

Psychomotor seizures, arterio-venous malformation and the olfactory reference syndrome

A CASE REPORT

B. K. TOONE

A case history is reported of a 44-year-old man with a 6-year history of psychomotor seizures. For the past year he had described persistent olfactory hallucinations of an unpleasant nature which he referred to himself. In many respects these symptoms conform to the pattern observed in the olfactory reference syndrome, a recently described but apparently quite discrete psychiatric disorder; there were, however, certain atypical features. At a later stage unilateral anosmia was noted. Radiological examination then demonstrated an arterio-venous malformation in the right frontal lobe. The relationship between the malformation and the psychomotor seizures, and the implications that each has for the development of an olfactory reference syndrome are fully discussed.

Key words: Anosmia – arterio-venous malformations – automatisms – hallucinations.

During the first third of this century descriptions of a syndrome characterized by a persistent morbid preoccupation with personal odour at times approaching levels of delusional intensity appeared in a number of case reports in the French and German literature. Various descriptive titles were applied (*Souchanoff* (1906), *Birnbaum* (1915)) and classifications attempted (*Reichardt* (1918), *Mayer-Gross* (1932)), but a recognition of the syndrome as a discrete nosological entity was delayed until the publication of a short series of case studies by *Videbech* (1967) who introduced the term "olfactory paranoid syndrome". A later survey (*Pryse-Phillips* (1971)) emphasized the distinctive characteristics of the syndrome: unlike schizophrenia the concern with body odours remains the dominant symptom throughout, the patient does not regard the smell as an imposition by some external agency but holds himself solely to blame. His response is correspondingly contrite and apologetic. The olfactory reference syndrome, to use this author's terminology, may also be distinguished from depressive disorders in which the affective disturbance is primary. As this extensive survey provides the most detailed description of the syndrome, the title suggested by *Pryse-Phillips* will be the one used here.

The case reported here is of interest partly because there are certain atypical features which may reflect the presence of an underlying organic lesion, but also

because it introduces the possibility that the development of an olfactory reference syndrome may in part be dependent on certain physical as well as psychological factors.

CASE REPORT

The patient, a 44-year-old male upholsterer, was first referred to a psychiatrist at the age of 38 for the investigation of episodes of altered consciousness during which, according to eye witnesses, he would look blank, talk irrelevant, fumble with his hands and on occasions indulge in bizarre behaviour. Once he picked up a lighter and attempted to set fire to the Black Jack table at a nightclub; another time he was found unscrewing a door in a hotel bedroom for no apparent reason. At the time he seemed unaware of what was going on. His only complaint was of amnesic spells, lasting for up to 15 min and on two of these occasions he had been incontinent of urine. Such episodes occurred every 1-2 months. There was no previous record of psychiatric illness in the patient or his family and his birth and upbringing were quite unexceptional. He was described as an easy-going but rather egocentric man. He was at times inclined to drink rather heavily. He changed jobs frequently but was rarely unemployed. He had been married and had two children but was now divorced.

No abnormality was found on examination. A diagnosis of temporal lobe epilepsy was made and this was supported by EEG investigation. Although a routine waking EEG was normal, a sleep recording showed numerous rhythmic sharp wave forms in the mid-temporal regions bilaterally though more marked on the right. A bitemporal-sphenoidal EEG showed the same sharp waves at the sphenoidal electrodes. Routine haematological investigations, blood WR, plain chest and skull X-rays were all normal. Psychometric testing indicated a full scale IQ of 116 on the WAIS (verbal 118; performance 110). Performance on the Wechsler Logical Memory test, Rey-Osterreith test, Walton Black Modified Word Learning test and Wisconsin Card Sorting test, was not impaired. His score on the Benton Visual Retention test was less than expected taking his overall IQ into account, but this was not thought to be of any significance.

