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Anatomical Variation of Branchial Clefts: A Retrospective Study

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This retrospective study was conducted to define the relationship of between existences of the bronchial cysts and fistulas with gender of patients as well as review of literature. Data were collected from the records of total 149 patient files (63 male and 86 female) with including history of having branchial cleft anomalies (cysts and fistulas), that admitted in ENT Department of Imam Khomeini and Apadana Private Hospitals between 1998 and 2008. The analyzed characteristics include gender of patient, anatomical location and type the anomaly. The most frequent type of branchial cleft anomalies in cyst and fistula disorders was second branchial with the values of 85.71% in 21 male patients, 37.5% in total 48 patients; 76.19% in 42 male patients, 31.68% in total 101 patients and values of 62.97% in 27 patients, 35.41% in total 48 cases; 76.27% in 59 female patients, 44.55% in total 101 cases, respectively. There was no significant difference in frequency ($p>0.05$) between male and female patients as well as the locations of anomalies in neck. The overall frequency of the second branchial cleft in different types of disorders such as cysts and fistulas in Southwest region of Iran is the highest, which was similar to international findings. In summary, anatomical location and type of congenital neck masses help narrow the differential diagnosis.

Key words: Cyst, fistulas, sinus, bronchial cleft

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INTRODUCTION

Structures between the developing head and the heart such as the face, neck, oropharynx and the larynx, develop from the branchial apparatus. There are six branchial arches; the last two are rudimentary. Caudal to each of the four arches is an internal pouch lined with endoderm (Faerber and Swartz, 1991). The 5 week embryo is characterized by the presence of four branchial clefts, of which only one contributes to the definitive structure of the embryo (Vazquez *et al*., 1995). The clefts form a cavity lined with ectodermal epithelium, (cervical sinus) but with further development this sinus disappears. The dorsal part of the first cleft penetrates the underlying mesenchyme and gives rise to the external auditory meatus (Carlson, 2004). The epithelial lining at the bottom of the meatus participates in formation of the tympanic membrane (Carlson, 2004). Active proliferation of mesenchymal tissue in the second arch causes it to overlap the third and fourth arches. Finally it merges with the epicardial ridge in the lower part of the neck and the second, third and fourth clefts lose contact with the outside (Sadler, 2006). Branchial anomaly may result from the abnormal persistence of branchial apparatus remnants and they present around each of the developed brachial derivates. Such congenital abnormalities have been traditionally classified as cysts, sinuses, or fistulas (Mandell, 2000; Haba *et al*., 2000). First branchial cleft anomalies are intimately associated with the external auditory canal and the parotid gland (Haba *et al*., 2000). Second branchial cleft anomalies are found along the anterior border of the sternocleidomastoid muscle and most commonly present just lateral to the internal jugular vein at the level of the carotid bifurcation (Lee *et al*., 2006). Anomalies of the third and fourth branchial clefts are relatively uncommon and the distinction between third and fourth branchial anomalies remains controversial, primarily because both lesions similarly present around the perform sinus (Mandell, 2000). Branchial anomaly typically present in infancy and childhood but can be diagnosed for the first time at any age. Definitive treatment for branchial cyst, sinus and fistulas is complete surgical excision. If not treated surgically, these lesions do not spontaneously regress and often result in recurrent infections. The timing of surgery is dependent on the age of the patient and the presence of infection (Mandell, 2000). The frequency of branchial anomaly as original article and some case report study has been reported from many countries (Schroeder *et al*., 2007; Rajshekhar and Gurucharam, 2007; Cagdas and Taner, 2006; Choo *et al*., 2002) but no such study has been reported from Iran. This retrospective study was conducted to define the relationship of presentation of the cysts and fistulas were derived from branchial cleft with gender of patients and also compared with available literatures.

