
Conventional Contrast Study in Demonstration of Branchial Cleft Fistula: A Report of Three Cases with Review of the Literatures.

Darunee Boonjunwetwat*

Varunee Stapatayawong**

Abstract:

Anomalies of the branchial apparatus are one of the common benign congenital neck mass. We have reported three cases of the branchial cleft fistula. One is the anomaly of the second branchial cleft and the other two cases are the anomaly of the third ones. Clinical presentations, radiographic contrast studies, revisions of the anatomy, embryology and differential diagnosis of the mass are included.

Key words: Branchial Cleft fistulae : radiographic contrast studies. Revisions of Anatomy, Embryology and differential diagnosis. 3 cases report.

Abnormal remnants of the branchial cleft and pouch is one of the common benign congenital neck mass. (2) Most branchial anomalies which present as fistulae, sinuses and cysts are considered to be derived from second branchial cleft remnants. Third and fourth branchial cleft sinuses are rare. (5)

MATERIALS AND METHODS

Cases Report:

Case 1: A 34 years old man presented by a right neck mass with purulent discharge from a skin opening adjacent to the mass. The mass was noted since he was 4 years of age. He also had a feeling of discharges dripping in the right side of the pharynx corresponding to the neck mass. Physical examination revealed a cystic mass about 4×3 cm. in size with a small skin opening at the inferoposterior aspect of the mass situated at the

lower part of the anterior border of sternocleidomastoid muscle. There was a purulent discharge coming out from the right tonsillar fossa when squeezing the cystic mass in the neck. The fistulogram was performed by injection of the contrast medium via the skin opening. The study showed the contrast medium filling the tract which is about 1 cm. in diameter and connected with the right tonsillar fossa of the oropharynx. (Fig. 1)

An operation was performed. The cystic mass was found beneath the sternocleidomastoid muscle with an external opening at the lower one third of the anterior border of this muscle. The fistulous tract was passing cephalad superficial to the common carotid artery and extending between the internal and external carotid artery with an internal opening into the right tonsillar fossa. Total excision was done and the final diagnosis was a second branchial cleft fistula.

* Chulalongkorn University Hospital.

** Bangkorklam Municipality Hospital.

Case 2: A 22 year old woman came with a complaint of sore throat and a left neck mass for 7 days. She has had the same symptoms repeatedly on and off for about 10 times since she was 10 years old. Recurrent suppurative thyroiditis was the provisional diagnosis. Her neck mass had disappeared after the inflammation subsided by medical treatment. She was treated with antituberculous drugs for two years without improvement. Incision and drainage of the neck mass was also performed three times in the past. Physical examination revealed a left neck mass, size about 4×4 cm. with signs of inflammation. The mass moved with swallowing. Surgical scars were also noted over the mass. After having been treated with antibiotics, the inflammation and the mass disappeared. Indirect laryngoscopy, later performed, was normal. Esophagogram revealed a tract originating from the apex of the left pyriform sinus and terminating in the area of the left lobe of thyroid gland, as shown in fig. 2.

Operation was performed with complete ligation of the tract.

Case 3: A 23 years old woman presented with a tender neck mass for 4 months. Previously, she had this same symptoms twice, three years and one year ago respectively. On each occasion, she was treated by antibiotics and drainage of pus with disappearance of mass and complete recovery of the inflammatory symptoms. Physical examination revealed left inflammatory thyroid nodule. The diagnosis of recurrent suppurative thyroiditis was given. The indirect laryngoscopy and fiberoptic laryngoscopy were performed but no abnormality were detected in the glottis or pyriform sinuses. No fistulous tract or opening can be demonstrated. Later, esophagogram was performed and a sinus tract was demonstrated at the left pyriform sinus terminating at the area of left lobe of thyroid gland as shown in fig. 3.

Operation was performed with complete ligation of the tract.

Discussion:

The branchial apparatus consists of a series of six mesodermal arches that are separated from each other externally by ectodermally lined branchial clefts and internally by endodermally lined pharyngeal pouches. (Fig. 4) By the end of the fourth weeks of gestation, four well defined pairs of branchial arches are visible externally. Shortly there after, the second arch increases in thickness and proliferates caudally to meet the enlarging epicardial ridge of the fifth arch. The second, third and fourth branchial clefts became enclosed in an ectodermally lined cavity, the cervical sinus of His. Eventually, this sinus becomes obliterated by the apposition and fusion of its walls, yielding a smooth uniform contour to the external surface of the neck. The endodermal derivatives of the pharyngeal pouches then migrate to their final

position. (5) Differentiation of the branchial apparatus to the structures of the neck are shown in table 1.

