

Case Report

High Resolution Ultrasonography Evaluation of Branchial Cleft Sinus / Fistulas

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Abstract: Diagnosis and differentiation of branchial cleft anomalies rest on higher imaging modalities such as computed tomography (CT) or magnetic resonance imaging (MRI). However, anatomical knowledge and meticulous characterization of anomaly on high resolution ultrasonography (HRUS) can clinch the diagnosis in most cases. Anomalies of the second branchial cleft account for 90% of the developmental abnormalities of the branchial apparatus. However, complete second arch fistulae are rare and comprise 2% of all branchial anomalies. We report here a case of branchial cleft fistula type 2 diagnosed on HRUS.

Keywords: Branchial anomalies, Cyst, Sinus, F istula.

INTRODUCTION

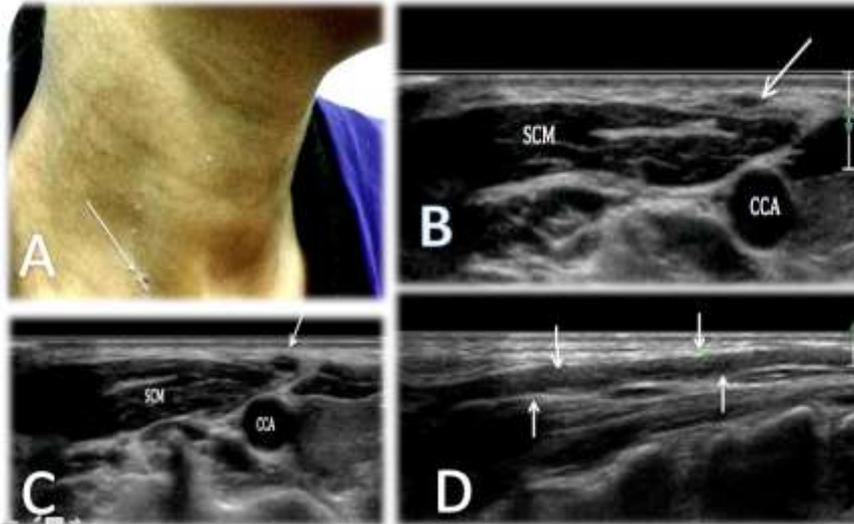
Anomalies of the second branchial cleft account for 90% of the developmental abnormalities of the branchial apparatus [1, 2]. However, complete second arch fistulae are rare and comprise 2% of all branchial anomalies. Branchial cysts are more common (80.8%) than branchial fistulae. Diagnosis and differentiation of branchial cleft anomalies rest on higher imaging modalities such as computed tomography (CT) or magnetic resonance imaging (MRI). The branchial cleft cysts or fistulas embryologically have been classified into various types and anatomical knowledge can thus, be useful in differentiating these types on HRUS.

Most branchial anomalies arise from the second branchial apparatus. Incomplete obliteration of the cervical sinus plays an important role in this process. If no communication occurs with inner mucosa or outer neck skin, trapped arch remnants form a *cyst*. *Sinus* anomalies have, by definition, a communication with the external skin or pharyngeal mucosa and end as a blind tubule or sacculle within mesenchymal tissue. The word *fistula* is applied to the anomaly when both external and internal openings are present.

CASE REPORT

A 18 years old female patient with complaints of discharging sinus from birth in neck was referred by the ENT department of our institute for ultrasonography

neck (Fig. 1A). On examination there was a painless sinus in the right antero-inferior aspect of the neck with serous discharge. Patient was subjected to HRUS neck using 5-12 Mhz linear probe, the opening of the sinus tract was seen anterior to the sternocleidomastoid and deep to platysma muscle (Fig. 1B & 1C). On scanning longitudinally, there was a sinus tract running cranially, anterior to sternocleidomastoid muscle (Fig. 1D). The sinus tract was anterior to common carotid artery (Fig. 2A) and on tracing further it coursed between internal carotid artery (ICA) and external carotid artery (ECA) (Figure 2B). The sinus tract then dipped into the deep neck tissues. There was no evidence of any cystic or localized collection in the deep tissues of neck (Fig. 2C). On basis of sonographic findings, the provisional diagnosis of a Branchial Cleft Sinus/Fistula Type 2 was made. A confirmatory sinogram of the discharging sinus was then advised, where a non-ionic water-soluble contrast was injected through external opening in the anterior aspect of the neck on the right side (Fig. 3A). A fistulous tract was demonstrated opening into oropharynx (tonsillar fossa region) above pyriform sinuses and there was free flow of contrast into laryngopharynx and oesophagus with regurgitation of contrast into oral cavity (Fig. 3B & 3C) confirming a Branchial Cleft Fistula Type 2. The patient was advised for surgery by the ENT surgeons. Subsequent follow up was uneventful.