In view of the late onset of epilepsy the need for more detailed radiological investigation was discussed with the patient but permission for this was refused. He was treated with combined anticonvulsants and the frequency and severity of the attacks diminished although they never disappeared altogether. This state of affairs persisted for 5 years.

Six months prior to the present admission a new symptom appeared. The patient began to notice that he gave off a peculiar, burning odour (like old leaves or an old bonfire). This followed him around wherever he went and he assumed that he must be responsible for it as no other explanation was forthcoming. His awareness of the odour was to a considerable extent situationally determined; although he could from time to time perceive it when he was alone, he became immediately more aware of it when he was in the presence of others, particularly if they were strangers. Moreover if someone in his presence sniffed or sneezed he was instantly conscious of the odour. He began to watch for any suggestion on the part of others that they had become aware of the smell and were offended by it. He felt that people were avoiding him. He said that when he entered a room someone would straight away open a window. On one occasion, on entering a public house, the publican turned on the extractor fan; he took this as indubitable evidence that the odour that he gave off was also discerned by others and he took to shunning all company and to drinking by himself. He also began to use an amount of deodorant that he later admitted to be excessive in an attempt to counteract the smell, although he did not attempt to explain this in terms of failure of personal hygiene or biological function. He badgered his friends and relatives, demanding to know if he smelt, and refusing to believe them when they attempted to reassure him. With his brother he almost came to blows on one occasion. Although prone to misjudge the actions of others, ideas of reference, drawn from verbal sources, were notably absent. Snatches of overheard

conversation might arouse his suspicions but did not lead to more specific misinterpretations.

Over the same period he noted other disturbances in his sense of smell. From time to time naturally perceived odours were experienced with an intensity that was quite unexpected: a woman's perfume or a smell of baking bread as he passed a bakery. Pleasant odours were as likely to be enhanced as disagreeable ones. He also noticed that at other times his sense of smell was definitely deficient and odours which he would have expected to perceive went unnoticed. Still more remarkably he found that on some occasions he perceived pungent odours which on further investigation proved to be completely unfounded. On entering a public house he was overwhelmed by the agreeable aroma of a Sunday joint in preparation. He complimented the landlord only to be informed that no cooking was taking place on the premises.

These symptoms became increasingly intrusive and eventually he reckoned that for about one quarter of his waking hours he was acutely aware of powerful odours, particularly those which he attributed to his own body. He continued to have occasional psychomotor seizures. These would occur without any warning and upon recovery were experienced only as an unaccountable gap in time. At no point was there an olfactory component and their occurrence was quite unrelated to the persisting disturbance in olfaction described above.

On examination the only abnormal finding was a virtually complete right-sided anosmia for coffee, vanilla, camphor and aniseed. These substances when presented to the left nostril were readily identified. Full neurological examination was negative. No bruit could be detected over the front of the head and no abnormal scalp vessels were to be seen. The optic fundi were normal.

Investigations. Routine haematological investigations, serum folate and B 12, liver function tests, plain skull and chest X-rays and routine waking EEG showed no abnormality. A Gamma scan showed a large lesion in the medial and lower part of the right frontal lobe and a carotid arteriogram showed an arterio-venous malformation in the same area (Fig. 1). The psychometric tests confirmed results obtained on the previous occasion.

Management. It was felt that, in the absence of any symptoms suggestive of an intracranial haemorrhage, surgery was not justified. The patient was told that his olfactory experiences were not based on any objective reality; in part they were due to the presence of an abnormal circulation in a part of his brain and the effect this might have on the way he perceived things. He showed some surprise but was willing to accept this explanation. However, at follow-up 4 months later he admitted that he still had some doubts and had continued to use an excessive amount of deodorant to eliminate any unpleasant body odours.

