

PRACTICE OBSERVED

Research in Progress

A new syndrome from general practice?

D H JUDSON

Not all "research" leads to publishable papers, and potentially useful hypotheses and data may be lost forever because the researcher can never get them into a paper. Scientific editors are naturally unemphatic about publishing unfinished work, but perhaps they should make exceptions where circumstances dictate that researchers can never hope to finish their work. These circumstances are very often limited time and resources. Our general practitioner advisers have convinced us that we should try publishing some unfinished work and see what happens. Perhaps a reader somewhere will be able to take up the ideas in the paper and develop them. We are therefore publishing this paper by Dr Judson, and to begin the process of taking the work further have asked two other researchers to comment on Dr Judson's work.

For 15 years, first in urban Yorkshire and then in rural Orkney, I have been in pursuit of what I believe may be a distinct syndrome that may be caused by a particular virus. The common feature of the syndrome, which may present in several different ways and in which different symptoms can predominate, is a characteristic eruption on the soft palate. I first began to think that I might be identifying a distinct syndrome in the late 1960s, and in 1969 I observed a small epidemic of 215 cases in a practice of 2600 patients. At that time I attempted to define the syndrome clinically and find a cause. These attempts have continued ever since, and I now have observations on many more cases.

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The syndrome

Patients usually present with a sore throat or a cough with or without fever and gastrointestinal symptoms. Abdominal pain—often associated with fever but rarely with vomiting or diarrhoea—may be the chief complaint and may mimic appendicitis. These main symptoms may be associated with vague ill health, depression, or headache, or, indeed, patients may present with only these vague symptoms. In 215 cases 104 had sore throats, 94 coughs, 94 headaches, 65 rhinorrhoea, 54 fever, 42 abdominal pains, 36 diarrhoea, 24 vomiting, 21 pain in the legs, 21 backache, 18 earache, and 18 nausea.

Cases occur sporadically but mostly in the last quarter of the year, and I have seen the one epidemic in 1969. Usually the illness lasts about five days, and I guess that it has an incubation period of about seven days. Relapse can occur, and the second illness lasts for three to four days. Most cases occur in people under 50.

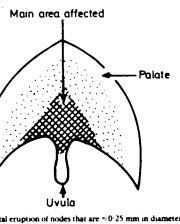
The characteristic sign that links all cases is the palatal eruption. It is not easily seen in direct light and is best viewed with indirect light, and in children with the tongue extruded. The eruption consists of tiny nodules which look like vesicles (but it is difficult to be certain). These are 0.25 mm or less in diameter. They occupy a defined area (see figure) and are distributed evenly and symmetrically on either side of the midline. Usually they appear in the first week of the illness and are often present when the patient is first examined. With time they become more prominent as they develop a fine erythematous ring round their bases and become less translucent. Over one to three weeks the vesicles flatten and disappear. The eruptions do not cluster or become confluent, appear on only one side, or ulcerate. They may extend over the hard palate or remain limited to the posterior portion of the soft palate on either side and anterior to the uvula. The eruptions never extend on to the uvula, which can, however, become oedematous. In acute reactions the whole palate may become erythematous and oedematous, and petechiae can be seen. Occasionally a solitary ulcer 2-3 mm across and with erythematous margins appears on the periphery of the pattern formed by the palatal eruption. Dusky to pink reddening of the anterior tonsillar pillars may be present. The tonsils are often more pink than usual but do not have the appearance of tonsillitis. The lymph nodes of the neck are not affected but the tonsillar lymph nodes may be enlarged and tender. The mucous membrane of the mouth, the posterior pharynx, and the tongue are all normal.

Little else is found on examination. A few patients have conjunctival injection, and one or two have had a generalised skin rash. This has appeared one to three days after the start of the illness and may be macular or

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rubelliform. It starts in the flexures or around the neck and covers the trunk and limbs. The only other findings of abdominal tenderness to those who have abdominal pain.

