. Short term consequences of radio iodine treatment include dry mouth and eyes and pain in salivary glands. Long-term consequences of thyroid cancer treatment include increased risk of kidney problems (treatment of hypothyroidism) and osteoporosis. Thyroxine replacement known to decrease experienced quality of life above and beyond the consequences of thyroid cancer.
No evidence in favour of or against surveillance	
Availability of screening and follow-up can serve to alleviate anxiety over the effects of radiation on health of the exposed populations. This may also mean, however, that once implemented, attempts to scale back monitoring can increase anxiety and stress of those affected. Whilst this anxiety and stress is real it may be based upon a distorted view of the risks faced. Those screened can also experience anxiety over test results and social anxiety from fear of discrimination both from receiving screening and from positive test findings. Universal screening can serve to detect thyroid malignant cancer at an earlier stage. In the case of the Fukushima cohort, at the point of FNAB testing, 20% of children showed suspicious and malignant lesions, compared to 60% of children in international retrospective hospital cohorts. Children in hospital cohorts would need to present with signs or symptoms suspicious of disease. The children from the Fukushima cohort can be thought of as generally being detected with sub-clinical disease. This disease may behave substantially different to that disease detected in the hospital cohorts and extrapolation from hospital cohort data may not be reliable. Therefore, more evidence on the impact of early detection on long-term outcomes is needed to support any decision to adopt screening for detection of subclinical disease†. Detection of other thyroid conditions during screening. There is the risk of over diagnosis and treatment of these conditions with the resulting additional monetary, physical and emotional burden to children and those providing care for them.	

^ Sensitivity defined as # true-positive/(# true-positive + # false-negative). *Specificity defined as # true-negative/(# true-negative + # false-positive). ֍ Based on Japanese prevalence rates between 2001 and 2010, published by Katanoda and Kamo (2016), Japanese Journal of Clinical Oncology, 1–3. †Francis et al. and American Thyroid Association Guidelines Task Force. (2015). Thyroid, 25(7), 716–759.

the direction of effect, measure that effect in natural units if possible and value those natural units if feasible (McIntosh et al., 1999; McIntosh et al., 1999). This method provides a more comprehensive description of features important to decision-making than the more commonly used cost-effectiveness analysis, predominant in previous studies of economic evaluations of health surveillance programmes [e.g. Wang, 2015, Herida et al., 2016]. This approach is applied in this study and results are presented in Table 3.

Taking into account the potential range of costs for detection and treatment resources (online supplementary material), the expected cost per child of a surveillance system involving ultrasound screening was estimated to range between €283 and €570 over the duration of the screening and treatment window (Table 3). This estimate assumed 70% of the target population is screened. Lower participation rates resulted in lower costs; at 50% participation rates the expected cost per child was estimated to lie between €202 and €407; at 30% participation, the expected cost per child was €121 to €244. At very low levels for the uptake of surveillance (not modelled) the cost per child would increase as substantial diseconomies of small scale arise (infrastructure has to be put in place but very few children use it). The expected cost per child of a surveillance system involving primary care presentations only (i.e. 'opportunistic case finding') ranged between €89 and €220.

The main drivers of the higher cost per child of the ultrasound surveillance alternative compared to the opportunistic case were the costs incurred over successive rounds of follow-up tests and the smaller number of children having to receive monitoring and treatment in the opportunistic case finding alternative. Sensitivity analyses (online supplementary material), however, showed that the efficiency of the ultrasound screening programme was sensitive to the participation rates achieved, as well as the number of children who would receive the more costly diagnostic procedures such as FNAB in the opportunistic case finding alternative.

Differences in the rate of detection between the ultrasound surveillance alternative and the opportunistic detection alternative were not estimated as part of the analyses. This is because it was difficult to determine from available data the magnitude of the potential screening bias associated with a thyroid cancer surveillance programme after a radiation accident. The necessary information to detect screening bias includes knowing the population prevalence rates of disease before and accident and these are not always available, even for comparable populations to those affected by the accident. Screening effects from surveillance can result in overtreatment arising from the detection of disease that has not yet progressed (and may never progress) to a clinically meaningful stage. The negative effect from overtreatment from a surveillance programme is greater in circumstances where a lower dose of radiation is received and hence where the level of exposure may not be sufficient to cause disease. Given this uncertainty, the justification of a screening programme for childhood thyroid cancer on the basis of early detection requires more evidence on the impact of early detection on long-term outcomes (Francis, 2015).

4. Discussion

The economic evaluation of health surveillance systems can be an important factor in informing the decisions about the nature and extent of health monitoring after a radiation accident. The evaluation performed here focused on the introduction of a paediatric thyroid ultrasound screening programme for those exposed to a low dose of radiation after a radiation accident. It illustrated the potential costs and impacts of undertaking a sophisticated ultrasound screening programme in a scenario where the target population has been exposed to low radiation doses. The result suggests that in such a scenario, the 'cons' to ultrasound thyroid screening are more evident than the benefits to such a programme. A more appropriate approach may therefore be to make available thyroid screening, with appropriate information provision and support, to those affected by the nuclear accident that request it, as proposed in the SHAMISEN recommendations for health surveillance after a radiation accident (Ougthon et al., 2017). Ultimately, however, implementation will depend on what decision-makers view as the most relevant potential benefit from screening.

