IJMS
Vol 31, No 1, March 2006

Case Report

Preservation of Facial Nerve in Surgery of First Branchial Cleft Anomaly: Five Cases of Duplicated External Ear Canal

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Abstract
We report five cases of duplicated external ear canal with especial attention to its histology and surgical care of facial nerve for their complete resection. These five patients were selected from 50 cases of first bronchial cleft anomalies including atretic external ear canals, preauricular fistula, cyst and sinuses. The characteristic histopathology and clinical features such as external opening around the auricle and upper neck above the level of the hyoid bone were not observed in other patients. There were no other abnormalities in the real external ear canal (EEC) and auricle. Three of them were connected to the natural EEC with an opening, hence producing ear discharges. Histologic appearances of well formed canal included a circumferential epithelial lined up and canal consisting of stratified squamous epithelium with skin adnexes. A remarkable cartilaginous skeleton (type 2 of first branchial cleft anomaly according to Work's classifications) was also observed. All of them were treated surgically, with preservation of the facial nerve and excising the superficial parotid gland along with the ductal opening and the tract.
Iran J Med Sci 2006; 31(1): 50-52.

Keywords ● First branchial cleft ● anomaly ● duplicated external canal

Introduction

Duplicated external ear canal is a rare congenital condition. It is a form of presentation of firs branchial cleft type 2 anomaly.[1] All of this abnormalities may have an external opening which is located from ear lobule down to the hyoid bone in the neck.[1,2] Type 2 anomaly of the first branchial cleft has been reported more frequently.[4,5] Some of these cases may have an opening to the external ear canal, in this case the patient will complain from ear discharge. The tract of this fistula in type 2 of first branchial cleft anomaly will have skin adnexes and cartilaginous structure which differentiate it from type 1 first branchial cleft anomaly.[1]

Our cases were from type 2 anomaly. Pathologic findings confirmed complete structures of external ear canal with complete circumferential ring having normal skin and adnexes with cartilaginous component. Therefore, according to the classification of Vokurka the name of duplicated external ear canal was applied to these patients because they had their own normal external ear canal.[6] Patients' facial nerves had a distinct course in most of the cases.

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This study presents two main surgical points (loop formation of facial nerve and opening into the normal ear canal) with complete pathological structures of the fistula tract. However, as Work has pointed out they may have an abnormal route,[1] and it makes the results of this study unique in that two patients with abnormal loop formation around the fistula tract. Therefore, those surgeons who deal with these abnormalities should be careful in not hurting facial nerve.

Patients and Methods

The significant clinical remarks, the courses and the outcomes of the surgical intervention of five cases of duplicated external ear canal are presented in Table 1. We have to point out to the external opening and discharge as the chief complains presenting ever since their birth. This opening was above the hyoid bone in all the reported cases. We excluded such a case that was treated incompletely elsewhere while we completed the resection. This case did not meet our criteria for presentation here.

Table 1: Clinical conditions of patients (#) with duplicated external ear canal (EEC) openings and their relation to facial nerve (FN)

#	sex	Internal end	Relation to FN	FN	D
1	M	C at 1/3 EEC	loop formation	Intact, weak	-
2	F	O at 1/3 EEC	Inferomedial	Intact	+
3	F	C at 1/3 EEC	Inferomedial	Intact	+
4	F	O at 1/3 EEC	loop formation	Intact	+
5	M	O at 1/3 EEC	Medial	Intact	-

C= closed; O= opened; D= discharge

Three out of our five patients had an internal opening into the natural external ear canal (at 1/3 of the external canal), while two of them had blind internal end closed at the natural external ear cartilage. Although, the dye did not pass through EEC of the last two patients they did not complain about ear discharge.

During the surgery an excision of the anomalous opening and the tract was accomplished along with the superficial parotidectomy by preserving the main nerve trunk and its branches. In two out of five patients during the process of dissection we encountered with the loop of facial nerve around the tract which was the hallmark of surgical hard work as previously emphasized by Work.[1]

Histopathologic slides prepared with H&E staining delineated circumferential duct surrounded by normal parotid tissue externally and squamous epithelium lining inside with impacted desquamated material and debris (Fig 1). The duct (duplicated external ear canal) had a complete ring of cartilaginous skeleton all through its course to the internal end. The post-operative course was uneventful except for minimal weakness in one patient which subsided gradually. None of these patients developed recurrence or discharges up to follow-up of three years.

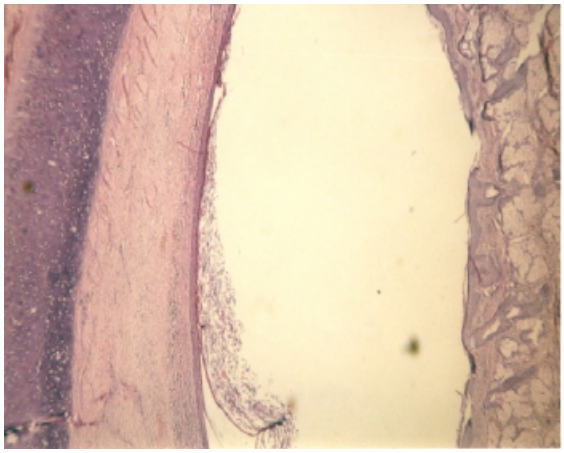

Fig 1: Cross Section of duplicated external ear canal (H/E; X 400)

Results

Here we present five cases of duplicated external ear canal while preserving the facial nerve. This abnormality is unique in that all cases of first branchial cleft anomaly had a circumferential (canal like) pattern. The specimen examine for complete excision helped us to expect an uneventful recovery and long term assurance for no recurrence. We had to do a complete excision, preserve the facial nerve, and close the opening end while it was connected with the natural ear canal.

Discussion

The review and the report of retrospective study of five cases of isolated first branchial cleft anomaly in the form of duplicated external ear canal presented here comprises the clinical course and the surgical accomplishment of a clean surgical excision with especial attention to the course of the facial nerve branches and the internal opening. Abnormal remnants of the first branchial cleft are rare congenital anomalies that have persisted through fetal life up to the delivery and present clinically from birth to adulthood.[3,5] They present as cyst or tract formation with external opening (sinus) or internal opening (fistula). The internal opening may locate at the external ear canal or even into the middle ear cavity.[2,3] They may also show other syndromes and non syndromic findings.[4,6,7]

These cases are unique in that they were presented with well formed duplicated external ear canal without other types of malformations as reported by others.[8-10] The remarkable surgical findings in these cases included the abnormal course of the facial nerve exploration using surgical microscope and nerve locator. The histopathological survey placed our cases into type 2 category of the anomalous of the first branchial cleft.

Conclusion

We have to insist on the knowledge and skill of the medical surgeons in early detection, referral and thorough clinical, surgical and histopathological surveillance to manage this abnormality more efficiently.

Acknowledgment

The authors would like thank the personnel of audiovisual service of Ghaem Hospital of Mashad University of Medical Sciences, for their sincere hard work in preparing and converting slides and digital pictures into printed materials.

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