



# Complete Second Branchial Cleft Fistulas: A Clinicosurgical Experience

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**Abstract** A complete second branchial fistula is very rare and has an internal opening at the tonsillar fossa and an external opening at the lower third of the sternocleidomastoid (SCM). Patients commonly present with persistent or intermittent mucoid or mucopurulent discharge from an external opening. The diagnosis is most often clinical and radiological investigations are rarely needed. Treatment of choice is complete surgical excision. The aim of this article is to aware young ENT surgeons of the various clinical and intraoperative surgical findings that can be encountered while dealing with these cases. This observational study was done for a period of 10 years. A total of 20 cases of fistula were included which intraoperatively had a complete track from tonsillar fossa to neck. Excision of the tract was carried out via combined transcervical and transoral approach under general anaesthesia using two incisions in stepladder pattern. Each patient was seen after one year of surgery to assess for any recurrence. Different findings of patients including age/sex at surgery, initial presentation, family

history, laterality of the fistula tract, Intraoperative surgical findings, complications, and recurrences. were noted. Of the 20 patients, 13 (65%) were females and 7 (35%) were Males. Most common complaint was fistulous opening with intermittent discharge (15 patients; 75%). Branchial cleft fistulae more commonly affected the right neck (14 patients, 70%) among unilateral cases and 2 patients (10%) had bilateral fistulae. No patient had associated congenital anomaly/syndrome, family history or visible opening in tonsillar area. Glossopharyngeal nerve was identified in 12 cases and track was seen passing lateral to it except in one case. The internal opening of track was seen over posterior tonsillar pillar in 15 cases (75%) while in 5 patients the track was seen entering tonsillar tissue or bed. Tonsillectomy was done in 5 cases while not done in 15 cases where track was seen entering posterior pillar. All patients were seen at one year follow up. No recurrence was seen at one year of follow up. Complete second branchial cleft fistulae are rare. They are usually right sided and unilateral. The track passes between carotid bifurcation and invariably passes lateral to both glossopharyngeal and hypoglossal nerves. Track usually ends at the posterior tonsillar pillar. Tonsillectomy is not routinely indicated. Recurrences are not typically seen.

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## Introduction

Von Baer first described branchial apparatus and its anomalies were first described by Von Ascheron [1]. The anomalies of Branchial cleft are formed due to failure of embryonic structures to obliterate during development [2]. These anomalies are the second most common pediatric congenital

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head and neck masses, accounting for approximately 20% of cases [3]. Ninety to ninety five percent of the branchial apparatus anomalies are attributed to anomalies of the second branchial cleft [4]. They can present as true fistulae, sinus tracts, complete isolated cysts, or cartilaginous remnants [5]. However, complete second arch fistula with internal opening into the tonsillar region is rare and comprises 2% of all branchial anomalies [6].

Branchial fistulae are formed due to the abnormal persistence of the embryonic second branchial cleft [2]. These fistulae are lined with epithelium. A complete second branchial fistula has an internal opening at the tonsillar fossa and an external opening at the skin in the line of the anterior border of the lower third of the sternocleidomastoid (SCM) [2]. The tract of these fistulae is described as coursing over both the glossopharyngeal (IX) and hypoglossal (XII) nerves, then traversing between the internal and external carotid arteries [7].

Affected patients commonly present within the first two decades of life [8]. Patients present with persistent or intermittent mucoid or mucopurulent discharge from an opening in the lateral aspect of the neck following mostly after an upper respiratory tract infection [8]. There is slight female preponderance. Usually, they are unilateral and more so involve right side frequently. Bilateral branchial fistulas have been documented [9].

The diagnosis is most often clinical and radiological investigations are rarely needed. Contrast fistulogram if performed delineates the tract and it is often the commonest investigation available. A complete fistula demonstrable by a fistulogram is uncommon [10]. Methylene blue injection in the fistula tract facilitates to delineate the course of the tract, and the presence of methylene blue in the throat can also definitively confirm a fistula [11]. Occasionally, thick secretions or granulation tissue may obstruct the fistula tract thereby giving a false negative methylene blue test [8].

