

The role of ipsilateral tonsillectomy in the extirpation of branchial cleft anomalies- A retrospective monocentric analysis over 13 years

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Abstract

Introduction. Branchial cleft anomalies (BCA) can occur as sinuses, fistulas or cysts. They arise from the first, second, third or fourth pharyngeal cleft due to non-fusion or subinvolution. Mostly, located in Robbin's neck-level II, BCA clinically present as a painless compressible swelling, cutaneous draining sinus, or fistula. Surgical treatment is the gold standard to prevent recurrence, though the necessity of ipsilateral tonsillectomy is discussed. **Study Design.** We conducted a retrospective comparative cohort study. **Setting.** Monocentric. **Methods.** In retrospect, data was collected from patients, that were admitted with the diagnosis BCA between 2006 and 2020 in an academic tertiary care center. **Main outcome measures.** 160 patients met inclusion criteria, the data was further evaluated, the focus was set on the occurrence of recurrence with or without tonsillectomy. **Results.** Recurrence of BCA was observed in 2 out of 160 surgically treated patients (1,25%), one of them with simultaneous tonsillectomy, the other without. **Conclusion.** A statistically significant difference in the recurrence-rate between these two groups (with/without tonsillectomy) could not be shown. The performance of an ipsilateral simultaneous tonsillectomy in the surgical workup of BCA cannot be recommended at the basis of our data.

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Key points:

- Branchial cleft anomalies (BCA) occur as sinuses, fistulas or cysts, arising mostly from the second branchial cleft represent 40-95%
- To effectively treat second, third and fourth cleft BCA, total surgical excision is recommended
- A strict differentiation between sinuses, cysts or fistulae is necessary to guarantee the optimal choice of surgical technique and approach
- In the existence of residual tracts leading to the tonsillar fossa, beside the need of extirpation of the tract itself, the necessity of ipsilateral tonsillectomy to prevent recurrence is discussed
- After total excision BCA recurrency-rates ranges from 0 to 4% and within our data recurrency occurred 1,25%, with no clarity due to the necessity of tonsillectomy

1 Introduction

During the embryological development, in the fourth week of gestation, six pairs of arches, clefts and pouches form the branchial or pharyngeal apparatus. Every arch consists of a cartilaginous element, muscular component, a corresponding branch of the aortic arch and a cranial nerve. These components later form various structures in the head and neck and due to non-fusion or subinvolution can result in branchial cleft anomalies (BCA).[1, 2] These BCA can occur as sinuses, fistulas or cysts and are present at birth, although maybe symptomatic until later in adulthood. [2, 3] BCA comprise about 20% of congenital lesions in children and arise from the first, second, third or fourth pharyngeal cleft [2]

Whereas first branchial cleft anomalies can be divided into Work type I (preauricular and lateral to the facial nerve) and Work type II (mandibular angle/submandibular and medial/or lateral to the facial nerve), third branchial anomalies present in the middle and lower third of the sternocleidomastoid muscle (SCM). The fourth BCA are extremely rare (1%) and normally present in the middle portion of the SCM.[2]

BCA arising from the second pharyngeal cleft are the most common and represent 40-95%. [2, 4] They are usually located in the lateral neck anterior and medial to the SCM and can have contact to the ipsilateral pharyngeal, explicitly the tonsillar region. [2, 4, 5] The majority of BCA present as cysts between the age of 20-40, in younger age (<5 years) sinuses and fistulas are more common.[4]

Clinical presentation of BCA

Clinically, the majority of BCA present as a painless compressible swelling, draining sinuses, or fistulae situated at the anterior border of SCM in line between the mandibular angle and clavicle. [6] Presenting patients may report a variety of duration and periods of waxing and waning of the neck swelling. Acute size increase can occur due to upper respiratory tract infections. [4] Secondary infections and inflammation can occur, therefore neck abscesses are possible. [7]

