the germ of an idea for explaining how malformations are produced by chromosomal aberrations. Otherwise this branch of pathology is shrouded in complete darkness. I must also apologize for presenting so many peculiar and mysterious diagrams. However, if they have been entertaining, though incomprehensible, I shall feel that I have been excused for asking your attention for such a long time on subject matter which may seem remote from medicine. At one of the best meetings of the Harveian Society that I ever attended, the chief performer was an expert in prestidigitation, and I only wish that I could have been as entertaining as he was on that occasion: only very modest conjuring can be performed with dermatoglyphics.

To demonstrate this perhaps I may be permitted to end up with an anecdote. Some years ago a stranger from an eastern country came to my room at University College. He produced, rather furtively, some palm prints and asked me to give an opinion about the person from whom they had been taken. I explained that I did not tell fortunes or estimate character; but I said that they probably came from a normal male, and I assured him that they were not the prints of a certain well-known type of imbecile. "That is good," he said, "because he is our prime minister."

Further Information on the Identity of 47,XYX Males

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Since the report by Jacobs *et al.* (1965) of an unusual number of males with the chromosome complement of 47,XYX at the Scottish State Hospital, considerable interest has been taken in the frequency of such males among men in maximum security hospitals and in prisons. Jacobs *et al.* drew attention to the unusual distribution of height of males with two Y chromosomes. Taking advantage of this, Casey *et al.* (1966) showed 12 out of 50 males of 183 cm. (6 ft.) or more in height from the English maximum security hospitals of Moss Side and Rampton to have a 47,XYX complement, 4 out of 50 similar males to have this abnormality from the maximum security hospital at Broadmoor, and 2 out of 24 males of 183 cm. or more who were serving intermediate term sentences (six months to five years) in Nottingham Prison also to have this complement.

The study at the Scottish State Hospital showed the XYX males to have no obvious abnormality of sexual development or any other important physical abnormality save perhaps for a tendency to myopia (Price *et al.*, 1966; Jacobs *et al.*, 1968). Our more recent experience of such males, ascertained among penal groups or in hospitals for the mentally subnormal, has been that these men also are normally developed sexually. It is necessary, however, to add the caveat that, from the very nature of the groups in which they are found, these men are often single and it is seldom that there is information on their fertility.

By the end of 1967 we had identified 25 men who either had a 47,XYX complement or were chromosome mosaics in whom this cell line predominated in lymphocyte cultures. One was

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a man referred to hospital with hypogonadism and bilateral talipes equinovarus, and the information on him was published by Court Brown *et al.* in 1964 (Case 82/62). The next nine cases came from the State Hospital at Carstairs, and the details of these are given by Jacobs *et al.* (1968). This communication deals with the remaining 15 males with a 47,XYX sex chromosome complement or who are mosaics in whom the XYX cell line is the dominant one. Its primary intent is twofold—to underline the apparently normal sexual development of these individuals and to emphasize that they are found in various groups and not just in maximum security hospitals or in prisons. This paper shows that while many of those described have a criminal record, a finding biased by the method of ascertainment of some of them, the XYX male may not have such a record though he may pose behavioural problems.

Patients

The 15 abnormal males have been identified in various groups, and with one exception (Case 289/67) all have been found in chromosome surveys. These surveys include studies of liveborn male babies, of new entrants to the Scottish Borstals, of men in the Scottish prisons and in one Young Offenders Institution, of males in an epileptic colony, and of men in hospitals for the mentally subnormal and for mental disease, while one case (364/67) was found in a survey of men who had been given thorotrast intra-arterially for diagnostic purposes many years ago. Formal reports of these surveys will be published later, and this paper deals specifically with the physical and mental features of the abnormal males and gives information on their behaviour. The only male not identified

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in a survey was referred from a psychiatric clinic because of the combination of aggressive and delinquent behaviour and excessive height. The case numbers quoted are those held by the cases in the Medical Research Council's Registry of Abnormal Karyotypes.

