Brit. J. Psychiat. (1977), **131**, 468–77

XYY Males

Some Clinical and Psychiatric Aspects Deriving from a Survey of 1,811 Males in Hospitals for the Mentally Handicapped

By HENRY HUNTER

A survey of 1,811 mentally handicapped males in eighteen hospitals is described. Males discovered with supernumerary Y chromosomes numbered 15, of whom 12 were 47,XYY. Some of the physical, social, psychological and psychiatric findings are described and compared with groups matched for IQ and height.

The main psychiatric findings are diminished intelligence, retardation in development of secondary sexual characteristics, and poor emotional control leading to inadequate social adaptive patterns which are described and discussed.

Introduction

The first XYY male was described in 1961 (24), and subsequently sporadic cases in children and adults were published (2, 6, 7, 15, 23, 25, 26) until the first survey of a Special Hospital (i.e. maximum security hospital) population (13). The incidence of genotype 47,XYY in newborn male populations has been found to be 1 in 700 (22) in a single survey, with a collective incidence of 1·8 per 1,000 (22).

Special Hospital surveys by Jacobs *et al* (13) and Casey *et al* (4) showed a prevalence of 2·9 per cent and 3·7 per cent respectively. Surveys of subnormality hospitals have provided a prevalence of 1·3 per cent 47,XYY males (1) in a 233 adult male population and a nil prevalence (17) in another 694 male unit.

The present investigation forms part of a larger study of sex chromosome aneuploids among the residents of all hospitals for the mentally handicapped within the (former) Sheffield Hospital Region. This area approximates to that of the East Midlands of England together with South Yorkshire, and has a population of some 4·5 million. The number of hospital units included in the survey was 18, and their total male population numbered 1,811 comprising both children and adults.

The survey was commenced in 1968 and completed late in 1972.

Method

Interphase nuclei from buccal mucosae were stained with quinacrine dihydrochloride and were fluoresced according to the method of Pearson and Bobrow (18). All positive and questionable slides were repeated, and subsequent positive results were confirmed by blood culture, using a modification of the method of Moorhead *et al* (16). A minimum of thirty cells were examined.

Results

All males, both children and adults, resident in the eighteen hospitals were successfully included in the survey. Of the 1,811 males examined, those with supernumerary Y chromosomes numbered 15 (0·8 per cent); 12 47,XYY, 2 48,XXYY and 1 48,XYYY. The 48,XXYY and 48,XYYY males have been described elsewhere (12). No children with Y chromosome aneuploidy were discovered.

In height, 1 XYY was 5 ft 10 in and the remainder were over 5 ft 11 in tall; 7 were over 6 ft.

The IQ of the 12 XYY ranged between 46 and 90.

https://doi.org/10.1192/bjp.131.5.468 Published online by Cambridge University Press

TABLE I

IQ= -49	50–59	60–69	70–79	80–89	90–99	Total
1	1	3	5	1	1	12

At the time of investigation the admission category of the XYY to hospital was as follows: 6 informal, 1 Section 6 (MD Act), 1 Section 9 (MD Act), 3 Section 60 (MHA) and 1 Section 26 (MHA).

Discussion

Prevalence

This survey is the largest so far to be published. The overall prevalence of 0·8 per cent shows a marked increase in the number of males with supernumerary Y chromosomes over the 0·18 per cent expected in the normal male population. The prevalence of males with XYY genotype in this survey is 0·7 per cent. Although this represents a concentration of some 3·7 times more than expected, it is far less than the 20·4 times concentration found in the Special Hospitals (4).

The prevalence of XYY males in different hospitals for the subnormal varies considerably. The results of four hospital survey reports in England and Scotland are summarized in Table II.

TABLE II

	N	XYY	%
Scotland:			
Gogarburn Hospital (8) ..	370	0	0
N.E. England:			
Prudhoe Hall (8)	597	0	0
N.W. England:			
Coleshill Hall (1)	233	3	1·29
East Midlands:			
Sheffield Hospital Region	1,811	12	0·7

This distribution is not thought to be geographical or genetic in origin but due to selectivity of admissions. For example, Coleshill Hall Hospital tends to admit a large proportion of patients with behaviour disorder (1). In this survey, of the 12 XYY cases, 8 cases came from two hospitals—Aston Hall and Balderton, both of which tend to admit a high proportion of cases from the Courts (Table III).

