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Clustering of anophthalmia and microphthalmia

No clustering has been found—but a link seems to exist with population density

Papers p 905

A few years ago public concern was raised in England by the appearance of apparent clusters of cases of anophthalmia and microphthalmia.[1] The pesticide benomyl, and later on its derivative carbendazim, was suspected to be the cause of the alleged clustering. In response to a press campaign the government in 1993 commissioned further research, although without clearly indicating the direction for the investigations. In this week's issue Dolk et al publish the results of that research (p 905).[2] Although they did not confirm clustering, their results raise further, interesting, questions.

Microphthalmia is a general term used to describe a broad range of improperly developed, small eyes in newborn children. One end of the range is marked by babies with complete absence of eyes—that is, anophthalmia—whereas at the other end are cases that are rather arbitrarily diagnosed because no clear cut border exists between mild microphthalmia and small normal eyes. Often the eye abnormality is part of a syndrome and is accompanied by other clinical features. Individual cases may differ widely in their cause. Specific genetic factors, such as chromosomal abnormalities and inherited mutations in developmental genes,[3] may form the underlying cause. Or the disorder may result from environmental influences on fetal development, such as exposure to certain infectious agents or teratogenic chemicals.[4-6] Cases without any obvious cause are generally ascribed to a combination of environmental factors and genetic susceptibility.

Three fundamental questions can be asked about the cases that occurred in England. Firstly, could benomyl cause the disorder? Secondly, is there convincing evidence for a clustered prevalence of anophthalmia and microphthalmia? Thirdly, is there any obvious link between benomyl and the regions of increased prevalence? Animal studies have already shown that benomyl can induce anophthalmia and microphthalmia.[7] The doses used, however, were about 1000 times higher than the expected dose received by farm workers handling the pesticide. Taking this into account, the alleged clustering could simply be explained by assuming that people in the areas of clustering had been exposed to higher concentrations of benomyl than those in the surrounding regions. Alternatively, the areas of clustering might harbour subpopulations with a higher genetic susceptibility to the compound. In principle, these potential explanations could be investigated, but before embarking on such research one would like to answer the second question.

In this respect the results of Dolk et al are of fundamental importance. In fact, Dolk et al were not able to show statistically significant regional variation in the presence of anophthalmia and microphthalmia in England between 1988 and 1994. Moreover, no convincing evidence of localised clusters was obtained when mild cases and cases with a known cause were omitted. Although the results depend on the intrinsic limitations of epidemiological approaches to the concept of clustering, as outlined in the commentary (p 910), they indicate that further research based on the alleged clustering is probably bound to fail and will not provide useful data. In this respect the government's earlier dilemma of whether to pursue the relation between pesticides and clustering or to investigate the major causes of anophthalmia and the possibilities for prevention seems to have been resolved.

The results of Dolk et al suggest that there no longer seems to be a serious reason for public concern. Indeed, some may wonder whether all the commotion has been for nothing. This is certainly not so. Any suspected increase in prevalence of disorders such as anophthalmia, which interfere with life expectancy or quality of life, demands proper attention. This is especially so where clustering of cases is suspected, since this might serve as an efficient tool to unravel the causes of the disorder. Few will doubt that the observation of an increased incidence of cancer in the Chernobyl region would eventually have led to the detection of its cause. Furthermore, epidemiological studies often unearth unexpected findings or new insights—as with the association between socioeconomic status and the prevalence of neural tube defects and the subsequent links to diet and folic acid.[8,9] For this reason Dolk et al's finding of an inverse relation between the prevalence of anophthalmia and population density merits further investigation.

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Ageing costs

Evidence to royal commission emphasises need for explicit standards and funding

debate of the age

Providing and paying for long term care for Britain's ageing population is an urgent issue. The fastest growing sector of the population is the over 80 year olds. By 2030 a third of the population will be of pensionable age.[1] Last December the government set up a royal commission to produce costed options for the next 50 years and report back. The commission has accumulated a mountain of opinion and evidence from some 2000 individuals and special interest groups concerned with care of the elderly. Last week it held its final public hearing to give them the opportunity to hammer home their messages.

Much consensus was evident. Radical organisational change and new methods of financing are needed to reverse the effects of policies which have left old and vulnerable people hostage to confusing, inadequate, inequitable, and poorly coordinated services. The onus on elderly people to demonstrate what they can't do rather than what they can in order to get help is one of many perverse incentives that need to be removed. Independence should be seen as the goal and more resources directed towards prevention and early intervention to help keep people in their own homes. The current approach of plunging in late with a battery of expensive services before arranging (cheaper) institutional care should be a last resort, not the preferred option. Elderly people and their carers should have better information and more choice about what services are provided, when, and by whom.

Much current dissatisfaction is to due the divide between health services (which are free) and social services (which are not).[2] Cost shunting between agencies has resulted in many people having to pay for what by any reasonable definition is really nursing (and hence health) care.

The argument for structural change to achieve an integrated and coordinated service is widely accepted. The government's recent discussion document *Partnership in Action*, which advocates joint working, pooled budgets, and a lead authority has been welcomed.[3] Most organisations representing elderly people also share the view that housing should be an integral part of community care. Whether the various agencies should be controlled by a new overarching community care agency, as advocated by the charity Age Concern, is debatable, but all agree that a single point of access and delivery is essential. Information and advice needs to be available, free of charge, from the same body that is responsible, and accountable, for assessment and delivery of multidisciplinary services. At present assessment of "need" and the response to it is arbitrarily determined by individual local authorities—hence the wide geographical variations. The case for setting nationally agreed methods of assessment and criteria for eligibility for services is strong.

Most dissension arises over money. All agree that more funds are needed, and the estimates are daunting. Two years ago the Rountree Foundation emphasised that totally new methods of funding long term care for elderly people had to be introduced and that, until they kicked in, the cost to the taxpayer for continuing care would be around £540m a year.[4] "The trouble is, the figures are largely speculative,"said Sir Stewart Sutherland, chairman of the commission. "We don't know what current costs are, nor which models of care provide best value for money." There needs to be a consensus, it was suggested, on what constitutes good quality long term care for elderly and disabled people. Ideally, national standards for care in all the different settings in which it is provided should be defined, costed, and made explicit.

Broad agreement seems to exist that, subject to means testing, individuals should bear the costs of accommodation, food, and certain domestic services while the state should pay for health, personal, and social care. What then are the main options for raising revenue to fund the state's services? Few favour increasing taxes. Many see a long term care insurance scheme as the answer. A common view is that people should pay weighted premiums into a compulsory scheme throughout their lives. In return their long term care would be secured and they would know exactly what they were entitled to. Means testing seems inevitable but remains controversial.

Another thorny issue for the commission is what to do to safeguard the vast army of unpaid, largely female carers. Their contribution is acknowledged to be crucial and will remain so. If they were paid for what they do it would cost the exchequer an estimated £8bn a year.[5] They will not easily be replaced by new recruits, for the working patterns and expectations of young women have changed. The commission is not short of advice, and it has only eight weeks left to marshall its thoughts if it is to meet its target publication date of early January 1999. Few reports will have been so eagerly awaited.

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