CAPILLARY ANGEIOMATOSIS OF THE PAROTID GLAND (PARTIAL EXCISION).

BY CLEMENT NICORY, LONDON.

WITH A PATHOLOGICAL ACCOUNT AND REMARKS

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THE condition to be described is of interest from both a clinical and a pathological point of view. The patient, a male infant, age 9 months, was seen by me as an out-patient at the Evelina Hospital, London. The child was of normal development, and had given no difficulty since birth. When it was six weeks old the mother accidentally noticed a round lump, about the size of a marble, in the right parotid region. The swelling was said to have gradually increased, and when seen there was a marked prominence on the right side of the face, about the size of a bantam's egg, corresponding anatomically with the parotid gland. The skin over its most prominent part showed a slight bluish-red tint produced by a few small superficial vessels. There were no signs of inflammation, and the skin was freely movable over the elevation. On palpation the tumour felt uniformly soft and elastic, but without definite fluctuation. It was not movable on the deep structures, and yielded no pulsation.

From within the mouth the tumour gave a similar impression. The opening of Stenson's duct appeared normal; no fluid was seen to exude on applying firm pressure to the mass. There was no history of constitutional disturbance, or pain; and the child seemed very little concerned during examination. The neighbouring lymphatic glands were not enlarged, and there was no pyrexia.

The diagnosis was, a tumour in connection with the parotid gland. The mother was anxious that something should be done to reduce the increasing disfigurement. The patient was admitted under Mr. H. S. Clogg, by whose kind permission I am able to publish the following account of the operation.

A slightly curved incision with its concavity upwards, 3 inches in length, was made from just behind the lobule of the right ear, half an inch above and almost parallel with the lower border of the mandible. On reflecting the skin, a non-adherent bluish mass of nævoid appearance was exposed, though separable only with difficulty from the deeper structures. The external carotid artery was seen entering the lower part of the tumour; and only after clamping it and securing numerous bleeding points was hæmostasis obtained. The whole of the parotid gland appeared to be involved, and part of the retro-mandibular portion had to be left owing to its proximity to the large vessels. Stenson's duct and branches of the facial nerve were unavoidably divided. The incision healed by first intention; the child made good progress, though when discharged the affected side was still slightly larger than the left, and the facial paralysis produced at the operation was still present. The nævoid condition of the skin which was noticed on admission had disappeared. Whether the origin of the angioma is to be associated with the incipient function of the gland, it is impossible to determine.

In the case of the parotid the ptyalogenous function is present in the new-born child; in that of the submaxillary, ptyalin secretion is not established under the age of two months.¹ The mucinogenous function of the latter gland is probably later.

Since the observations cited by Halliburton, further investigations have been carried out in the same direction, with the following additional results: (1) The secretion of ptyalin before term. Moll² obtained saliva containing ptyalin from a parotid fistula in

a child prematurely born at 7 months. (2) The increased output of ptyalin after starch feeding. Finzi³ found the amylolytic power was the same up to the age of six months, whether the food contained starch or not; after six months the power was increased if the food contained starch; the property was, moreover, highest at midday.

The following results have been recently obtained by myself at the Evelina Hospital, London. The saliva was collected by giving infants plugs of gauze (weighing 500 mgrms.) to suck, the amount of the saliva being estimated by the increase in weight of the plugs. The latter were extracted with distilled water, and the action of the fluid tested on starch solution. Ptyalin was found present shortly after birth in the saliva of infants prematurely born at 32, 34, and 36 weeks. The quantity of the ferment was very small; but it gradually increased during the first year; after this no appreciable change was demonstrable. In the premature infant of 34 weeks, 1 part of the ptyalin extract took 98 minutes to reduce an equal part of 1 per cent starch solution to the achromic point; whereas at full term (40 weeks) it took only 23 minutes; three months after birth, 11 minutes; and eight months after birth, 1 minute. The action was more pronounced in strong infants than in weak. After the age of 4 months the saliva of children who had received starch in their diet contained more ptyalin than that of those who were having none.

PATHOLOGICAL NOTE AND REMARKS BY PROFESSOR S. G. SHATTOCK.

Figure 346.—To the eye the portion removed exhibited the familiar structure of a salivary gland, the lobules being quite discrete, and but loosely held together by connective tissue.

FIG. 346.—A section of the angiomatic parotid gland described, enlarged about six times, showing the persistence of its normal lobulated construction.