MATERIALS AND METHODS

Patients and data: A total 149 patient's data collected from the records of the ENT Department of Imam Khomeini and Apadana Private Hospitals. The information about patients with branchial cleft anomalies recorded during the 10 year period between 1998 and 2008, were retrieved, reviewed and analyzed. This study was approved by the Institutional Ethics Committee of Ahwaz Jondishapour University of Medical Sciences (AJUMS). Written consents were taken from all the parental participants of this study. The preoperative diagnosis was based on history, clinical examination and radiology reports and postoperative diagnosis was based on the intra-operative findings and postoperative histopathology. The presence and location of a skin opening, the course of fistula tract in relation to surrounding neurovascular structure and the presence and location of mucosal opening all helped defined the diagnosis. The diagnosis of anomaly accompany with branchial cleft was categorized based on the suspected branchial cleft of origin and then further subdivided into cyst, sinus and fistula. The main analysis outcomes were gender of the patients, location and type the branchial cleft anomalies.

Statistical analysis: The data were analyzed statistically by SPSS 13.0. Association between the type and location of branchial cleft anomalies and sex variable of patient has done using Chi-square test. A p-value less than 0.05 were accepted statistically significant.

RESULTS

Out of 149 subjects, 48 cases (21 male and 27 female) with cystic anomalies and 101 (42 male and 59 female cases) with fistula anomaly have been detected. The age range was from 3 months to 13 years (mean age of 5±1.8 years).

The majority of observed cyst in neck among two genders was derived from second branchial cleft (85.71% in male patients, 37.5% in total 48 patients; 62.97% in female patients, 35.41% in total 48 cases) (Table 1). Figure 1a-c show a 25 year-old female patient with a cyst abnormality. The difference between two gender statistically was not significant ($p>0.05$). The cyst reviewed here were also almost equally distributed between left and right sides of neck ($p>0.05$). Bilateral cysts were seen only in few cases. The frequency of cyst

Table 1: Number and depended frequencies of cysts type in study group (n = 48)

	Male (n = 21)					Female (n = 27)				
	Unilateral					Unilateral				
Origin	Right No. (%)	Left No. (%)	Total No. (%)	Bilateral No. (%)	Total No. (%)	Right No. (%)	Left No. (%)	Total No. (%)	Bilateral No. (%)	Total No. (%)
B.C.I	1(4.76)	1(4.76)	2(9.53)	-	2(9.52)	2(7.40)	5(18.51)	7(25.91)	1(3.70)	8(29.63)
B.C.II	5(23.80)	10(47.61)	15(71.43)	3(14.28)	18(85.71) *	4(14.81)	9(33.33)	13(48.14)	4(14.81)	17(62.97)*
B.C.III	1(4.76)	-	1(4.76)	-	1(4.76)	-	1(3.70)	1(3.70)	-	1(3.70)
B.C.IV	-	-	-	-	-	-	1(3.70)	1(3.70)	-	1(3.70)†
Total	7(33.33)	11(52.38)	18(85.72)	3(14.28)	21(100)	6(52.08)	16(43.75)	22(81.49)	5(18.51)	27(100)