Computed tomography (CT) scan and magnetic resonance imaging (MRI) of the neck are helpful to produce cross-sectional images of organ and other internal body structures. They are useful mainly in delineating the relationship of surrounding neurovascular structures to the lesion [8].

Treatment of choice for branchial fistula is complete surgical excision of the fistulous tract by using combined transoral and transcervical approach. Incisions for the excision could be Hockey Stick-type or Stepladder incision [8]. The standard surgery for a second branchial arch fistula is the stepladder approach. It is done through two incisions in the neck that gives exposure of the fistula tract with less tissue dissection. The higher incision must be bigger than the lower to avoid the damage to important neurovascular structures [8]. Ipsilateral tonsillectomy has been advocated by some authors given the fact that the tract terminates at the tonsillar fossa.

Most frequent complication of the surgery is recurrence, occurring in 3% of fresh cases to up to 20% in second surgical attempts especially when only external approach is used [6]. However, no recurrence rate is reported after using combined transoral and transcervical approach [8].

The rarity of these complete second branchial arch fistulae's demands that the young ENT surgeons should be made aware of all the aspects related to these fistulae's. The aim of this article is thus to aware young ENT surgeons of the various clinical and intraoperative surgical findings that can be encountered while dealing with these cases so that they do such cases perfectly without any fear of complications/recurrence.

## Methods

This observational study was done in the department of ENT at a tertiary care teaching hospital for a period of 10 years from June 2012 to June 2022. A total of 20 cases of complete second branchial fistula were included in this study with the following inclusion and exclusion criteria's.

### Inclusion Criteria's

- Patients of any age group
- Intraoperative confirmation of fistulous track from neck to tonsillar area
- Unilateral or bilateral second branchial cleft fistulae

### Exclusion Criteria's

- Patients with a history of previous neck surgeries and neck trauma
- Patients with external opening but without a complete track

A preoperative fistulogram was done in some cases to reveal the extent of the tract. CT or MRI was not done in any case. Excision of the tract was carried out via combined transcervical and transoral approach under general anaesthesia. Intraoperatively, methylene blue dye was injected through the external opening followed by canalization with a lacrimal probe.

Two incisions were given to remove fistulous track in all except in one case. First incision was circumscribed taken around the fistulous opening. Then the fistulous tract was separated from the skin and fascia over it. External fistulous opening was intermittently pulled to delineate the track. A second incision (stepladder dissection) a bit bigger than the first incision was taken in the upper part of the neck at the level of the hyoid bone over the anterior border of the sternocleidomastoid. The fistulous track was pulled up

and delivered through the upper incision. Meticulously the fistulous tract was followed over the hypoglossal nerve and between the carotid bifurcation. Track was further followed up to tonsillar area. Glossopharyngeal nerve was encountered during deep dissection in some cases while further following the track up to oropharynx. The track was pulled via oropharynx and delivered through mouth in all cases. Dissection was done by closely following and adhering to the tract in all our cases.

The mucosal opening was closed with 3 o vicryl sutures wherever need was felt. All patients underwent procedure with the same surgical technique. All patients were put on standard postoperative treatment plan which included post operative antibiotics and anti-inflammatory agents with suture removal on 7th day. First follow up of patients was between 1st to 2nd week and few further follow ups were done as an when patient reported. Each patient was seen after one year of surgery to assess for any recurrence.

Basic demographic data were collected. Data collected included age/sex at surgery, initial presentation, family history, laterality of the fistula tract, Intraoperative surgical findings, complications, and recurrences. The data collected is summarised in two descriptive tables.

## Results

Total number of patients fulfilling the inclusion and exclusion criteria were 20. Of the 20 patients, 13 (65%) were females and 7 (35%) were males (Table 1).

Average age at time of surgery was 13 years (range 4–25 years). Majority of patient (13,65%) were in the age group of 10 to 20 years.

External opening of fistula was located in almost all cases in lower neck except one case where it was at upper one third (Fig. 1).