Figure 347.—A microscopic study yields the following results: With few exceptions the proper glandular structure of each lobule is replaced by a close network of capillaries. In all the lobules affected, one or more patulous ducts, with their typical columnar-celled lining, are distributed in the capillary basis; in some there are, in addition, small groups of the secreting cells, these different elements being widely disparted in the vascular neoplasm; the stages from partial to complete replacement are readily traceable. Small, ill-defined groups of lymphocytes lie here and there between the capillaries; these correspond in position with the gland-ducts and cells of the alveoli. The interlobular connective tissue presents no angiomatic abnormality. The capillaries composing the mass which replaces the lobules are so

tortuous and compressed that they can scarcely be traced in detail, except by the partial persistence of the lumina, or the narrow lines of red cells within them. As studied in favourable spots the vessels, where patulous, consist of a single layer of endothelium, and they lie in apposition; where broader, the intervals contain a few connective cells, with accompanying fibrillæ.

Remarkable and little known as the condition is, it is not unique. There is in the Royal College of Surgeons, London, a circumscribed angioma of similar kind, and in the same position, thus described in the recent Catalogue of General Pathology.⁴

A circumscribed angioma, 5 cm. (2 in.) in chief diameter, from the parotid region of an infant. It has a definitely lobulated construction, and to the naked eye closely resembles the proper salivary gland, in the lobules of which it has arisen. Microscopic examination shows that each lobule of the gland is replaced by a plexus of capillaries with a minimum of intervening tissue. In each there are widely-scattered remnants of the proper glandular elements.

To these may be added a third example of intrasalivary angioma, but in this instance involving the submaxillary, and not the parotid, gland. The history of this case,

which was under the care of Mr. Raymond Johnson, and operated upon by Mr. G. E. O. Williams in 1911 (to both of whom we are indebted for leave to publish it) is as follows :—

The patient, a child, age 4 months, presented a rapidly-enlarging swelling in the neck ; on examination it was found to be a fairly-well circumscribed tumour, the size of a Tangerine orange, below the left angle of the lower jaw. After excision it was thought to be a nævoid lipoma ; it was of dark-purple colour, and without a definite capsule. Many tortuous and dilated vessels were distributed through its substance. In microscopic structure I find this specimen to be a perfect replica of the two just described, the lobules of the gland being equally discrete. A few of the latter are normal ; in some, the

FIG. 347.—Showing a single lobule of the angiomatous gland. It is completely pervaded with a close plexus of capillaries, in the midst of which lie some of the ducts. The septa are uninvolved in the angiomatosis. ($\frac{3}{8}$ obj.)

angiomatous element involves only part of a lobule ; in others, the whole, the volume of these being much augmented. The ducts and groups of secreting cells are dispersed amid the vascular plexus, as they are in the other two examples. The vessels themselves are simple capillaries with mere traces of intervening tissue. In certain spots the tortuous, thin-walled channels are abnormally wide or ectatic, though still capillary in structure. The interlobular septa are intact.

Lymphocytic foci are much less pronounced than in the first of the cases referred to.

In regard to all these examples of salivary angiomatosis, it may be observed that the general histogenesis is of the same kind as that traceable in the development of a

capillary angioma of the subcutaneous fat, where the lobulation and circumscription of the neoplasm may be as pronounced as is that of the lobules of the fat in which it arises. In the museum of the Royal College of Surgeons there are contained the two following specimens :—

1425.1. (*General Pathology*).—A lobulated angioma which was removed from the shoulder of an infant. The growth is of flattened oval form, 3.8 cm. ($1\frac{1}{2}$ in.) in chief diameter, and is composed of a series of closely apposed but discrete lobules. Microscopic examination shows it to consist of a close network of capillaries with a minimal amount of intervening connective tissue. Scattered through the lobules there are small groups of unilocular fat-cells, the marked lobulation of the tumour being explained by its having arisen in the lobules of the subcutaneous tissue.

1426.1. (*General Pathology*).—A flattened oval angioma, which was removed from under the skin over the breast of a child 9 months old. A small portion of the skin which was involved in the tumour and discoloured during life, has been removed with it. The growth is distinctly circumscribed, and lobulated in construction. Bristles have been put unto certain of the larger vessels which lie in the interlobular connective-tissue. Histologically, the lobules consist of a plexus of capillaries, in the midst of which fat-cells occur in varying numbers. In the case of some lobules the structure consists almost solely of adipose tissue, proving that the lobulation of the tumour is due to the origin of the angioma in the original fat of the part.

The recognition of this origin of subcutaneous nævi in the lobules of the subcutaneous fat dates from the time of Virchow, and is concisely described in the incomparable *Krankhaften Geschwülste* (Angioma). He observes : “The subcutaneous nævus presents most frequently the form of a tumour, and this is sufficiently well limited, though not surrounded with a special capsule. In general the aspect of this form depends on the mother tissue from which it proceeds ; and as the fatty subcutaneous tissue presents as a rule a lobulated disposition, so the nævus also takes a kind of lobulated structure.”

