

An Autopsy Case of Branchiogenic Carcinoma

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Branchiogenic cysts arise from the remnant of the branchial apparatus. It is no doubt that primary carcinoma arising in a branchiogenic cyst does exist as a pathological and clinical entity. A 45-year-old man complained a painless mass in the left lateral neck. There were no other symptoms. Postmortem examination revealed a huge 11 by 10 by 10 cm ulcerated, partly necrotic and hemorrhagic tumor. Microscopic picture exhibited epidermoid carcinoma composed in part of a trabecular pattern of large, polyhedral cells with faintly granular and acidophilic cytoplasm. Intercellular bridges were observed but not prominently. Occasionally giant cells, with giant nuclei or multiple nuclei, were seen. Individual cell keratinization was frequent, but keratin pearls were not found. Mitotic figures were numerous. Special stains failed to reveal any cytoplasmic inclusions. There have been only a few cases reported that showed histological evidence of carcinoma arising in a branchiogenic cleft cyst. In this case the author could not demonstrate cancer arising in the wall of a branchiogenic cyst, and no remnant of the branchial apparatus were found. Exhaustive studies failed to demonstrate a primary site for the cancer in other areas. Clinically the tumor was soft and fluctuant, and serous bloody fluid was removed by puncture. These findings suggest the possibility of branchial cyst origin.

Primary branchiogenic carcinomas are extremely rare. In 1861, LANGENBECK described a primary squamous cell carcinoma involving the neck which had no relation to other organs. In 1882, VOLKMANN reported a primary carcinoma in the neck which he believed arose from remnants of the branchial arches, and for this reason called it "Branchiogenes Karcinom". Up to the present time, numerous case reports of branchiogenic carcinoma have appeared in the literature, but most of these cases have been diagnosed merely because of failure to demonstrate a primary site of cancer in other areas. MARTIN and co-authors¹⁾ stated that the only absolute proof of the existence of such a specific

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tumor as a branchiogenic carcinoma would be the histological demonstration of cancer arising in the wall of a branchiogenic cyst. There have been only a few cases reported that showed histological evidence of carcinoma arising in a branchiogenic cleft cyst. In the case reported here we could not demonstrate the cyst wall but I believed the tumor to be a primary carcinoma possibly arising in a branchiogenic cleft cyst.

Case Report

A 45-year-old man gave a history of having had a painless mass in the left side of his neck for about ten days prior to examination. The patient had no other symptoms, but he was concerned because the swelling appeared to be gradually increasing in size, and he was seen in the Saiseikai Hospital in Nagasaki on March 21, 1961.

Physical examination revealed an essentially normal male except for a 2 cm mass below the angle of the mandible on the left. The skin over the mass was freely movable. No other masses could be palpated in the neck. A tentative diagnosis of tuberculous lymphadenitis was made, and X-ray therapy was given. The mass decreased in size following the therapy.

In July, the mass was noted to have increased in size again, was hard, tender and fixed. One week before admission, the mass measured about 8 cm in diameter was soft, and fluctuant. Fifty five ml of serous bloody fluid was removed by puncture.

He was admitted on Sept. 8, 1961, at which time the physical findings were essentially the same as one week before. The white cell count was 7000 per cu. mm. The skin over the mass was ulcerated and drainage was present. By the end of September drainage material became more cloudy in character. On Nov. 5, 1961, a biopsy was taken from the mass and a diagnosis of undifferentiated carcinoma was made (by ABCC, Nagasaki). He developed anorexia, malaise and difficulty in swallowing. On Nov. 30, 1961, he became unconscious and died.

Autopsy was performed at ABCC, Nagasaki on Nov. 31, 1961. Postmortem examination revealed a huge 11 by 10 by 10 cm ulcerated, partly necrotic and hemorrhagic tumor in the left lateral neck. The base of the tumor was attached to but did not involve the left common carotid artery. The tumor did not appear to arise from the neck organs and every effort was made to locate a possible site for a primary lesion outside of the neck. Careful examination of the ear, nose, nasopharynx, pharynx, hypopharynx larynx, base of tongue, thyroid gland, salivary gland and esophagus revealed no lesion.

The sectioned surface of the tumor was made up largely of hemorrhagic necrotic material, but there were a few nodules of pale yellow,

fleshy tissue.

An additional finding of interest which could not be explained was the presence of extensive hemorrhage in the epicardium, myocardium and endocardium. The heart resembled hearts after cardiac massage and possibly external massage had been attempted though there is no history of this. The left pleura was densely adherent. There were numerous calcifications in the upper lobe of the left lung. The parenchyma of both lungs was edematous and fluid could be expressed from the cut surface. The kidneys weighed 170 gm each and showed mild arteriolar nephrosclerosis.

