EPULIS IN THE NEWBORN

BY

F. A. LANGLEY and J. DAVSON

From the Departments of Obstetrics and Gynaecology and of Pathology at the Victoria University of Manchester

(RECEIVED FOR PUBLICATION, JULY 16, 1949.)

Epulis in the newborn is a comparatively rare condition and, although such tumours have been reported in the American and continental journals, the only reference in the British literature is that given by Willis (1948), who, discussing the status of 'myoblastoma', refers to papers by Ceelen (1931), Meyer (1932), and Crane and Tremblay (1945), describing cases of congenital epulis.

Case Reports

Case 1. K.G. was a female infant who, at birth, was found to have a small mass attached to the left lower alveolar margin by a narrow neck. Ten days later the rounded solid mass, measuring approximately 1 cm. in diameter, was removed, and when sectioned it was pale and firm.

Histology. Much of the outer surface of the tumour was covered with squamous epithelium, but in one part this had been lost by ulceration and replaced by fibrinous exudate. Where the epithelium was intact the tumour cells lay closely beneath but were clearly separated from the basal epithelium (fig. 1). The tumour cells were large and mainly polyhedral in shape measuring up to 36 μ; others were more elongated and very occasionally spindle-cell in shape. The more or less centrally placed nuclei were slightly oval, measuring 6-10 μ, with well defined nuclear membranes. No mitoses were seen. The cytoplasm of the cells was filled with fine acidophile granules having no special arrangement so that the cells somewhat resembled xanthoma cells (fig. 2); but Sudan stains on frozen sections failed to demonstrate the presence of lipid. The cytoplasmic granules stained blue with Heidenhain's azan and faintly pink with muci-carmine. Sections stained by Gomori's silver technique showed that the cytoplasm contained fine granules sometimes arranged as rods, at other times as circles (fig. 4), and also that each cell was surrounded by a distinct reticulin framework (figs. 3 and 4).

In sections stained by Heidenhain's iron haematoxylin to demonstrate myofibrils it was found that the cytoplasmic granules failed to retain the stain after normal differentiation, and no longitudinal or cross-striated myofibrils could be demonstrated.

Case 2. This baby, C.V., was admitted to the Duchess of York Hospital for Babies, Manchester, at the age of two days, having had a normal delivery at full term; the birth weight was 7½ lb. On admission she was seen to be a normal-looking, healthy baby apart from two pedunculated tumours arising from the upper alveolus to the left of the mid-line. The anterior tumour measured 4 cm. by 1 cm. and the posterior one 1·2 cm. by 0·8 cm. No other abnormality was found. The tumours were removed under local anaesthesia at the age of four days. The tumours were firm in consistency and the cut surfaces were uniformly grey. The child was discharged two days later and made a straightforward recovery. She was seen in the out-patient department at the age of three and a half months; at this time radiographs of the long bones and skull were taken but no abnormality was found. This investigation was made because at this time the true nature of the tumour was not understood and on account of the histological picture a diagnosis of multiple xanthomatosis was considered possible. The child was seen again at nine months of age; her weight was 24 lb. 8 oz. and she had infantile eczema. She was seen for the last time one month later when the eczema was still present. There was no recurrence of the tumour.

Histology. The outer surface of the tumour was covered with squamous epithelium. This was separated from the tumour by a thin zone of connective tissue from which coarse strands descended into the substance of the tumour. No true lobulation was produced but these coarse strands broke up into fine fibres which surrounded each tumour cell. This pericellular reticulum was readily seen in the silver-stained preparations. The cells of the tumour were identical in morphology with those in Case 1.

Case 3. This child, H.M.S., was brought to the out-patient department of the Duchess of York Hospital, Manchester, at the age of five days. A pedunculated lump, about the size of a large pea, arose from the central position in the upper jaw. Next day the lump was removed under local anaesthesia in the out-patient department. The baby has been seen subsequently at four and six months of age. There is no sign of local recurrence of the tumour, and, apart from being slightly over-fat, she is in excellent health.

