



Study of 54 cases of the branchial apparatus anomalies: Our experience with review of literature

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Abstract:

Intricate knowledge of the embryology of the branchial cleft system is fundamental for successful management of branchial remnants. Branchial remnants can present as sinus, cyst or fistula either unilateral or bilateral. There is no predilection for any side (right or left) and gender. The second branchial remnants are the most commonly seen anomaly followed by first, third and fourth. Computed tomography and fistulogram are helpful for anatomical location, relations and to narrow down differential diagnosis. Complete surgical excision is the treatment of choice to prevent recurrences.

Key words: Branchial cleft anomalies; branchial cyst; branchial fistula; branchial remnants; lateral neck swelling.

Introduction

It is important to understand the developmental anatomy and ontogeny of the branchial apparatus for a surgeon to accurately predict the course of branchial cleft remnant [1]. Branchial remnants may present in the form of a cyst, sinus or fistula. If a portion of the cleft fails to involute completely, the entrapped remnant forms an epithelium-lined cyst which is called branchial cyst.

A branchial sinus is a blindly ending pouch with a single opening. Branchial fistula is applied to the remnant when both external and internal openings are present. Depending on their anatomical location, branchial remnants are classified into first, second, third and fourth anomalies. These lesions do not spontaneously regress and often result in recurrent infections, hence require complete surgical excision [2]. The aim of this study is to describe our

experience with 54 patients diagnosed to have branchial remnants, to analyze our methods of diagnosis & management of these lesions and review of literature.

Material and methods

The retrospective and prospective data of 54 patients admitted in ENT Department of our institute from 2005, who were diagnosed of branchial remnants were collected. Analysis of age, gender, origin, surgical procedures and post-surgical evaluation was done. Diagnosis was based on history, clinical examination, radiology reports, intraoperative findings and histopathology. All patients were followed up for atleast six months for any recurrence; patients who had recurrence were reoperated. The branchial remnants were categorized according to site of origin and then into cyst, sinus or fistula.

Results

Our study of 54 patients (28 females and 26 males) had 56 branchial remnants. 46 patients were below age 13 with youngest age of presentation being 8 months old and oldest being 48 years old. Second branchial apparatus anomaly was seen in 42 patients followed by first (10 patients), third (2 patients) and fourth (2 patients) (figure 1). Of these, 27 were on left, 25 on right and 2 patients were having lesions bilaterally. Branchial cyst was more common (37 patients) followed by branchial sinus (15 patients) and branchial fistulae (4 patients) (figure 2). The junction of the upper and middle third of sternocleidomastoid muscle was the most common site for branchial cyst and in one case branchial cyst was found arising from crypta magna of the palatine tonsil (figure 3). Five patients had prior surgical intervention as incision and drainage somewhere else, suggesting misdiagnosis. All patients underwent complete surgical excision after treating infections if any. Two patients with branchial sinus and one patient with branchial fistula required re-exploration.

Discussion

The branchial apparatus is transient structure that is present from weeks 4 to 7 of foetal development [3]. It consists of series of mesodermal arches that are separated from each other externally by ectodermally lined branchial clefts and internally by endodermally lined pharyngeal pouches. By the end of fourth week of gestation, four well defined pairs of branchial arches are visible externally; fifth and sixth arches are small and cannot be seen on embryonic surface. As development proceeds, the

second arch increases in thickness and proliferates caudally to meet enlarging epiperocardial ridge of the fifth arch. As a result the second, third and fourth branchial clefts become enclosed in an ectodermally lined cavity called as cervical sinus of His. Eventually this sinus becomes obliterated by opposition and fusion of its wall yielding a smooth uniform contour to external surface of neck. The endodermal derivatives of pharyngeal pouches including the thymus, parathyroid glands and ultimobranchial body, then migrate to their final position [4]. Incomplete closure of branchial pouches or failure of obliteration of the branchial grooves will result in branchial remnants anomalies.

First branchial cleft anomalies is a rare congenital malformation of the head and neck with an incidence of less than 10% of all branchial cleft defects [5,6] and Rajshekar and Gurucharan [7] presented a rate of 18%. In our study 18% branchial remnants are derived from first branchial cleft. Second branchial cleft anomalies are the most common of branchial remnants [8], although the incidence is variable. In our study, 75% branchial remnants are derived from second branchial cleft. The data of this study about third and fourth branchial remnants confirms previously established rarity of these anomalies [9,10]. In our study, 7 %branchial remnants are derived from third and fourth branchial cleft. Male and females were equally affected by branchial remnants in this study .There is no proclivity of the position of branchial remnants to one side (right or left). However ,Third and fourth branchial remnants have strong predominance for the left side due to the asymmetry of the transformation of the fourth branchial arch to form aorta and innominate arteries [10]. In our study, the locations of all third and fourth branchial anomalies were also on left side.