Historically, the first example of salivary angioma recorded in this country is that by Gascoyen.⁵ The patient was a man, age 44. The tumour was congenital, and latterly had come to occupy the whole of the parotid region. There was no pulsation or bruit in the mass, which became tense and livid under excitement. Other nævi were scattered over the body and limbs. After death (which occurred from suffocation) several nævi were found studding the surface of the intestines ; one of these originated in the submucosa. The parotid tumour consisted of tortuous and dilated blood-vessels embedded in the gland, the structure of which was plainly seen filling up the intervals between their ramifications. The vessels “appeared to be the capillary veins of the gland enormously distended, which opened into large pouches, many of the dilatations being filled with phleboliths, some as large as a horse bean.” Of this specimen no histological examination was made, and finer details are altogether wanting.

Two further instances are referred to by Virchow⁶ as having been possibly parotid angiomata.

One of the patients was a woman, age 29. The angioma, which involved the parotid region, was chiefly if not solely of the arterial kind, the vessels being fed from the temporal artery. An attempted operation was followed by such hæmorrhage that no excision was carried out ; and no detailed information as to the anatomical lesion of the parotid gland is forthcoming.

The other case was a boy, age 8, who presented a swelling of the parotid and adjoining area ; the tumour projected into the mouth in the neighbourhood of Stenson's duct. It had been noticed only for a year, and from its rapid growth and uneven surface it was regarded as malignant in character. After preliminary ligation of the common carotid it became considerably paler ; during its removal the distended condition of the numerous and enlarged offsets of the carotid excited surprise. No details regarding the condition of the parotid gland are recorded.

There is at present attending St. Thomas's Hospital a further case of angioma, implicating the right parotid region ; but as the condition is being treated by means of

radium, exact pathological details are as wanting in this as in the three last referred to. The patient was admitted under Mr. Corner in December, 1920, with a large naevoid swelling affecting the mucosa of the right cheek, extending forwards as far as the angle of the mouth, and posteriorly surrounding the orifice of Stenson's duct. The tumour, which had been slowly increasing since birth, bulged between the dental arches, and externally led to a protrusion on the right side of the face from the ear to the mouth. The suprajacent skin was uninvolved.

Two tubes of radium were inserted into the swelling—for $7\frac{1}{2}$ hours on Dec. 12, and for 10 hours on Dec. 18—but with little result. Two phleboliths about the size of peas were subsequently felt in the mass, the formation of which was thought to have followed the insertion of the radium tubes.

Figure 348.—On tracing the history of the case, it was discovered that the patient had previously been admitted when two years of age, under Mr. H. H. Clutton, with the note that he had been born with a naevus in the cheek, which had been increasing. At the date of admission in March, 1904, there was a large, somewhat compressible, swelling on the inside of the cheek; the diagnosis was, "Cavernous angioma of the cheek". No operation was carried out.

The early age at which the disease was noticed in each of the three cases which were fully examined microscopically, may be finally pointed out: (1) Parotid, 6 weeks; (2) Parotid, infant; (3) Sub-maxillary, 4 weeks; but, as already remarked, whether the origin of the angiomatosis is in any way related to the nascent function of the gland is a problem beyond determination. In Gascoyen's case (male, age 44) the disease was congenital. And the same is true of that at present attending St. Thomas's Hospital (male, age 18).

The gross enlargement of the veins in both of these seems to indicate that, with the lapse of time, a primary capillary angiomatosis of the gland is succeeded by an increase in the size of the afferent and efferent vessels; a result which might be anticipated in view of the increasing demand for blood entailed by the growth of so vast a capillary plexus in the glandular substance.

FIG. 348.—A photograph of the last case referred to, of angioma involving the right parotid region and extending thence to the cheek and lips.

REFERENCES.

- ¹ HALLIBURTON, *Chemical Physiology*, 1891, citing Schiffer, Korowin, Zweifel.
- ² MOLL, *Monats. f. Kinderh.*, 1905.
- ³ FISZI, *Rev. Hyg. et Méd. Inf.*, viii, No. 3, 224.
- ⁴ *Catalogue of Gen. Pathol. Roy. Coll. of Surg. Lond.* By S. G. SHATTOCK, with the assistance of C. F. BEADLES.
- ⁵ GASCOYEN, *Path. Soc. Trans. Lond.*, 1860, xi, 237.
- ⁶ VIRCHOW, *Canstatt's Jahresbericht*, 1889, iv, 312.