Search for a cause

In 30 consecutive cases virus culture was attempted from throat swabs, palatal scrapes, and faecal specimens. Specimens were refrigerated and taken to the laboratory on the same or the next day. Culture was attempted in cell cultures and suckling mice. No viruses were grown. Throat swabs were



also cultured for bacteria but none were grown. Serology was also performed on paired specimens of sera for Coxsackie virus group B1-5, adenovirus, respiratory syncytial virus, myxovirus A, B, V, and S, herpes virus, pituitovirus, Coxsackie herpes, Myxoplasm pneumoniae, and cytomegalovirus. Two patients showed evidence of infection with *M. pneumoniae* and two of infection with myxovirus A.

Discussion

After 15 years of work on this condition I have no proof that it is a distinct clinical entity and I have not succeeded in isolating a cause. But I do believe that the characteristic palatal eruption makes it likely that this is a distinct entity, although I have seen a similar eruption in two patients infected with myxovirus A and in two infected with *M. pneumoniae*. But in all of these cases the course of the illness was not typical, and I hypothesize that if these two organisms had been causing the syndrome then they would have been isolated more often.

A wide range of viruses cause respiratory and gastrointestinal problems, and Coxsackie viruses and echoviruses can cause lesions in the mouth. I believe that this syndrome may be caused by a particular virus, but with my limited resources and present geographical isolation I am unable to find the cause. I have chosen to publish these very incomplete results in the hope that somebody may be able to take them further.

Dr DANIEL REID:

Dr Judson describes a condition that is seeking a cause. His problem is reminiscent of those faced with hand, foot, and mouth disease before Coxsackie viruses (commonly type A16) were implicated or the "stapped cheek syndrome" before the parvoviruses were isolated.

Before being sure that Dr Judson is describing a distinct clinical entity more evidence needs to be put forward—and it is likely that this evidence is readily available from Dr Judson's records. Although it is tempting just to await results from laboratory tests, much can be done in the meantime—after all, during the cholera epidemic in 1854 John Snow showed that the disease had an

infective aetiology, many years before Koch isolated *Vibrio cholerae* from patients with cholera in 1883. Dr Judson's description is largely clinical, but there are microbiological data which could throw much light on Dr Judson's puzzle.

Answers to the classical questions: Who? Where? and When? are always helpful. Dr Judson states that most of his cases are aged under 50, but are the patients infants, children, or adults (if so, what do they work at)? Are there any family connections? What about sex differences? In children are the reports related to the home or the school? In adolescents might kissing be a mode of spread as in infectious mononucleosis? In adults might the workplace be implicated? Are there any common factors that may link cases, such as contact with animals? Perhaps Dr Judson has charted the cases over his 15 years of study. Do cases occur every year? If not what is the interval when no cases occur? Most of the cases occur in the last quarter of the year—why should this be; are they related to the reopening of schools or the gathering together of persons in groups for dances, bingo, etc? Although a viral aetiology has been unsuccessfully sought, what about help from other laboratories to get an indication of an infective cause? Is the white cell count raised and what about the erythrocyte sedimentation rate? The answers to questions such as these should help to record the facts about this condition.

Thereafter lies the vexed problem of "association" or "causation." It is especially persuasive if the observed association between the facts is (a) strong (for example, scrotal cancer was 200 times commoner in chimney sweeps than in other workers), (b) consistent, (c) specific (for example, brucellosis in persons drinking infected milk), and (d) plausible (although this may be difficult if the association is too far ahead of its time for the scientific community). "What was discovered yesterday is very clear, what is discovered tomorrow is obscure." (Despite searching over 60 books of quotations, aphorisms, and phrases neither we nor Dr Reid can identify the source of this quotation: we will be pleased to hear from any reader who can.—Ed, BMJ.)

Dr Judson appears then to have some support for his belief that he has identified a distinct syndrome. His findings are certainly consistent ("all cases have a palatal eruption") over a long period of observation (15 years), they are plausible of an infective aetiology as the symptoms and signs are certainly what one might expect following an infection. This is supported by the occurrence of an outbreak of 215 cases (and 8% attack rate) in his practice during 1969. Dr Judson should take heart from Sir Austin Bradford Hill in his presidential address to the section of occupational medicine of the Royal Society of Medicine in 1965: "All scientific work is incomplete—whether it be observational or experimental. All scientific work is liable to be upset or modified by advancing knowledge. That does not confer upon us a freedom to ignore the knowledge we already have or to postpone the actions that it appears to demand at a given time."—Director, Communicable Diseases (Scotland) Unit, Ruchill Hospital, Glasgow G20 9NB.

