

# Head and neck fistulas of congenital or infectious etiology: retrospective analysis of 23 cases

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## Abstract

**Objective:** In this study, we aimed to conduct a retrospective analysis of head and neck masses initially presenting with fistulas.

**Methods:** A total of 23 patients with head and neck fistulas who admitted to the otorhinolaryngology department of our tertiary care center between January 2011 and May 2012 were retrospectively reviewed. Age, sex, and duration of symptoms were noted from the case records. The side and site of the lesion and the opening of fistula were noted. Co-morbidities, post-operative complications and histopathological diagnosis were classified.

**Results:** Of the 23 patients with head and neck fistulas, (12 males, 11 females) the average age was 26.52±14.1 (range: 9 to 74) years. Of the 23 lesions, the most prevalent lesion was branchial fistulas (n=13, 56.52%) followed by thyroglossal fistulas (n=7, 30.43%), dermoid fistulas (n=2, 8.7%) and tuberculosis (n=1, 4.4%). Drainage (n=23, 100%) was consistently observed as the initial symptom in all patients while swelling (n=22, 95.6%) was the second most common presenting symptom.

**Conclusion:** Correct diagnosis is essential to avoid inadequate surgery and multiple procedures for head and neck fistulas of congenital or infectious etiology.

**Keywords:** Branchial, thyroglossal, congenital, fistula, head and neck.

## Özet: Konjenital veya enfeksiyöz etiyojili baş ve boyun fistülleri: 23 olgunun retrospektif analizi

**Amaç:** Bu çalışmada başlangıçta fistülle gelen baş ve boyun kitlelerinin retrospektif analizini gerçekleştirmeyi amaçladık.

**Yöntem:** Üçüncü basamak merkezimizin KBB Ana Bilim Dalına Ocak 2011 ila Mayıs 2012 arasında kabul edilmiş baş ve boyun fistüllü toplam 23 hasta retrospektif olarak gözden geçirildi. Olgu kayıtlarından yaş, cinsiyet ve semptomların süresi ile birlikte lezyon yeri ve tarafı, fistülün ağzı da not edildi. Komorbiditeler, postoperatif komplikasyonlar, histopatolojik tanı sınıflandırıldı.

**Bulgular:** Baş ve boyun fistülleri olan 23 hastanın (12 erkek, 11 kadın) yaş ortalaması 26.52±14.1 (aralık: 9–74) yıl idi. Yirmi üç lezyon arasında en yaygın olanları brankiyal fistüller (n=13, %56.52), ardından tiroglossal fistüller (n=7, %30.43), dermoid fistüller (n=2, %8.7) ve tüberküloz (n=1, %4.4) gelmekte idi. Hastaların hepsinde başlangıç semptomu sürekli olarak drenaj (n=23, %100) iken, şişlik (n=22, %95.6) ise ikinci sıklıkla en çok görülen semptom idi.

**Sonuç:** Konjenital veya enfeksiyöz etiyojili baş ve boyun fistüllerinde yetersiz cerrahi ve çoklu girişimlerden kaçınmak için doğru tanı esastır.

**Anahtar sözcükler:** Brankiyal, tiroglossal, konjenital, fistül, baş ve boyun.

Congenital cysts and sinuses of the neck are common mostly in the pediatric population. These lesions can be safely excised usually not requiring a detailed preoperative workup.<sup>[1]</sup> However, rare lesions such as fistulas of the first cleft and cysts of the fourth pouch may be misdiagnosed. Congenital midline anomalies such as thyroglossal duct cysts and dermoid cysts are embryologically different from malformation of the lateral cervical region. Branchial

anomalies are further classified as cysts, sinuses, and fistulas. Cysts are proposed to be entrapped remnants of branchial cleft or sinuses. Sinuses are remnants of cleft or pouches, and fistulas occur as a result of the persistence of both pouch and cleft.<sup>[2]</sup> Maldevelopment of branchial apparatus have been implicated in various anomalies of the head and neck region. The importance of understanding the anatomy and pathology of branchial apparatus is in

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applying the knowledge during surgery, and handling complications.

Management of the congenital fistulas of the neck is possible by focusing on the specific lesion, and appropriate surgical treatment relies on a precise preoperative diagnosis. This discussion presents an overview of the embryology, pathophysiology, and diagnostic modalities for congenital neck fistulas. Additionally, we also described the current principles of surgical management. Although these anomalies are generally easily treated, accurate preoperative diagnosis and appropriate surgical therapy are essential to prevent recurrence and to ensure optimal cosmetic outcomes.<sup>[3]</sup>

## Materials and Methods

### Study design

A total of 23 patients, aged between 9–24 years, with neck fistulas who admitted to the department of otolaryngology of our tertiary care center between January 2011 and May

2012 were retrospectively reviewed. Age, sex, and duration of symptoms were noted from the case records. The side and site of the lesion and the opening of fistula were noted. All patients underwent routine pre-operative blood examination. Co-morbidities, post-operative complications and histopathological diagnosis were classified.

