

Bilateral variation of the parotid gland in cadaver: a case report

Serpil Cilingiroglu Anli¹ and Zuhul Kazak²

¹Department of Anatomy, Kirikkale University Faculty of Medicine, Kirikkale, Turkey

²Department of Anatomy, Kirikkale University Faculty of Dentistry, Kirikkale, Turkey

ABSTRACT

Bilateral variation of the parotid gland is an anatomically rare entity. In this study, we present a 91-year-old female cadaver with aplasia of the parotid gland on the left side of the face and hypoplasia of the parotid gland on the right side of the face. The accessory parotid glands on both sides were of normal size, while bilateral submandibular glands were found to be larger than normal. The surgical anatomy of the parotid gland is very important because this region of the face is a complex structure intertwined with important vessels and nerves. Patients with aplasia, atresia, agenesis, or hypoplasia of the parotid gland should be diagnosed early and proper treatment must be planned. Consequently, this case was found to be remarkable in terms of the coexistence of parotid gland aplasia and hypoplasia.

Keywords: parotid gland, major salivary glands, aplasia, hypoplasia, facial nerve

Abbreviations:

L: length

H: height

W: width

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INTRODUCTION

The parotid gland is the largest of the three pairs of great salivary glands adjacent to the oral cavity, which is located on the posterior lateral side of the buccal region and in the preauricular region. The parotid gland duct emerges from the superior-anterior edge of the gland, runs alongside the masseter muscle, and eventually opens into the oral cavity. The accessory parotid gland is on the trajectory of the parotid duct with a frequency of 20%. The saliva secreted from the parotid glands plays a role in softening of food, lubrication, enhancement of taste, and cleansing of the oral cavity; therefore, it contributes to immunity, digestion, and maintenance of homeostasis.¹⁻⁴ Another role of the parotid gland is to create a safe area for the facial vessels

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Corresponding Author: Serpil Cilingiroglu Anli, MD, PhD

Department of Anatomy, Kirikkale University Faculty of Medicine, Yenisehir, Ankara Road 7. km. Kirikkale University Campus, Yahsihan, Kirikkale, 71450, Turkey

Mobile phone: +90 535 719 46 38, Office: +90 318 333 50 00, Fax: +90 318 333 50 11, E-mail: serpile@kku.edu.tr

and nerve structures during the movements of the mandible. This situation makes it important to protect these structures during surgical interventions on the parotid gland.

The parotid gland begins to form from the oropharyngeal ectoderm in the 6th week of intra-uterine life. Meanwhile, the neighboring mesoderm forms lymphatic vascular networks and lymph nodes that are surrounded by the un-encapsulated parotid parenchyma.^{4,5} The parotid parenchyma develops epithelial-mesenchymal interactions that are required for the normal development of the epithelium.⁶ The developmental failure of this process leads to various malformations of the parotid gland, such as aplasia, atresia, agenesis and hypoplasia.^{1,7-9} Anatomically, aplasia describes the defective development or congenital absence of an organ or tissue, while atresia describes an abnormally-closed or absent orifice or duct. Agenesis, on the other hand, does not describe a developmental dysfunction during embryonic growth, but rather defines the lack of organ development due to the absence of primordial tissue. Finally, hypoplasia describes the underdevelopment or incomplete formation of a tissue or organ.⁹ Parotid gland agenesis may be partial or total, unilateral or bilateral, and may occur in conjunct with or in isolation from developmental problems in other major salivary glands. Unilateral absence of the parotid gland is a very rare entity with very few cases documented in the literature.^{8,10} In addition, hypoplasia of the parotid gland is reported exceedingly rarely.⁹

In this case report, it was observed that the left-side parotid gland of a 91-year-old female cadaver was absent, and hypoplasia of the parotid gland on the right side of the face was also noted. This situation is rarely described in the literature, and such anatomical variations may considerably alter the diagnosis and treatment of patients.

