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Second branchial cleft anomalies: a review of 12 cases

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ABSTRACT

Background: It is a combined retrospective and prospective study of 12 cases of branchial cleft anomalies presented to our hospital. We analysed them for their age and sex incidence, side prevalence, according to clinical features and treatment outcomes.
Methods: Thorough history was taken and examination was done and these cases are investigated with CT scan, MRI scan and treated with surgical excision.
Results: Cyst is seen in 7 (58.33%) patients while one case of sinus and one (8.33%) of fistula,1 case of co-existing sinus and fistula (8.33%), and one Bilateral sinus and 1 bilateral fistula (8.33%) were seen. Overall incidences of branchial anomalies are more (40%) in 11-15 year age group. Overall incidence of branchial anomalies is more in male (66.66%) than female (33.33%). 4 cysts were right sided and 3 were left sided. Sinus and fistula were seen one case having right sided and 1 case having bilateral anomaly. Patients having branchial cysts presents most commonly with neck swelling and pain while patients having fistula and sinus presents commonly with discharging opening over anterior neck. 1 case of recurrence found in cyst on follow up and no recurrences were seen in sinus and fistula.
Conclusions: Cyst is the most common anomaly, most commonly affected age group is 11-15 years, male are more commonly affected, these anomalies are more prevalent on right side, and there are very less chances of recurrence after surgical excision.

Keywords: Cyst, Fistula, Sinus, Branchial

INTRODUCTION

Branchial fistulas and cysts and sinuses are anomalies of embryonic development. Approximately 20% of all paediatric cervical masses are due to branchial anomalies.[1] "Branchia" - Greek word for gill – this name is given to these structures due to their resemblance to gills of fishes. Branchial arch consists of core of mesenchyme covered externally by ectoderm and internally by endoderm.

There are six branchial arches from which important structures of head and neck develops. Out of them fifth is rudimentary and sixth arch disappears. These anomalies are types into first, second, third and fourth anomalies according to their location.

First branchial fistula is rare - less than 5 percent. External lower opening in the neck varies in position but lies on a line from the tragus to the hyoid bone and may look like a small skin-lined pit.[2] It is present since birth and may become obvious by squamous or epithelial discharge from it. The upper end –variable, anterior to the tragus, the opening may open in the floor of the external auditory canal, or may form a duplicated canal under the external auditory canal. Work has divided them into two types according to the presence or absence of mesothelial elements within the wall.

Second arch cysts are located along the anterior border of the sternocleidomastoid muscle along its upper one third.

Second arch fistulae and sinuses are much the commonest of the branchial arch anomalies (95 percent).[3] Third and fourth branchial anomalies are quite rare. There are four types of second branchial anomalies (Figure 1):

Type 1: Located along the anterior border of SCM at the junction of the middle and lower one thirds, deep to the platysma and cervical fascia.

Type 2: Lie in contact with great vessels (most common).

Type 3: Pass medially between ICA and ECA, extending towards the lateral pharyngeal wall and lying above the glossopharyngeal and hypoglossal nerves and below the stylohyoid ligament.

Type 4: Very rare and located next to the pharyngeal wall, medial to the great vessels at the level of the tonsillar fossa. It may extend superiorly up to skull base.

The second branchial cleft fistula presents as a congenital opening on the lower neck anterior to the sternomastoid muscle. The track of the second arch fistula is directed upwards and medial to mass between the internal and external carotid arteries. The upper end communicates with the pharynx through the tonsil. These fistulae nearly always leak clear or mucoid fluid from the lower opening. Opening of the third and fourth anomalies are related to the piriform fossa.

Our study is a combined retrospective and prospective study of 12 cases of branchial cleft anomalies presented to our hospital. We analysed them for their age and sex incidence, side prevalence, according to clinical features and treatment outcomes.

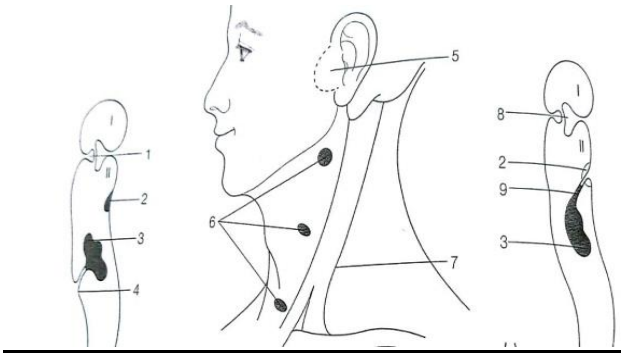

Figure 1: Site of branchial fistula opening.

METHODS

Our study is a combined retrospective and prospective study of 12 cases of branchial anomalies presented to department of ENT, MP Shah Government Medical College and GG Hospital Jamnagar from 2014 to 2017. In all cases, thorough history was taken and complete examination of head and neck was done. All routine blood investigations were done. And cases of cysts were investigated with ultrasound and sinus and fistula were investigated with CT/MRI and sonogram (Figures 4, 5 and 6). In case of infected cyst, it was first treated with injectable antibiotics and then operated upon. In cases with branchial anomalies associated with other congenital anomalies were excluded. Elliptical incision was given in case of sinus or fistula and skin crease incision was given in case if cyst. In almost all cases of fistula, tonsillectomy was done by standard intraoral approach. Data collected was analyzed by mean values and simple percentage methods.