Microscopic examination of the mass revealed an epidermoid carcinoma composed in part of a trabecular-like pattern of large, polyhedral cells with faintly granular and acidophilic cytoplasm. Nuclei were variable in size and had one to several nucleoli. The chromatin pattern was vesicular with a fairly constant nuclear membrane. Occasionally giant cells, with giant nuclei or multiple nuclei, were seen. Individual cell keratinization was frequent, but keratin pearls were not found. Mitotic figures were numerous. In other areas of the tumor there were foci in which cell outlines were sharply defined but the cytoplasm appeared empty. In some other areas, the degree of variation of the cells was greatly increased and most of the tumor cells appeared spindle-shaped.

Special stains failed to reveal and cytoplasmic inclusions in the tumor cells; Periodic acid-Schiff-stain, Best's mucicarmin (for production of mucin), Smith-Dietrich's lipid stain (for phospholipid), Sudan III stain (for nonspecific lipids and neutral fat), Bodian's protargol silver stain (for argyrophilic granules) were all negative. Intercellular bridges were observed however they were not prominent and a hematoxylin stain for phosphotungstic acid was of rare help in demonstrating them. Snook's reticulum stain exhibited reticulum fibers arranged around group of tumor cells or irregularly interspersed between the tumor cells.

COMMENT

It is known that malignant tumors may arise from any tissue in the body. The location of this tumor and its histologic picture had some resemblance to tumors classified as nonchromaffin paraganglioma. In 1961, MARSHALL and HORN²⁾ reported a comparative study on nonchromaffin paraganglioma in which the comparison was made on clinical, morphological and limited histochemical basis; The tumors of paraganglia gave a positive reaction for nonspecific lipids and neutral fat, the phospholipid stain also positive but PAS reaction was negative. The reaction described in 1957 by HAMPEL and LATTES³⁾ with the Bodian's

protargol technique also could show argyrophilic granules in the non-chromaffin paraganglioma. In our case, nonspecific lipids and neutral fat stain, phospholipid stain, Bodian's protargol silver stain were all negative.

It is generally believed that branchial cysts arise from the remnants of the branchial apparatus. Branchial cysts appear as circumscribed nodular swellings on the side of the neck. Most frequently they occur close to the anterior border of the sternocleidomastoid muscle. They vary in size from that of an olive to an orange and in some instances fluctuate in size. These cysts are usually lined by stratified squamous epithelium, pseudostratified ciliated columnar epithelium, or epithelium of both types.

There can be no doubt that primary carcinoma arising in a branchiogenic cyst does exist as a clinical and pathological entity.

In 1958, STRONG and SOMMERS⁴⁾ reported two interesting cases with histological data in which multiple malignant tumors occurred and epidermoid carcinoma arising in a branchiogenic cleft cyst was found.

In 1959, COLLINS and EDGERTON⁵⁾ presented a case of squamous cell carcinoma arising in the wall of a squamous epithelial-lined lateral cervical cyst with definite histological evidence of carcinoma arising in this cyst.

WILLIS⁶⁾ in his "Pathology of tumors" reported that of 64 consecutive cases of epidermoid carcinoma of the facio-cervical region, he has made the diagnosis of possible branchial carcinoma in only two instances. He emphasized that he has seen no other acceptable reports except for the case reported by COLLINS and EDGERTON.

In our case we could not demonstrate cancer arising in the wall of a branchial cyst, and no remnants of the branchial apparatus were found. But exhaustive studies failed to demonstrate a primary site for the cancer in other areas. Clinically the mass was soft and fluctuant, and serous bloody fluid was removed by puncture. These findings suggest the possibility of branchial cyst origin.

SUMMARY

A case of epidermoid carcinoma with histological data arising in the left lateral neck is presented. The diagnosis of primary branchiogenic carcinoma should not be made except in cases in which there is both gross and microscopic evidence of a true branchial cyst and carcinoma is found within the cyst wall. In our case we could not demonstrate cancer arising in the wall of a branchial cyst and no remnants of the branchial cyst were found.

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Legends for Figures

- Fig. 1. Macrograph of a tumor in the left lateral neck of a 45-year-old man. The tumor had been growing rapidly with ulceration on the surface.
- Fig. 2. Histologic appearance of the tumor at biopsy. The invasive and irregularly-shaped nests of atypical epidermoid cells are noticed. No pathological changes in the epidermis were found. H-E stain.
- Fig. 3. Microphotograph of the tumor. The epidermoid carcinoma composed in part of a trabecular pattern of large and polyhedral cells with faintly granular and acidophilic cytoplasm was shown. H-E stain.
- Fig. 4. Microphotograph of the tumor. Note keratinization of the individual cell. H-E stain.