CASE REPORT

During the general studies performed in the macroscopy laboratory at the Department of Anatomy of Kirikkale University Faculty of Dentistry, a 91-year-old female cadaver was dissected. There was no macroscopic pathology and asymmetry in the head of the cadaver, which was fixed with 10% formalin. In the medical history of the subject, there were no diseases or syndromes that could be associated with variations of the parotid gland. This study was approved by the non interventional scientific research ethics committee of Kirikkale University (No. 2021.03.12).

Superficial structures were removed starting from the anterior edge of the auricula in accordance with routine dissection guidelines. The vessels and nerves in the area were preserved and the existing adipose tissue was carefully removed. In the intraoral evaluation of the cadaver, the majority of the primary teeth were present in good condition except for a 3-unit fixed partial denture. Parotid papillae ridges on the inner surface of the buccal mucosa were also noted. Metric data belonging to the visible gland sections were determined by linear measurements made with a digital caliper sensitive to 0.01 mm (Asimeto). We discerned hypoplasia of the parotid gland on the right side of the face (Figure 1). The superficial lobe of the parotid gland extends anteriorly and partially upward, over the musculus masseter and adjacent to the angulus mandible. The capsule of the gland partially bisected the superficial lobe portion, almost at its midline. The superficial lobe and each of the two separated segments of this lobe were quadrangular in shape. The dimensions of the right superficial lobe of the parotid gland were: length (L), 53.86 mm; height (H), 25.23 mm; width (W), 4.21 mm. On the right side, a deep lobe structure directed towards the retromandibular fossa from the posterior part of the mandible was noted. The dimensions of the right deep lobe of the parotid gland were: L, 30.54 mm; H, 23.94 mm; W, 11.17 mm. The accessory parotid gland, localized in the middle of the front edge of the right gland with close proximity, was also noted (Figure 1). This accessory lobe also had

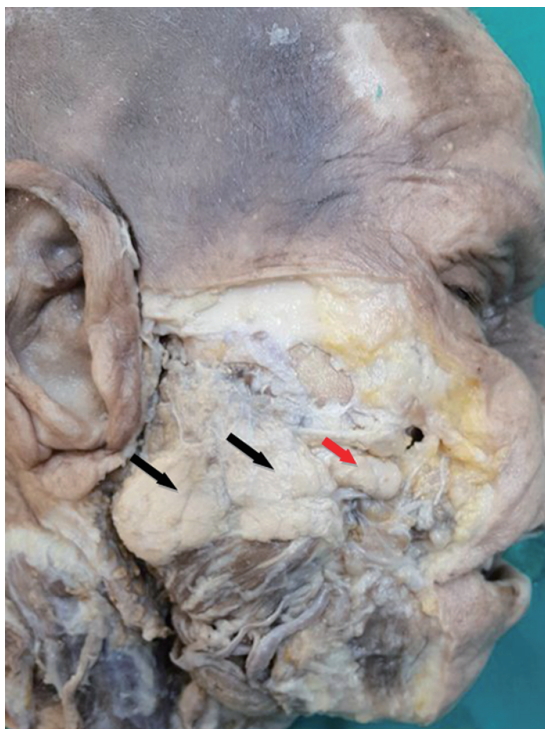


Fig. 1 Hypoplasia of the parotid gland (↘) and the accessory parotid gland structure (↗) (right side)

a quadrangular shape; maximum dimensions measured as the L, H, and W of the parotid gland were 18.29 mm, 9.07 mm, and 5.08 mm, respectively. We observed the right parotid duct to originate from a relatively deep site of the accessory parotid gland.