History and physical examination are the most important elements in the diagnosis of branchial remnants. Computed tomography is non invasive tool to confirm diagnosis of branchial remnants and helpful in guiding surgical therapy, particularly with the more complicated cases [11,12]. Fistulogram or sonogram (figure 4) is helpful in delineating the course of branchial fistula especially in third and fourth branchial remnants [13,14].

Complete surgical excision is necessary as untreated lesions are prone to have recurrent infection and abscess formation [15,16]. Identification and preservation of important anatomical structures like facial nerve, recurrent laryngeal nerve and major neck vessels are

important. Bailey (1933) mentioned stepladder incisions (figure 5) as a standard technique for removal of branchial remnants especially when lesions are long [17]. Methylene blue was used into the fistulous tract to facilitate intra operative identification of the tract. After identification of the fistula or sinus tract, proximal end was closed with pursestring suture to prevent recurrence.

Conclusion

Intricate knowledge of the embryology of the branchial cleft system is fundamental for successful management of branchial remnants. Branchial remnants should be considered in differential diagnosis of lateral neck lesions, especially in children and young adults presenting with a swelling or a tumour in the lateral part of the neck. It is important to understand the developmental anatomy and ontogeny of the branchial apparatus for a surgeon to accurately predict the course of branchial cleft remnant to avoid multiple incision and drainages which may lead to recurrences. The second branchial remnants are most commonly seen anomaly followed by first, third and fourth. Radiographic studies like CT and fistulography are helpful to determine size, depth and the course of branchial remnants. The objectives of therapy should be a complete surgical excision on the first attempt itself.

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Figure 1: Frequency of types of branchial remnants

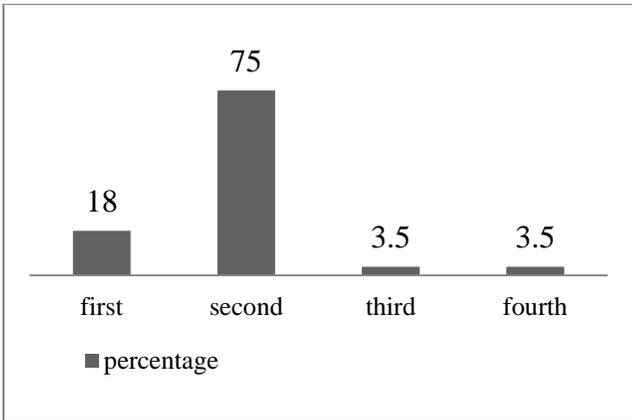


Figure 2: Frequency of cyst, sinus and fistula

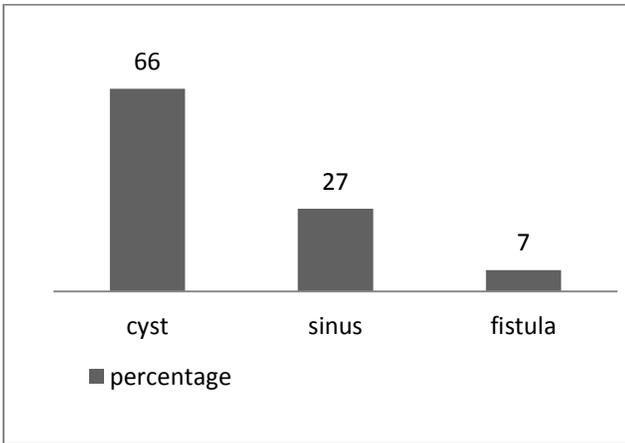


Figure 3: 9 year old female patient had right branchial cleft cyst (→) which was found arising from crypta magna of the palatine tonsil (o)

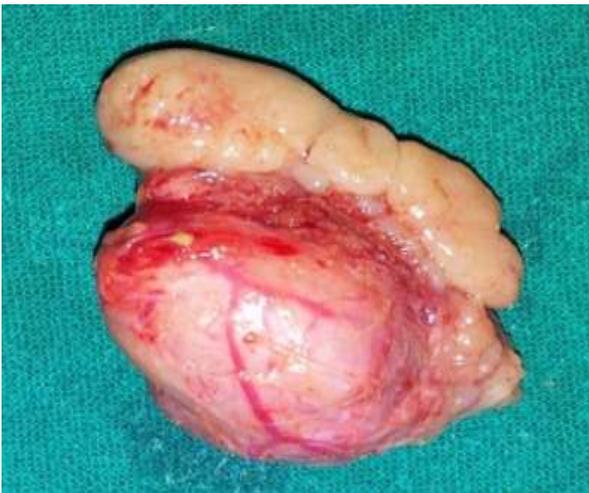


Figure 4: Sinogram of a 5 year old male patient who had recurrent history of discharging from a small opening on the right side of the neck



Figure 5: Figure showing step ladder incision used for removal of second branchial cleft sinus of 12 year old male