Although rare, bilateral second branchial cleft cysts have been reported [8] and in some patients this is part of the branchio-oto-renal syndrome (BOR), an autosomal dominant disorder. [9] BOR or Melnick-Fraser-Syndrome [10] symptoms include hearing impairment, cup-shaped pinnae, preauricular pits, branchial fistulae and renal anomalies. [4] Even though there is a positive predictive value in preoperative diagnosis of BCA, cystic neck masses should presumed malignant. [11]

Treatment of BCA

To effectively treat second, third and fourth cleft BCA, total surgical excision is recommended. [12-14] A strict differentiation between sinuses, cysts or fistulae is necessary to guarantee the optimal choice of surgical technique and approach. [14] Within the treatment of fistulae or draining sinuses, a cutaneous excision of the duct opening is recommended. In the situation of the existence of residual tracts leading to the tonsillar

fossa, beside the clear indication of extirpation of the tract itself, the necessity of ipsilateral tonsillectomy to prevent recurrence, is discussed. [5, 15-18] Overall, BCA-recurrence is stated up to 4%. [19-21]

Objectives

The aim of our analysis is to evaluate the need of ipsilateral tonsillectomy within the surgical treatment of BCA due to the recurrence rate in surgical treated BCA.

2 Material and Methods

The work has been reported in line with the STROCSS criteria in its updated version.[22] The trial has been registered under researchregistry7772, “The role of ipsilateral tonsillectomy in the extirpation of branchial cleft anomalies- A retrospective monocentric analysis over 13 years”.

Institutional Review Board review and data protection

The study is stated as exempt due to IRB approval and EU data protection regulations. Our retrospective chart review fits the exempt criteria. The research involves the collection of existing data, documents, records, pathological specimens or diagnostic specimens and the data is recorded in an anonymous manner such that subjects cannot be identified directly or through identifiers linked to the subject.

Study population

The study population was derived from the electronic database of all consecutive patients who admitted to the tertiary academic ENT department with the diagnosis of BCA between 2006 and 2020. (see Figure 1)

Data collection and statistical analysis

We retrospectively evaluated patient charts, operation protocols and pathological reports. Relevant data were collected: gender, age, location of BCA (Robbin’s neck-level and side), tonsillectomy yes/no, histopathological differentiation, duration of follow up and recurrence of BCA. Nominal scale data was described with frequency, ratio scale data over median and standard deviation. We performed a purely descriptive data analysis using the software IBM SPSS Statistics 26.

Preoperative and intraoperative workup

Operative indication was based on anamneses, prior infections /neck swelling, clinical presentation and ultrasound or a MRI/CT to state the diagnosis of a BCA. In case of an existing fistula, excision of the skin duct and preparation along the tract with full extirpation was performed. Unilateral tonsillectomy was only performed, when a tract to the tonsillar fossa could be identified.

By way of illustration, a second branchial cleft cyst in a female pre-tonsillectomized patient was operated over a modified neck dissection approach and transoral transection of the tonsillar region. The cyst had contact with the tonsillar fossa on the right side. (see Figure 2 and Figure 3)

3 Results

The data of 160 patients (48,75% female; 51,25% male) included, comprised a median age of 35 years [3M;83yrs]. The grouped age distribution is shown in Table 1. We could integrate 17 patients (10,6%), with a lateral branchial cleft fistulae, whereas the rest of 143 patients (89,4%) included, had a lateral branchial cleft cyst. Within the BCA, 54,37% were located on the left, and 45,63% located on the right side.

Due to Robbin’s neck level [23] the most of BCA were located in the Level II (76,3%), followed by Level III (16,2%), Level I (2,7%), IV (2,0%) and V(1,4%), whereas 1,4% couldn’t be associated with a concrete Level.

