

Brankial Yarık Anomalileri: Klinik Deneyimimiz ve Literatür Taraması

Branchial Cleft Anomalies: Our Clinical Experience and Literature Review

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ÖZ

GİRİŞ ve AMAÇ: Brankial yarık anomalileri embriyolojik gelişimin nadir anomalilerindedir. Fetal gelişim sırasında brankial aparatın gelişiminin tamamlanmamasına bağlı olarak gelişir ve kist, sinüs traktı, fistül veya kıkırdak kalıntısı olarak gözlenebilir. Bu çalışmanın amacı, kliniğimizde opere edilen brankial yarık anomalilerin klinik özelliklerini ve cerrahi tedavi sonuçlarını gözden geçirmektir.

YÖNTEM ve GEREÇLER: Brankial yarık anomalisi tanısı alan ve 2007-2017 tarihleri arasında opere olan 62 hasta retrospektif olarak incelendi. Tüm hastaların dosya kayıtları yaş, cinsiyet, klinik prezentasyon, semptomların süresi, tetkikleri, patolojik sonuçları ve takibi açısından incelendi.

BULGULAR: Çalışma süresi boyunca kliniğimizde brankial yarık anomalisi tanısı ile tedavi edilen 62 hastanın 40'i (%64.5) kadın, 22'si(%35.5) erkekti. Ortalama yaş 26.5 ± 12.9 iken en genç hasta 7 en yaşlı hasta 71 yaşındaydı. Hastalarda en sık izlenen başvuru şikayeti 53(%85.5) hastada izlenen boyunda şişlikti. 62 hastadan 20'i (%32.3) birinci, 42'i (%67.7) ikinci brankial yarık anomalisiydi. Üçüncü ve dördüncü brankial yarık anomalisi izlenmedi.

TARTIŞMA ve SONUÇ: Baş boyun yapılarının gelişiminde brankial aklar önemli rol oynar. Bu yapıların anormal gelişimi, boyunda kist, sinüs veya fistül gibi ileriki dönemde ortaya çıkan farklı anomalilerin oluşumuna yol açar.Brankial yarık anomalilerinin tedavisi cerrahi eksizyondur.

Anahtar Kelimeler: brankial yarık anomalileri, kist, fistül

ABSTRACT

INTRODUCTION: Branchial cleft anomalies are rare anomalies of embryological development. It develops due to incomplete development of branchial apparatus during fetal development and can be observed as a cyst, sinus tract, fistula or cartilage residue. The aim of this study is to evaluate the clinical features and surgical treatment results of branchial cleft anomalies operated in our clinic.

METHODS: A total of 62 patients diagnosed as the branchial cleft anomaly in our clinic and operated between 2007-2017 were evaluated retrospectively. The records of all patients were examined in terms of age, gender, clinical presentation, duration of symptoms, examinations, pathological results, and follow-up.

RESULTS: 40 (64.5%) of the 62 patients who were treated with the diagnosis of branchial cleft anomalies were female and 22 (35.5%) were male. The mean age was 26.5 ± 12.9 and the youngest patient was 7 and the oldest patient was 71 years old. The most common complaint in the patients was neck swelling in 53 (85.5%) patients. 20 (32.3%) of 62 patients have first and 42 (67.7%) second branchial cleft anomaly. The third and fourth branchial cleft anomalies were not observed.

DISCUSSION and CONCLUSION: Branchial arches play an important role in the development of head and neck structures. Abnormal development of these structures leads to the formation of different anomalies such as cysts in the neck, sinus or fistula in the future. The treatment of branchial cleft anomalies is surgical excision.

Keywords: branchial cleft anomalies, cyst, fistula

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INTRODUCTION

Branchial arches form embryological precursors of face, neck, and pharynx. Branchial arches develop between the fourth and seventh week of gestation and form embryological precursors of muscle, blood vessels, bone, cartilage and mucosa of the ear, face, neck and pharynx(1). In total, the six double-branchial arches develop with craniocaudal alignment on both sides of the pharyngeal structures. The fifth branchial arch is usually rudimentary or does not show any improvement, eventually, a total of five double arches form adult structures(1,2).

Branchial cleft anomalies (BCA) occur due to the maturation disorder of the branchial apparatus during fetal development(3). It is classified as first, second, third and fourth arch anomalies depending on anatomic location(4). These anomalies may be in the form of a fistula which develops between the sinus, skin, and pharynx, combining the epithelial cyst, skin or pharynx with a blind pouch in the neck not connected with the skin or pharynx(5).

BCA develops from the second branchial arch (BA) by 90%. The first BA anomalies are 8-10%; the third and fourth BA anomalies are very rare(6,7). Patients usually present with nonspecific symptoms such as swelling of the neck, recurrent infection, flix from the skin. Although BCA is suspected with clinical history and physical examination and radiological imaging methods, the definitive diagnosis is made by surgical excision and pathological examination(5,8).

The aim of this study is to present the clinical features, diagnostic methods and surgical treatment of branchial anomalies operated in our clinic.

