'Nuns, virgins, and spinsters'. Rigoni-Stern and cervical cancer revisited

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The view that nuns have a very low risk of cervical cancer is questioned. The historical evidence for this view is reviewed, from the beginning of the eighteenth century to the present. An estimate of the actual mortality rate from cervical cancer suggests that risk of death from this neoplasm among nuns is little different from that among the general female population.

Summary. The view that nuns have a very low risk of cervical cancer is questioned. The historical evidence for this view is reviewed, from the beginning of the eighteenth century to the present. An estimate of the actual mortality rate from cervical cancer suggests that risk of death from this neoplasm among nuns is little different from that among the general female population. It is recommended that nuns should not be excluded from cervical cytology screening. When symptoms which might suggest cervical cancer arise in such women, full gynaecological assessment is necessary.

Present Department of Health policy regarding provision of cervical screening suggests that such screening is not necessary for 'nuns, virgins, and spinsters' (Warden 1990). This policy is supported by the widely accepted view that cervical cancer is extremely rare among such women. Only if this view is correct is it reasonable to dispense with cervical screening in such women. If it were to be shown that such women had a lower, but still significant risk of cancer, then such an omission would be unreasonable.

Historical review

Rigoni-Stern

Answering a reader's question in the British Medical Journal, Drife (1984) states, regarding cervical cancer, that '... it is now well documented that the disease is rare in nuns and common in prostitutes. ... a connection between intercourse and cervical cancer was apparently first suggested in 1842.'

Almost any paper on the epidemiology of cervical cancer mentions the work of Rigoni-Stern, a surgeon in Padua in the mid-nineteenth century, who appears to have had an amateur interest in epidemiology.

At the 4th Congress of Italian Scientists, during a meeting of the surgical sub-section, Dr Gandolfi of Modena presented a paper on cirrhosis and cancer. Afterwards, a number of points concerning the epidemiology of cancer were made by Rigoni-Stern and although these were not included in the official report of the conference, Rigoni-Stern subsequently expanded his ideas and published these in a regional medical journal (Rigoni-Stern 1842).

Although his work is often quoted in the context of the epidemiology and aetiology of cervical cancer, it is important to remember that during his time the diagnosis of 'cancer of the womb' was not separated into carcinoma of cervix and corpus. Thus, it was impossible for Rigoni-Stern to make any comments on the incidence of deaths from cervical cancer. He did, in fact, find four deaths from cancer of the womb among 1288 deaths in nuns. Based on the numbers of deaths from cancer of the womb in the general population, six would have been expected (Skrabanek 1988). There is a suggestion of considerable under-reporting of this cancer, both in nuns and in the general population.

The observations made are not of statistical significance.

In fact Rigoni-Stern was not so foolish as to draw any of the conclusions from these data that have been ascribed to him subsequently. He limited his observations on cancer of the womb to noting that the ratio of deaths from breast cancer to deaths from cancer of the womb was far greater in nuns (and single women) than in other women. This observation (of an excess of breast cancer in nuns)
had already been made almost 150 years earlier by Ramazzini (1713). This would be expected on the basis of modern knowledge and is associated with the protective effect of pregnancy against breast cancer.

The quality of Rigoni-Stern's work is poor by modern standards. His paper is littered with errors of arithmetic. He adduces importance to differences which we would not now regard as being statistically significant. Interesting as Rigoni-Stern's observations were, it is somewhat surprising that his work is quoted as being authoritative. It is alarming that it is almost universally misquoted. How such misquotation comes about is difficult to understand. Although the paper is written in Italian, there is no lack of clarity about his findings. Perhaps part of the explanation of this misquotation lies in the antiquity and obscurity of the journal in which the paper was published. It seems that once Rigoni-Stern's findings were misinterpreted many subsequent authors have merely accepted the accuracy of earlier quotations. Two observations support this view: when the paper is quoted the title of the reference is often incomplete; the British Library, who do not hold a copy, have no record of any UK library holding a copy and had no record of having obtained a copy for any reader. (Recently his paper and the reference to it were published in full English translation, Rigoni-Stern 1987). It would be interesting to learn who first misquoted Rigoni-Stern and who was, therefore, responsible for the genesis of the myth. Skrabanek & McCormick (1989) have described this type of phenomenon as 'the Bell-man's fallacy', whereby if an untruth is repeated often enough it becomes accepted as true.

Versluys

Versluys--a post-war Dutch epidemiologist--presented cancer-mortality statistics from the Netherlands for the years 1931 to 1935 (Versluys 1949). Among the general female population 4.91% of cancer deaths were due to carcinoma of the cervix. Deaths from cervical cancer were more common in married women than unmarried (5.92% vs. 2.33%). Versluys also calculated age-specific rates: these revealed, as expected, increasing rates in older women. The incidence of deaths from cervical cancer in single women in younger age groups (under 60 years of age) being about a quarter that in married women, and about half the incidence in older women.