Histology. About half of the surface of the tumour was covered by squamous epithelium; over the remainder of the surface the epithelium
Fig. 1.—The tumour cells can be seen lying below the basal layer of the buccal epithelium; stained with haemalum and eosin. ×200.

Fig. 2.—The tumour cells show some variation in shape, and contain abundant granular cytoplasm; stained with haemalum and eosin. ×290.

Fig. 3.—A silver impregnation stain shows that each cell is surrounded by a reticulin fibre mesh; stained with Gomori's reticulin stain. ×200.

Fig. 4.—A higher magnification showing the numerous argyrophilic granules in the cytoplasm of the cells; stained with Gomori's reticulin stain. ×800.
had been lost and replaced by fibrinous exudate. The body of the tumour was separated from the epithelium by a thin zone of connective tissue into which the inflammatory process extended. The cells of the tumour showed identical appearances with those of Cases 1 and 2.

Discussion

The three neoplasms occurring in the jaws of newborn infants showed identical morphological characteristics. The tumours were unencapsulated and the cells lay beneath and distinct from the overlying buccal epithelium. The tumour cells were large and polyhedral with central, rather dark, nuclei, and the cytoplasm was packed with fine, somewhat eosinophilic, granules. The cells thus had a superficial resemblance to xanthoma cells. Each cell was separated from its neighbour by a network of reticulin fibres. No lipid was present in the tumour cells. The cytoplasmic granules stained blue with Heidenhain's azan and faintly pink with muci-carmine. No myofibrils were present. Similar tumours have been found attached to the maxilla by Massin (1894), Füh (1902), Kleine (1929), Volkmann (1929), Ceelen (1931), Meyer (1932), Leroux and Delarue (1939), Kratochvil (1941), Lascano-Gonzalez (1941), and Crane and Tremblay (1945), and to the mandible by Olivier (1895), Schorr (1906), Abrikossoff (1931), Jorge et al. (1932), and Crane and Tremblay (1945). The age distribution of patients with these tumours is interesting; of fifteen maxillary tumours (including ours) all except one was found in the first few days of life, but Kratochvil (1941) does not give the age of his patient. Of the eight mandibular tumours six were found in the first few days of life, one of Abrikossoff's (1931) cases was aged 36 years, and the other 55 years. Of the eleven cases of maxillary tumour whose sex is given, ten were female, and of the eight cases of mandibular tumour all, except Abrikossoff's two, were female. The three cases reported here were all female infants. Thus of twenty-two cases reported to date, in which the sex is known, nineteen were females.

Judged by the subsequent history, these tumours are benign; no recurrence was found after six months in Kleine's first case or after a year and a half in his second case. Massin's was well after one year and no recurrence was found after two years by Olivier (1895), Füh (1902), and Meyer (1932), and of three years by Jorge et al. (1932). Crane and Tremblay (1945) found no recurrence in their first case, and we found none in our three cases. This is in agreement with the general histological uniformity of the tumour cells and the absence of mitoses.

Origin of the tumours. Three main theories have been proposed to account for the origin of this type of tumour. It has been suggested that they may arise from muscle, misplaced dental anlage, or basal epithelium.