Fi. 1 (A-D): [A] A 18 years old female patient with a sinus opening on right antero-inferior aspect of neck(arrow).[B][C] HRUS images showing the opening of the sinus tract(arrow) anterior to the sternocleidomastoid muscle. [D] a sinus tract (marked by arrows) seen running cranially, anterior to sternocleidomastoid muscle.

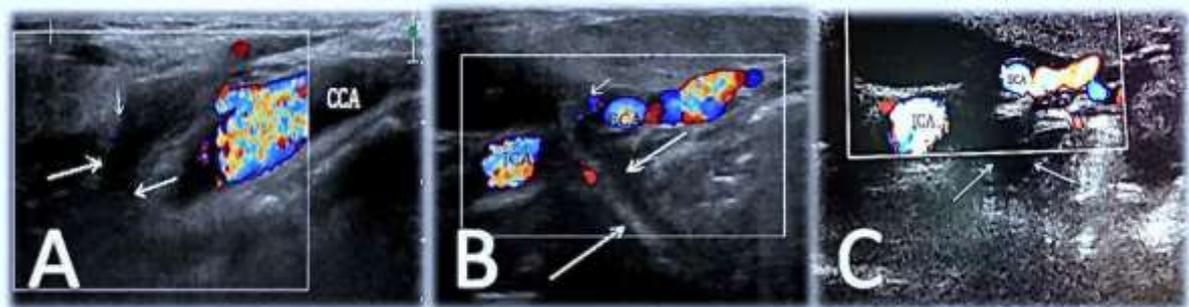


Fig. 2 (A-C): [A] HRUS image showing sinus tract (arrows) coursing cranially anterior to common carotid artery. [B] Sinus tract (arrows) coursing in between internal carotid artery (ICA) and external carotid artery (ECA). [C] Sinus tract (arrows) dipping into the deep neck tissues. No cystic or localized collection in the deep tissues of neck.

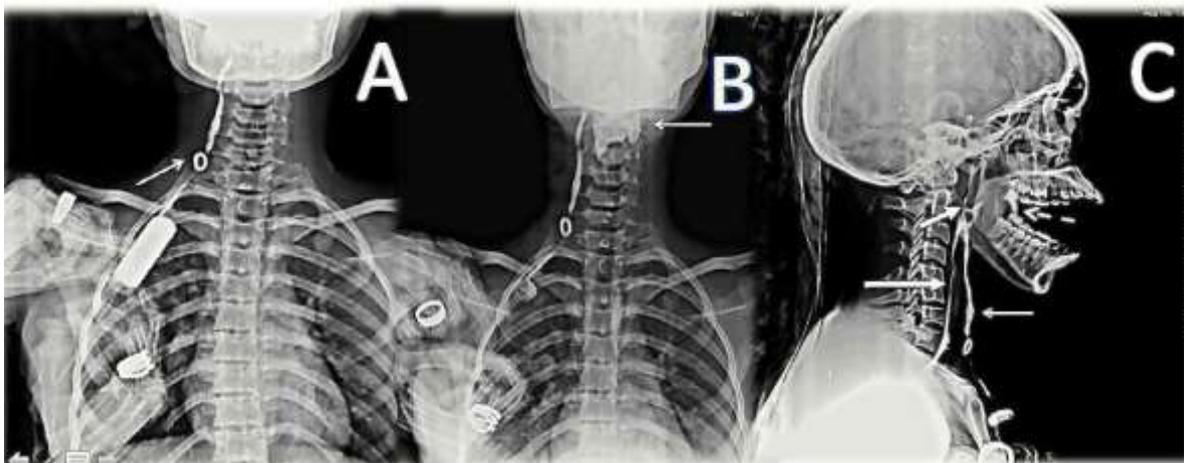


Fig. 3(A-C): Sinogram shows [A] Sinus tract outlined by contrast coursing cranially on right side of neck. [B] A fistulous communication opening into oropharynx (tonsillar fossa region) above pyriform sinuses (arrow).[C] The fistulous tract opening into tonsillar fossa (arrow) there was free flow of contrast into laryngo-pharynx and oesophagus (bold arrows) with regurgitation of contrast into oral cavity(dashed-arrow).