When looking at the two BCA cohorts, within the fistula group, ipsilateral tonsillectomy was performed in 6 out of 17 patients (35,3%) and in 2 out of 143 patients within (1,4%) the branchial cleft cyst group. So, overall 8 out of 160 patients (5%) underwent ipsilateral simultaneous tonsillectomy.

Due to recurrence rate, we found relapses in 2 surgically treated patients within the branchial cleft cyst group (1,4%) and none within the branchial cleft fistula group, with a mean follow up of 31 months (26,9% readmission rate). Within the branchial cleft cyst group, 1 out of 2 patients underwent ipsilateral tonsillectomy, the other had no tonsillectomy. We could not prove a statistically significant difference in the recurrence-rate between the groups with or without tonsillectomy.

4 Discussion

The aim of this work was to figure out, whether the recurrency rate of BCA, where a tonsillectomy was performed, was lower than those BCA, where no tonsillectomy was performed. If this were the case, a simultaneous tonsillectomy during the extirpation of BCA would have been recommendable. In general, the recurrence rate of BCA after surgical excision is low. Within our data, in 2 out of 160 (1.25%) patients, we found BCA recurrency after surgical treatment. Both recurrency cases were evident within extirpated branchial cleft cysts (N=143), no recurrence occurred within the branchial cleft fistula group (N=17).

In literature, BCA recurrency-rates ranges from 0 to 4%. [13, 14, 21] Due to the low recurrency-rate within our data, a consistent conclusion cannot be drawn. To statistically achieve that, at least 30 recurrences would have been necessary, corresponding to a total number of cases of 2150.

The analysis of our data depicted, that tonsillectomies were performed in a reluctant manner. Tonsillectomies were solely performed in cases, where a fistula ended in the tonsillar fossa. This conservative behaviour is explained by the risk of postoperative bleeding, which is described in an occurrence-rate between 1,9% and 6% after tonsillectomies. [24, 25] Given the fact that recurrence-rate of BCA is lower to the risk of postoperative bleeding in tonsillectomy, the standardized ipsilateral tonsillectomy should be avoided. In our opinion, even in an residual tract in contact to the tonsillar fossa, tonsillectomy can be avoided due to the risk/benefit ratio, in accord with other authors. [5, 15-18]

Moreover, our data showed that 22.2% of patients that were initially suspected to have a BCA, in fact had a different diagnosis, that was of either benign or malignant histopathology. Eventhough we excluded those patients from our analysis we think, the reason for this was, that many of the patients were referred to the ENT department by either general practitioners or resident ENT specialists, that do not have the proper equipment to run necessary diagnostics. Further, preoperative diagnosis of cystic lateral neck masses can be crucial.

A weakness of our study is a certain loss-of-follow-up, which cannot be numericized. The underlying cause is the chosen study design. We do not know, whether all treated patients in the tertiary academic ENT department were readmitted to the same hospital in case of BCA recurrency. Furthermore, there is a chance, that some patients still relapse in the future. These factors could explain, why the recurrence rate within our data is lower than in literature about this topic.

5 Conclusion

The performance of an ipsilateral simultaneous tonsillectomy in the surgical workup of BCA cannot be recommended at the basis of our data due to the risk/benefit ratio.