Dr R. Cameron, Dr W. L. J. Ed. Medical services and clinical trials, 2nd ed. London: Oxford University Press, 1974, p. 121.
2 Bradford Hill A. The environment and disease: associations or causation? *Proceedings of the Royal Society of Medicine* 1965; 58: 399-400.

Dr D W MACLEAN:

New clinical entities are established from time to time. Hand, foot, and mouth disease is a recent example. A minor self-limiting entity may pass undifferentiated because it is minor and only comes to notice through a committed researcher. This paper may represent one of these occasions but other possibilities must be considered.

In his paper on hand, foot, and mouth disease Meadow described "minor disease" as a separate entity in 18 patients who were more ill and who did not have the peripheral rash. The oral manifestations, however, of this are basically lesions of the anterior mouth and therefore may be dismissed from this differential diagnosis.

Herpangina, usually due to viruses of the Coxsackie A group, may cause fever, sore throat, headache, and abdominal pain. The lesions are macular, papular, and then vesicular, and affect the soft palate, especially at its free edge. The lesions also affect the anterior tonsillar pillar, posterior pharynx, and hard palate. This is one condition of the posterior oral cavity that must be considered but which does not quite fit the description.

The description that may equate best is that of Steigman *et al* for the condition they named acute lymphonodular pharyngitis.¹ Twelve of the 15 cases were associated with Coxsackie A10 virus. The present series differs in that cough and rhinorrhoea were features, the uvula was spared, and occasionally lesions ulcerated. There are, however, many similarities, including the occasional conjunctival involvement. Carter published three photographs of acute lymphonodular pharyngitis, showing clearly the identical site shown here in the figure.² One of the patients suffered from rubella and from measles. Carter concluded that these palatal nodules cannot be thought of as pathognomonic of enteroviral infections alone. [Judson's patients included two each with evidence of infection by *M. pneumoniae* and influenza virus A.]
Specimens were submitted for virological examination from 30

patients. There must surely have been a high probability that some isolations would have been obtained if a virus of the Coxsackie A group had been the predominant causative organism. Nevertheless, the serological testing did not cover those viruses commonly implicated in oropharyngeal syndromes, and a case still stored might be justifiable to test a sample for signs of infection by, for example, Coxsackie A10 virus.

Dr Judson is now in an isolated rural practice. Should he diagnose further cases (clinical photographs might help to elucidate the problem. An Orkadian island might well have a visiting naturalist, expert with a camera.—Senior lecturer, Department of General Practice, Lettison House, Edinburgh EH8 9DX.

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- 3 Steigman LR, Dasec FW, Rhodes AJ. Acute lymphonodular pharyngitis: a newly described condition due to coxsackie A virus. *J Hyg* 1962; 41: 311-6.
- 4 Carter P. Lymphonodular pharyngitis. *J Pediatr* 1946; 28: 1107-15.