*Indicates most frequent type; †: Indicates less frequent type

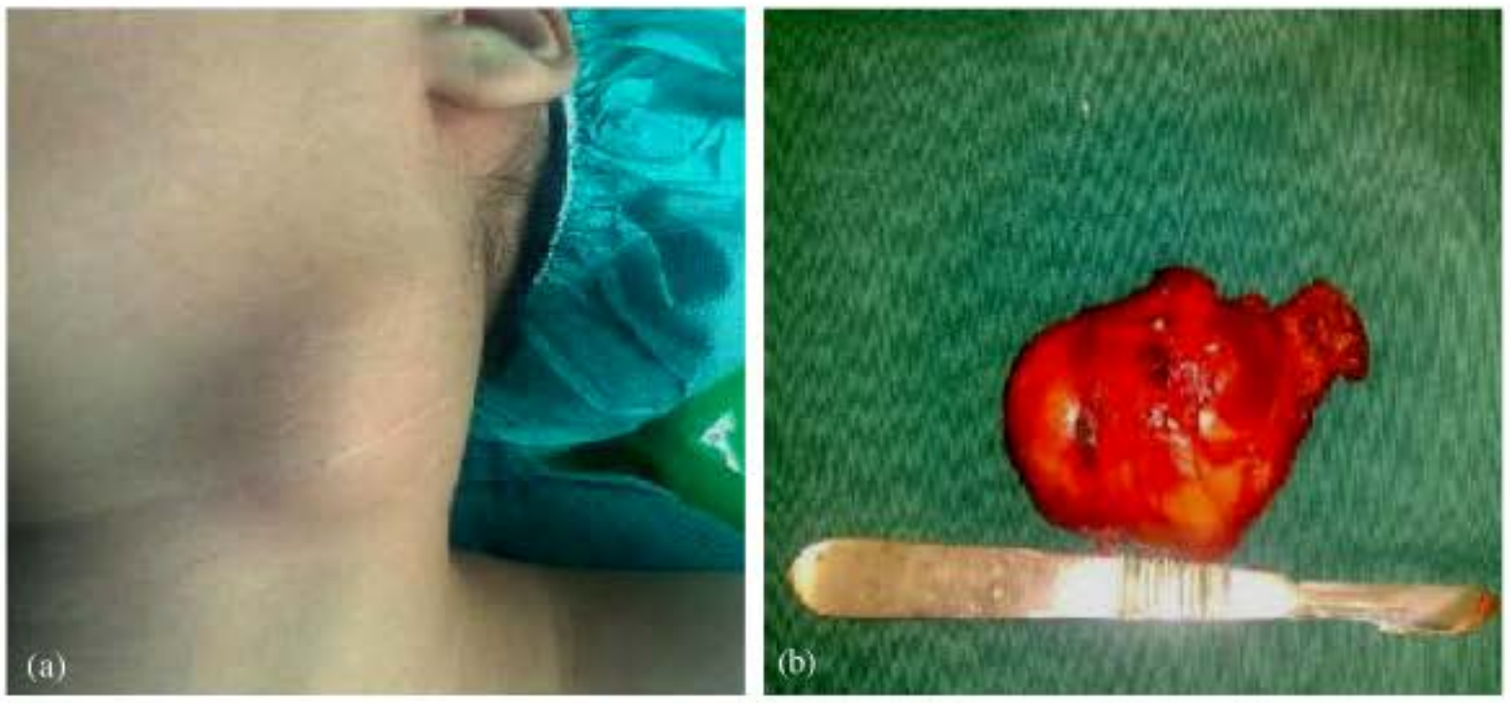

Fig. 1: (a) Left sided branchial cyst in a 25 female patient, (b) surgical extracted cyst and (c) axial CT scan from the cyst mass

derived third branchial cleft was very rare in male but in female cases the rare cyst was derived from the fourth cleft.

The frequency of fistula in neck among different genders was derived from second branchial (76.19% in male patients, 31.68% in total 101 patients; 76.27% in female patients, 44.55% in total 101 cases) (Table 2). Figure 2a-c show bilateral fistula in a 34 year-old male patient and second type branchial fistula extended from neck to external auditory meatus in a female 19 year-old patient and stapp-ladder surgical incision. The difference between two gender statistically was not significant ($p>0.05$). The fistula reviewed here were also equally distributed between left and right sides of neck ($p>0.05$). Bilateral cyst were seen only in few cases. The frequency of fistula derived third branchial cleft was zero in male but in female cases the frequency was zero for fistulas which derived from fourth branchial cleft.