The dissection of the left side was advanced from the surface to the deep tissue. During dissection, it was observed that the tissue in the region where the parotid gland should have been located was in fact filled by extension of the local adipose tissue (Figure 2). The left side parotid gland was not present in the preauricular region (Figure 3). We widened the dissection anteriorly, preserving the facial branches of the facial nerve. A glandular tissue was present at the usual site of the accessory parotid gland (Figure 3). The left parotid duct structure originating from the deep face of this glandular tissue extending into the oral cavity was determined. The left accessory gland was found to be larger in size than the accessory gland on the opposite side of the face. The largest dimensions as the L, H, and W of the left accessory parotid gland were: 20.68 mm, 13.39 mm, 4.28 mm, respectively.

It is well established that submandibular glands provide the majority of salivary secretion. We extended the dissection in the direction of the submandibular triangle in order to evaluate the functional status of these glands. Submandibular glands were present on both sides with larger than usual dimensions. The maximum dimensions measured as the L, H, and W of the right/left submandibular gland within the submandibular triangle were 39.06 mm / 44.59 mm, 25.98 mm / 28.72 mm, and 21.35 mm / 17.72 mm, respectively. In the study, no parotid gland was seen in the left parotid gland space in the axial plane, which was visualized by non-contrast CT. In the localization indicated by the arrow in the right parotid gland cavity, an appearance compatible with hypoplasia of the parotid gland was detected (Figure 4).

Bilateral variation of the parotid gland



Fig. 2 Adipose tissue filling the parotid gland area (left side)



Fig. 3 The accessory parotid gland structure (↘) (Left > Right) and the absence of the parotid gland (left side)

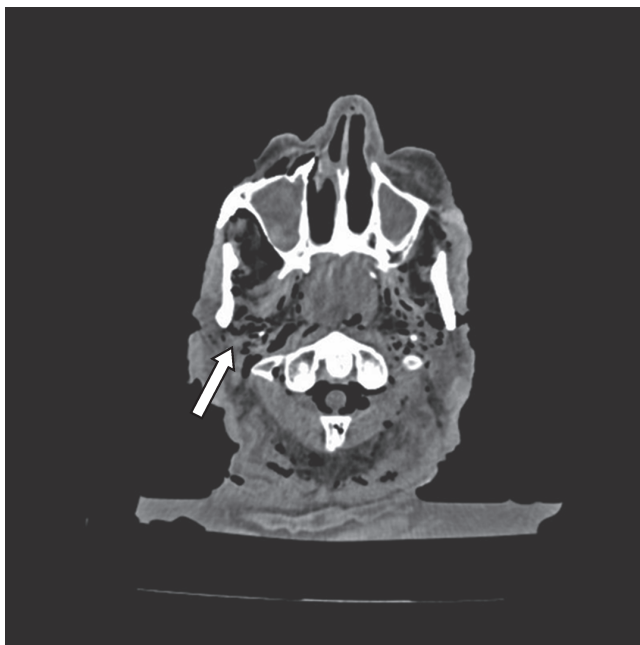


Fig. 4 The absence of the parotid gland and hypoplasia of the parotid gland on CT imaging. The hypoplastic parotid gland is marked with a white arrow in the right parotid gland cavity.

DISCUSSION

The absence of the parotid gland is an anatomically rare anomaly in humans. According to the literature data, it is evident that very few cases have reported unilateral or bilateral gland aplasia.^{5,8,10,11} A cadaver study showed that such an anomaly was described in the form of bilateral parotid gland aplasia in an adult male. Another study published in 1927, which assessed a male cadaver, showed both aplasia and hypoplasia in the parotid gland.¹² Our female cadaver demonstrated similar anatomical variations to the case reported by this study. It is extremely difficult to determine the incidence of unilateral gland aplasia, which is often asymptomatic.^{8,11} However, in some cases, facial asymmetry can be seen due to unilateral gland aplasia, and this can be clinically descriptive.^{10,11} In addition, cases with aplasia of the parotid gland on one side of the face may also have hypertrophy of the parotid gland on the opposite side, and in these cases, the findings may be misinterpreted as a tumor.¹⁰ Rarely, the retromandibular parotid space on the side of the face with parotid aplasia may be filled with adipose tissue.^{9,13} When the literature was reviewed, it was reported that half of the 22 cases with unilateral absence of the parotid gland were male and half were female, and the absence of the gland was seen more frequently on the right side of the face.⁵ Compared to the literature, our advanced-age female case had a symmetrical face before dissection. In the dissection of the same side of the face, we observed that the left parotid gland was absent and the entire retromandibular space was filled with adipose tissue. It was also observed that there was hypoplasia of the parotid gland on the contralateral side.