In each branchial arch, an aortic arch connecting the ventral and dorsal aortas developed. The first two pairs are involuted, by the time the sixth pairs is formed. The artery of the first persists forming the common and proximal part of the right subclavian artery. The fifth arch arteries involuted and the sixth pair forms the pulmonary arteries. (Fig. 6)

The anomalies are classified into sinus, fistula and cyst. Sinus is an incomplete tract that usually open externally and rarely internally. Fistula communicates to both internally and externally. Cysts have no communication neither internally nor externally. On the basis of the precise location of the anomalies, they can be classified according to their proposed pouch or cleft of origin.

1. First branchial anomalies.

It may originate anywhere along the nasopharynx, middle ear cavity or external auditory canal and external anteriorly or posteriorly to the pinna, or below the angle of mandible, involving the parotid gland or lying medial or superficial to it. (Fig. 7)

2. Second branchial anomalies.

The external opening is typically along the anterior border of the sternocleidomastoid muscle around the junction of the middle and lower third. Deep to platysma, along the carotid sheath, it then passes deep between the internal and external carotid arteries. After crossing over the hypoglossal and glossopharyngeal nerve, it then extends upward to end near the tonsillar fossa. (fig. 8)

3. Third branchial anomalies.

It passes from an external opening in the lower neck along the line of the anterior border of the sternocleidomastoid muscle, ascending in relation to the carotid sheath and passes deep posterior to the internal carotid artery between the glossopharyngeal nerve above and hypoglossal nerve below, piercing the thyrohyoid membrane to enter the pyriform sinus. (Fig. 9)

4. Fourth branchial anomalies.

It arises internally from the apex of pyriform sinus, penetrating the thyrohyoid membrane behind the fold of the internal laryngeal nerve. On the left, the fistula would loop around the aortic arch and on the right around the subclavian artery. Thereafter, the tract ascends to the common carotid artery passing over the hypoglossal nerve and descends to an external opening in the lower neck along the anterior border of sternocleidomastoid muscle. (Fig. 10)

The clinical presentation of the patient with first branchial cleft anomalies is a recurrent periauricular swelling, a sinus in the periauricular region, a mass in the external auditory canal, a dimple or depression in the floor of the canal, granulations or polyps in the floor of the canal or a chronic discharge from the ear in a normal tympanic membrane. (7)

The second branchial cleft anomalies, patients came with a history of recurrent discharge or infection of a sinus in the lower part of the neck, or a neck mass.

The rare third and fourth branchial anomalies presented with recurrent neck abscess or recurrent acute suppurative thyroiditis (usually on the left side)

The patient may present with only a cystic neck mass without other symptoms. So we must differentiate them from other conditions. These included:

1. Thyroglossal duct cyst - usually midline.
2. Parathyroid cyst - rare, near inferior border of Thyroid.
3. Thyroid cyst - related to thyroid, anterior to carotid artery, and internal jugular vein.
4. Cervical thymic cyst - lower neck.
5. Cystic hygroma - usually posterior to carotid artery and internal jugular vein.
6. Cystic metastasis - usually in the posterior triangle, behind carotid artery and internal jugular vein.

7. Thyroid neoplasm - rare.

8. Dermoid cyst - usually upper neck.

9. Teratoma and cystic neuroma - symptoms related to nerve involvement.

Conclusion:

We presented three cases of branchial cleft fistula, with preoperative radiographic contrast studies showing the fistulous tract connecting the upper digestive tract and the skin opening. These studies had help the surgeon in identifying the fistulous tract and its course and planning of the operation can be done prior to surgery.

Esophagogram is useful for demonstrating the tract leading to proper diagnosis and thus proper management.