Table I contains data on heights and weights, on sexual development, and on any physical abnormalities. All these males were examined cytologically by means of cultures of lymphocytes, and Table II provides information on the distribution of chromosome counts together with a record of the karyotype for each case. It is to be noted that two men were chromosome mosaics. In Case 364/67 12 of the 15 cells with 45 chromosomes or less did not contain a Y chromosome, while four of the five cells with 46 chromosomes had only a single Y chromosome. This case is certainly a 45,X/47,YYY mosaic, and he could also have a third line of 46,XY cells. In Case 405/67 all eight cells with a count of 46 chromosomes had a normal male complement, and the case is presumed to be a 46,XY/47,YYY mosaic. Finally, the question of mosaicism was considered in Case 317/67. In the first blood sample there were four cells with 45 chromosomes and in three of these both Y chromosomes were absent, suggesting the possibility of a 45,X cell line, but a repeat sample failed to show any more such cells. A further complication is that 4 of the 10 cells with 46 chromosomes had a 46,XY complement, so that the presence of a normal male line is also possible. In our present state of ignorance about XYY males and the possible effects of age on their chromosome counts, we prefer to regard this man as having a 47,YYY complement, though this diagnosis may be modified in the light of subsequent experience.

Case Histories

Summaries are given below of the data on the cases in the order of their arrangement in Tables I and II, and each history indicates the manner of ascertainment of the relevant case.

TABLE I.—Clinical Information

Case No.	Age in Years	Height (cm.)	Weight (kg.)	Sexual Development	Physical Abnormalities
290/67	0	45*	2.1	Normal infantile	None
289/67	13	202	82.7	?	?
255/67	16	189	76.4	Normal	None
76/66	17	169*	62.7	Normal	Bilateral genu varum
341/67	17	199	73.2	Normal	Bilateral gynaecomastia
404/67	32	170	86.4	Normal	Congenital heart disease
364/67	34	183*	66.7	Normal	None
356/67	38	182*	84.1	Normal	Myopia
342/67	38	185	?	?	?
140/67	45	186	80.3	Normal	None
395/67	48	184	87.3	Normal	Multiple facial sebaceous adenomata
340/67	52	198	78.6	Normal	None
394/67	53	185	92.7	Normal	None
317/67	53	188	76.4	Normal	Varicose ulceration
405/67	62	185	69.5	Normal	Previous varicose ulceration

* No restriction on height in ascertainment.

TABLE II.—Cytogenetic Data

Case No.	Age in Years	Cells With Chromosome Counts of					Total Cells Analysed	Karyotype
		<45	45	46	47	48		
290/67	0	—	—	—	30	—	30	47,YYY
289/67	13	1	1	2	26	—	30	47,YYY
255/67	16	—	—	1	29	—	30	47,YYY
76/66	17	—	1	3	26	—	30	47,YYY
341/67	17	—	—	—	30	—	30	47,YYY
404/67	32	—	—	1	28	1	30	47,YYY
364/67	34	1	14	5	80	—	100	45,X/47,YYY*
356/67	38	—	—	2	28	—	30	47,YYY
342/67	38	—	—	1	29	—	30	47,YYY
140/67	45	—	—	—	30	—	30	47,YYY
395/67	48	—	—	—	30	—	30	47,YYY
340/67	52	—	1	3	26	—	30	47,YYY
394/67	53	—	—	1	29	—	30	47,YYY
317/67	53	1	4	8	36	1	50	47,YYY*
405/67	62	1	—	8	57	—	60	47,YYY*
					41	—	50	46,XY/47,YYY*

* See text for discussion on mosaicism.

Case 290/67.—Aged 0 years. Identified in survey of liveborn male babies. Gestational age 35 weeks. Co-twin female with a 46,XX chromosome complement.

Case 289/67.—Aged 13. Referred to psychiatric clinic at 11 years because of delinquency, anxiety symptoms, and school truancy. Described as very aggressive. Combination of this and height reason for study. Normal chromosome complement both parents. No other data.

Case 255/67.—Aged 16. Identified in survey of males 178 cm. (70 in.) or more in hospital for mentally subnormal. Educationally subnormal school from 11 to 15 years. Convicted of assault at 15 years and sent to approved school. Referred to present hospital at 16 years. Average intelligence. No behavioural problems in hospital.

Case 76/66.—Aged 17. Identified in survey of all males referred for Borstal training in Scotland during a 12-month study. Mother died when patient aged 2 years; deserted by father. Adoptive mother died when patient aged 13. Convicted of breaking and entering at 14, sent to approved school. Similarly convicted at 15; admonished. Convicted of malicious damage and theft at 17; sent to Borstal. Homosexual. I.Q. 86.