Most common complaint was fistulous opening with intermittent discharge (15 patients; 75%) followed by only fistulous opening in 4 patients (20%) and recurrent abscess in 1 patient (5%) (Table 1).

Branchial cleft fistulae more commonly affected the right neck (14 patients, 78%) among unilateral cases and 2 patients (10%) had bilateral fistulae (Fig. 1).

Fistulogram (Fig. 2) was done in only 9 patients (45%). Methylene blue was injected in all cases intraoperatively and entered tonsillar area in only 15 cases (75%).

Length of fistulous track intraoperatively ranged from 5 to 13 cm with a mean of 8 cm (Fig. 3). In all cases Fistulous track was passing between external and internal carotid and crossing lateral to hypoglossal nerve (Fig. 3). Glossopharyngeal nerve was identified in 12 cases and track was seen passing lateral to it except in one case (Fig. 3). Tract immediately related to the anteromedial aspect of the internal

**Table 1** Showing clinical characteristics of patients

Clinical characteristics of patients	No	Percentage
Sex		
Males	13	65
Females	7	35
Age at presentation		
Range: 4–25 yrs		
Mean Age: 13		
0–9 years	6	30
10–20 years	13	65
21–30 years	1	5
Location of Fistulous opening		
Junction of middle and lower one third/lower one third	19	95
Upper one third	1	5
Presenting complaints		
Fistulous opening with intermittent discharge	15	75
Only fistulous opening	4	20
Recurrent abscess	1	5
Laterality		
Unilateral	18	90
Right	14	78
Left	4	22
Bilateral	2	10
Visible oropharyngeal opening	0	0
Associated Congenital syndrome	0	0
Family history	0	0
Fistulogram done	9	45

jugular vein (IJV) in all cases. No visible opening was seen in oropharynx (Table 2).

The internal opening of track was seen over posterior tonsillar pillar in 15 cases (75%) (Fig. 2) while in 5 patient the track was seen entering tonsillar tissue or bed. Tonsillectomy was done in 5 cases where the tract was seen to enter tonsillar bed/tissue (Table 2).

Two incisions (step ladder) (Fig. 2) were given in 19 cases (95%) while one case whose external opening was in upper one third neck was managed by single incision.

None had any intraoperative complications while one had postoperative wound infection and one post operative severe pharyngitis. No recurrence was seen at one year of follow up. No patient had associated congenital anomaly/syndrome, and/or family history.

## Discussion

The branchial apparatus consists of arches, pouches, and grooves [7]. Second branchial apparatus anomalies constitutes 95% of all branchial anomalies [12]. A fistula is the

**Fig. 1** Showing external openings of three unilateral (two in lower third neck and one in upper third) and one bilateral fistula



result of failure of obliteration of both groove and a pouch [7]. This causes a communication between skin and mucosa. Complete fistulas are rare [7]. Anget et al. [13] in his literature review of branchial cleft fistulae found that complete second branchial cleft fistulae are rarely encountered.

Females (13,65%) outnumbered males (7,35%) in our study. Similar female predominance were noted in studies by Maddalozzo et al [7] and Chandler and Mitchell [14]. Male to female ratio was 1:1.86 in our study while Simpson [15] in contrast reported Male/female ratio of 3:1, which is almost similar to ratio of 2.7:1 in a series by Rattan et al. [16]

Although branchial fistula may occur at any age but commonly, they present in first or second decade of life. Average age at time of surgery in our study was 13 years (range 4–25 years). Majority of patient (13, 65%) were in the age group of 10 to 20 years. Average age in a study by Abhita Reddy et al. [17] was 4.1 years (4 months–19 years) while in a study by Bist et al. [8] age range was 6 to 17 years. A study at one pediatric hospital by Maddalozzo et al. [7] found that the average age at surgery was 36.2 months (6–131 months).