### Statistical analysis

Data were analyzed using the Statistical Package for Social Sciences (SPSS Inc., Chicago, IL, USA) software (version 10.0 for Windows). Continuous variables were presented as mean±SD. Parametric tests were applied to data of normal distribution, and non-parametric tests were applied to data of questionably normal distribution.

### Results

The patient data were shown in Table 1. The average age of the 23 patients with neck fistulas, (12 males, 11 females)

**Table 1.** An overview of data gathered from our series.

Patient no.	Age	Gender	Comorbidity diagnosis	Histopathological complications	Postoperative complaints	Chief
1	13	M	-	BF	-	S, D, P
2	28	M	-	Tbc	-	S, D, P, F
3	9	M	-	BF (PAF)	-	S, D
4	27	F	Hypothyroidism	DF	-	S, D
5	44	F	Diabetes mellitus	DF	-	S, D
6	20	M	-	BF (PAF)	-	S, D
7	29	F	-	BF	-	S, D, P
8	27	F	-	TGF	-	S, D, F
9	74	F	-	BF (PAF)	Infection	S, D, F
10	62	M	Hypothyroidism	TGF	Recurrence	S, D
11	42	F	Hypothyroidism	TGF	-	S, D, P
12	21	M	-	BF	-	S, D, F
13	16	M	-	BF (PAF)	-	S, D
14	17	M	-	TGF	-	S, D
15	32	M	Hypothyroidism	BF	Recurrence	S, D
16	35	F	Diabetes mellitus	BF	-	S, D
17	24	F	-	BF (PAF)	-	D
18	16	F	-	TGF	-	S, D, F
19	17	M	-	BF (bilateral)	-	S, D, P
20	16	F	-	TGF	Infection	S, D, F
21	15	M	-	BF (PAF)	-	S, D
22	17	M	-	TGF	-	S, D
23	19	F	-	BF (PAF)	-	S, D, P

BF: branchial fistula; D: drainage; DF: dermoid fistula; DM: diabetes mellitus; F: fever; P: pain; PAF: preauricular fistula; S: swelling; Tbc: tuberculosis; TGF: thyroglossal fistula

was  $26.52 \pm 14.1$  (range: 9 to 74) years. Male to female sex ratio was 1.09:1 with a slight male predominance. Of the 23 lesions, maximum incidence was of branchial fistulas (56.52%) followed by thyroglossal fistulas (30.43%).

Branchial fistulas (n=13, 56.52%) were frequently detected, and 7 of them were pre-auricular fistulas (n=7, 30.43%) located on the lateral cervical region (Fig. 1). Thyroglossal fistulas (n=7, 30.43%) (Fig. 2) and dermoid fistulas (n=2, 8.69%) located in the midline. One case was diagnosed as fistula of tuberculosis (n=1, 4.34%). Only one case with branchial fistula presented bilaterally. In this bilateral case, excision of the masses was performed in a two-stage operation three months after the first intervention.

Main symptoms at initial admission were swelling and drainage from a cervical mass. The other symptoms include pain and/or fever. Swelling and discharge were the presenting symptoms in all 23 cases. Six patients complained of pain regardless of etiology (26.08%).

Postoperative infection was the main postoperative complication encountered in 2 cases (8.69%). In addition to surgery, systemic antibiotics (amoxicillin-clavulanate) were administered to these cases for three days. In 1 patient (4.34%), diagnosis of cervical tuberculosis fistula was estab-

lished. Anti-tuberculous treatment consisting of rifampicin and isoniazid were given for six months.

Co-morbidity is another parameter in our study. The most common co-morbidity was hypothyroidism (regardless of etiology) and diabetes mellitus. Four patients had hypothyroidism (17.3%) and 2 patients had diabetes mellitus (8.6%).

Operation type consisted of Sistrunk for thyroglossal duct fistulas (n=7, 30.43%) while complete surgical excision was the selected choice of treatment in the rest of cases.

In two cases with branchial fistula, additional pre-operative systemic antibiotics (amoxicillin/clavulanate 2 g/day orally) were administered due to acute infection. Such patients were taken to the operating room one week after antibiotic treatment. In one case with mycobacterial lymphadenitis, anti-tuberculous treatment was administered after histopathological diagnosis.

Recurrence was observed during postoperative recovery in 2 cases (8.69%) requiring re-operation within 12 months.

## Discussion

Understanding the anatomical or clinical features of each lesion is a 'must' to make a right diagnosis and to avoid



Fig. 1. Typical view of a left preauricular fistula.



Fig. 2. A thyroglossal fistula with drainage.

recurrences of head and neck fistulas.<sup>[4]</sup> A high index of suspicion and clinical awareness as well as thorough physical examination can lead to a definitive diagnosis. Branchial anomalies should be involved in the differential diagnosis of any unexplained masses in the head and neck area or recurrent neck spaces infections. Radiological investigations should be used to evaluate neck lesions of congenital origin. The extent of the lesion can be defined by CT and/or MRI. The relation of the facial nerve to the first cleft anomaly can be assessed by imaging methods.<sup>[5]</sup>

Branchial cysts may present as circumscribed lesions or have tract-like extensions on one side.<sup>[6]</sup> According to anatomical location, second cleft cysts can be classified into four groups which are type I occurring under the superficial aponeurosis of the neck, type II located just to the front of the large vessels, type III usually between the branches of the carotid bifurcation, and type IV occurring beneath the large vessels and pharyngeal wall. After a complete surgical excision recurrence is uncommon.<sup>[7]</sup> Our cases of branchial fistulas are in type II, and no recurrence occurred after resection.