MATERIALS and METHODS

The records of 62 patients who were operated in our clinic with the diagnosis of BCA between 2007-2017 were retrospectively reviewed. Patients who did not receive surgical treatment were excluded from the study. The symptoms duration of patients, the complaints of the patients, the preoperative imaging methods and fine needle aspiration biopsy (FNAB) results were examined. Postoperative pathological results were evaluated.

Statistical Analysis

Mean, standard deviation and minimum-maximum values of the demographic data of patients were calculated. In the significance of statistical results, a value of $p < 0.05$ was accepted. For statistical analysis, IBM SPSS Statistics for Macintosh, Version 20 (IBM Corp., Armonk, New York) was used.

RESULTS

During the study period, 40 (64.5%) of the 62 patients who were treated with the diagnosis of BCA in our clinic were female and 22 (35.5%) were male. The mean age was 26.5 ± 12.9 and the youngest patient was 7 and the oldest patient was 71 years old. When the symptom duration of the patients is questioned, it was determined that in 17 (27.4%) patients since the birth, in 23 (37.1%) patients less than 1 year, in 18 (29%) patients between 1 to five years, the symptoms were observed (Table 1).

Table 1. Demographic data and symptom duration of the cases

Age	26.5 ± 12.9
Gender	40 (64.5%) female 22 (35.5%) male
Symptom duration	17 (27.4%) since birth 18 (29%) 1 to 5 years 23 (37.1%) less than 1 year

Most of the patients present with a complaint of 85.5% (53) neck swelling, the second group had the pain rate of 50% (31) and third place, the discharge was observed at the rate of 37.1% (23). 30 (48.4%) patients had a history of recurrent infection (Table 2). The fistula was detected in 27 (43.5%) patients in preoperative physical examination. When the patients with fistula are classified according to the type of branchial cysts, 12 (19.4%) patients had 1. BCA, 15 (24.2%) patients had 2. BCA.

Table 2. Symptoms of the cases

Symptom	n %
Neck swelling	85.5% (53)
Pain	50% (31)
Discharge from fistula	37.1% (23)
Recurrent infection	30 (48.4%)

When the lesion side of the patients was evaluated, in 31 (50%) patients, the branchial cleft anomaly was observed in the right side, in 25 (40.3%) patients in the left side and in 6 (9.7%) patients bilateral anomaly was detected. No statistically significant difference was found between the sides.

When the preoperative imaging methods of the patients were examined, it was determined that ultrasonography (USG) was requested from 50 (80.6%) patients, a computed tomography (CT) from 4 (6.5%) patients, magnetic resonance imaging (MRI) from 20 (32.3%) patients. Fistulography was performed preoperatively to 8 (12.9%) patients. Preoperative FNAB was performed in 34 (54.8%) patients. Aspiration biopsy results in 28 (82.3%) of these patients were reported as squamous type material and were reported primarily in the branchial cyst direction. The aspiration result in 7 (17.7%) patients were reported as non-diagnostic

When the patients were classified in terms of branchial anomalies, 20 (32.3%) out of 62 patients were in the first and 42 (67.7%) in the second BCA. Third and fourth BCA were not observed. When evaluated according to the first BCA Work calcification, 65% (13) was evaluated as type 1 and 35% (7) as type 2. In all of the patients classified as Type 2, the branchial anomaly was on the surface of the facial nerve (Table 3).

Table 3. Branchial anomaly types

Types	N (%)	N (%)
First, type 1	13 (65%)	20 (32.3%)
First, type 2	7 (35%)	
Second	42 (67.7%)	42 (67.7%)
Third	-	-
Fourth	-	-

Total surgical excision was performed in patients with the branchial anomaly. Intra-operatively in all patients, for the purpose of follow-up of the tract, total excision of the lesion and prevention of recurrence, staining with methylene blue was performed. Recurrence was detected in 2 (3.22%)

patients. Surgical treatment was repeated in patients with recurrence. The most frequent complication during the postoperative period was wound site infection with 12.9% (8). Wound site dehiscence was the second most common complication with 8.1% (5).

DISCUSSION

Branchial apparatus occurs in the fourth week of embryonic life and plays an important role in the development of head and neck structures. There are grooves (ectoderm), arches (mesoderm) and sacs (endoderm) in each branchial apparatus pair. Branchial complex anomalies are caused by developmental abnormalities of branchial apparatus during the embryological period (9). Although they are usually congenital anomalies that occur in childhood, they can also be observed as a mass or infection in the neck in adult life (8,10). Many different theories about the development of branchial anomalies have been developed. Although different development theories such as branchial apparatus theory, cervical sinus theory, thymopharyngeal theory, inclusion theory are produced, the most acceptable is the incomplete involution of the branchial apparatus (4).