Among 197 cancer deaths in nuns, Versluys found five deaths from cervical cancer--against an expected 4.6. These represented 2.5% of cancer deaths in nuns. His account is unclear but it appears that the 'expected mortality' from cervical cancer was based on an age-adjusted rate and that a correction may have been made for the nuns being older than the general population. Versluys' study found cervical cancer to be no less common in nuns than in other single women. The incidence in single women being about half that in married women.

Gagnon

Fabien Gagnon was a French-Canadian gynaecologist who believed that pre-existing cervicitis was a major factor in the aetiology of cervical cancer. At the time of his work (Gagnon 1950) a number of authors were reporting the success of destructive treatment for cervicitis in preventing cervical cancer. These reports describe series of hundreds of women, mostly young, followed for relatively short periods of time. The incidence or death from cervical cancer at the time of these studies was of the order of one per 10 000 women per year. The incidence of death from cervical cancer among a small population of young women followed up for a few years would be expected to be less than unity. However, on this basis Gagnon and his contemporaries were able to convince themselves of the merit of their approach.

Gagnon was responsible for the provision of gynaecological care to a number of Roman Catholic convents. In order to test his hypothesis that cervicitis was an aetiological factor, he decided to study this population of nuns, whom he assumed would not have been at risk of cervicitis as they would not have been sexually active. Due to his position he was able to analyse the death records, going back many years, of a number of religious orders. He studied an 'annual average population of 13 000 covering a 20-year period'. He admitted that data were inadequate in 3500 women, but found no deaths from cervical cancer in the remainder. Subsequently, he made a study of the records of various histology departments in the same catchment area. Not all histology departments in the catchment area were included. Cancers not biopsied would not have appeared in these statistics at all. Nevertheless histology department statistics revealed three cervical cancers in nuns and Gagnon assumed that these represented cases from the same population. Gagnon found 19 cases of cancer of the uterine corpus against three of the cervix. Unlike Versluys (1949) who compared the actual incidence to that expected. Gagnon used a ratio of cervical: uterine cancers. According to various sources he claimed that this ratio should be 6:1, whereas in his study the ratio
appeared to be inverted. This ratio was obtained from the general population and so ignores the possibly increased risk of corpus cancer found in nulliparous women (such as nuns). Gagnon's study revealed at least three cases of invasive cervical cancer among an 'annual average population' of nuns of 13 000 over a period of 20 years. This is an annual mortality rate from cervical cancer of 11.5 million nuns. At the time when Gagnon carried out his study, the mortality rate from cervical cancer among white women in North America was less than 100 per million women (Gardner & Lyon 1974). On the basis of Versluys' observations the rate among single white women would be expected to be about half of this. Kessler (1981) quotes rates among white women of 35 and 13 per 100 000 for ever-married and never-married respectively. Clearly the rate of cervical cancer mortality among nuns, found by Gagnon did not differ significantly from that among other single women. The incidence among nuns was about one third that in ever-married women. Even Gagnon accepted that his estimate was likely to he artificially low due to the possible errors out-fined above. This makes his conclusions seem even more dubious.

**Logan**

Logan was chief medical statistician at the General Register Office, in London—the predecessor of time Office of Population Censuses & Surveys (OPCS). He related marriage and child-bearing to cancers of the breast and uterus (Logan 1953). Overall cancer of the uterus (cervix and corpus combined) was slightly less common in single women. Surprisingly cancer of the uterine corpus had a similar frequency in both groups, whereas cervical cancer was twice as common in married women. However, it is note-worthy that, of all cancers of the womb, 60% in single women and 45% in married women were of unspecified site. With regard to the numerically large group of 'unspecified' cancers,, Logan felt that 'since they mainly represent an ill-defined mixture of cancer of two sites . . . it would serve no useful purpose to discuss them in detail.'