Hypoplasia of the parotid gland is vanishingly rare, even when compared to the rarity of parotid gland aplasia. The extremely rare disorder of aplasia or hypoplasia of the parotid glands is probably caused by a block during organogenesis, a situation whose etiopathogenesis is not

explained clearly.^{9,14} Variations of the parotid gland can be isolated occurrences or may be associated with other conditions. These conditions include the following: variations of other major salivary glands, first and second branchial arch anomalies, Down syndrome, Klinefelter syndrome, Levy-Hollister syndrome, mandibulofacial dysostosis, and hemifacial microsomia.^{1,5,8,10,12,13,15} In a study evaluating parotid gland sizes by ultrasonography in 50 patients with healthy salivary glands, it was determined that the size of the parotid gland was not statistically dependent on age, and the parotid glands were measured as 46.3 ± 7.7 mm in the horizontal axis and 37.4 ± 5.6 mm in a transverse axis.¹⁶ In our study, when we evaluated the dimensions of the right parotid gland, the dimensions of the deep lobe showed marked hypoplasia. Additionally, the L measurement of the superficial lobe was normal, while the H and W measurements were found to be well below average.

The accessory parotid gland, which resembles the histological structure of the parotid gland, can be seen with parotid gland aplasia on the same side.¹ Average sizes of the accessory parotid gland are: longest diameter (mm) 15.8 ± 4.8 (range, 3.8–29.4) and shortest diameter (mm) 5.0 ± 1.4 (range, 2.0–10.4).¹⁷ In our case, these glands were bilaterally normal in size.

In a study on the subject, the dimensions of the submandibular glands were found as follows: L, 35 ± 5.7 mm; cloth depth, 14.3 ± 5.7 mm; extension in frontal scanning, 33.7 ± 5.4 mm.¹⁶ Considering literature data, the submandibular glands on both sides of our case were found to be hypertrophic and the left submandibular gland was slightly larger than the right. In most of the cases with unilateral absence of the parotid gland presented in the literature, the opposite parotid gland is painless but larger than normal.^{8,10,11} When the medical history and intraoral findings of the cadaver are evaluated together, we were able to define that the findings concerning the left parotid gland were conclusive for isolated unilateral aplasia, while the right parotid gland demonstrated findings conclusive for hypoplasia.

CONCLUSIONS

The parotid gland is closely associated with many vascular and nerve structures in the region. We reported a very rare case with unilateral parotid gland aplasia who also had hypoplasia of the parotid gland on the contralateral side. In addition, the accessory parotid glands were of normal size bilaterally, and submandibular gland hypertrophy was detected on both sides. In this case, we have mainly seen parotid gland variations together with various differences in vessels and nerves. It is very important for dentists, otolaryngologists, and head and neck surgeons to know the functional response and anatomical evaluation of the vascular and nerve structures in the parotid gland and also structures with close proximity outside of the gland. In live cases whose literature data were examined, reports of parotid gland aplasia were very rare and studies describing parotid gland hypoplasia were vanishingly rare. Our case report involving this advanced-age female cadaver was remarkable in that both aplasia and hypoplasia of the parotid gland were seen together.

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CONFLICTS OF INTEREST

None declared.