6 References

1. Bagchi, A., et al., *Branchial cleft cysts: a pictorial review*. Pol J Radiol, 2018. **83** : p. e204-e209.

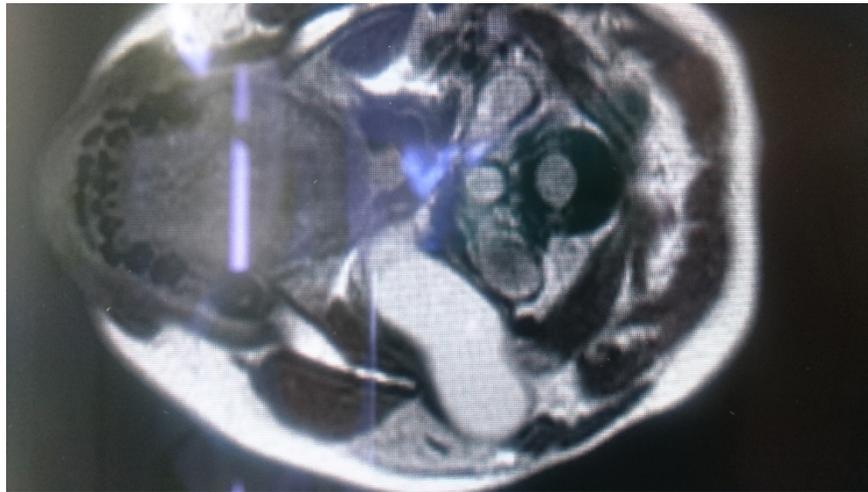
2. Coste, A.H., D.H. Lofgren, and C. Shermetaro, *Branchial Cleft Cyst*, in *StatPearls*. 2020, StatPearls Publishing
3. Papadogeorgakis, N., et al., *Branchial cleft cysts in adults. Diagnostic procedures and treatment in a series of 18 cases.* Oral Maxillofac Surg, 2009. **13** (2): p. 79-85.
4. Muller, S., et al., *Second Branchial Cleft Cyst.* Head Neck Pathol, 2015. **9** (3): p. 379-83.
5. Cheng, J. and L. Elden, *Management of pediatric second branchial fistulae: is tonsillectomy necessary?* Int J Pediatr Otorhinolaryngol, 2012. **76** (11): p. 1601-3.
6. Acierno, S.P. and J.H. Waldhausen, *Congenital cervical cysts, sinuses and fistulae.* Otolaryngol Clin North Am, 2007. **40** (1): p. 161-76, vii-viii.
7. Magdy, E.A. and Y.A. Ashram, *First branchial cleft anomalies: presentation, variability and safe surgical management.* Eur Arch Otorhinolaryngol, 2013. **270** (6): p. 1917-25.
8. Bolman, R.M., *Bilateral branchial cleft cyst.* Am J Surg, 1946.**71** : p. 96-9.
9. Jalil, J., F. Basheer, and M. Shafique, *Branchio-oto-renal syndrome.* J Coll Physicians Surg Pak, 2014. **24** (5): p. 367-8.
10. Fraser, F.C., et al., *Genetic aspects of the BOR syndrome—branchial fistulas, ear pits, hearing loss, and renal anomalies.* Am J Med Genet, 1978. **2** (3): p. 241-52.
11. Guldfred, L.A., B.B. Philipson, and C. Siim, *Branchial cleft anomalies: accuracy of pre-operative diagnosis, clinical presentation and management.* J Laryngol Otol, 2012. **126** (6): p. 598-604.
12. Prosser, J.D. and C.M. Myer, 3rd, *Branchial cleft anomalies and thymic cysts.* Otolaryngol Clin North Am, 2015. **48** (1): p. 1-14.
13. Li, L., et al., *The utilization of selective neck dissection in the treatment of recurrent branchial cleft anomalies.* Medicine (Baltimore), 2019. **98** (33): p. e16799.
14. Spinelli, C., et al., *Branchial cleft and pouch anomalies in childhood: a report of 50 surgical cases.* J Endocrinol Invest, 2016.**39** (5): p. 529-35.
15. Reddy, A., T. Valika, and J. Maddalozzo, *Definitive surgical management for second branchial cleft fistula: a case series.* J Otolaryngol Head Neck Surg, 2020. **49** (1): p. 55.
16. Yilmaz, I., et al., *Complete fistula of the second branchial cleft: case report of catheter-aided total excision.* Int J Pediatr Otorhinolaryngol, 2004. **68** (8): p. 1109-13.
17. Zhu, G.C. and D.J. Xiao, *[Transoral resection of partial fistula wall to treat incomplete second branchial fistula: a case report].* Lin Chung Er Bi Yan Hou Tou Jing Wai Ke Za Zhi, 2017.**31** (16): p. 1298-1299.
18. Kajosaari, L., et al., *Second branchial cleft fistulae: patient characteristics and surgical outcome.* Int J Pediatr Otorhinolaryngol, 2014. **78** (9): p. 1503-7.
19. Chih-Ho Hong, M.R.C., MD; , *Branchial Cleft Cyst Treatment & Management*. 2020, Medscape.
20. Reiß, M., *Facharztwissen HNO-Heilkunde*. 2009, Heidelberg: Springer Medizin Verlag. 1170.
21. Meijers, S., et al., *A Systematic Literature Review to Compare Clinical Outcomes of Different Surgical Techniques for Second Branchial Cyst Removal.* Ann Otol Rhinol Laryngol, 2022. **131** (4): p. 435-444.
22. Agha, R., et al., *STROCSS 2019 Guideline: Strengthening the reporting of cohort studies in surgery.* Int J Surg, 2019. **72** : p. 156-165.
23. Robbins, K.T., et al., *Standardizing neck dissection terminology. Official report of the Academy's Committee for Head and Neck Surgery and Oncology.* Arch Otolaryngol Head Neck Surg, 1991.**117** (6): p. 601-5.