TABLE III

Group	Patient population	XYY males N	Patients on section orders N
Aston Hall ..	265	4	41
Balderton ..	256	4	15
Glenfrith ..	435	3	18
St Catherine's	225	0	7
Harmston Hall	425	0	28
Sheffield ..	205	1	20
	1,811	12	129

Too great reliance must not be placed on these figures. Many other factors, e.g. type of offence, age, social background, admission and discharge policy will distort their prevalence within any hospital.

Height

Since all but one of the males with a supernumerary Y chromosome were over 5 ft 11 in tall, clinically, height remains a good marker, especially in the presence of dull normal, borderline or subnormal intelligence.

Intelligence

Penrose (19) suggested that 'there is a tendency for all sex chromosomal errors to reduce intelligence more or less proportionately to the quantity of the deviation from the normal karyotype'. In the case of XYY males there is little deviation in their phenotype, and one would not expect great variation due to an additional Y chromosome, for it is thought to contain little genetic material of importance other than that of testicular formation.

The evidence of a concentration of XYY males in subnormality hospitals, including Special Hospitals for the subnormal helps to suggest that the presence of the supernumerary Y chromosome may tend to diminish intelligence in some cases. Casey *et al* (3) showed that in Special Hospital populations at Rampton and Moss Side the IQs of the XYY genotypes were $79·4 \pm 3·2$ and $72·5 \pm 3·8$ respectively. In this survey the mean IQ was $70·0 \pm 12·3$ supporting the suggestion that in some XYY males there is a tendency for the IQ to be diminished by at

https://doi.org/10.1192/bjp.131.5.468 Published online by Cambridge University Press

least one standard deviation from the general population mean. However, in some cases the supernumerary Y chromosome was associated with diminished intelligence to severely subnormal level of more than three SD from the mean.

The reduction of IQ may have been influenced by hormonal variation (19); it was not possible to measure hormone levels of these XYY subjects.

The Verbal and Performance IQ of 9 of the 12 XYY males were obtained, another had an IQ of 46 on the Stanford Binet. Two were not available for testing (Table IV).

TABLE IV

	WAIS full scale	Verbal	Performance
Case 1	90	88	95
Case 2	77	85	74
Case 3	68	64	57
Case 4	76	67	77
Case 5	71	59	88
Case 6	46*	—	—
Case 7	61	57	71
Case 8	68	76	67
Case 9	74	65	89
Case 10	74	76	75

* Stanford Binet.

There is a tendency for Performance to exceed Verbal scores as found in psychopathic personalities, but the number is too small to draw firm conclusions.

Social factors

An attempt was made to ascertain the social status of the father and whether the family provided an apparently socially stable cohesive unit. Difficulties were experienced in forming such judgements in that some patients had been hospitalized for long periods and relatives had dispersed; and, in general, previous hospital files were inadequate in their social histories. Wherever possible recent local Social Services reports and opinions were sought and in some cases obtained. In one case the elderly parents refused to cooperate saying that they had had enough disgrace brought upon them by their son. Evidence of overt breakdown of the family group was sought in histories of parental divorce or separation, enforced sibling separation, etc. With this relative poverty of detailed knowledge, only a rather broad social assessment could be made as to whether the family group appeared to have maintained itself and thus provided a cohesive background.

An attempt was also made to evaluate whether the family had social problems of such a nature or degree as to need help by one or more Social Service agencies. It is against this scenario of relatively unsophisticated assessment of the social factors that the contents of Table V need to be interpreted.

This table illustrates a wide variation in the social status of the families. Moreover, no evidence emerges of obvious social maladjustment in the sibs or of breakdown of the families as

TABLE V

	Father's occupation	No. of siblings	Antisocial behaviour in other siblings	Family cohesive	Family with social support
1	Self employed	2	No	Yes	No
2	Army regular	2	No	Yes	No
3	Self-employed	2	No	Yes	No
4	School caretaker ..	3	No	Yes	No
5	Labourer	4	No	Yes	No
6	Old-age pensioner ..	3	No	Yes	No
7	Coal carter	5	No	Yes	No
8	Unemployed sick ..	9	No	Yes	No
9	Miner	2	No	Yes	No
10	Chronic sick	5	No	Yes	No
11	NK	2	NK	NK	NK
12	NK	2	NK	No	Yes

(NK = Not known).

https://doi.org/10.1192/bjp.131.5.468 Published online by Cambridge University Press

units. Only one or possibly two families were given support by Social Service agencies.