Table 2: Number and depended frequencies of fistula type in study group (n = 101)

	Male (n = 42)					Female (n = 59)				
	Unilateral					Unilateral				
Origin	Right No. (%)	Left No. (%)	Total No. (%)	Bilateral No. (%)	Total No. (%)	Right No. (%)	Left No. (%)	Total No. (%)	Bilateral No. (%)	Total No. (%)
B.C.I	2(4.76)	4(9.52)	6(14.28)	1(2.38)	7(16.66)	6(10.16)	3(5.08)	9(15.25)	3(5.08)	12(20.33)
B.C.II	8(19.05)	20(47.61)	28(66.66)	4(9.54)	32(76.19) *	19(32.20)	24(40.67)	43(72.87)	2(3.40)	45(76.27)*
B.C.III	1(2.38)	1(2.38)	2(4.76)	-	2(4.76)	-	-	-	1(1.70)	1(1.70)
B.C.IV	-	1(2.38)	1(2.38)	-	1(2.38)€	1(1.70)	-	1(1.70)	-	1(1.70)†
Total	11(41.86)	26(46.51)	37(88.08)	5(11.92)	42(100)	26(44.05)	27(45.72)	53(89.77)	6(10.16)	59(100)

*Indicates most frequent type; †: Indicates less frequent type

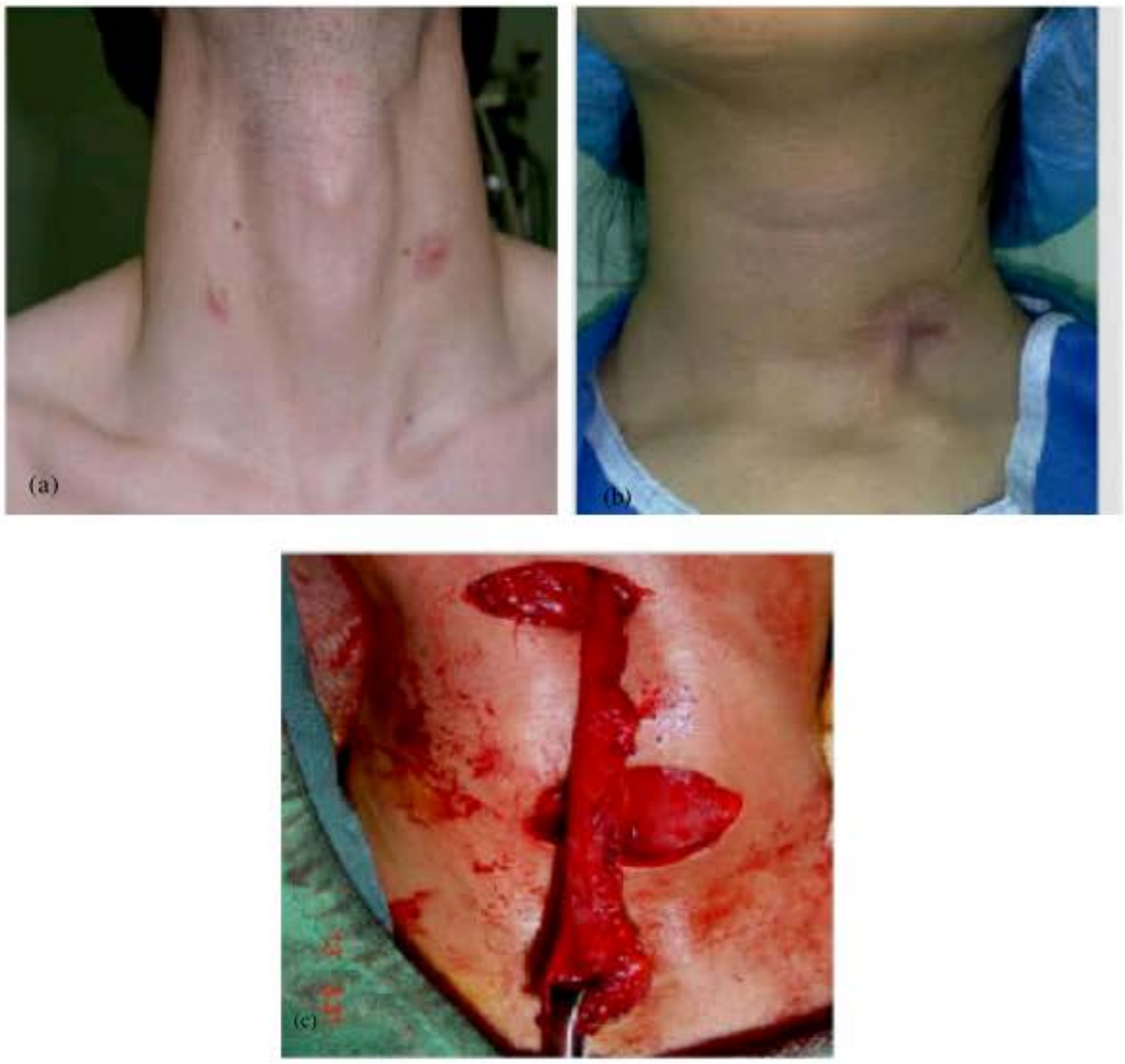

Fig. 2: (a) Bilateral fistula in a 34 year-old male patient, (b) second type branchial fistula extended from neck to external auditory meatus in a female 19 year-old patient and (c) stapp-ladder surgical incision

DISCUSSION

Branchial cleft remnant accounts for the majority of branchial cleft anomalies. Clinically they can present as different morphologic patterns of cyst (a tract with no opening), sinus (a tract with one opening), or fistula (a tract with two openings). A sinus forms when a branchial cleft or branchial pouch fails to obliterate and subsequently communicates with either the skin or the mucosa, respectively , of the upper airway. A fistula results when both a branchial cleft and a pouch fail to obliterate, causing a continuous communication between the mucosa and the skin. A cyst is created when a branchial cleft remnant forms an epidermis-lined space without a communication to the skin or mucosa (Schroeder *et al*., 2007). 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