Tumours composed of cells similar to those found in congenital epulides have been found in the tongue by Abrikossoff (1926). Crane and Tremblay (1945) quote fifty-eight other cases, and Willis (1948) shows illustrations of two. In some, but not all, such tumours of the tongue, some cells show longitudinal and cross striations and transitions to undoubted muscle fibres. Similar cells have been found in muscle tumours in other sites, e.g. in the arm (Geschickter and Maseritz, 1939). Such tumours have therefore been classed as 'myoblastomas' by Abrikossoff (1926, 1931) and by Crane and Tremblay (1945), although the latter felt that no definite conclusion as to their myoblast origin could be drawn. In the early stages of muscle development in the embryo there is a phase when the myoblasts contain numerous cytoplasmic granules (Goldweski, 1902; Weed, 1936), and it is from these granules that myofibrils are thought to develop. The granular cells of the lingual 'myoblastomas' are considered to be homologous to these embryonic myoblasts. However, as Willis (1948) points out, true myoblasts of the embryo do not at any stage resemble Abrikossoff's 'myoblasts'; the granular cytoplasm and situation of the tumour cells suggests merely a superficial similarity. Willis also remarks on the similarity between these granular cells and the regeneration sprouts described by Clark (1946) in damaged muscle; hence, he suggests, the granular cell 'myoblastomas' of the tongue and other tissues are not neoplastic but degenerative or regenerative lesions of muscle fibres. Abrikossoff himself, in 1931, suggested that some 'myoblastomas' are derived from damage to muscle and that others are embryonic in origin. The absence of muscle from the sites of these epulides in the newborn renders their origin from degenerative or regenerative changes in muscle very unlikely. Further, the striking uniformity in histological structure of these epulides and the absence of muscle-like elements makes it probable that they may not be of the same nature as the lingual tumours.

The theory that these tumours are developed from misplaced dental anlage was proposed by Massin (1894). Kleine (1929) has examined this theory and rejected it. Attractive as the theory is, we have been unable to see any resemblance between the cells of the tumour and those of dental rudiments. Kleine (1929) himself proposed the view that these tumours arise from the basal epithelium of the oral cavity possessing embryonic potency. We have been unable to find evidence to support this view. The tumour cells lie clearly separated from the overlying basal epithelium in all three cases, and the fact that each cell is separated from its neighbour by a reticulin fibre mesh further argues against the epithelial origin of these tumours.

The tinctorial properties of the granules in the tumour cells are interesting but do not throw much light on the origin of the tumour. Schorr (1906) reported that, after fixation in alcohol, the granules stained red with eosin, yellow with picric acid, and
violet with polychrome methylene blue and thionin. Kleine (1929) in his first case found that the granules stained partly blue and partly red with mucicarmine, and with Heidenhain’s azan they stained distinctly blue. This he took as evidence that the granules were mucoid. In our cases the granules had similar tinctorial properties. Since, however, mucoid granules can occur in both epithelial and connective tissue cells, these staining reactions cannot be taken to indicate the epithelial nature of the tumours, and as the individual cells are surrounded by a fine reticulum, it is more likely that they are mesenchymal.

Jorge et al. (1932) called their three tumours ‘angiolinfangioma xantelomatosa’. They stressed the vascularity of their tumours, which was not a conspicuous feature of the other congenital epulides described in the literature. Moreover, although no specific stains for fat were used, their description suggests that fat was present in the tumour cells. It seems possible that the epulides of Jorge et al. (1932) are different from the others of this group, but the cells of many of these tumours give an impression of a xanthomatous nature on first examination.

In conclusion we admit that we have been unable to contribute any further positive evidence towards elucidating the histogenesis of congenital epulis, but recognize that clinically and morphologically these neoplasms constitute a distinctive entity. There appears no definite justification for calling these tumours myoblastomas, and we suggest that the term ‘congenital epulis’ be retained until further evidence of their histogenesis is forthcoming.

Summary

Congenital epulides in three infants are described. The tumours consisted of large round or polyhedral granular cells separated from each other by a fine reticulum.

The twenty-three similar tumours previously described are briefly reviewed.

The theories of origin of these tumours, namely muscular, misplaced dental anlage, basal epithelium, and lymphatic endothelium, are briefly discussed. None appears to be adequate.

It is suggested that these congenital granular epulides are mesenchymal in nature.

We wish to thank Professor S. L. Baker for helpful criticism, and Mr. F. Ward for the microphotographs.

References

— (1931). Ibid., 280, 723.
Schorr, G. W. (1906). Ibid., 39, 82.