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Radiology in Focus

Radiological appearance of primary branchial cleft cyst carcinoma

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Abstract
The hypothesis that primary branchiogenic carcinoma originates from a branchial cleft cyst is controversial. Many reports regarding primary branchiogenic carcinoma failed to provide sufficient evidence to distinguish it from metastatic cervical lymph nodes arising from previously unrecognized primary tumours. The radiological appearance of malignant transformation from a branchial cleft cyst has not been reported previously in the English literature. A radiological study is presented that confirms the primary branchiogenic carcinoma. The management in suspected cases would be wide surgical excision of the tumour including ipsilateral radical neck dissection followed by radiation therapy.

Key words: Carcinoma; Branchial region; Radiology; Pathology

Introduction

Branchial anomalies in the head and neck region can exist as one of three types: sinus, fistula, or cyst. They are due to the persistence of vestigial remnants of a branchial cleft or pouch. Most branchial cleft cysts are located along the anterior border of the sternocleidomastoid muscle as are the cervical lymphatics. Because neoplasms from branchial cleft cysts are located in the same region anatomically as metastatic cervical lymph nodes, diagnosis is controversial and confusing both clinically and pathologically (Shaw, 1970; Batsakis and McBurney, 1971; Wolff *et al.*, 1979; McCarthy and Turnbull, 1981).

Martin *et al.* (1950) proposed criteria for the confirmation of primary branchial cleft carcinoma: (1) The cervical tumour must occur somewhere along the anterior border of the sternocleidomastoid muscle. (2) The histological appearance of the growth must be consistent with an origin from tissue known to be present in branchial vestigia. (3) The patient must have survived and have been followed by periodic examinations for at least five years without the development of any other lesion which possibly could have been the primary tumour. (4) There should be histological demonstration of a cancer developing in the wall of an epithelial-lined cyst situated in the lateral aspect of the neck. He stated that the fourth criterion was most important in confirmation of primary branchial cleft cyst carcinoma.

Since the 1950 landmark paper of Martin *et al.*, only 10 patients have totally fulfilled their four criteria of primary branchial cleft carcinoma in our review of the English literature (Park and Karmody, 1992; Singh *et al.*, 1998). However, there has been no report about a radiological study of the progression of malignant transformation for confirming the diagnosis of primary branchial cleft carcinoma. This case report describes radiological findings characteristic of primary branchiogenic carcinoma.

Patient and methods

Case report

A 58-year-old man was seen with a two month history of a slowly-growing mass measuring 5 × 6 cm in the right lateral neck region anterior to the sternocleidomastoid muscle and a complaint of neck pain with reddish overlying skin. The neck mass had been noted two years ago with radiological confirmation of a branchial cleft cyst (Figure 1). The mass was oval shaped and of low density. He was recommended to have the mass excised, but had not received surgery. The cystic mass disappeared spontaneously but recurred intermittently. However, three months before admission, the mass started growing slowly and became harder. He was admitted to the department of general surgery and treated with antibiotics after a diagnosis of an infected branchial cleft cyst. He complained of worsening symptoms. The mass was evaluated by computerized tomogram (CT) scan which showed a low density mass with thick and irregular rim enhancement on the right (Figure 2).

The patient consulted our ENT department and fine needle aspiration cytology was recommended and squamous cell carcinoma highly suspected. Thereafter, general physical examination, chest and gastrointestinal studies were performed. The appropriate head and neck examination including thorough endoscopic studies with random biopsy were performed. No abnormal finding was

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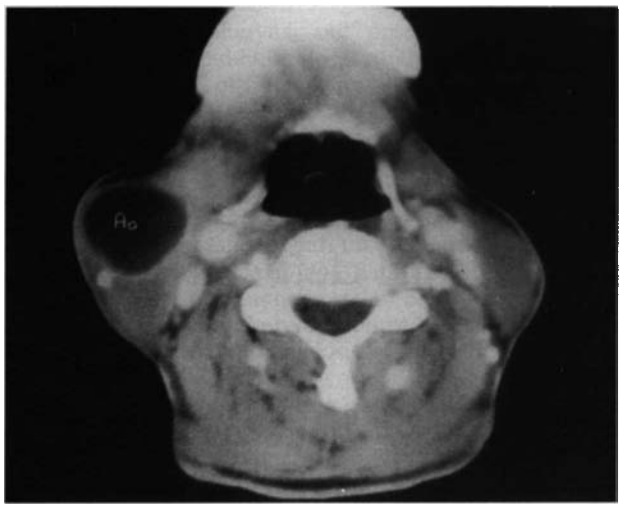

FIG. 1

Contrast-enhanced CT scan at level of hyoid bone shows oval shaped low density mass on right. No peripheral rim enhancement is noted. Mass is located adjacent to the anterior border of the sternocleidomastoid muscle and anterolateral to the carotid sheath.