Branchial cleft fistulae are commonly seen to be unilateral involving right side more frequently. Branchial cleft fistulae similarly in our study was more commonly affecting

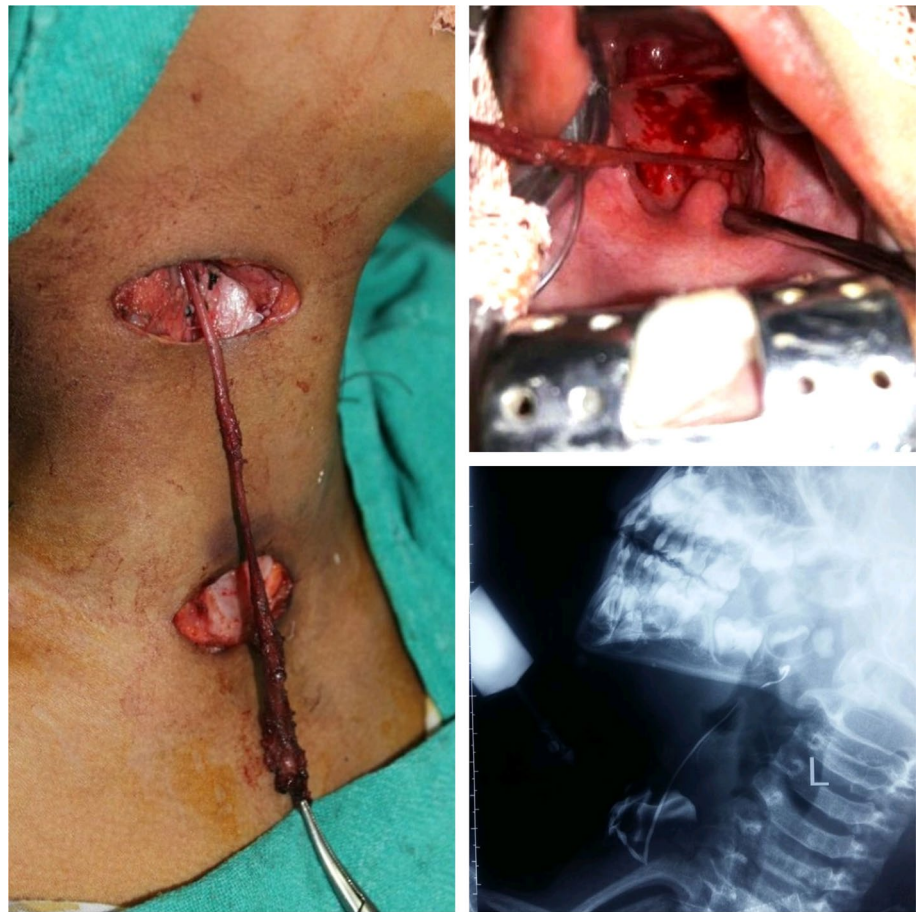
the right neck (14 patients, 78%) among unilateral cases. In a series by Bist et al. [8] all cases involved right side while Abhita Reddy et al. [17] found right side involvement in (54 patients, 81%),

Bilateral second branchial cleft fistulae are even rarer than unilateral ones. Two of our patients (10%) had bilateral fistulae. Maddalozzo et al [7] in their study on the 28 patients with complete second branchial cleft fistulas found only 3 (10.7%) had bilateral fistulae. Kajosaari et al. [18] in a recent review of 255 cases of second branchial anomalies from six studies found that 15% of the sinuses/ fistulae were bilateral.

Symptoms include intermittent mucopurulent discharge from the anterior aspect of neck and recurrent attacks of inflammation following an attack of upper respiratory tract infection. Incision and drainage may be needed for cellulitis or abscess formation on rare occasions [8]. The external opening may be seen to move upwards with deglutition. Probing the tract may sometimes produce palpitation, cough, pallor and vomiting because of tracts close proximity to the vagus nerve [19]. In our study the most common complaint was fistulous opening with intermittent discharge (15 patients; 75%) followed by only fistulous opening in 4 patients (20%) and recurrent abscess in 1 patient (5%). In a series by Rattan et al. [16] two of their



**Fig. 2** Showing stepladder incision, Fistula being pulled via posterior tonsillar pillar and Fistulogram



cases were infected who were treated with antibiotics before surgery.