(Triglia *et al*., 1998; Ford *et al*., 1992) and Rajshekhar and Gurucharam (2007) presented a rate of 18% but Agaton-Bonilla and Gay-Escoda (1996) has reported rates as high as 25% (1996). In this study 9-29% of branchial cleft cyst is derived from first branchial cleft but the frequency of fistula presented higher rate of 16-20% that were little bit high comparing to other reports worldwide. Second branchial cleft anomalies are the most common of the branchial anomalies (Schroeder *et al*., 2007; Chaudhary *et al*., 2003). In the current series, the majorities (62/149 = 41.61) of branchial cleft cysts and fistula were derived from the second cleft; this is similar to earlier findings of Schroeder *et al*. (2007) and Chaudhary *et al*. (2003). The data of this study about third and fourth branchial cleft anomalies confirms the previously established extreme rarity of these anomalies (Liberman *et al*., 2002; Lin and Wang, 1991). Males and females were equally affected by branchial anomalies in this study. This is similar to earlier findings of Schroeder *et al*. (2007). The anomalies reviewed here also equally distributed the right and left sides of the neck. However, it has been reported that third and fourth branchial anomalies have a strong predominance for the left side due to the asymmetry of the transformation of the fourth branchial arch to form the aorta and the innominate arteries (Lin and Wang, 1991). But in the current series, there was not any difference in the location of third and fourth branchial anomalies.

In conclusion, frequency of the second branchial cleft in different types of disorders such as cysts, sinus and fistulas in south west Iranian children is the highest, which was similar to international findings. In summary, anatomical location and type of congenital neck masses help narrow the differential diagnosis. The treatment of first branchial cleft anomalies requires understanding of the various specific types of lesions (cyst, sinus, or fistula) and their relationship to the facial nerve. Successful surgery mandates complete resection and facial nerve preservation. Both of these goals are best accomplished by initial dissection and exposure of the facial nerve, followed by a safe excision of the anomaly.

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