Case 341/67.—Aged 17. Identified in survey of males 178 cm. (70 in.) or more in a Young Offenders Institution. Had been at educationally subnormal school. Convicted of theft at 12 years, and similarly on several subsequent occasions. Last conviction, theft of car. Mentally subnormal.

Case 404/67.—Aged 32. Identified in survey of all males in hospital for mentally subnormal detained under Mental Health Act, 1959. Educationally subnormal school 9 to 16 years and then to approved school. Detained in present hospital at 17 years. No convictions. Repeated absconder. Occasionally threatens patients. I.Q. 59.

Case 364/67.—Aged 34. Identified in survey of male patients who had had intra-arterial thorotrast for investigation of intracranial haemorrhage. Severe occipital headache and loss of consciousness at 11 years. Spontaneous recovery. Later study failed to identify cause of haemorrhage. Normal schooling, leaving at 15 years. Worked as errand boy, then apprentice electrician, and now as trained electrician. No behavioural disturbances. Tends to faint easily. E.E.G. normal. Married, with two normal sons, both having a 46,XY chromosome complement. Average intelligence.

Case 356/67.—Aged 38. Identified in survey of males in an epileptic colony. Ordinary education, leaving school at 15 years. Worked in bakery and as caretaker of village hall. Admitted to colony at 21 years. History of fits since infancy. Behaviour unreliable and irresponsible. No criminal record.

Case 342/67.—Aged 38. Identified in survey of males of 178 cm. (70 in.) or more in Scottish prisons. Convicted of malicious mischief. No further data available.

Case 140/67.—Aged 45. Identified in survey of males of 178 cm. (70 in.) or more in hospital for mentally subnormal. Admitted to hospital at 7 years as mentally subnormal, but at 11 years sent to educationally subnormal school. Convicted about 25 times and imprisoned seven times for theft; full details not available. Admitted present hospital under court order. Frequent absconder. I.Q. 76 (Stanford-Binet).

Case 395/67.—Aged 48. Identified in a survey of males of 183 cm. (72 in.) or more in hospital for mentally subnormal. Said to have had fits until seven years. Attended educationally subnormal school. Detained in present hospital at 16 years. No convictions. Not aggressive to patients, and participates in hospital social activities. Employed outside hospital. I.Q. 42 (Terman-Merrill).

Case 340/67.—Aged 52. Identified in survey of males of 183 cm. (72 in.) or more in hospital for mentally subnormal. Early history unknown. Worked for 20 years as labourer and later by scrounging and scavenging. Convicted of petty thefts at 36 and 37 years and detained in present hospital. Causes annoyance to patients through interference with activities, but not aggressive. I.Q. 62 (at 19 years).

Case 394/67.—Aged 53. Identified in survey of males of 183 cm. (72 in.) or more in hospital for mentally subnormal. Early history unknown. Attended elementary school, but educationally backward. Detained in hospital for mentally subnormal at 19 years after conviction for indecent assault on young girl. On licence from 22 to 24 years. At 24 years convicted of theft of female

clothing and detained in Rampton Special Hospital. Transferred to present hospital at 36 years. Described as aggressive with abnormal sexual propensities—homosexuality, fetishism, and indecent exposure. I.Q. 78 (Terman—Merrill).

Case 317/67.—Aged 53. Identified in a survey of males of 183 cm. (72 in.) or more in a hospital for mentally subnormal. Epilepsy from infancy until about 13 years. Attended educationally subnormal school. Attempted work as factory hand. At 17 years without visible means of support and detained in present hospital. No criminal record. At commitment said to be detached, solitary, and subject to attacks of excitement and moodiness. Has been vindictive and upsetting to others. Behavioural problems diminishing with increasing age.

Case 405/67.—Aged 62. Identified in a survey of males 183 cm. (72 in.) or more in a hospital for mental disease. Early history unknown. Appears to have led vagrant life with casual labouring jobs. Has been drinking for about 20 years. Attempted suicide twice. Convicted for breach of peace twice. Admitted present hospital at 60 years from police station, charged with being drunk and disorderly. Discharged but readmitted informally one year later in same circumstances.