Branchial cleft anomalies such as branchial sinuses; cysts and fistulas are mostly found in pediatric population.<sup>[8]</sup> About 96–97% of these anomalies are unilateral, and only 2–3% have a bilateral presentation; the bilateral occurrence is higher in familial cases.<sup>[6]</sup> In our cases, one patient has bilateral branchial fistula but no familial history. The treatment protocol for such lesions is surgical excision. Antibiotics are only used to treat infections in the tract. Surgical excision is a definitive treatment; however, surgery should be avoided during the period of acute infection. If present, surgical drainage of the abscess is indicated along with concurrent antibiotherapy.<sup>[9]</sup>

Occasionally, a branchial anomaly may be a part of branchiootorenal (BOR) syndrome, which is defined with branchial arch anomalies, hearing loss and renal malformations. This genetic syndrome with autosomal dominant transmission has first been described by Melnick et al. and Fraser et al.<sup>[10,11]</sup> We had no BOR syndromic patients in this study, but BOR should be kept in mind for the patients with branchial fistulas that can be associated with external ear anomalies and/or a history of hearing loss and similar findings in other family members.

Lymphadenopathy due to tuberculosis is commonly encountered in endemic areas, and the cervical region is the most common site of tuberculous adenitis. The term “scrofula” refers to this condition. The tuberculous adenitis can be found at other sites such as axillary, intrathoracic, intra-

mammary, intraabdominal (mesenteric lymph nodes or paraaortic), and occasionally inguinal sites.<sup>[12]</sup> In our study, the fistula was in the supraclavicular region and the patient has neither systemic symptoms nor tuberculosis history. Both tuberculosis and HIV prevalence has been reported to be higher in countries that have a higher incidence of tuberculous lymphadenitis. During recent years, it has been found that tuberculous lymphadenitis is common in the third-fifth decades of life and it consists of a predominantly female population. The lymph nodes can be infected by *Mycobacterium* through hematogenous route or by a local extension from tuberculous infection of adenotonsillar tissue.<sup>[13]</sup>

Thyroglossal duct cysts are the most common congenital neck masses with a typical presentation as a painless cystic mass or fistula in the region of hyoid bone at or near the midline. Due to the process of embryogenesis, thyroglossal duct cysts can be found at the base of the tongue to the lower midline of the neck. A midline location and close association with the hyoid bone is the only nearly universal components of these lesions.<sup>[14]</sup> Functional thyroid tissue can be contained in the thyroglossal duct remnants. Most thyroglossal duct cysts present during the first five years of life. Due to its close relation to the hyoid bone and foramen caecum, the cyst moves in craniocaudal direction with swallowing. These patients typically have fluctuations in the size of the neck mass. Although most thyroglossal duct remnants present as cystic masses, up to one-quarter of these lesions present as a draining sinus tract in the midline. The thyroglossal duct sinus occurs due to the spontaneous rupture of the cyst. Yet, fistulous communication between the skin and foramen caecum has been reported very rare.<sup>[15]</sup> In our cases, there might have been a rarely patent thyroglossal tract, which manifested as a cyst in childhood, which on rupture resulted in the formation of a fistula.

Dermoid cysts and fistulas account for up to 25% of midline cervical anomalies. Dermoid fistula typically present as painless, superficial subcutaneous masses in the anterior neck, but can occur in other locations, such as the occipital and frontal scalp. Most anterior neck lesions occur in relatively close proximity to the hyoid bone and are frequently misdiagnosed as thyroglossal duct pathologies.

Unlike thyroglossal duct cysts, dermoid cysts do not move with swallowing and tongue protrusion as they lack mesodermal attachments and have a superficial location. Because dermoid cysts have no connection to the oropharynx, infection is rare. Cyst rupture may, however, occur due to trauma or enlargement, and results in granulomatous

inflammation of the surrounding skin and soft tissues. It may be difficult to differentiate between an infected thyroglossal duct cyst and a ruptured dermoid. In these cases, fine needle aspiration with cytology and culture may be useful. Neck fistulas are common pathological disorders of neck surgery. Our study involves fistulas of neck including thyroglossal fistulas, tuberculous fistula, branchial fistulas and dermoid fistulas. Limitations of our study are retrospective design, small sample size, and the absence of definitive criteria for the selection of patients for this method. However, we hope that this series highlights the clinical features of neck fistula and main diagnostic and therapeutic steps to be taken.

## Conclusions

Head and neck fistulas are relatively common pathologies that can be treated successfully with the appropriate surgical method. Correct diagnosis is essential to avoid insufficient surgeries and or second procedures. The surgical approach needs to be tailored to the suspected arch of origin of the anomaly. Definitive excision is essential for good outcomes.

**Conflict of Interest:** No conflicts declared.

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