24. Gross, J.H., et al., *Predictors of Occurrence and Timing of Post-Tonsillectomy Hemorrhage: A Case-Control Study*. *Ann Otol Rhinol Laryngol*, 2021. **130** (7): p. 825-832.
25. Mösges, R., et al., *Hemorrhage rate after coblation tonsillectomy: a meta-analysis of published trials*. *Eur Arch Otorhinolaryngol*, 2011. **268** (6): p. 807-16.

Figure 1 Study population and inclusion/exclusion algorithm



Figure 2 intraoperative picture of lateral second branchial cleft cyst extirpation



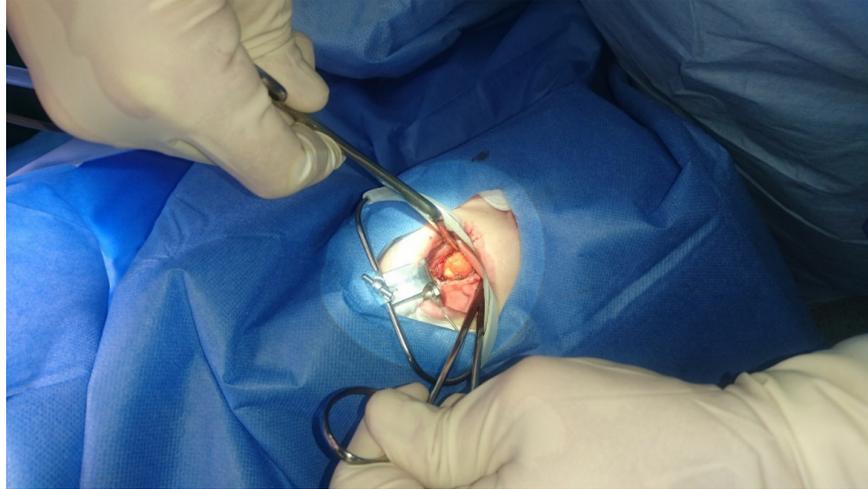


Figure 3 MRI and intraoperative situs of a large second branchial cleft cyst in contact with the tonsillar fossa

Age group	%
0 to 10 years	9,3% (N=18)
10 to 20 years	17,1% (N=33)
20 to 30 years	17,1% (N=33)
30 to 40 years	13,5% (N=26)
40 to 50 years	22,8% (N=44)
50 to 60 years	9,3% (N=18)
60 to 70 years	6,7% (N=13)
70 to 80 years	2,6% (N=5)
>80 years	1,6% (N=3)
	[?]=100%

Table 1 grouped age distribution in BCA