Discussion

The study of males with a 47,YYY complement has passed through two phases and is now in its third phase, which is likely to last for a long time. The first phase was characterized by the discovery of such abnormal males among those whose chromosomes were examined because they showed various physical abnormalities of development or, as in the instance of the original case reported by Hauschka *et al.* (1962), because they were found during the study of individuals related to others known to have abnormal chromosome complements. By 1965 there were no more than about a dozen examples recorded of the YYY male in the world's literature; these showed no consistent pattern of phenotype, and it appeared possible that such individuals might be very rare. These cases are summarized by Balodimos *et al.* (1966).

The preliminary publication by Jacobs *et al.* (1965) introduced the second phase, when these authors reported the finding of unusual numbers of YYY males during a chromosome survey of patients at the Scottish State Hospital. There were two important findings in this study, the first being that the men showed no developmental abnormalities (Price *et al.*, 1966) and the second that their distribution of height was quite significantly different from that of men with a 46,XY complement from the same hospital (Jacobs *et al.*, 1965, 1968). In fact, they were on average about 15 cm. (6 in.) taller than the men with a single Y chromosome, and about one in four of the men in the hospital of 183 cm. (6 ft.) or more had an YYY complement. These findings were confirmed by Casey *et al.* (1966) from the examination of men of 183 cm. or more at the English special hospitals of Moss Side, Rampton, and Broadmoor. This second phase, therefore, was quite short, and it was characterized by the demonstration of YYY males in these maximum security hospitals in frequencies which had to be regarded as quite unusual, even though nothing was known at the time of the frequency of such males in the liveborn population. Finally the men identified in this way showed no physical abnormalities, were on average usually tall, and, as suggested by the work of Price and Whatmore (1967), they appeared to have a genetically determined predisposition to aberrant behaviour.

In the present and third phase the natural history of the 47,YYY males is being elucidated through their identification in chromosome surveys, often restricted to males above a certain height, and through the detailed examination of the physical, mental, and behavioural features of those identified. Surveys are in hand of liveborn babies, of boys and men in various penal groups, and of patients in hospitals for the mentally subnormal and the mentally diseased, and of males of tall stature in the ordinary population. It is becoming clear that certain other

groups will repay investigation, such as boys at educationally subnormal schools and at approved schools and disturbed young males, especially those with a history of delinquency, seen in psychiatric clinics.

The cases in this communication have been reported by us with the purpose of focusing attention on the variety of groups in which the 47,YYY male has now been found and on the essential normality of his sexual development. As our data show it is also apparent that some of these males are chromosome mosaics, as were Cases 364/67 and 405/67. The former case was found by accident in the ordinary population, and there is no reason to doubt but that he is a socially well-adjusted man. It would, of course, be unwise to conclude that his normality bore any relation to his mosaic state, but the general question is raised of to what extent the presence of a second cell line may modify the effects of a 47,YYY line.

It remains the case that all the YYY males found by us since our work at the State Hospital at Carstairs have shown no abnormality of sexual development as judged by inspection of the external genitalia and palpation of the testicles, though there were two cases which we have not been able to examine (Cases 289/67 and 342/67). A number of physical disabilities are listed in Table I, and one man had gynecomastia, one had congenital heart disease, one was myopic, one had multiple sebaceous adenomata on his face, and three had varicose veins, for two of which there was evidence of present or past varicose ulceration. With the small numbers of cases it would be unwise to draw conclusions from these data.

Excluding the baby, the shortest YYY male in this communication is the Borstal boy who was 169 cm. (Case 76/66), and he could well continue to grow for a few years. Another male (Case 404/67) was 170 cm. in height at 32 years. He had congenital cyanotic heart disease, and this may have affected his growth. However, the shortest male in the Carstairs survey was only taller by 1 cm. and he had no physical abnormalities. While at present there is a tendency to restrict studies of adult males by height—for example, to those of 183 cm. or more—it should not be overlooked that this restriction will lead to an important proportion of 47,YYY males not being identified. As yet there are too few cases recorded to examine the distribution of height in detail, but it is worth noting the heights of the 12 YYY males we have now identified in circumstances in which there has been no restriction on height and who are over 15 years of age. These include the single case reported by Court Brown *et al.* (1964), the nine cases reported by Jacobs *et al.* (1968), and Cases 76/66 and 356/67 in this report. The heights of these males range from 169 to 188 cm., with a mean of 180 cm., or about 5 ft. 11 in.