DISCUSSION

This case report prompts a number of questions. Firstly does the syndrome described here meet the criteria for a diagnosis of olfactory reference syndrome as defined by *Pryse-Phillips* (1971)? The diagnosis may be made on the basis of positive criteria: olfactory hallucinations or delusions should be present and should refer to an odour, usually unpleasant, which the patient believes to be his own and for which he assumes responsibility. This should be the principal complaint and the cardinal feature of the illness at its inception and throughout its subsequent course. Other psychiatric conditions such as schizophrenia, affective disorders and temporal lobe epilepsy in which olfactory hallucinations may occur should be excluded.

This patient's symptoms conformed to the picture described above in most

respects. But in two important details they differ. The content of the hallucination and the interpretation placed upon it varied; most commonly the perceived odour was unpleasant, was referred to the patient's own body and became marked during stressful circumstances. Occasionally, however, the hallucination occurred in an emotionally neutral situation, concerned odours which were not necessarily disagreeable and which were referred by the patient to the outside world. Although the latter type of experience is not a feature of the olfactory reference syndrome it is also highly atypical of the other conditions in which olfactory hallucinations occur. The patient experienced the hallucination as a normal and thoroughly commonplace event and placed no special significance upon it. It was only the response of others who did not share his experience that led him to recognize its unusual nature. In this the characteristics of the phenomenon differ from olfactory hallucinations which occur in schizophrenia and temporal lobe epilepsy. The patient also noticed that on occasions his sense of smell was quite deficient. The possibility that this unusual feature may be related to the presence of an underlying organic lesion will be commented on later.

In other, perhaps less distinctive ways, this patient's history departs from the classical description of the olfactory reference syndrome. Numerous authors have commented on the premorbid personality and their observations have shown a remarkable consistency. Such patients are notable for their moral scrupulousness but also for their keen sense of failure (*Landis & Bolles (1946), Schneider (1959)*). *Langfeldt (1951)* has referred to the "Sensitive-Reaction type" and has suggested a constitutional basis. In this patient's history there is evidence of a somewhat turbulent personality, an unstable work record, a broken marriage and a tendency to drink heavily, but little to suggest the presence of character traits of the kind described above. On the contrary he was said to be of a care-free, easy-going disposition although somewhat egocentric. Finally his ready acceptance of the explanation of his symptoms is very much at variance with the intractable beliefs held by patients with the olfactory reference syndrome. However, it must be admitted that some reassurance was required from time to time as his doubts were prone to return.

However, notwithstanding these reservations, a diagnosis of olfactory reference syndrome, albeit with some atypical features, comes closest to summarizing the psychiatric symptoms reported here. In that case three separate diagnoses have been made: olfactory reference syndrome, psychomotor epilepsy and a frontal arterio-venous malformation. Are these conditions aetiologically independent of one another or are they in some way related? There are grounds for supposing that the psychomotor seizures are symptomatic of the vascular malformation. It is recognized that lesions of the frontal lobes may produce features more commonly associated with temporal lobe pathology (*Schneider et al. (1961)*). *Hill & Driver (1962)* have drawn attention to the fact that ictal automatisms may be initiated by epileptic foci in the frontal and orbito-frontal areas. Moreover epilepsy is not uncommonly a first manifestation of arterio-venous malformations. However, the relevance of these two conditions for the later development of an olfactory reference syndrome is more speculative. Expanding lesions,