TABLE 1

Differentiation of the Branchial Apparatus

Location	Cleft(Ectoderm)	Arch(Mesoderm)	Pouch(Endoderm)
First	External ear canal	Mandible, muscles of mastication, fifth CN., malleus and incus	Eustachian tube, tympanic cavity, mastoid air cells
Second	Cervical sinus of His	Muscles of facial expression, body and lesser horns of hyoid, CN 7,8	Palatine tonsil
Third	Cervical sinus of His	Superior constrictor muscles, internal carotid artery, CN 9, greater horn and body of hyoid	Inferior parathyroid thymus, pyriform fossa
Fourth	Cervical sinus of His	Thyroid and cuneiform cartilage, CN 10, aortic arch, and right subclavian artery, part of laryngeal muscles.	Superior parathyroid, Apex of pyriform sinus
Fifth and sixth	None	Portions of the laryngeal muscles and skeleton, inferior pharyngeal constrictor muscle, CN 11	Parafollicular "C" cells of the thyroid gland.

CN = CRANIAL NERVE

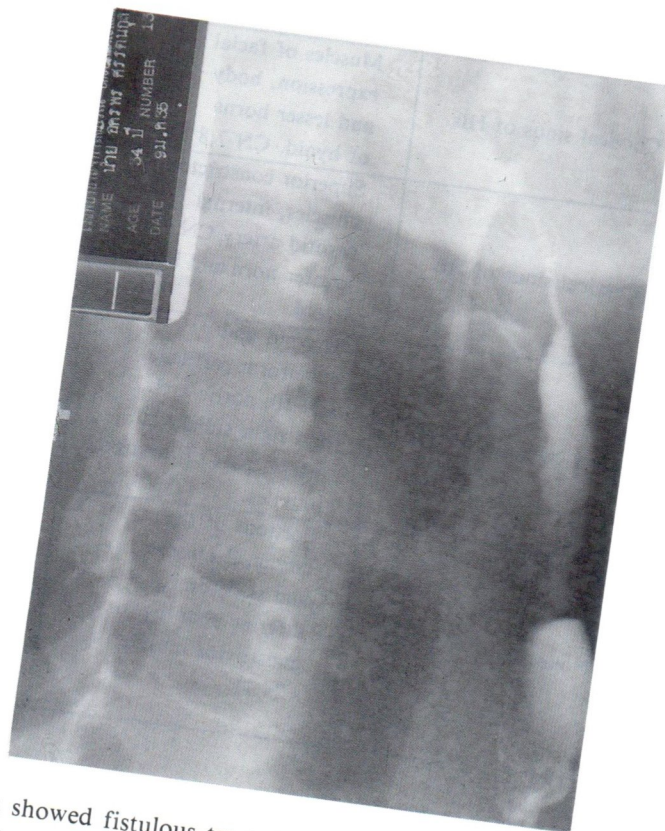
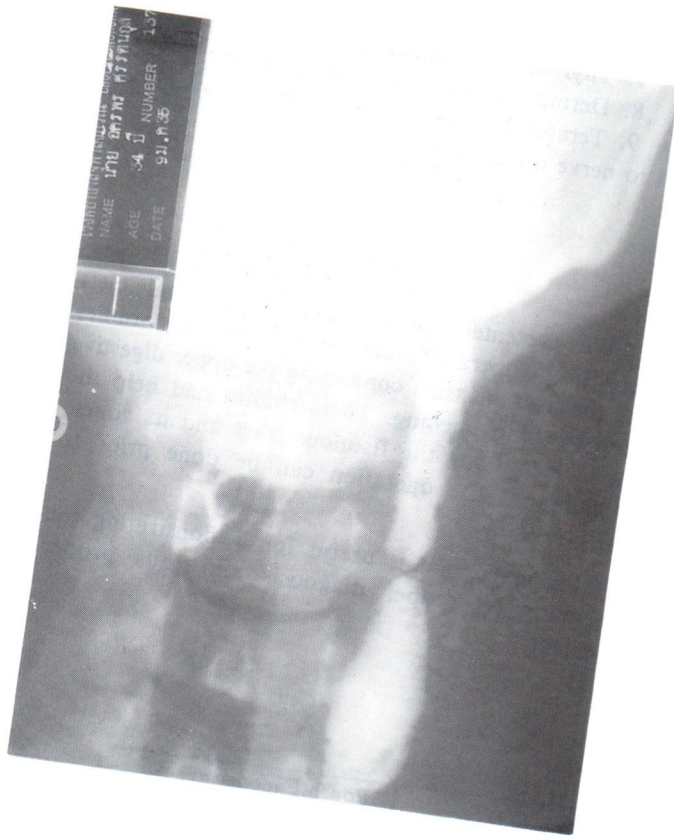


Fig.1 Case 1 Fistulogram showed fistulous tract connected with right tonsillar fossa (AP, lateral and left anterior oblique view).