We now have reliable information on the ages of both parents at the births of 18 of our YYY males. Though this is a small number, it seems worth while examining the data

TABLE III.—Edinburgh Data on Parental Ages at Birth of 47 YYY Males, Excluding Mosaics

Case No.	Age at Ascertainment	Parental Ages	
		Mother	Father
82/62	23	19	27
120/65	31	27	30
122/65	32	32	35
123/65	39	20	22
132/65	17	23	30
134/65	31	41	41
141/65	28	23	22
150/65	22	28	34
152/65	28	32	37
76/66	17	30	30
290/67	0	25	23
289/67	13	29	36
255/67	16	24	26
341/67	17	32	33
356/67	38	29	31
342/67	38	28	30
140/67	45	36	37
317/67	53	27	33
Mean age ..		28.1	30.9

Case 82/62, Court Brown *et al.* (1964).
Cases 120/65—152/65, Jacobs *et al.* (1968).

(Table III) for evidence of an effect of paternal age. By analogy with the findings for 47,XXX females in which there is evidence for the risk of the conception of such females increasing with maternal age (Penrose, 1964), one might expect to find comparable evidence for the paternal age in respect of 47,XYY males, and this should be demonstrated through an increase in the difference between the means of the paternal and maternal ages. As can be seen from Table III the mean paternal age is 30.9 years and the mean maternal age 28.1 years, with the difference of the means being 2.8 years. This certainly accords with information quoted by Penrose (1956) which gives a mean paternal age in England of 30.9 years, a mean maternal age of 28.6 years, and a difference of 2.3 years. So far, therefore, these limited data show no evidence for an effect of paternal age.

It is clear that 47,XYY males are to be found in at least some hospitals for the mentally subnormal. All present cases were identified in two such hospitals, both of which are particularly concerned with behavioural disorders, and some of these males were committed under a court order. There may well be some variation between hospitals in the frequency of XYY males depending on admission policy. Nevertheless the 47,XYY complement has now to be regarded as yet another form of sex chromosome aneuploidy which can be associated with mental subnormality. Another feature of some of these males identified as mentally subnormal is that while they may show behavioural disorders they need not have criminal records. A further and perhaps new point arises from considering Cases 340/67 and 405/67. Both men were essential chronic drifters and vagrants, and if their histories are reliable they appear to have just managed to maintain themselves at a low level in society without infringing the law until fairly late in their careers, and the infringements then were trivial. For example, Case 340/67 was charged with the theft of a milk bottle and a shirt, and was committed to his present hospital at 37 years; while Case 405/67 was committed at 61 years after two charges of breach of the peace. These two cases may represent a class of males with an XYY complement who can remain in ordinary society, but at a low socio-economic level, for some considerable time before they come to the notice of the authorities. Perhaps it should not be overlooked that Case 405/67 was a mosaic.

Unfortunately it will be many years before we are in a position to assess the risks contingent on an XYY complement in a liveborn baby with any precision. It may be that the major proportion of these babies develop important behavioural disturbances, but this remains to be seen. In the meantime it is clear that such males are to be found in a variety of situations, and even our present limited knowledge suggests that their study may be a stimulus to specialists of several disciplines, including geneticists, psychiatrists, prison administrators, criminologists, and lawyers.

Summary

Increasing attention is being paid to the raised frequency of 47,XYY males in penal institutions and in maximum security hospitals. This paper reports information on 15 males who have either a 47,XYY complement or who are mosaics with the XYY line predominating. These cases are exclusive of earlier cases reported from the M.R.C. Clinical and Population Cytogenetics Research Unit and from the Scottish State Hospital. It is stressed that there is increasing evidence for these abnormal males being found in groups other than penal groups, and particularly in some hospitals for the mentally subnormal. The report also underlines the fact that these men in the main appear sexually to be normally developed, though information on fertility is scanty, and that there is apparently no characteristic physical phenotype shown by those with a 47,XYY complement.

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