Polydactyly and psychosis

Five cases of co-occurrence

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Background Abnormalities presumed to occur during foetal life have been associated with schizophrenia. Polydactyly is a developmental abnormality but no previous association has been reported between polydactyly and functional psychotic illness.

Method Individuals with both polydactyly and a functional psychosis were ascertained during a study of familial schizophrenia.

Results Five such individuals were ascertained in the course of assessing 234 individuals with familial psychosis, giving a rate of polydactyly in the sample of around 10 times the general population rate.

Conclusions This study provides preliminary evidence that polydactyly is over-represented in individuals with familial schizophrenia and related psychotic illnesses.

Schizophrenia is associated with subtle neurodevelopmental abnormalities (Murray, 1994) and an excess of minor physical anomalies (O'Callaghan *et al*, 1991), probably caused during foetal development. Polydactyly, the duplication of fingers and/or toes, is also a developmental abnormality, and although development of the skeletal and nervous systems are closely related, we know of no previous report of polydactyly and functional psychosis co-occurring. In this paper we describe five cases where polydactyly and functional psychosis co-occur. An initial case was noted by A.M.C., following which we sought further cases during a study of sibling pairs affected by schizophrenia.

CASE HISTORIES

An Indian male, 64 years old, had post-axial (ulnar duplication) polydactyly on all limbs. His paternal grandmother also had an extra digit on all limbs, but his father was unaffected. He had nine siblings, of which the youngest sister was reported to have extra toes bilaterally, and to fear contamination, but without psychotic symptoms. At 56 years, the subject developed delusions which continued chronically; he also suffered depressive symptoms, and one episode of manic symptoms. DSM–IV diagnosis (American Psychiatric Association, 1994) was psychotic disorder not otherwise specified.

An Afro-Caribbean/Caucasian male, 37 years old, had post-axial polydactyly on both hands. His father had extra fingers bilaterally. He had four siblings: one brother also had extra fingers, and another brother had an extra digit on all limbs, the latter brother had an unknown number of daughters with polydactyly. His mother had an unknown psychiatric illness, possibly psychotic, and the brother with four extra digits suffered from schizophrenia. At 26 years, the subject developed hallucinations and illogical speech, which continued chronically. The diagnosis was schizophrenia.

A Caucasian male, 33 years old, had post-axial polydactyly on the left hand. His mother and maternal aunt had extra fingers bilaterally, and both suffered from depression. He had three siblings, of which one brother had schizophrenia; also his paternal uncle had manic depressive illness, but neither has polydactyly. At 20 years, the subject suffered labile moods, then delusions which responded to medication. The diagnosis was psychotic disorder not otherwise specified.

An Afro-Caribbean male, 37 years old, had post-axial polydactyly on the left hand. There was no family history of polydactyly. His mother had schizophrenia. He had five siblings, including one brother who had schizophrenia. At 25 years, the subject developed hallucinations, delusions and formal thought disorder, which continued chronically. The diagnosis was schizophrenia.

A Caucasian male, who was 41 years old, had pre-axial (radial duplication) polydactyly on the right hand. There was no family history of polydactyly. He had two brothers, one had schizophrenia, the other committed suicide. At 21 years old, the subject developed hallucinations and delusions, which continued chronically. The diagnosis was schizophrenia.

DISCUSSION

The incidence of polydactyly is around 2/1000 births (Nelson *et al*, 1996), but that of post-axial polydactyly is higher in Blacks than Whites (3.6/1000; Taeusch *et al*, 1991). The five probands were ascertained in the course of assessing 234 individuals with schizophrenia or related functional psychoses from 124 families, nine individuals being Afro-Caribbean or of mixed ethnicity. Thus, polydactyly and psychosis co-occur in this sample around 10 times more frequently than expected (P=0.0003).

These data should be viewed with caution because the sibling sample was not epidemiologically-based, demographic variables were not controlled for, and this was a chance finding in the course of measuring a range of clinical variables, so replication is required. However, there was no prior expectation of an excess of individuals with polydactyly that might have introduced an ascertainment bias. The number of probands

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is too small to know whether their clinical features constitute a distinct syndrome, but all were male, four out of five had post-axial polydactyly, and four out of five had a chronic illness. The finding, therefore, provides preliminary evidence of an over-representation of polydactyly among individuals with familial schizophrenia and related psychoses.

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