Control groups

Two control groups, one for height and one for IQ were established from patients within the same hospital and if possible from the same ward as the XYY men. Selection was made randomly by the administrative staff of the hospital. Because of their extreme height and tendency to have IQs higher than the hospital population surveyed, difficulty was experienced in some cases in obtaining controls of close equivalent measurement. In general, the ten controls for height were in hospital because of their poor intelligence. Two XYY men were not available and no controls were sought.

Family factors

Using the same criteria mentioned previously, and with the same qualification to their interpretation, evidence was sought as to the control groups' family cohesiveness and whether Social Service support had been provided.

The families of the height control group, except for one, seemed socially cohesive. Within the IQ control group only five of the ten control families were considered socially cohesive, and only five had not received Social Service support. In this control group, eight had other siblings of whom in three of their families there were other delinquent siblings (Table VI).

The picture that emerges is that the XYY men in this study tend to be 'the black sheep of the family', similar to those described in a maximum security hospital XYY population (20).

Impulsiveness

A strong clinical impression gained by psychiatric examination and by discussion with families and nurses was that these XYY men showed little forethought and acted impulsively. These traits might reflect in early years as truancy and later as abscondence or poor employment records.

Truancy

From hospital notes a record of whether truancy had occurred and its frequency was extracted for the ten XYY men and their controls. In the case of the two XYY men without controls the notes reported frequent truancy for one, but there was no mention of truancy for the other.

Truancy frequency was scored 0 = Nil, 1 = Truancy reported on two or less occasions, and 2 = More than twice. The results are tabulated in Table VII.

These results indicated a markedly greater incidence of truancy among XYY children compared with their controls, though the numbers are too small for the trend to reach a statistically significant level.

Abscondances

Once in hospital the XYY group tended to abscond more than other patients. Because of the paucity of some hospital files it proved impossible to estimate the abscondance frequency in this XYY group. Discussion with their doctors and nurses revealed in some cases a marked tendency to abscond on impulse without planning, usually after a fairly trifling incident. In Special Hospitals, in both Scotland and England, reports show significantly more abscondances by XYY men than among controls (4).

Work record

Of the 12 XYY men, 5 had never been employed; in two cases this was because of low IQ and the other three had histories of

TABLE VI

	N	Families with siblings	Antisocial behaviour in other siblings	Family apparently socially cohesive	Family with social support
XYY ..	10	10	0	10	0
Control height	10	10	1	9	1
Control IQ ..	10	8	3	5	5

https://doi.org/10.1192/bjp.131.5.468 Published online by Cambridge University Press

TABLE VII

Truancy	XYY	Control height	Control IQ
0	4	7	7
1	1	2	1
2	5	1	2
Total	10	10	10

long periods in hospitals. Of the remaining 7 men, 1 had held a labouring job for over two years, 2 had several jobs for up to a maximum of three months, 1 had a job for two weeks and left for no reason, and 3 had held numerous jobs for very short periods of a week or so. From most employment they had been dismissed for inconsistent work, incompetence or fighting; on many occasions, they had just left.

Antisocial behaviour

(a) *Age of commencement.* Examination of the XYY case records showed that there was a tendency for unusual or abnormal behaviour patterns to be initiated at or around the beginning of the teens or earlier. To illustrate this, some early case histories are as follows:

Case 1 Early years, stealing, destructive to property. Between 12–16 years, expelled from three boarding schools, assaults on parents and siblings.

Case 2 Aged 8 years wandered off, arson at 10 years.

Case 3 Aged 9 years, beyond parental control.

Case 4 Aged 10 years, stealing money, unruly.

Case 5 Aged 13 years, stealing bicycles, sexual assault.

Case 6 Aged 14 years, sexual offence (girl 7 years).

Case 7 Aged 10 years, beyond parental control.

Case 8 Aged 5 years, described as very excitable, over-active, very immature, like a two-year-old emotionally.

(b) *Type of offence.* Comparison of the age and type of first offences with their controls shows that the poor behaviour of XYY males tended to bring them before the Courts earlier and more frequently than their controls (Table VIII). Both the XYY men without controls were shown from their histories to have committed several offences in childhood. One had not been brought before the Courts, and the other was treated similarly until aged 24, when he was admitted to hospital after being found guilty of wilful damage.