Fig.2 Case 2 Esophagogram showed sinus tract originating from the apex of left pyriform sinus and terminating in the area of left lobe of thyroid gland. (AP, lateral left anterior oblique view)

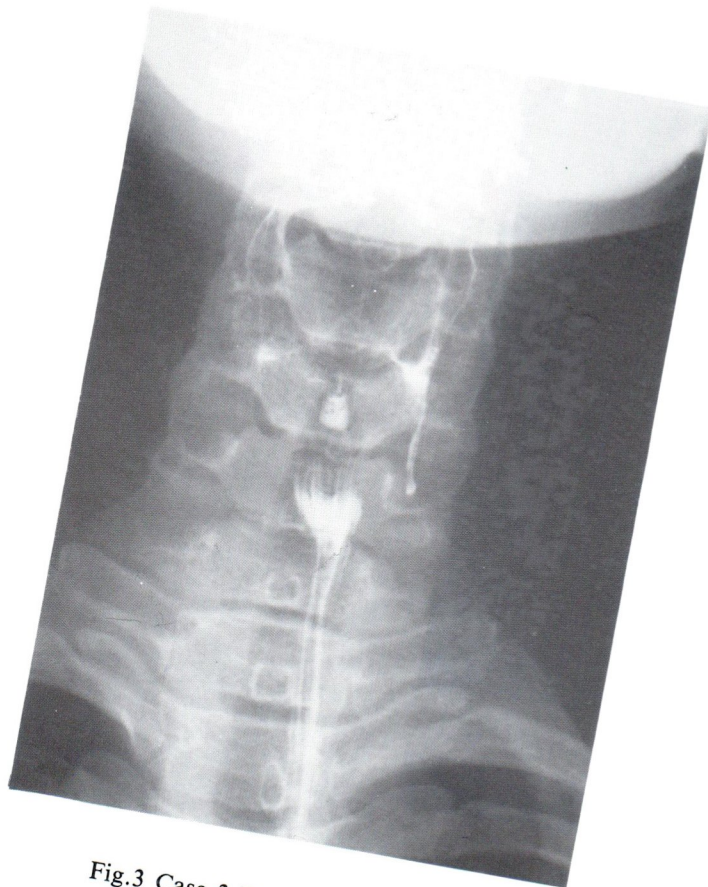


Fig.3 Case 3 Esophagogram showed a sinus tract from left pyriform sinus terminating at the area of left lobe of thyroid gland. (AP and left anterior oblique view)

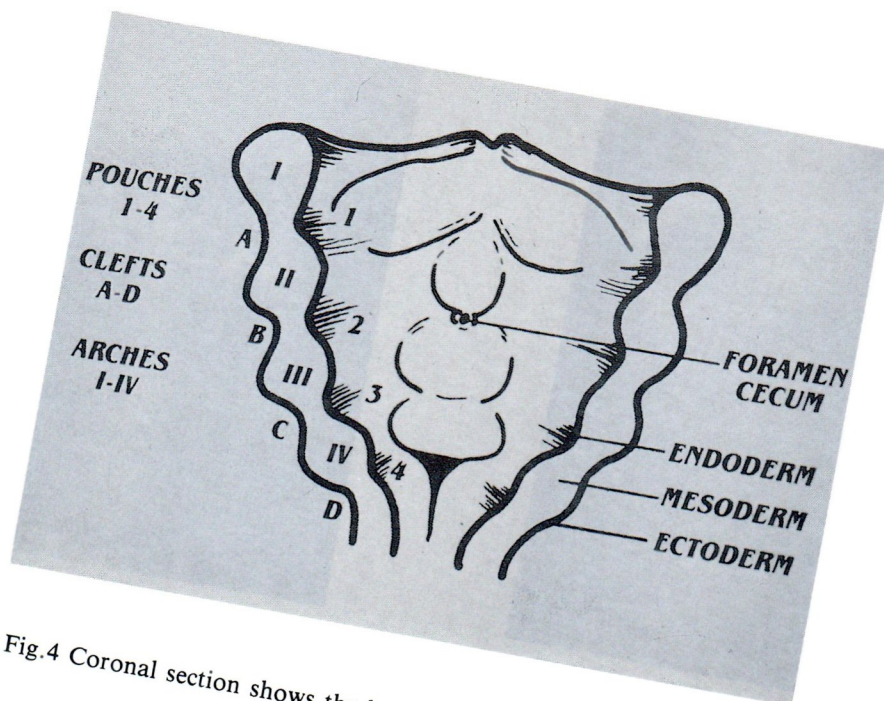


Fig.4 Coronal section shows the branchial pouches, clefts and arches.