**Towne**

Janet Towne, a Chicago based gynaecologist. examined the role of child birth in cervical cancer (Towne 1955). She believed that 'If the role of pregnancies in carcinogenesis of the cervix is of etiological significance, then other variables connected with marriage or sexual congress must he partially associated as causative agents in the nonparous group of patients. If such a partial correlation exists, Gagnon's report of immunity of celibate women to cervical cancer would establish a relationship between this malignancy and a given factor. In other words, as cervical cancer occurs in women who have had sex but no children, some other aetiological factor than childbirth must exist. If women who have never had sex did not get cervical cancer, then the occurrence of cancer in sexually active nulliparous women would probably be due to a sexual factor. She conducted a retrospective analysis of known cases of cervical carcinoma over a 21-year period, from a radiotherapy unit. Of 574 cases, 16.3% were in nulliparous women, 6.4% occurred in unmarried women, 0.52% in members of religious orders. This represented three cases among nuns. Towne calculated a rate for cervical cancer in nuns of l in 1000. This would imply a rate of a third that in parous married women. However, this statistic is arrived at by rather dubious means. The denominator used was the number of nuns examined in the 21-year period (rather than the population from which they were derived). All cases were histologically squamous carcinomas. In a further part of her study, she examined medical records from convents with an 'annual average population' of 10 000. The idea of comparing the incidence of such an 'annual average population' with the rate in the general population, as was done by Gagnon, takes no account of the age and social class structures of the two differing population bases. An unspecified number of cases were disregarded 'because accurate pathologic information could not be obtained', but despite this she discovered three further definite cases. Although Towne's findings are often quoted as supporting those of Gagnon, she in fact stated 'Thus, the results would appear to be quite different from those of Gagnon, in that 6 nulliparous virginal women were recorded with cervical carcinoma. She concluded that 'cervical malignancy can arise in women irrespective of virginity or parity'. She showed that the ratio of cancers of the cervix to those of the corpus was inverted in nulliparous and virginal women. This observation clearly makes nonsense of any study-such as Gagnon's-which calculates a ratio, rather than an incidence rate, as was done by Versluys. In her discussion she states 'to disseminate the idea that celibate or nulliparous women never develop cervical malignancies would be erroneous. Such misinformation creates the impression that it is unnecessary to suspect these women of or examine them for, carcinoma of the cervix.' In the context of screening, this might be extended to imply that such misinformation should not be
allowed to exclude such women from the cervical smear programme.

**Stocks**

Sir Percy Stocks, a senior research fellow of the British Empire Cancer Campaign, made use of the distinction that was made in national data from 1950 onwards of the specific site of 'cancers of the uterus'. He was thus able to study factors related to the incidence of cervical cancer, which had not previously been possible for England & Wales. In the years 1950 and 1951 there were 5287 deaths from cervical cancer (Stocks 1955).

Designating a value of 100 for 'all married women', comparative mortality ratios (this comparative mortality ratio is identical to what is now described as standardized mortality ratio [SMR]) for all ages were 43 for single women, 84 for childless married women, 103 for married women with children and 125 for divorced or widowed women. Thus the rate for single women was half the rate for childless married women, and just less than half the rate for parous women.

**Fraumeni**

Joseph Fraumeni and epidemiological colleagues at the US National Cancer Institute studied 31,658 white Catholic nuns in order 'to clarify the role of marital status in human carcinogenesis' (Fraumeni et al 1969).

Their study covered 41 religious communities for the years 1900 to 1954. They excluded all nuns who were not 'white, native-born, never-married'. Also excluded were nuns who 'had performed household or manual duties, were nurses or had served at foreign missions'. As a comparison group they used the total female population of Massachusetts (1900-1913) and the white female population of the US (1914-1954). The former group was used as the necessary data were unavailable for the whole US population until 1914. In fact, data on the incidence of specific gynaecological cancers were not available for these comparison groups. Estimates were obtained by the simple--but invalidated--method of assuming that the ratio of cancers at specific sites would be the same before 1930 as it was in 1930 to 1934, and calculating estimated specific rates for each genital site, based on the total for all genital cancers. The authors concede that this approach is not entirely accurate, it being known for instance that the proportion of uterine and cervical tumours varied considerably during this time. (In 1914 uterine tumours represented 91.6% of deaths from all genital cancers, and ovarian tumours 5.5%; in 1931 these values had changed to 83.0% and 13.1% respectively--clearly the proportions did not remain fixed during this time.)

As would have been expected, nuns suffered from an excess mortality from breast cancer. Except in the older age groups they had fewer deaths from malignancies of the genital tract. Almost 11% of deaths from genital cancer among nuns were due to cervical tumours, compared to more than half in the comparison group. However, 'comparison is further obscured by the high proportion of uterine cancers of unspecified origin among nuns and, to a lesser degree, the general population'. At best this seems to indicate that the physicians responsible for diagnosis and death certification may not have pursued diagnosis as fully as in lay women: at worst it may indicate a deliberate vagueness of diagnosis in many individual cases. As in many other studies, if only a proportion of 'unspecified site' diagnoses were more properly attributable to cervix, any difference between the two populations might disappear.

Fraumeni et al. (1961) concluded that 'The experience of nuns resembled that for all single women, who have a low rate for cancer of the cervix and high rates for cancers of corpus uteri and ovary.' They felt that this risk was probably hormonally mediated, reflecting the endocrine changes due to parity. Though also suggesting that 'an environmental factor related to coital experience appears responsible for the increased risk of cervical cancer among married women'.