REFERENCES

- 1 Goldenberg D, Flax-Goldenberg R, Joachims HZ, Peled N. Misplaced parotid glands: Bilateral agenesis of parotid glands associated with bilateral accessory parotid tissue. *J Laryngol Otol.* 2000;114(11):883–885. doi:10.1258/0022215001904257.
- 2 Nadershah M, Salama A. Removal of parotid, submandibular and sublingual glands. *Oral Maxillofac Surg Clin North Am.* 2012;24(2):295–305, x. doi:10.1016/j.coms.2012.01.005.
- 3 Kochhar A, Larian B, Azizzadeh B. Facial nerve and parotid gland anatomy. *Otolaryngol Clin North Am.* 2016;49(2):273–284. doi:10.1016/j.otc.2015.10.002.
- 4 Olsen KD, Quer M, De Bree R, et al. Deep lobe parotidectomy-why, when, and how? *Eur Arch Otorhinolaryngol.* 2017;274(12):4073–4078. doi:10.1007/s00405-017-4767-5.
- 5 Teymoortash A, Hoch S. Congenital unilateral agenesis of the parotid gland: A case report and review of the literature. *Case Rep Dent.* 2016;2016:2672496. doi:10.1155/2016/2672496.
- 6 Cutler LS, Gremski W. Epithelial-mesenchymal interactions in the development of salivary glands. *Crit Rev Oral Biol Med.* 1991;2(1):1–12. doi:10.1177/10454411910020010101.
- 7 Antoniadis DZ, Markopoulos AK, Deligianni E, Andreadis D. Bilateral aplasia of parotid glands correlated with accessory parotid tissue. *J Laryngol Otol.* 2006;120(4):327–329. doi:10.1017/S0022215106000338.
- 8 Martin-Granizo R, Garca-González D. Unilateral agenesis of the parotid gland: A case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2004;98(6):712–714. doi:10.1016/j.tripleo.2004.04.008.
- 9 Sun L, Sun Z, Ma X. Hypoplasia of the parotid gland: computed tomography sialography diagnosis. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 2013;116(4):e297–e301. doi:10.1016/j.oooo.2013.06.022.
- 10 Daniel SJ, Blaser S, Forte V. Unilateral agenesis of the parotid gland: an unusual entity. *Int J Pediatr Otorhinolaryngol.* 2003;67(4):395–397. doi:10.1016/s0165-5876(02)00375-0.
- 11 Almadori G, Ottaviani F, Del Ninno M, et al. Monolateral aplasia of the parotid gland. *Ann Otol Rhinol Laryngol.* 1997;106(6):522–525. doi:10.1177/000348949710600615.
- 12 Ferguson MM, Ponnambalam Y. Aplasia of the parotid gland in Down syndrome. *Br J Oral Maxillofac Surg.* 2005;43(2):113–117. doi:10.1016/j.bjoms.2004.01.001.
- 13 Lee BH. Unilateral agenesis of the parotid gland associated with a pleomorphic adenoma in the ipsilateral buccal space. *Jpn J Radiol.* 2010;28(3):224–226. doi:10.1007/s11604-009-0401-9.
- 14 Kubo S, Abe K, Ureshino T, Oka M. Aplasia of the submandibular gland. A case report. *J Craniomaxillofac Surg.* 1990;18(3):119–121. doi:10.1016/s1010-5182(05)80327-0.
- 15 Yilmaz YF, Titiz A, Yurur-Kutlay N, Ozcan M, Unal A. Congenital bilateral parotid gland agenesis in Klinefelter syndrome. *J Craniomaxillofac Surg.* 2010;38(4):248–250. doi:10.1016/j.jcms.2009.09.006.
- 16 Dost P. Ultrasonographic biometry in normal salivary glands. *Eur Arch Otorhinolaryngol.* 1997;254(Suppl 1):S18–S19. doi:10.1007/BF02439713.
- 17 Ahn D, Yeo CK, Han SY, Kim JK. The accessory parotid gland and facial process of the parotid gland on computed tomography. *PLoS One.* 2017;12(9):e0184633. doi:10.1371/journal.pone.0184633.