We wish to thank Dr. M. D. Sanders for the neuro-ophthalmological opinion and for the retinal photographs.

REFERENCES

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Malignant Nonchromaffin Paraganglioma

This case is reported because of its rarity at this age, and also because of the response to cytotoxic drugs.

Case Report

A 5-year-old boy who had previously enjoyed good health was referred to hospital with a complaint of cough for one month. His family doctor had also heard a cardiac murmur. The cough was irritating and nonproductive. Examination confirmed the presence of the murmur, early systolic in timing, and heard loudest at the 2nd and 3rd left interspaces and in the left scapula region. Clinical examination of the chest revealed no abnormality; the only other physical sign being a small hard lymph node in the right supraventricular region.

A chest x-ray showed collapse of the left lower lobe and widening of the mediastinum. He was admitted in February 1970 with a working diagnosis of tuberculous hilar lymph nodes giving rise to compression of the bronchus.

Investigations showed Hb 12·4 g/100 ml; WBC 11,000, normal differential; ESR 33 mm/hr; Heat test, negative; ECG, a moderate degree of left ventricular hypertrophy; blood pressure 100/60 mmHg, both arms and legs; catecholamine excretion levels within normal limits; IVP no abnormality.

During the next 10 days the clinical signs altered; initially collapse of the left lower lobe became more obvious, and then obstructive emphysema of the whole of the left lung appeared. On 9 February 1970 bronchoscopy was performed (Mr. D. J. Waterston). This showed a normal right main bronchus but the left main bronchus contained easily bleeding granulation tissue. Biopsy was taken from this, and the lymph node in the neck was also removed.

Histology. The tumour was composed of rounded groups of cells bounded by reticulin and collagen fibrils. The cells were large and had granular cytoplasm. The nuclei were large with marked nuclear pleomorphism and mitotic figures. There was necrosis of individual cells scattered through the tumour.

The patient was treated at St. Bartholomew's Hospital by irradiation (2500 rads in 30 days to neck and mediastinum), with relief of symptoms, but the cough returned after one month and x-ray showed a large mass in the region of the left main bronchus. Thoracotomy revealed collapse of the left lower lobe, and inoperable tumours at the hilum and at the tracheal bifurcation.

On 22 April 1970 treatment with intravenous vincristine, 2 mg/m² was started. On 10 June, intravenous cyclophosphamide 300 mg/m² was added. Injections were given fortnightly until September, when the chest was radiologically clear, and thereafter monthly. The cardiac murmur had disappeared by February 1971. In the following September he had varicella and rubella, but in October he appeared well. He had gained 3650 g in weight and grown 8 cm in height since falling ill 21 months before. When last seen in February 1972 he was well.

Discussion

The ganglia and paraganglia in relation to the autonomic nervous system both derive from neural crest cells. The former are part of the sympathetic chain while the latter are either (1) chromaffin-positive and endocrine-secreting, or (2) chromaffin-negative and functioning as neuro- or chemoreceptors (Haber, 1964). The nonchromaffin paraganglia have also been called 'chemodectoma' by Mulligan (1950) for this reason. Boyd (1937) described the embryology and phylogenetic development from nerves supplying the gills, which explains the distribution around nerves and vessels derived from branchial arches.

Smithers and Gowing (1965) reviewed 28 cases of chemodectoma in the region of the aortic arch; of these, 11 had survived some years after surgical excision. The others had died with varying degrees of complications attributable to the tumour. Both multicentric origin and metastatic spread have been reported (Smithers and Gowing, 1965;
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Haber, 1964; Brown, Burton, and Dahlin, 1967), though metastases are considered rare. They often remain asymptomatic until the size of the tumour produces pressure symptoms (Haber, 1964). In our case, metastasis to local nodes was proven. Surgical excision is the favoured form of treatment (Brown et al., 1967), though the vascular nature of the tumour and its proximity to vital structures may make this impracticable as in our case. A poor response to irradiation is to be expected (Gillis, Reynolds, and Merritt, 1956).

To our knowledge, this is the first reported case in which cytotoxic drugs have been used. To date it seems to be successful. The dosing schedule is an arbitrary one, designed to recognize that these tumours are slow growing and that metastatic lesions have sometimes appeared years after the primary excision.

Summary

A case of malignant nonchromaffin paraganglioma is described in a 5-year-old boy. Retroperitoneal examples of this tumour are very rare, and this is probably the youngest patient recorded. The successful response to cytotoxic drugs is also hither-to unrecorded.

References


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Evaluation of Dip Inoculum Urine Culture

Infants and young children cannot always be persuaded to pass urine at a given time, and do not respect laboratory hours. Moreover, once a urine sample has been collected, there is frequently a delay before its arrival in the laboratory for conventional plating. In an effort to overcome some of these problems the dip inoculum method of urine culture has been advocated (Mackey and Sandys, 1965; Arneil, McAllister, and Kay, 1970; Mabeck and Mabeck, 1970; Wille, Scharer, and Bickel, 1970). This communication reports a small trial in paediatric inpatient and outpatient practice.

Method

The Uricult dip-slide was used with MacConkey medium on one side and nutrient agar on the other. Fresh, clean urine samples were obtained from 73 infants and children of both sexes. The dip-slide, which was inoculated immediately, was then dispatched together with the clean urine sample to the bacteriology laboratory. The urine sample was then plated in the routine fashion, and plate and dip-slide incubated for 18 hours at 37 °C. After incubation, all the slides were read by one bacteriologist, while the routine plates were read independently. The time interval between inoculation of the dip-slide and plating of the paired urine samples was known accurately for 15 of the 73 specimens.

TABLE

Comparison of the Results from Dip-slide and Routine Cultures

<table>
<thead>
<tr>
<th>Routine Culture</th>
<th>Dip-slide Culture</th>
</tr>
</thead>
<tbody>
<tr>
<td>NG</td>
<td>18</td>
</tr>
<tr>
<td>SG</td>
<td>1</td>
</tr>
<tr>
<td>NSG</td>
<td>36</td>
</tr>
<tr>
<td>SG</td>
<td>2</td>
</tr>
</tbody>
</table>

Note: NG, no bacterial growth; NSG, no significant growth; SG, significant growth.

Results

The results are shown in the Table. Dip-slides and routine cultures were interpreted as showing no bacterial growth (NG), no significant growth (NSG), or significant growth (SG). There was complete agreement in 63 cases (86%); the dip-slide gave 7 falsely positive SG's (9.6%) and 2 falsely negative NSG's (2.6%).

The mean time interval between dip-slide and routine plate inoculation in the 15 cases was 2 hours 30 minutes, with a range from 1½ hours to 4 hours. There was complete agreement between dip-slide and plate in 14 of these cases; in one case, where the interval was 4 hours, the dip-slide showed SG while the routine culture showed NSG.

Discussion

The present study largely confirms previous work in that the dip-slide culture gave comparable results to routine cultures (Mackey and Sandys, 1965; Arneil et al., 1970; Mabeck and Mabeck,
Malignant nonchromaffin paraganglioma.

P De Buse

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