The complexity of a surveillance system, and the very limited evidence available to inform a model of thyroid screening for children, however, required a number of simplifying assumptions that imposed limitations to the analyses. These need to be addressed in the future in order to strengthen the economic evidence to justify screening. The ability to conduct an informative economic evaluation of health surveillance systems after a radiation accident hinges on the availability of data resources to estimate effects and costs of the surveillance activities. The majority of studies comparing outcomes and costs of health surveillance programmes have taken a very limited view of what costs and benefits are relevant to consider. They have tended to use a single measure of clinical effectiveness as an outcome measure and considered costs falling on the service provider and, albeit less commonly, those falling on the patient (Wang, 2015; Herida et al., 2016). Types of direct medical resource use and costs from implementing a health surveillance system include consultations with clinicians, tests, medications and surgical procedures. Hospital patient care costs applicable to an economic evaluation of a health surveillance programme may be available from national databases that systematically record and report this information. Examples of cost databases include the UK National Health Service's National Schedule of Reference Costs[1], the DRG price estimates for hospital treatment in Germany[2], Spain's Hospital Costs from the National Health System ("Sistema Nacional de Salud")[3], and the RIVM Cost of Illness database 2013 from the Netherlands[4]. The World Health Organization (WHO) also collates information on costs of health-related activities relevant to a surveillance system through its WHO Global Health Observatory website[5]. In the absence of this type of information, active data collection over a period of time may be necessary. In addition to costs, measures of medical and non-medical service use may be available from linking existing health monitoring registries to health care use data resources or by embedding service utilisations questions in larger epidemiological studies [Craig et al., 2006 p. 29]. In other instances, the relevant data (e.g. type and number of inpatient and outpatient hospital services used, hours of carer time, etc.) would have to be collected.

At the same time, very few studies have undertaken a cost-utility or a cost-benefit analyses (World Health Organization, 2005; Husereau, 2013) that include other direct costs and indirect costs to surveillance participants and to other members of society that are directly or indirectly affected by the surveillance activities, the latter including for example those whose care suffers as health resources are redirected on to surveillance, or cuts to social housing provision or transport government spending required to fund the surveillance system.

Similarly, on the side of effectiveness of health surveillance systems, it is widely recognised that one of the potential effects of health surveillance for populations is the psychological impact (see the accompanying SHAMISEN paper on this special issue). One aspect can be the reassurance associated to regular monitoring. On the other hand, the perceived risks that have prompted surveillance may cause anxiety and distress. Whilst tools exist to measure and value these benefits and, in economics parlance, dis-benefits, they have not yet been used in the economic evaluations of public health surveillance systems. Further research is needed to measure and value these psychological effects of surveillance in a way that could be incorporated into economic evaluations of surveillance programmes, which should include the uncertainty surrounding the unpredictability of psychosocial responses to an accident (World Health Organization, 2005; OECD/NEA Expert Group, 2015; Radiation Protection Division, 2014).

[1] https://www.gov.uk/government/publications/nhs-reference-costs-2014-to-2015

[2] http://www.drg.german-hospital-service.com/

[3] https://www.msssi.gob.es/estadEstudios/estadisticas/inforRecopilaciones/anaDesarrolloGDR.htm

[4] https://www.costofillness.nl

[5] http://www.who.int/gho/en/

5. Conclusion

This study briefly summarized available data on costs and resource use of a series of health surveillance programmes carried out after the Chernobyl and Fukushima Dai-Ichi nuclear plant accidents and reports a simple economic evaluation modelling exercise of a hypothetic childhood ultrasound thyroid cancer screening programme. Post-accident health surveillance programmes were costly to implement for local and national governments. The economic evaluation of the childhood ultrasound thyroid screening program suggests that other approaches to thyroid health surveillance may be more effective and efficient in addressing the physical and mental health consequences after an accident. Our work highlights the role that an economic evaluation framework can have in organising and synthesising data on the benefits, costs and potential harms of a health surveillance programme. The work emphasises the need for further more rigorous evaluations on the effectiveness and efficiency of health surveillance regimens after a radiation accident. As any other method that purports to inform decision-making, it is bound by the availability of "hard" data on the subject. As new evidence continues to evolve regarding, for example, asymptomatic disease outcomes and magnitude of screening effects, the balance sheet of the type presented in this article would be updated and would illustrate the revised trade-off that decision-makers need to consider. In our case study there were gaps in the evidence on the clinical relevance of disease detected. No data were identified on the value to individuals, the public and society as a whole of the health and other impact of surveillance (or no surveillance) and on the costs falling on children and their families of accessing surveillance services, as well as on the longer term impact on their physical and human capital development. Overcoming these and other data limitations discussed here is important to understanding whether health surveillance strategies implemented after a nuclear accident are effective and efficient.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Disclaimer

Where authors are identified as personnel of the International Agency for Research on Cancer / World Health Organization (IARC/WHO), the authors alone are responsible for the views expressed in this article and they do not necessarily represent the decisions, policy or views of the IARC/WHO.

Supplementary material

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