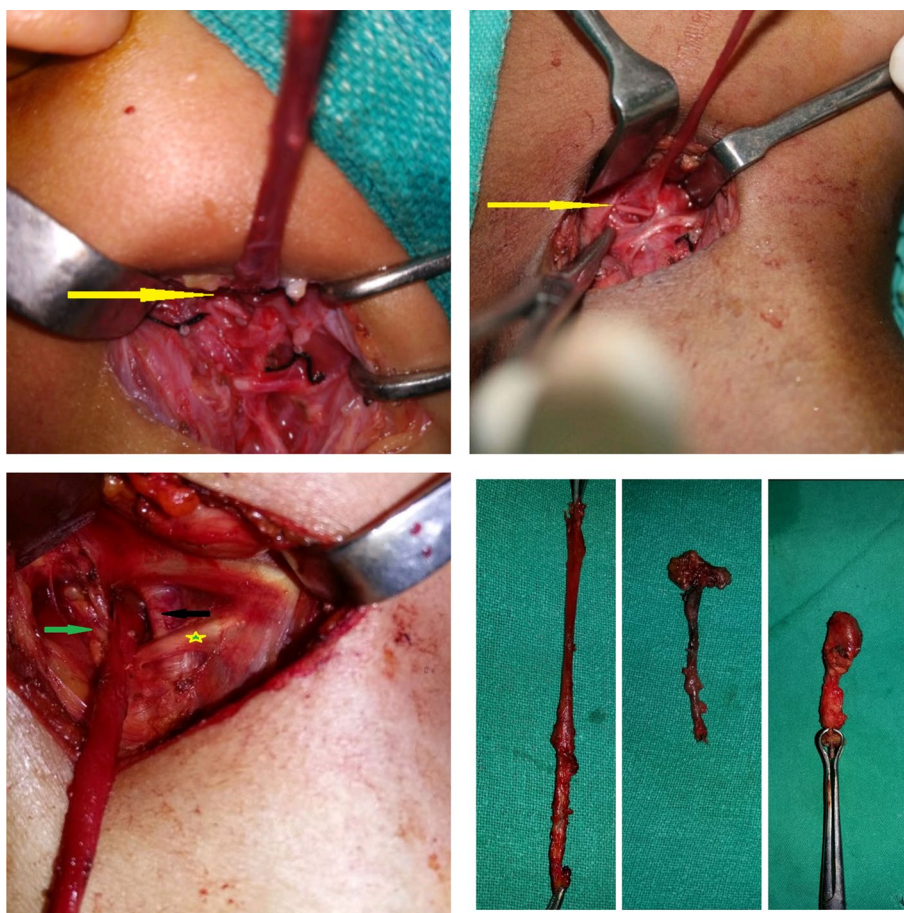
The length of tract can usually range from 1 to 8 cm. External opening usually presents in the lower third of the neck as a small skin pit anterior to the sternocleidomastoid muscle. External opening of fistula in our study was located in almost all cases in lower neck except one case where it was at upper one third. Length of fistulous tract in our study intraoperatively ranged from 5 to 13 cm with a mean of 8 cm. The length of the tracts has been reported to be of differently in different series like it was 8 cm to 14 cm [8], 6 cm [20] and 14 cm to 15 cm [21] in different reports. Maddalozzo et al [7] in their series of 28 patients found fistulous opening at the lower one-third of the SCM. Similarly in Bist et al. [7] series small opening was present at the junction of upper two-thirds and lower one-third of anterior border of sternocleidomastoid muscle in all the cases. The lining of the tract reflects its development. The fistulas are characteristically lined by stratified squamous epithelium in their external or the lateral part and columnar epithelium along their medial course [16].