100 YEARS AGO

Recent painful disclosures have, among other results, raised an important question which, in the present state of opinion, can be most readily discussed in the pages of a medical journal. We refer to the complete ignorance regarding the sexual vigors and the sexual functions which is permitted, and, indeed, sedulously fostered, by the ordinary education which boys and girls receive in this country. Not merely does our school system provide no information on these topics which so vitally concern the happiness of every individual, but the slightest allusion to the subject is to be rigorously prohibited, and perhaps branded as obscenity. The result is, that there is a great deal of ignorance on these questions, and a still greater amount of half knowledge, which is more dangerous than either total ignorance or the fullest information. We have the authority of Sir James Paget for the statement that some men grow up, and even marry, in complete sexual ignorance, and that, while this is rare in the male sex, it is extremely common among cultivated and refined women. The decent veil which we compare to those over everything concerned with the reproductive function serves, beyond doubt, some useful ends, and we trust the English people will always be characterised by their delicacy of thought and expression in this matter. But we are convinced that this secrecy, this "conspiracy of silence," has gone too far, and that it is productive of serious evils. We object, in the first place, to its unnatural. That our educational methods and social practice should train men, or more frequently women, to marry without knowing what marriage involves, is not merely unnatural, but may be the cause of much matrimonial unhappiness. Parents and school-masters act as if innocence in such matters would last for life, and as if knowledge were a crime. But a much more serious, because infinitely more common, evil is the objectionable mode in which sexual knowledge generally gets access to the mind. Instead of being conveyed in some plain and matter-of-fact manner, it is too often gained through the corrupting medium of low jest or obscene print. At the most emotional and plastic period of life, when new instincts are swelling up and causing great mental disquietude, we withhold from boys and girls the knowledge which nature is instinctively trying to impart, and we leave them to grope their way in darkness, or to seek illumination from some unauthorised source. Why do the young so often regard the sexual work with such fearful but so irresistibly curious? Not from mere depravity, as we often assume, but because they are thus unconsciously seeking information which they are unable to obtain elsewhere, and which are conscientiously bound to supply in some form which will enlighten the reason, without inflaming the imagination and exciting the passions. Sexual knowledge is not wrong, its tendency is not necessarily injurious, but our mistaken methods of secrecy have undoubtedly the most unfortunate effect of stimulating the imagination to the highest point. We know the fearful imagination of forbidden fruit, and the use of it is not to be simply because it is forbidden. This is a notable trait in human nature, and our attitude towards sexual knowledge, if we are disregarded, or rather acted in direct contravention of. The sexual function is naturally powerful, but we enormously increase its attraction for the young by labelling it as forbidden

It is usually easier to indicate a disease than to apply a suitable remedy, but we shall not conclude without venturing a few suggestions. First, let us glance at what is suggested in the very few books which touch upon the question. Many urge that parents should convey knowledge on these questions to their children, at the time of life when external signs and new sensations indicate that the sexual instinct is beginning to awake. But many, probably a majority of parents, are not well fitted to undertake such a duty. Our language is badly provided with the necessary terms, and the untrained parent, ignorant of anatomical expressions, would find it hard to convey the necessary information without incurring the suspicion, and, in his own mind, the reproach of indecency. Some advise that the family medical attendant should act *in loco parentis* in this matter, but we are certain that such an action would be highly disagreeable to the members of the profession. One suggestion alone seems to meet the case, but, fortunately, it meets it most thoroughly. Elementary anatomy and physiology should form an integral part of every education. We might begin by teaching boys and girls the bones and skeleton, the functions of the heart, stomach, etc., and then, when the suitable age arrives, the structure and functions of the sexual organs might be taken as the natural sequel of the previous portions of the course. In this way, the necessary knowledge would enter the mind naturally and simply, with no false shame on the one hand, and no fall to the imagination on the other. We are confident that an immense reform would thus be easily and quietly effected, and that much evil and suffering would be averted. We should thus convey, in the most natural and unobnoxious form, knowledge which we have no right to withhold, and we should remove the unwholesome fascination which our present habit of secrecy imparts to sexual questions. Certain it is that the stealthy approaches of vice are favoured by the existing system. It will often be found that there is a prevalent opinion that sexual immorality is to celibates a physical necessity, an attribute of manliness, and even a collateral or prevalent condition of health. This degrading error has been so vigorously denounced by the ablest of modern physiologists, that no one has any longer a pretext for entertaining or promulgating it. It has been the source of much evil, however, and wherever such an opinion is met, it must be energetically combated. There is an aspect of the question which cannot be overlooked, especially as recent revelations have thrown a lurid light upon it. It has been abundantly proved that young girls are often entrapped to their ruin in the most utter ignorance of sexual questions, and of the physical significance of the act to which they are enticed. This is surely a lamentable instance of property over-reaching itself. Innocent ignorance is always attractive, but, if it be the means of luring the innocent victim to her doom, it is surely most dangerous. How then is the girl, approaching to sexual maturity, to be made acquainted with the nature of her creature, and guarded against the temptations which the base impulses of passion? We commend this difficult question to the thoughtful consideration of our readers. In this respect, also, the mothers and the teachers have a very solemn duty, and it is to be regretted that, then, where, and by whom, it is best performed. (*British Medical Journal* 1885; ii: 303.)