TABLE VIII

	XYY males		Control height		Control IQ	
	First court offence		First court offence		First court offence	
	Age	Type	Age	Type	Age	Type
Case 1 ..		N*	22	Stole cars	18	Forgery
Case 2 ..	10	Arson, house-breaking		N	18	Stealing bicycles
Case 3 ..	19	Larceny of money	22	Sex assault		N
Case 4 ..	21	Indecent assault		N		N
Case 5 ..		N		N		N
Case 6 ..		N		N		N
Case 7 ..	13	Breaking and entering	30	Indecent assault		N
Case 8 ..	13	Sex assault		N	9	Breaking and entering
Case 9 ..	13	Sex assault	19	Theft		N
Case 10 ..	12	Trespassing		N	17	Stealing car False 999 calls
Total ..	7		4		4	

* No recorded court offences.

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Puberty and Psychosexual development

In this study an attempt has been made to assess the age of onset of puberty by obtaining, where possible, the age of commencement of shaving. Information was obtained from the patients, clinical notes, ward nursing staff and relatives. Of the 12 XYY men 11 were shaving; the 12th, although 19 years old, had not yet commenced. Two said they started at 15 years, 5 at 17 years, 1 at 22 years and in 3 no age of commencement could be obtained.

It is of interest to note that although 11 were shaving, one was said by the nursing staff not to need to do it, 3 shaving only twice a week and 1 only once weekly. The impression gained was that the onset of puberty in this group of males tended to be delayed to a greater or lesser extent in at least half of them.

An estimate of sex drive and orientation was attempted, using information provided from families, ward files and nursing staff in conjunction with clinical and psychiatric examination. Nurses who knew the ward and its population well were asked independently whether the patients actively sought encounters with either sex, or whether they tended to show lack of interest or passivity, as compared with their ward co-residents. The nurses were then asked to judge the patient's degree of interest or activity as a little or markedly below the general level of the ward's population, or noticeably or excessively above that level. There was a remarkable degree of conformity in the estimates expressed by the different nurses. The nursing observations were discussed and confirmed with the patients' consultant psychiatrist. The consensus estimates were expressed on a four-point scale, 2 being the modal or 'average level' of sex drive for the ward population, 1 slightly below this, 0 equates to no sex drive or interest exhibited by the patient; and similarly, 3 was above the general ward level to a marked degree and 4 to an excessive degree. This method of evaluating sex drive is subjective, but since it was made by professional observers who knew their patients, and since nearly all controls were from the same wards, it is felt that some comparison of the estimates can be made.

Sex orientation was estimated by the Kinsey Scale (14), where 1 represents total heterosexual and 6 total homosexual orientation (Table IX).

The modal value '3' on the sexual orientation scale may in this case reflect, not equal inclination towards males and females, but rather lack of orientation, possibly secondary to prolonged stay in hospital and sexual segregation.

The results show that in these XYY men there is a tendency to delay in development of secondary sex characteristics. Within the limitations of the assessment methods their sexual drive tended to be equal to or slightly below the level of their ward co-residents and where firm drive existed it tended to be heterosexual rather than homosexual in orientation.

Aggressiveness

The histories of the XYY men and their controls were examined for acts of aggression

TABLE IX

Patient	Sex drive			Sex orientation		
	XYY	Control height	Control IQ	XYY	Control height	Control IQ
1	2	1	2	1	2	2
2	1	1	3	5	4	5
3	0	3	1	3	3	2
4	2	2	0	1	5	0
5	1	2	1	3	2	3
6	0	0	0	3	3	3
7	1	2	0	3	3	3
8	2	1	0	2	2	3
9	3	2	2	1	2	2
10	1	1	1	2	3	3

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against either persons or property. In addition, using observations from patient interviews and from their nursing staff, an attempt was made to estimate and grade the XYY males and the controls for aggressiveness. In obtaining staff views care was taken not to suggest that aggressiveness was expected, and they were asked to compare the patient's behaviour with that of the other ward residents. If the staff described the patient as aggressive, this was teased out in further discussion as to whether the staff member was describing a general attitude of truculence or belligerence, verbal aggression or physical aggression. In each case several nurses' views were sought independently. In general, little weighting was given to the general attitudinal description and more to the description of the reaction to different amounts of stress by verbal or especially physical aggression, as compared with the behaviour of the other residents of that ward. The opinion of their consultant psychiatrists was also sought.