**Kinlen**

Kinlen, an epidemiologist based at the Cancer Research Campaign unit in Oxford, examined the hypothesis that meat and fat consumption might be related to cancer mortality. He studied members of religious orders who had dietary regimens of differing strictness (Kinlen 1982). He failed to demonstrate a significant association between these dietary factors and cancer of the large bowel or breast.

He was much less concerned with cervical cancer. He did, however, note a lower incidence of cervical cancer among nuns than expected from the general population (2 cases seen as against 9.7 expected, P <0-01). Deaths from cancer of the endometrium and of 'uterus site not specified' were grouped together and exceeded expectations for endometrium alone. It would seem that once again the low incidence of cervical tumours might, at least in part, be accounted for by an excess of
unspecified site tumours.

What is the actual situation?

The mortality rate for cervical cancer in the United Kingdom is 74 deaths per million women per year (Cancer Research Campaign 1990). In England it is 76.8/million/year, 95 - 8/million/year in Wales (Office of Populations Censuses & Surveys 1989). It tends to be accepted that cervical cancer is a relatively common cause of death in women. In fact cervical cancer is only the seventh most common cause of death from malignant disease among women, and as such is of similar frequency to cancers of the bladder, oesophagus, uterine corpus and leukaemias (OPCS 1989).

Given the above facts, and accepting that the number of nuns is relatively small, one would not expect vast numbers of deaths from cervical cancer among women in religious orders. In 1988 in England & Wales, there were 976 Anglican and 10 353 Roman Catholic nuns in religious communities (D. Skidmore, personal communication). Unfortunately no data are available concerning the age-distribution of this population. If the risk of cervical cancer among these women were assumed to be the same as among the general female population in the United Kingdom, then the expected mortalities from cervical cancer would be 0.84 deaths per year. During the years 1986-1988, inclusive, there were two deaths in female unmarried members of religious orders (viz., nuns) (R. L. McGowan, OPCS, personal communication). This gives an observed annual rate of 0.66 deaths--not significantly different from that expected from the above calculation.

Discussion

None of the studies referred to has actually shown cervical cancer among nuns to be the rarity that has been claimed. Many of the studies are let down by unreliable historical data, often referring to a base population which is frequently not stable and may be unknown. It is impossible to calculate rates unless reliable values for both denominator and numerator are available. Most of the studies have discovered among nuns a disproportionate number of tumours of unspecified site. It is wrong to assume that all or even most of these tumours are non-cervical.

A number of studies have shown that marital status may be a risk factor for cervical cancer, in particular that the disease is more common in married, rather than single women. The use of such relative risk factors ignores the absolute risk. The majority of cervical cancers occur in married (or once married) women rather than single (never married) women, because most women have been married by the age of 55 years, the peak incidence for cervical cancer being from 55 onwards. Malignancy of the uterine corpus is known to be associated with low parity. Nuns are thus more likely to develop such tumours. Calculations of the ratio of tumours of the uterine body to those of the cervix are therefore likely to be increased even if the incidence of cervical cancer remains unchanged.

This review of previously published studies of the incidence of cervical cancer among nuns shows that the rate of death from this tumour in such women is comparable to that among other single women, and may not differ markedly from the rate in the general population.

Where cervical cancer occurs in a nun it is tempting to ascribe this to a less chaste past than her sisters, perhaps before entering a religious community. This attitude may reflect an unfair and erroneous assumption and, in any case, is of no practical value. We do not know whether cervical cancer arises in virginal nuns, although the work of Towne and others suggests that it can. As we cannot be expected to know the intimate past history of all nuns, none should be considered immune to cervical malignancy.

Careful review of the evidence reveals that there is no substantial objective evidence for the dogma that cervical cancer deaths are rare among nuns. Having convinced medical practitioners of this dogma three potential harms might result. Firstly being convinced that a cervical cancer could not arise in a nun--unless she had enjoyed a less than chaste past--a doctor seeing a nun might be tempted to dismiss this as a possible diagnosis and thus fail to give her the most appropriate treatment. Secondly, for similar reasons, when certifying the cause of death lie might be led to record an incorrect diagnosis, either due to disbelief or to avoid a statement which might reflect on the deceased's character. In a number of studies of nuns the majority of deaths from cancer of the womb were recorded as 'site not specified' and it is possible that accepting the dogma could introduce a bias which would appear to support the dogma. Thirdly as cervical cancer is regarded as a rarity in nuns, it is considered unnecessary to offer them cervical cytology screening (Warden
1990). We are doing a grave disservice to more than 10 000 women, if it is believed that such screening might be effective in preventing cervical cancer. Nuns should be offered routine cervical smears.

When a nun has symptoms suggestive of cervical cancer, that diagnosis should be considered as seriously as for any other woman. Irregular vaginal bleeding, intermenstrual bleeding or recurrent vaginal discharge, should lead to a full pelvic examination under anaesthesia.

References:


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