The second branchial apparatus anomalies are the most common subtype. In the Bajaj et al. study, the second branchial arch anomalies were observed at 78%, different rates such as 50% in Prasad et al. study, 40% in Choi et al. study were determined. The first branchial arch anomalies were reported as 22%, 38.24% and 25% in the same studies, respectively (4,9,11). In our study, the second BCA was the most frequent with a rate of 67.7% and the first BCA with a rate of 32.3%. First BCA is divided into two according to the clinical and histopathological features according to the Work calcification. They are ectodermal cystic masses with squamous epithelium which does not contain type 1 cartilage and skin residue. It is usually located near the pinnae in the post-auricular fold and on the surface of the facial nerve. Type 2 cyst can be seen as a fistula tract or sinus. It may contain skin and cartilage residue. The tract may be superficial or deep in the facial nerve within the parotid salivary gland (12). In our study group, 20 (32.3%) patients

had 1 branchial arch anomaly and 65% (13) were classified as type 1 and 35% (7) type 2.

Ford et al. found that branchial arch anomalies appeared more on the right side(13). Likewise, in the study of Prasad et al., 55.38% right side incidence and 44.62% left side incidence were determined(4). In our study, branchial cleft anomalies were observed in 31 (50%) patients on the right side, in 25 (40.3%) patients on the left side and 6 (9.7%) patients had the bilateral anomaly. There was no statistically significant difference between the two sides.

In the study of Choi and Zalza, the most common complaint was flix, neck mass, recurrent infection; in Prasad et al study, the most common complaint was the mass in the neck with 61.76% (4,9). In our study, neck swelling observed as 85.5% (53) was the most common complaint.

Although anamnesis and physical examination are important for the diagnosis in BCA, imaging methods are important in preoperative evaluation. USG is helpful in defining the size of the cystic structure of the lesion. Due to the lack of exposure to radiation, it is a more preferred method in pediatric patients (5,14). CT and MRI methods are valuable in determining anatomical localization, identifying possible infection or abscess, and distinguishing malignant degeneration (4). The most common diagnostic method was USG with 80.6% in our study. CT was requested in 4 (6.5%) patients and MRI in 20 (32.3%) patients. Unlike other studies, it was observed that MRI was a more preferred method than CT(4). The reason for this can be explained as the use of malignant degeneration in adult patients.

In order to rule out malignant diseases in adult patients, FNAB can be applied for diagnosis. In Lee at al. study, it was reported that FNAB has a 100% diagnostic sensitivity in the 2. branchial cleft anomalies. In addition, it was reported that FNAB may provide better results than the traditional method with USG (15). In our study, FNAB was pre-operatively performed in 34 (54.8%) patients. Biopsy results in 28 (82.3%) of these patients were reported in the branchial cyst direction. The aspiration result in 7 (17.7%) patients were reported as non-diagnostic.

The fistulogram can be used to define the fistula anatomy preoperatively. It can be applied in terms of planning before surgery but may be difficult to apply in pediatric patients (14). In our study, 8 (12.9%) patients had preoperative fistulography.

The treatment of BCA is surgical excision. If abscess formation has not developed in the presence of acute inflammation, incision and drainage are not required, they can usually be controlled by medical treatment. Surgery can be performed 4-6 weeks after the treatment (4). For complete surgical excision, cyst, fistula, and tract should be kept intact, and a lacrimal probe can be placed in the trauma during surgery (16). In patients, staining with methylene blue intra-operatively is also a method used for complete surgical excision (4). In our study, the staining was performed with methylene blue intraoperatively for all patients.

This is the relationship with the facial nerve that should be considered during the first BCA. The branchial anomaly can be found in the surface or deep of the facial nerve. The relationship of the facial anomaly with facial nerve should be evaluated with preoperative tests and an operation plan should be performed (16). In our study, in 20 patients 1. BCA was present. In preoperative examination and intraoperative evaluation, all lesions were superficial to the facial nerve.

In the second BCA, it can be approached on the mass with transcervical incisions. A step-ladder incision can be performed to increase visualization if the tract is long or moving towards the pharynx (4). In our study, in 42 patients 2. BCA was detected. 18 of these patients were followed up with methylene blue and followed up to the region near the tract pharynx. It was not observed that the tract was opened to pharynx or tonsil loge in none of the patients.

Wound infection, wound healing, cranial nerve injury, internal jugular damage, persistent fistula formation, seroma or hematoma formation are the complications that may be observed after the surgery (4). The most frequent complication during the postoperative period was wound site infection with 12.9% (8). Wound site dehiscence was the second most common complication with 8.1% (5).

In the study of Ford et al. the recurrence after the surgery was 3%, in the study of Prasad et al. it was 1.2%(4,13). Although Prasad et al. found that methylene blue staining was sufficient for the detection of branching and branching in the proximal region, it was also more meaningful to prevent recurrence by dissecting the micro-shock or magnification loops in distal branching (4). In our study, the recurrence rate was 3.22% (2). In all of our patients, intra-articular staining with methylene blue was performed but no microscopic dissection was performed.

CONCLUSION

Branchial apparatus plays an important role in the development of head and neck structures. Abnormal development of these structures may cause abnormalities such as cyst, fistula, and sinus. The treatment of branchial anomalies is surgical excision. Because of its proximity to the vital organs in the head and neck, preoperative presentation of the fistula or sinus tract is important to prevent injuries that may occur during surgery.

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