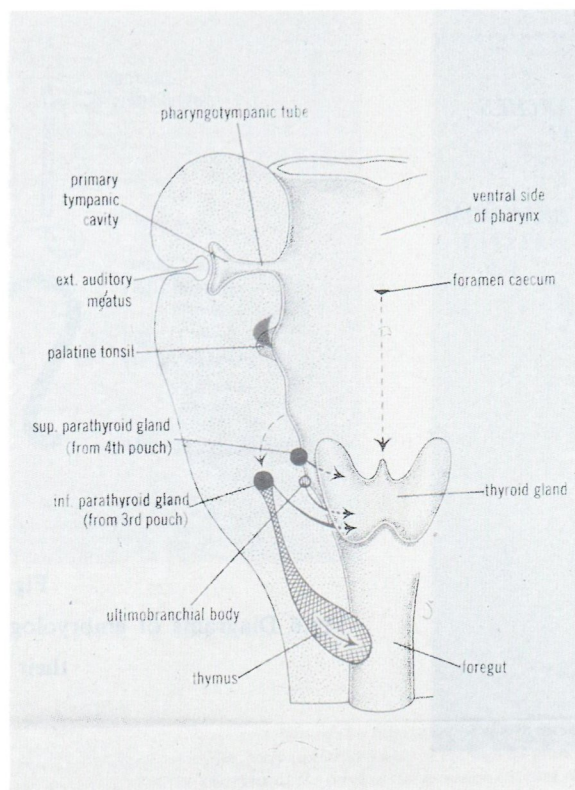


Fig.5 a

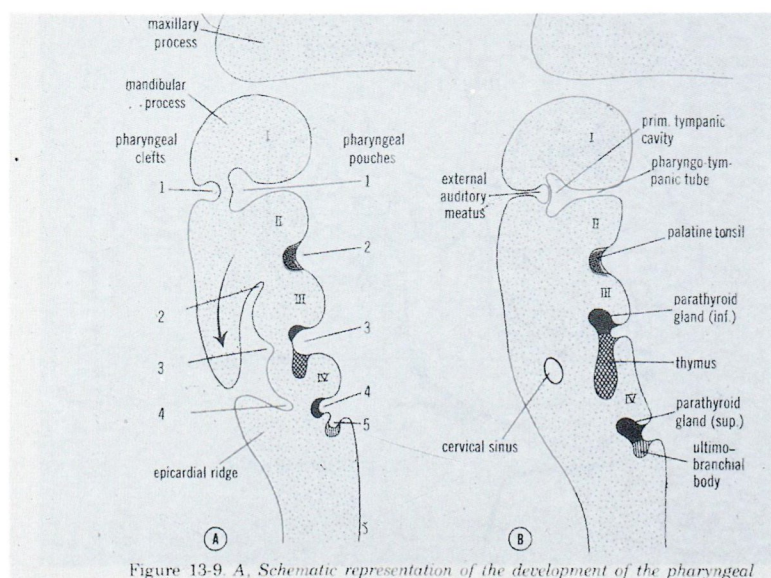


Figure 13-9. A, Schematic representation of the development of the pharyngeal

Fig.5 b

Fig.5 a and b Development of the pharyngeal clefts and pouches.