A



B

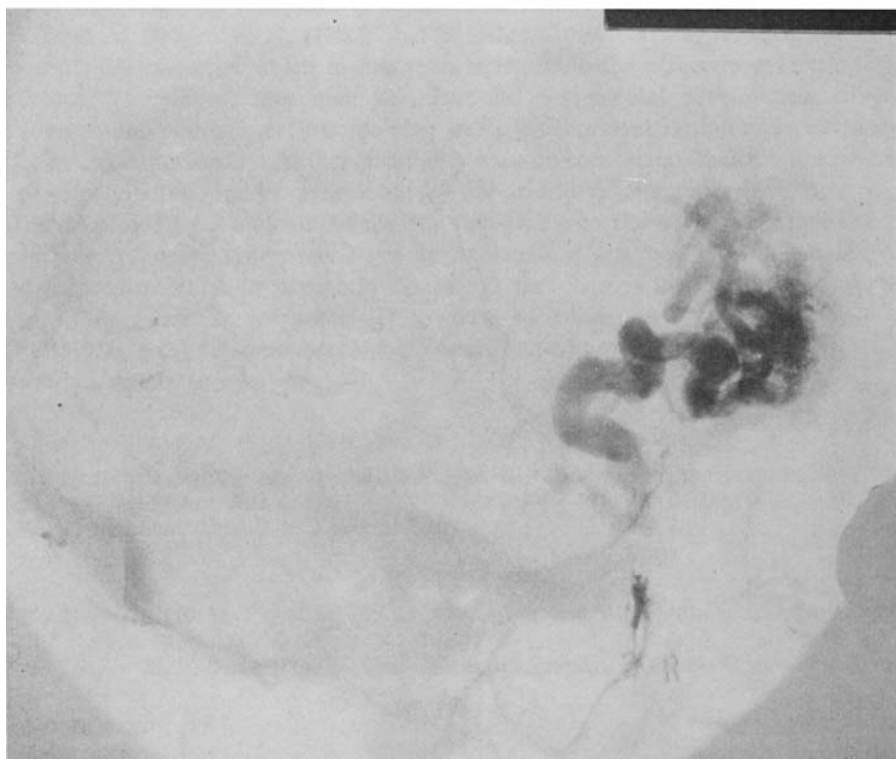


Fig. 1. Subtraction carotid angiography showing an arterio-venous malformation in the right frontal lobe. *A*, arterial phase of filling. *B*, venous phase of filling.

particularly olfactory groove meningiomas, may cause olfactory symptoms through pressure on the olfactory nerve but this usually takes the form of varying degrees of anosmia, or less commonly, parosmia. No cases have been described with olfactory hallucinations, or of olfactory delusional syndromes. This patient experienced, in addition to a persistent and disagreeable odour which he referred to himself, fleeting disturbances of olfactory perception which he attributed to sources outside himself and which were lacking in personal significance. That such symptoms could have an organic basis seems worth considering. If this was so their ephemeral nature suggests that they may be caused by ischaemia due to local vascular shunting rather than by a direct pressure effect.

Occasionally he noticed that his sense of smell was deficient. This is in accordance with the finding of unilateral anosmia and is likely to have an organic explanation.

Whether such a structural lesion could also be responsible for the psychiatric symptoms more typical of the olfactory reference syndrome is uncertain. Previous surveys (*Videbech* (1967), *Pryse-Phillips* (1971)) have not described any cases in which an organic aetiology was considered likely: on the other hand it is unusual for such patients to undergo extensive neuro-radiological investigation. Even so such an association must be considered distinctly uncommon.

It is worth considering whether certain forms of prior perceptual experience may directly influence the development of the psychotic illness. Following this approach *Pond* (1962) has suggested that the ictal experience of temporal lobe epilepsy may lead in time to emergence of chronic paranoid hallucinatory psychosis. This is unlikely to be the case here as the description of the ictal events lack any olfactory component. Of more relevance to this case are the observations of *Orbach et al.* (1952) that colostomy patients may develop olfactory paranoid syndromes which in many ways resemble the olfactory reference syndrome. Therefore rather than postulate the coincidental development of two uncommon disorders, each containing as its predominant symptom an abnormal olfactory experience, a further possibility may be considered, that the disturbance of olfactory function brought about by the arterio-venous malformation is capable of providing a biological substrate favourable to the development of the olfactory reference syndrome. Such an explanation assumes some degree of innate predisposition to psychotic illness but less than is usually the case in the olfactory reference syndrome. This would be consistent with the presence of certain atypical features, namely the uncharacteristic premorbid personality and the late age of onset.

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B. K. Toone, M.B.B.S.
The Maudsley Hospital
Denmark Hill
London SE5 8AZ
England