Figure 2: Branchial fistula.

Figure 3: Branchial cyst.

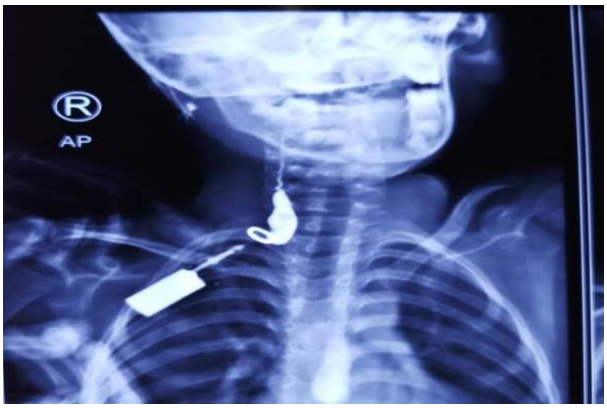

Figure 4: Sinogram-ap view.

Figure 5: Sinogram–lateral view.

Figure 6: Sinogram-oblique view.

RESULTS

Overall incidence of cyst is more (58.33%) than sinus and fistula whose incidence is equal (8.33%) followed by co-existing sinus and fistula (8.33%), Bilateral sinus and bilateral fistula (8.33%). Three patients came with bilateral branchial anomalies. One with bilateral sinus, another was bilateral fistula and one patient came having sinus with fistula.

Table 1: Comparative incidence of these anomalies

Anomaly	Incidence rate
	N (%)
Cyst	7 (58.33)
Sinus	1 (8.33)
Bilateral sinus	1 (8.33)
Fistula	1 (8.33)
Bilateral fistula	1 (8.33)
Sinus with fistula	1 (8.33)

Incidences of these anomalies are more in 11-15 year age group.

Table 2: Incidence in different age groups.

Age groups (in years)	0-5	6-10	11-15	>15
	N (%)	N (%)	N (%)	N (%)
Cyst	0 (0)	2 (28.56)	3 (42.85)	2 (28.56)
Fistula	0 (0)	2 (50)	2 (50)	0 (0)
Sinus	0 (0)	2 (50)	2 (50)	0 (0)
Total anomalies	0 (0)	6 (40)	7 (46.66)	2 (13.33)

Table 3: Overall sex wise distribution.

Sex	N=15
Male	10
Female	5

Table 4: Sex prevalence of different anomalies

	Male	Female
	N (%)	N (%)
Cyst	3 (42.85)	4 (57.14)
Fistula	4 (100)	0 (0)
Sinus	3 (75)	1 (25)
Bilateral anomaly	3 (100)	0 (0)

Cysts are seen more in female (57.14%) than male (42.85%). Fistula is seen almost exclusively in male (100%). Sinuses are seen more in male (75%) than females (25%).

Overall incidence of branchial anomalies is more in male than female.

Bilateral branchial anomaly has more male incidence.

Table 5: Sidewise distribution.

	Left	Right	Bilateral
Cyst	3	4	0
Fistula	0	1	1
Sinus	0	1	1
Sinus with fistula	0	0	1
Total side Incidence	3	6	3

Cysts are more commonly seen on right side (57.14%) than left side (42.85%).

Prevalence of sinus and fistula are equal bilaterally and on right side (50%) side.

Bilaterallity is seen equally in sinus and fistula.

Overall incidences of branchial anomalies are more on right side.

Table 6: Comparison between clinical features.

	Swelling	Opening over neck	Pain	Fever	Discharge	Superinfection with burst abscess
Cyst	6	Na	3	1	Na	1
Fistula	0	4	0	0	4	0
Sinus	0	4	0	0	4	0

Patients having branchial cysts presents most commonly with neck swelling while patients having fistula and sinus presents commonly with discharging opening over anterior neck.

One patient having branchial cyst presented with infected burst open abscess and fever.

Recurrence rate is more (14.28%) in cysts and no recurrences were seen in sinus and fistula. Overall recurrence rate is 6.66% among these branchial anomalies.

DISCUSSION

In our study, unilateral incidence is seen in 75% and bilateral incidence is seen in 25% cases which is compared to study by Teo et al is conclusive in terms of unilaterality and bilaterality.[4]

Male incidence is seen in 66.66% while female incidence is seen in 33.33% cases which is same as study by D'souza et al.[5]

In our study 6.66% recurrence rate is seen compared to study by Takimoto et al and Spinelli.[6,7]

In a study by Spinelli branchial fistula is most common second branchial anomaly followed by cyst, combined cyst with sinus.[7] In our study, the results are contradictory with above mentioned study. According to our study, cyst is most common followed by fistula and sinus.

CONCLUSION

The results of our study suggest that history, clinical examination and embryological anatomy are important factors in diagnosing branchial anomalies. Accurate diagnosis with sinogram and CT scan is necessary for planning of surgery. Proper surgical technique prevents recurrences and multiple surgical procedures. Overall incidences of branchial anomalies are more in 11-15 years age group. Overall incidence of branchial anomalies is more in males whereas cysts are common in female and fistula and sinuses are common in male. Overall incidences of branchial anomalies are more on right side. Recurrence rate is more in cysts and no recurrences were seen in sinus and fistula.

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