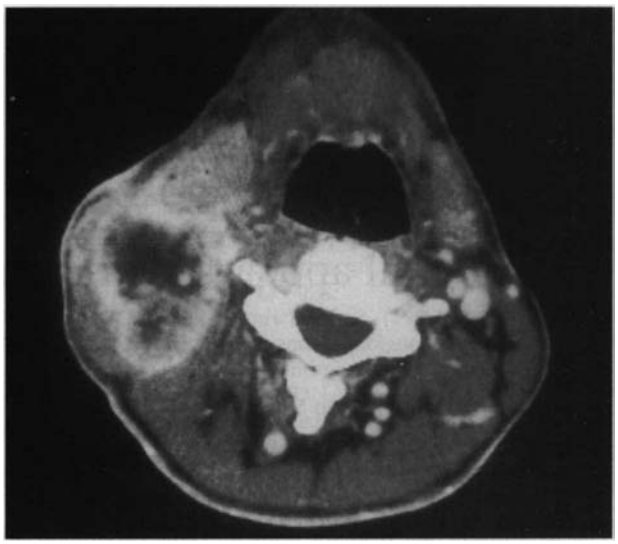

FIG. 2

Contrast-enhanced CT scan at lower level of hyoid bone shows a low density mass with a thick, irregular rim enhancement on the right. Fat planes adjacent to mass are abnormal.

detected. The neck mass was re-evaluated by CT scan and the cystic cavity was seen to be filled with an inhomogenous enhancing soft tissue mass that invaded the sternocleidomastoid muscle. There was also some thickening of the overlying skin and increased attenuation in the subcutaneous fat (Figure 3). The mass was tender and harder and fixed to the surrounding tissue.

We suspected a primary branchial cleft cyst carcinoma and performed wide tumour resection with radical neck dissection. The defect was reconstructed with a pectoralis major myocutaneous flap. The subsequent pathological diagnosis was squamous cell carcinoma contained within a branchial cleft cyst. The gross pathological findings, were that the main lesion was a whitish solid mass measuring $5.2 \times 4.3 \times 4.0$ cm. The mass revealed infiltrative growth to the adjacent soft tissue and central cystic cavities. Histopathologically, the central cystic lesion was lined with benign, keratinizing stratified squamous epithelium. Some foci of benign squamous epithelium were observed to evolve directly into an infiltrating, well-differentiated squamous cell carcinoma, demonstrating the malignant transformation of this lesion (Figure 4A). Most of the tumour was composed of invasive squamous cell carcinoma components (Figure 4B). The patient received 6500 cGy of radiation post-operatively and was followed up to 11 months after surgery without any evidence of recurrence of cancer.

Discussion

Khafif *et al.* (1989) disagreed with Martin's third criterion for the confirmation of primary branchial cleft carcinoma – a five-year follow-up without evidence of a primary tumour. He argued that Martin's third criterion cannot often be satisfied because patients may die of unrelated causes before this period has elapsed. Many patients receive post-operative irradiation, which may control an occult primary tumour. So he proposed two other criteria: 1) the absence of an identifiable primary cancer by a thorough evaluation, including endoscopy, CT scan of the head and neck, and appropriate biopsies; and 2) histological identification of a cystic structure partially lined by normal squamous or psuedostratified columnar epithelium with gradual transition to invasive squamous cell carcinoma. His suggestion may be the most up-dated criteria for diagnosis of primary branchial cleft carcinoma. In our case, the patient fully satisfied Khafif's two criteria and the tumour appears to be a primary squamous cell carcinoma arising from a branchial cleft cyst histologically, but the patient has not satisfied Martin's third criterion of a five-year disease-free survival because the patient has not yet been followed up for five years.