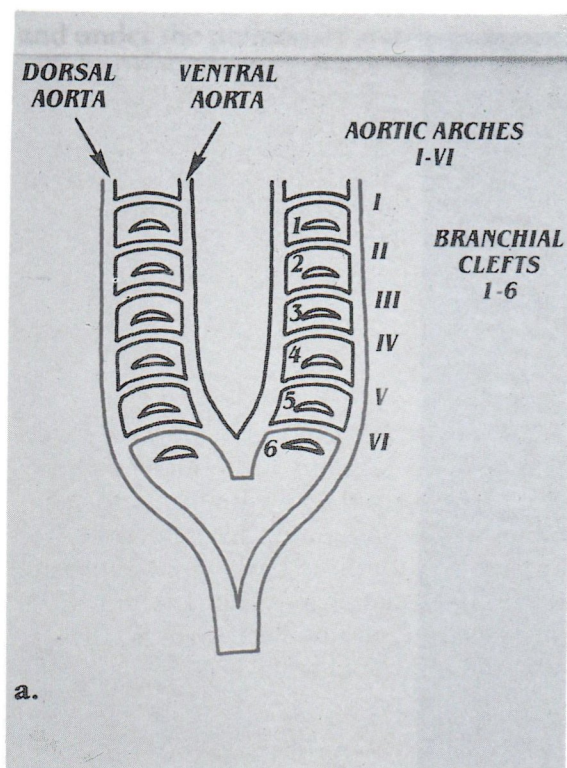


Fig.6 a

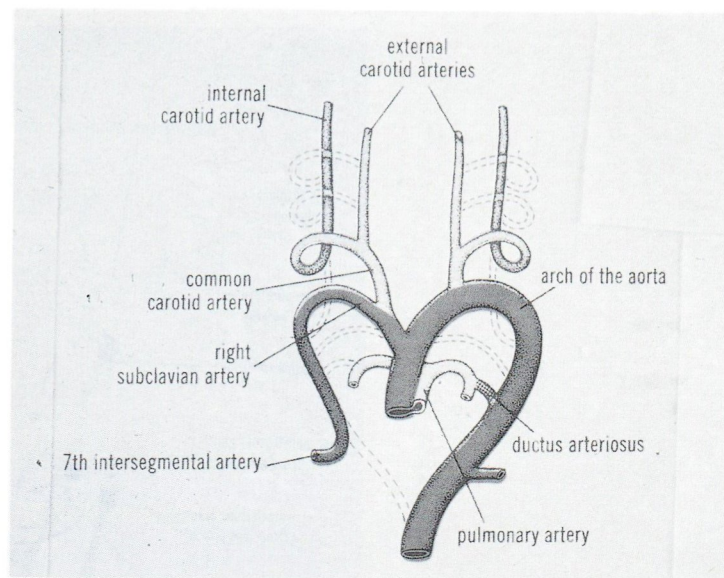


Fig.6 b

Fig.6 Diagrams of embryology of the aortic arch arteries and their relations to the branchial clefts.

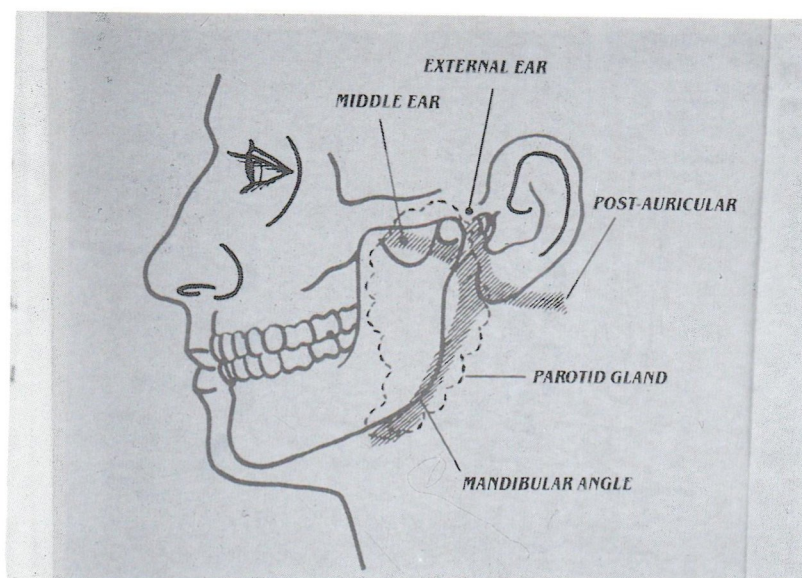


Fig.7 Diagram showed the course and relationships of 1st branchial anomalies.