From all these sources an agreed consensus opinion was formed for each individual on a four-point scale: 0 = None, 1 = some, 2 = ordinary or usual, 3 = marked and 4 = severe tendency to aggressive acts as compared with the general level of aggressive type activity of the ward residents. Although a level of aggressiveness is difficult to describe, there was no difficulty experienced by the staff, who seemed to understand what it meant in terms of their experience of the patients on their ward.

This type of evaluation is mainly subjective, but since it is founded on a consensus view of skilled observers who knew all their patients well and since in nearly all cases the XYY males and their controls were in the same close environment and subject to the same type of stresses, a comparison can be made between these different patients. Interpretation of the results, however, must be cautious in view of the method and circumstances surrounding the patients in their different institutions (Table X).

These results illustrate a marked tendency for the XYY men to be considered as more aggressive than their controls. The staff described the aggression as both physical and verbal, with a tendency more towards physical than verbal. Thus, out of the 8 noted for their aggression 5 would act out physically and/or verbally and 3 usually physically only.

TABLE X

	XYY	Control height	Control IQ
Case 1	3	1	0
Case 2	3	2	3
Case 3	3	3	0
Case 4	4	2	0
Case 5	2	1	2
Case 6	0	0	1
Case 7	3	2	2
Case 8	3	2	3
Case 9	3	1	1
Case 10	3	2	1

Where aggression occurred it tended to be open and against the person rather than surreptitious and against objects. These findings tend not to be confirmed by the results at the Special Hospitals (4). The criteria for assessment in the two studies are different, with recorded police offences in one and inclusion of clinical and nursing evaluation in the other.

When aggression against the person occurred it was usually against children, other patients and staff. A distinct impression was received that the aggression was directed against others who were smaller and less able to stand up for themselves, e.g. children, weaker patients, inexperienced student nurses and physically smaller parents. In three of them, aggression was only seen or made worse after alcohol.

Interpersonal relationships

At psychiatric interview all the XYY men tended to be suspicious, and some were belligerent. With a few a degree of rapport could be established and developed after several interviews. This pattern contrasts with 47,XXY cases previously described (10), with whom little rapport was possible.

A five-point scale was constructed to try to record observers' estimates of the interactions between the XYY men and their peer group. Appropriate nursing staff were asked to make assessments on the following rating scale: 0 = Avoids people, 1 = Does not avoid people, 2 = Makes non-specific relationships with peers, 3 = Makes specific friends, 4 = Makes deeper

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friendships. The results of 10 XYY men and their controls are shown in Table XI.

Although these figures are small, they do show a tendency in the XYY men, when compared with controls, to mix with others but not join with them and this tends to support a similar clinical impression. However, variation did occur and, as has been described, rapport was established with some of the men.

TABLE XI

Rating	XYY	Control height	Control IQ
0	1	1	1
1	5	2	2
2	3	6	6
3	1	1	1
4	—	—	—
Total	10	10	10

Some other clinical findings

All patients were physically examined, and in general few overt clinical differences between the XYY men and their controls were noted. However, four XYY men were noted as muscularly strong, whereas this feature had not merited report in any of the controls.

Three XYY men tended to perspire a great deal and exhibited marked tenseness well beyond what might have been expected in the circumstances. In these men these phenomena were observed regularly at repeated interviews.

Prognosis

It was possible to follow up some of the XYY men after hospital discharge. One, leaving hospital at age 30, had been in regular employment for about three years, another has worked regularly since leaving hospital at age 31, for some two and a half years. A third has returned to his family after many years in hospital; a fourth has maintained himself in work for the last two years after leaving hospital aged 19 years. In general, an impression was gained that with increasing age this group of XYY men will tend to exhibit less abnormal or unusual behaviour patterns and impulsiveness.

Conclusions

This paper reports on a special group of people, i.e. residents in hospitals for the mentally subnormal who have been surveyed for XYY aneuploidy. The prevalence found has been greater than in the general population, but less than that in Special Hospitals.

In the XYY individuals discovered and studied there do seem to be some behavioural or other defects. There appears to be some depression of intellectual development and a variable delay in development of psychosexual, emotional and social adaptative behaviour patterns. The delayed development can be seen in childhood as poor learning with a high truancy rate.