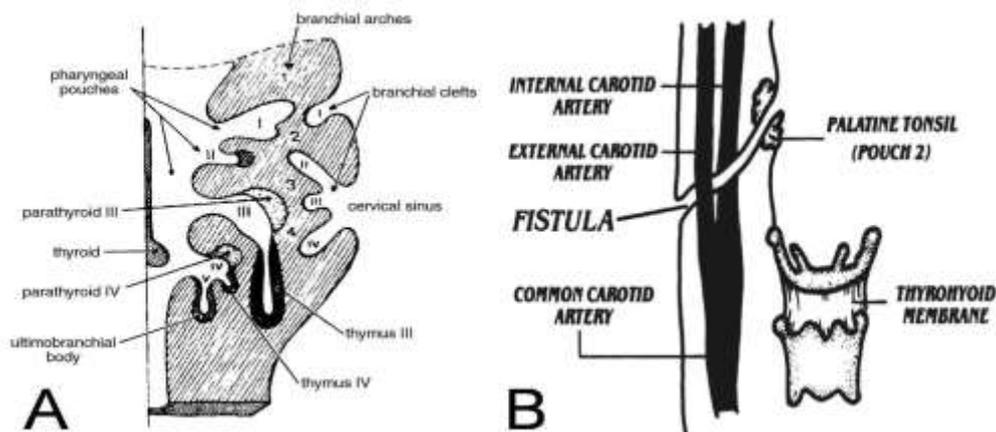


Fig. 4[A-B]: Schematic diagrams showing [A] differentiation of the pharyngeal clefts and pouches. [B] Course of a complete second ranchial fistula passing between the internal and external carotid arteries and extending to the palatine tonsil.

DISCUSSION

Complete second arch fistulae are rare and comprise 2% of all branchial anomalies. The fistula is more commonly seen in males (60%) and can range from 1 to 8 cm in length [3]. It can present at any age, more commonly in the first and second decade of life. Two to ten percent (2-10%) of them can be bilateral. When unilateral, 70% of them occur on the right side. Branchial cysts are more common (80.8%) than branchial fistulae [4].

Most branchial anomalies arise from the second branchial apparatus. A number of theories exist to explain the development of abnormalities within the branchial cleft. The most widely held belief is that incomplete obliteration of the cervical sinus plays an important role in this process. If no communication occurs with inner mucosa or outer neck skin, trapped arch remnants form a *cyst*. *Sinus* anomalies have, by definition, a communication with the external skin or pharyngeal mucosa and end as a blind tubule or sacculle within mesenchymal tissue. The word *fistula* is applied to the anomaly when both external and internal openings are present [5].

Embryology: By the end of 4th week of embryonic life, the brachial arches (derived from neural crest cells) and the mesenchyme (derived from the lateral mesoderms) are easily recognizable. Five pairs of ectodermal clefts (grooves) and five endodermal brachial pouches separate the six arches, with a closing membrane located at the interface between the pouches and the clefts [6]. The *first branchial cleft* normally gives rise to the Eustachian tube, tympanic cavity and mastoid antrum and contributes to the formation of the tympanic membrane. It is the only cleft to contribute to an adult structure, that is the external auditory canal. The second, third, fourth branchial clefts are part of an ectodermally lined depression known as the cervical sinus of His. As the second and the fifth branchial clefts merge with each other, the cervical sinus is obliterated. The second branchial pouch, lined by endoderm gives

rise to the palatine tonsil and the tonsillar fossa. The third branchial pouch forms the inferior parathyroid gland, thymus and pyriform sinus. The fourth branchial pouch forms the superior parathyroid gland and the apex of the pyriform sinus [6].

The branchial cleft cysts or fistulas can be classified as:

Type 1: Sinus tracts drain into the external auditory canal, and complete duplications of the canal can be seen. Best diagnostic clue: Cystic mass around pinna and external auditory canal (EAC) or extending from EAC to angle of mandible [7].

Type 2: Branchial anomalies associated with second arch derivatives are the most common (90-95%) [1].

A second branchial cleft fistula arises anterior to the sternocleidomastoid muscle (SCM). This fistula tracts deep to the platysma and runs superiorly between the internal and external carotid arteries. It passes superficial to the hypoglossal and glossopharyngeal nerves whereupon it empties into the tonsillar fossa [Figure 4A & 4B]. A sinus tract may empty into the tonsillar fossa or the skin anterior to the SCM [8].

Type 3: A third branchial cleft (7%) fistula arises anterior to the SCM and tract deep to the platysma. This fistula tract will pass posterior to the internal carotid artery and between the hypoglossal and glossopharyngeal nerves. The tract will run just superior to the superior laryngeal nerve and empty into the pyriform sinus.

Type 4: Courses from pyriform sinus apex caudal to superior laryngeal nerve, to emerge near the cricothyroid joint and descend superficial to the recurrent laryngeal nerve [9].

Surgery is usually not indicated if the fistula is asymptomatic. However, most are symptomatic and the surgical excision is carried out to avoid the risk of

recurrent infection and for cosmetic reasons. Sclerosing agents are seldom used today due to the associated inflammatory reaction and the risk of necrosis with perforation into the pharynx [1].

Differential diagnosis of second branchial cleft fistula include Type 2 branchial cleft sinus, Type 3 branchial cleft sinus/fistula, Type 4 branchial cleft sinus/fistula [9], orocutaneous fistula secondary to dental infection, salivary gland lesions and neoplasms [10], discharging sinus in the neck secondary to tubercular lymphadenitis [11].

CONCLUSION

Anatomical knowledge and high resolution sonography in concurrence with sinograms cannot only diagnose but also differentiate types of branchial cleft anomalies obviating the need of higher imaging modalities and clinching felicitous diagnosis of a branchial cleft anomaly.

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