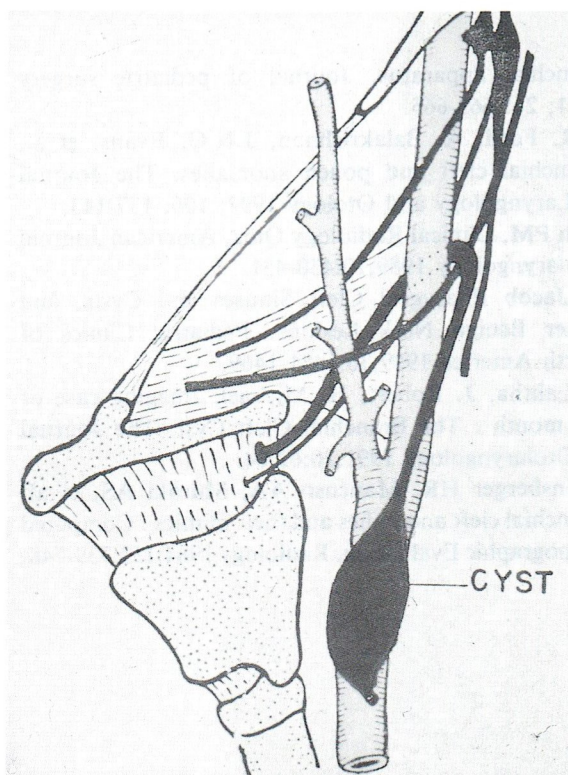


Fig.8 Diagram showed the course and relationship of 2nd branchial anomalies.

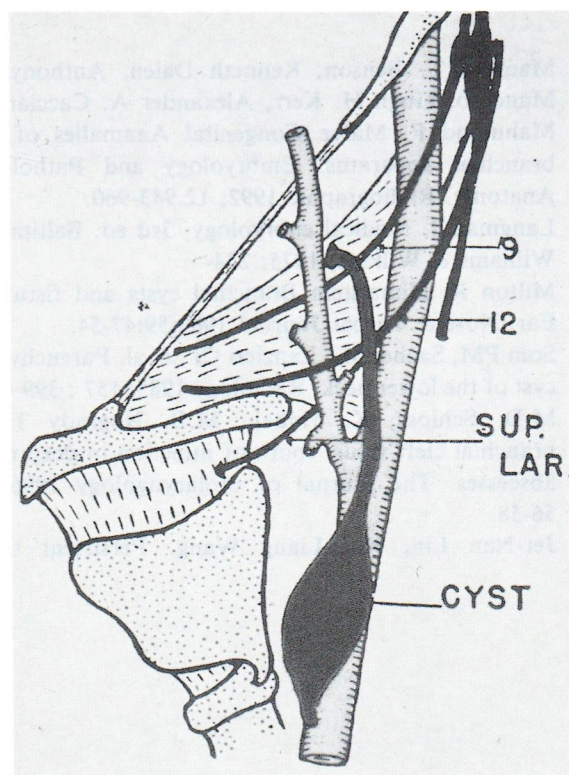


Fig.9 Diagram showed the course and relationship of 3rd branchial anomalies.

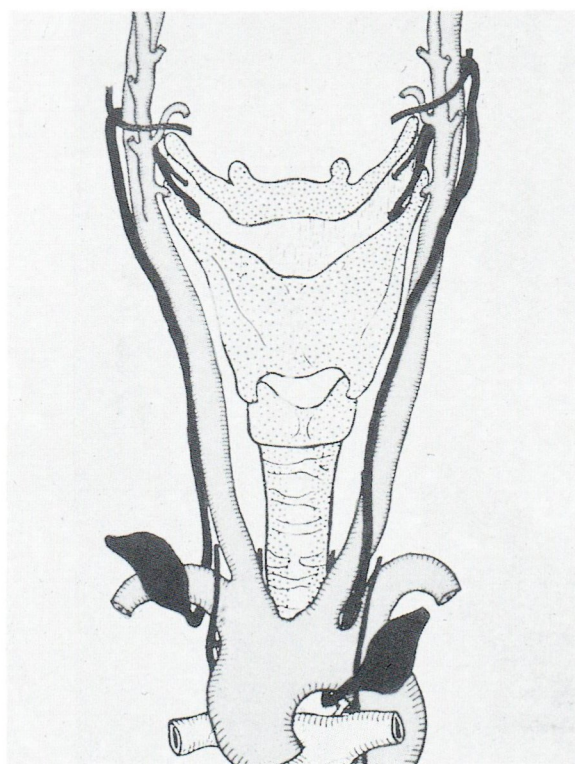


Fig.10 Diagram showed the course and relationship of 4th branchial anomalies.

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