Adolescence, a stormy period even in the normal, was made more difficult in these XYY males by their immaturity, delayed development of secondary sex characteristics and diminished intellect: poor frustration tolerance leads them to impulsive behaviour such as successively leaving employment and frequent abscondances. The impulsiveness can be of such a degree as to initiate an immediate motor type response, usually demonstrated by physical aggressive acts against the person. Alcohol will tend to loosen the subject's poor control, generating aggressive behaviour. Aggression also tends to be used in a bullying manner to the weaker and smaller, a pattern reminiscent of schoolboys.

Delayed maturation is also observable in these patients' emotional development, creating poor interpersonal relationships, further marred by their impulsiveness, aggressiveness, bullying behaviour and physical size.

In some the immaturity can be seen as a developmental retardation, for in time and with help acceptable social behaviour patterns emerge.

From what has been said so far it would be all too easy to argue for a genetic basis to the behavioural difficulties expressed by this group of XYY men, especially since they seem to have come from homes which provided no obvious gross disturbance or criminal background, and in which they were the 'black sheep'. Is there another aspect which might give a different view on these results?

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In the 4·5 million population in the Sheffield Region, about 2·0 million males could be expected. Since no evidence has been produced of early mortality in XYY men, one might expect a male population numbering 3,600 with this aneuploidy. This figure far exceeds the known cases in Special Hospitals, prisons and hospitals for the subnormal in the Region; this means that most men with 47,XYY aneuploidy are existing unknown in the community and presumably living normal, socially acceptable lives.

The family milieu of the XYY men was cohesive and revealed no antisocial activities in the other siblings, and the description of these homes as 'respectable' cropped up repeatedly. Because they came from 'respectable homes' there was evidence that some parents had higher expectations of their son than he could fulfil. His lack of attainments could not be understood or easily accepted, leading to the parents' bewilderment and their son's increasing frustration, which compounded the situation. In such homes the presence of a single non-conforming incumbent might well be beyond the experience of the other family members, and the parents reported difficulty in coping with situations. This was enhanced by their son's large size and greatly increased by a fear of 'what will the neighbours say', thus leaving the whole situation to be exploited by the bullying child. Submission to the demands of the XYY child would have tended to reinforce his abnormal behaviour patterns.

In attempting to reconcile these views a picture may begin to emerge that in the presence of XYY aneuploidy some males show, besides increased physical growth, a delayed development of secondary sex characteristics. In some there is also a delayed maturation of personality, emotional control and social learning associated with IQ levels lowered to a variable degree. In the presence of an overstressful environment or situation such persons may react in a childish or immature manner with impulsive behaviour of a physically intimidatory or overtly aggressive type, or by physically running away.

In the vast majority of XYY men the effect of the aneuploidy is minimal and their environment such that they develop into individuals indistinguishable by their behaviour from the normal. Of the smaller number of XYY men in whom the interaction with their environment is less helpful and more stressful, difficulties may arise, but there are indications that maturation and emotion control can develop with time and help.

From the present study insufficient evidence exists to establish psychiatric effects specifically due to the presence of an extra Y chromosome in the 47,XYY aneuploidy. Much more detailed investigation will be required to unravel the interaction between the XYY male and his environment.

Where XYY aneuploidy is discovered, general but discreet supervision might be given to the family. If special problems begin to develop then more detailed supervision and counselling may be needed for the family and the affected person.

Acknowledgements

I would like to express my thanks to Mr R. Quaife, B.Sc. who did the cytogenic examinations for the survey and for his invaluable help; to the late Dr K. O. Milner of Aston Hall Hospital; Dr A. A. Valentine of Glenfrith Hospital; Dr M. Quinn of St Catherine's Hospital, Doncaster; Dr E. Gemmell of Sheffield and Dr J. W. Garry of Harmston Hall Hospital who kindly allowed me to examine their patients, and to all the nursing staff in those hospitals who gave unstintingly of their time, knowledge and experience of their patients.

This work forms part of a larger research project financed by the Research Committee of the Sheffield Regional Hospital Board (now Trent Regional Health Authority). The equipment used was provided by a grant from the Mental Health Research Fund.

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(*Received 17 June 1976; revised 10 January 1977*)

https://doi.org/10.1192/bjp.131.5.468 Published online by Cambridge University Press