In all our cases Fistulous track was passing between external and internal carotid arteries crossing lateral to hypoglossal nerve. Glossopharyngeal nerve was identified

in 12 cases and tract was seen passing lateral to it except in one case where it was medial. Fistulous tract usually pass from an external skin opening at the anterior border of the sternocleidomastoid muscle, between the external and internal carotid arteries, then cross over glossopharyngeal and hypoglossal nerves. The upper portion of the fistulous tract then ascends superior to the hyoid bone and passes below the stylohyoid ligament [10]. It opens internally in the lateral wall of the pharynx in the region of the tonsillar fossa. We found all tracts passing anteromedial to jugular vein at its caudal two third. Infact this caudal two-thirds relationship to IJV should always be kept in mind. The operating surgeon must be aware of the anatomic relationship of the IJV to the second branchial cleft fistula so as ensure safety of IJV. This very fact is also emphasized by Maddalozzo et al. in his paper on second branchial cleft fistulae [7].

At the upper end, the tract opens in to the posterior tonsillar pillar, supratonsillar fossa, or on to the tonsillar surface [6]. The internal opening of tract was seen over posterior tonsillar pillar in 15 cases (75%) while in 5 patient (25%) the tract was seen entering tonsillar tissue or bed. We found majority of tracts end at posterior pillar of tonsil. Similar observation was made by Bist et al. [8] in their series of 5 patients whereby they also found that tract ended at Anterior

**Fig. 3** Showing tract passing between external (Black arrow) and Internal carotid (Green arrow) arteries and crossing lateral to both hypoglossal (yellow star) and glossopharyngeal nerves (yellow arrows). Also seen tracts of different lengths



face of the upper half of the posterior pillar of the fauces in 3 of their patients (60%). Tonsillectomy was done in 5 cases (25%) only in those 5 patients where tract entered tonsil tissue/bed. Contrary to common teaching we found tracts don't enter the tonsillar tissue but penetrates the posterior pillar thereby it is possible to preserve tonsil in majority of cases. In a study by Kajosaari et al. [18], 46% of second branchial cleft fistulae excision included ipsilateral tonsillectomy, performed by otolaryngologists. We are of the opinion that Ipsilateral tonsillectomy is not needed for complete removal, has no beneficial effect on the clinical outcome in the majority of the cases and recurrence rates are not affected by the tonsillectomy. Tonsillectomy on the other hand is needed in such cases for complete excision if the internal opening is in the crypta magna [22].

Treatment of choice for branchial fistula is complete surgical excision of the fistulous tract. Two surgical methods are commonly used: The stepladder method and the stripping method. Transcervical approach using a wide cervicotomy (hockey stick) is also a surgical option [8]. The classical standard surgery for a complete second branchial arch fistula is the stepladder approach originally described by Bailey in 1933 with two incisions in the neck that gives exposure of the fistula tract [23]. The higher

incision should be bigger than the lower one because higher up the fistula tract is deeper in location in the vicinity of important neurovascular structures. The stripping method whereby the stripping of a branchial fistula by passing the stripper inside the tract is done was described by Taylor and Bicknell in 1977 [24]. It is however not routinely used as it is a blind procedure carrying a higher risk of injury to the surrounding structures [24].

Two incisions (Stepladder) were given in 19 cases (95%) while in one case whose external opening was in upper one third, only single incision was used. Some surgeons have used single incisions in their series. Maddalozzo et al. [7] in their series of 28 patients only used single incision as opposed to the "stepladder" incision but did not pull the track via oropharynx rather ligated the tract above the hyoid bone. Rattan et al [16] excised fistulae with a single incision except in six children over the age of 10 years where a stepladder approach was used. Gupta et al. [25] in a study involving 10 pediatric patients also used single incision with significant success.

Methylene blue was injected in all cases intraoperatively and entered tonsillar area in only 15 cases (75%).Fistulogram was done in only 9 patients (45%). MRI and CT scan was not done in any of our case.