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Audit Reports

Generic prescribing: hospital reports to a general practice

Five hundred consecutive typewritten reports that had been received from the Newcastle hospitals by three doctors in an inner city practice with a list of 4000 patients, were analysed to determine the practice with regard to the use of generic names. The reports were written by 100 levels of medical staff from consultant to senior house officer and concerned patients who had either been admitted or been seen in an outpatient clinic. The three general practitioners in the practice adhere to a strict policy of generic prescribing except for proprietary combination preparations. Two hundred and seven (41.4%) of the 500 reports contained recommendations about drug treatment. Of the 337 drugs recommended, 227 (67.4%) were by generic name, 96 (28.5%) by proprietary name, and 14 (4.1%) were proprietary combination preparations.

One objection sometimes raised by general practitioners to generic prescribing is that their hospital colleagues often recommend drugs by their proprietary name. Yet all the Newcastle hospital pharmacists carry out a policy of generic substitution. The general practitioners may be unaware that their patients will have received alternative generic drugs (except for most proprietary

combination preparations) both as inpatients and from the outpatient dispensaries.

Although it has been government policy since 1960 to encourage doctors to prescribe generically, by 1976 only 10% of general practice prescriptions were by generic name. Hospital reports are known to be an important influence on the prescribing habits of general practitioners. If hospital doctors were to recommend drugs by their generic name they would achieve not only semantic and pharmacological accuracy but they might also persuade their general practitioner colleagues to prescribe generically, thus realising the considerable potential savings in drug costs nationally.¹ Such savings might dissuade this government, or a future one, from further restricting prescribing in the National Health Service.—I D VAN ZWANBERG, general practitioner and lecturer, department of family and community medicine, University of Newcastle upon Tyne. (*Accepted 21 August 1985*)

1 Informal Working Group on Effective Prescribing. Report to the Secretary of State for Social Services. London: Department of Health and Social Security, 1981. (Unpublished report.)
2 Anderson R, Reilly P. Generic alternatives in general practice. *Br Med J* 1984; 289: 1128-31.

Treatment of hypertension

In our semirural practice of 7150 patients the records of 305 patients who were being treated for hypertension were reviewed. 78 (26%) were aged over 65 and 48 (16%) had experienced appreciable side effects from the medication that had been prescribed. Risk factors, including family history of cardiovascular disease, smoking history, cholesterol concentrations, evidence of impaired glucose tolerance, and amount of exercise taken, had been looked for in 238 (78%) patients and checks for end organ damage, including examination of the fundi, biochemical screening, urine testing, electrocardiography, chest x ray examination, and any history of cerebrovascular accident, had been carried out in 253 (83%) before treatment was initiated. The final pretreatment blood pressure reading was below 180 mm Hg systolic or 105 mm Hg diastolic in only four patients. All the patients were being followed up at intervals of between three and 12 months.

Before the study was undertaken our impression had been that we were treating a disproportionately high number of our patients over 65, that our pretreatment assessment had been skimpy, and that our criteria for starting treatment had been too liberal, but that our control of blood pressure was largely satisfactory. Instead, what this audit showed was that our pretreatment assessment of risk factors

and the search for complications had been much better than we thought; the level of side effects was high, but, most important, our success in reducing high blood pressure was little better than that found by Heller and Rose among less well organised inner London practices in 1977; or that reported by Kurji and Haines² and Michalel³ in 1984 in north west London. Our records showed that less than half (41%) of the men under 45 and less than a quarter (23%) of the women under 45 who were being treated had normal blood pressures. In the 45-64 age group just under half (45%) had normal blood pressure readings on treatment, whereas in those over 65 just over a quarter (28%) were in the normal range.

It seems that general practitioners need to have greater determination in rejecting unacceptable blood pressure levels if the objectives of treatment are to be achieved.—TOM STEWART, general practitioner, Souting Common Health Centre, Souting Common, Reading RG4 9SW. (*Accepted 18 September 1985*)

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2 Kurji M, Haines AP. Detection and management of hypertension in general practice in north-west London. *Br Med J* 1984; 289: 933-6.
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