On the histological finding, branchial cleft carcinoma should be differentiated from cervical metastasis (Berstein *et al.*, 1976; Krogdahl, 1979; Thompson and Heffner, 1998). In cervical metastasis, lymphoid elements such as germinal elements may be present but not peripheral lobulation, internodular trabeculae, or perinodal sinuses. In branchial cleft carcinoma, dysplastic epithelium may be present next to invasive carcinoma and this supports the origin of the carcinoma as being from an epithelial-lined branchial cleft

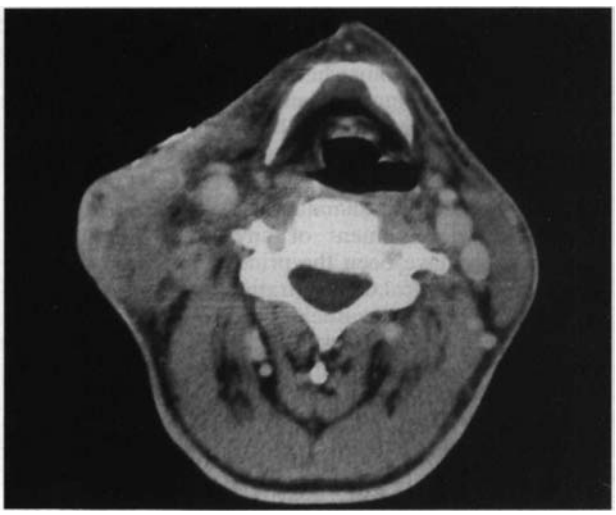

FIG. 3

Contrast-enhanced CT scan at level of hyoid bone shows indistinct, inhomogenous enhancing soft tissue mass that has invaded the right sternocleidomastoid muscle. There is also some thickening of the overlying skin and increased attenuation in the subcutaneous fat.

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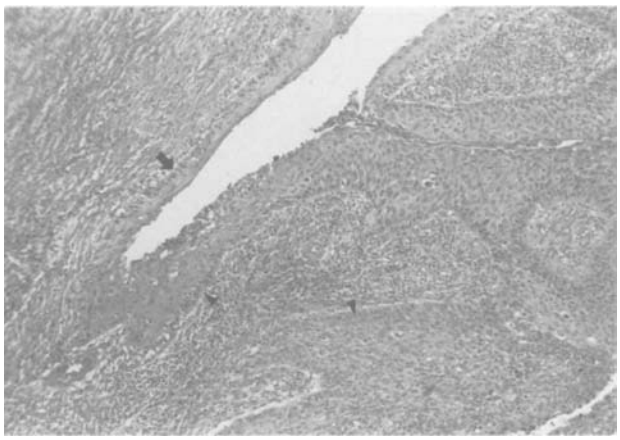

FIG. 4A

Benign stratified squamous epithelium (arrow) evolved directly to neighbouring overt well differentiated squamous cell carcinoma (arrow heads) (H&E; ×100).

cyst. This transitional zone of normal mucosal epithelium to dysplasia to carcinoma is most important for the confirmation of a primary branchial cleft carcinoma. Metastatic carcinoma of cervical lymph nodes from an upper aerodigestive tract would not have such a transition zone.

Generally, patients with a neck mass should have a CT scan of the head and neck. When a cystic mass has an irregular margin and heterogenous density, secondary infection and granulation tissue may be suspected. In primary branchial cleft carcinoma, a radiological study of malignant transformation from a branchial cleft cyst is usually not made because most branchial cleft cysts are excised following diagnosis. On radiological review of this malignant mass, the cystic cavity was seen to be replaced by enhancing soft tissue and showed invasion to the surrounding tissue including overlying skin.

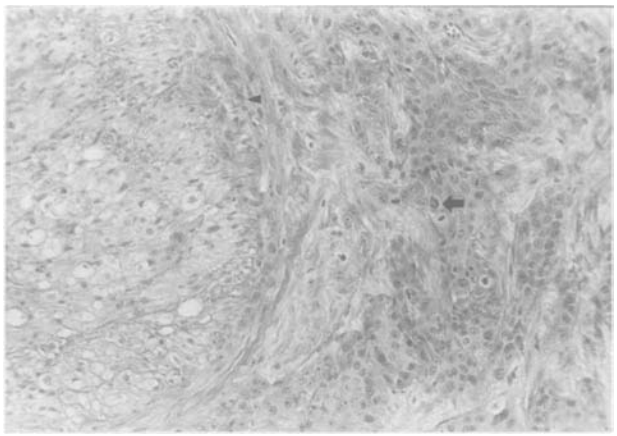

FIG. 4B

Photomicrograph showing the invasive squamous cell carcinoma with abnormal mitosis (arrow) and perineural invasion (arrow head) (H&E; ×200).

The treatment of branchial cleft carcinoma consists of surgery and radiation therapy. Wide surgical excision and neck dissection are recommended. When all lymph nodes are negative for tumour in the dissected specimen, radiation therapy may be reserved. However, radiation therapy should be recommended when patients refuse further lymphatic dissection or positive lymph nodes are detected following a neck dissection. Close follow-up examination should be performed for the remainder of the patient's life.

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