**Table 2** Surgical and post operative characteristics of patients

Surgical / Postoperative Characteristics of patients
Length of fistulous track
Range: 5 cm to 13 cm
Mean: 8 cm
No of patients in which Fistulous track was passing between external and internal carotid: 20 cases
Track In relation to glossopharyngeal nerve (identified in 12 cases)
Lateral: 11 cases
Medial: 1 case
Track In relation to hypoglossal nerve
Lateral: 20 cases
Medial: 0 case
Tract in anteromedial aspect of the IJV: All cases
Internal opening of track
Posterior tonsillar pillar: 15 cases
Tonsillar tissue /bed: 5 cases
Anterior tonsillar tissue: 0 case
Need of tonsillectomy: 5 cases
Incisions
Two incisions (step ladder): 19 cases
Single incisions: 1 case
Methylene blue injected into track intraoperatively entering tonsillar area: 15 cases
Complications
Intraoperative: 0
Post operative
Wound infection: 1 case
Recurrence at minimum 1 year follow up: 0 case
Pharyngitis: 1 case

Visualization of the fistula tract can be done through injecting a dye(methylene blue) or fistulogram. Fistulogram can trace the tract up to the internal opening. Routine contrast fistulogram may not be required in all patients [16]. CT fistulogram with reformatted images unambiguously delineates the relation of sinus tract to that of important structures of neck. It also helps in classifying the type of lesion, provides a roadmap for surgeon prior to surgery, and reduces the chance of recurrence [26]. We did not use CT fistulogram in our patients.

Methylene blue injection in the fistula tract facilitates to delineate the course of the tract, and the presence of methylene blue in the throat can also definitively confirm a fistula. However, if the fistula tract filled with methylene blue is punctured or injured accidentally, the spreading dye in the surrounding tissue may make the dissection work difficult [11]. Sometimes muscle relaxation or stagnation of secretion due to inflammation which will obstruct the fistula might give us a false negative study [27].

A positive family history of branchial anomalies may be present in 35% of the patients with complete fistula [6]. There are a few reports on heredity factor and these indicate that these may be inherited from the mother [15]. The

diagnosis of a genetic disorder has to be considered when a patient presents with bilateral second branchial cleft fistulae [11]. Branchio-oto-renal (BOR) syndrome is the most common syndrome associated with bilateral fistulae of the first and second branchial arches, and 66% of cases of bilateral second branchial cleft fistulae are correlated with BOR syndrome [11]. None of our patients had associated congenital anomaly/syndrome, and family history. Family history was seen in one of the study by Rattan et al. [16]

Most frequent complication of the surgery is recurrence [8]. Other less common complications include secondary infection and damage to nearby anatomic structures including facial, Internal jugular vein, recurrent laryngeal, hypoglossal, spinal accessory, and glossopharyngeal nerves. Haematoma and bad scar is a possibility [28]. The injury to these nerves can be prevented by closely following and adhering to the tract as we did in our cases. The reported incidence of recurrence rate was 3% where only external approach was used. This most probably is due to incomplete excision of the fistula tract. However, no recurrence rate is reported after using combined oral and transcervical approach [26]. None of our patients had any intraoperative complications while one had postoperative wound infection



and one post operative severe pharyngitis. No recurrence was seen at one year of follow up. Similarly no recurrence was seen in a report by Gong et al. [11], Maddalozzo et al. [7] and Bist et al. [8] while Agaton-Bonilla et al. [29] and Rattan et al. [16] found recurrence rate of 8.6% and 4% respectively.

## Conclusions

Complete second branchial arch fistulas are not routinely encountered by young ENT surgeons. These tracts are unilateral and frequently seen on right side. All such tracts pass between carotid arteries and almost always pass lateral to hypoglossal and glossopharyngeal nerves. Most of the times the internal opening of the tract is over the posterior pillar of tonsils. The best surgical approach is a step ladder combined transcervical and transoral. Ipsilateral tonsillectomy can be avoided in most of the cases where the tract opens at the posterior tonsillar pillar. Recurrences are rarely seen in clinical practice if whole tract is finally pulled and removed via oropharynx. Young ENT surgeons need not to fear for any complications if they get well versed with the clinicosurgical findings of these tracts.

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## Declarations

**Conflict of interest** There are no financial or non-financial interests that are directly or indirectly related to this work.

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