Hypoplasia of the parotid gland: computed tomography sialography diagnosis

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We report two unusual cases of non-syndromic hypoplasia of the parotid gland. The hypoplastic parotid gland mimicked a preauricular parotid tumor in one case and presented as an incidental image finding in the other case. Absence of the deep lobe and isthmus of the parotid parenchyma could be determined on axial computed tomography (CT) by revealing fat tissue composition of the parotid space. The underdeveloped superficial lobe of the gland was observed on three-dimensional CT sialography. (Oral Surg Oral Med Oral Pathol Oral Radiol 2013;116:e297-e301)

The parotid gland is the largest salivary gland and is anatomically divided into the superficial and deep lobes by the facial nerve and its branches. The superficial lobe extends anteriorly superficial to the masseter muscle and posteriorly to the retromandibular parotid space. Cranially the superficial lobe extends to the zygomatic arch and caudally to the angle of the mandible. The Stensen's duct emerges from the anterior superficial lobe of the parotid gland and courses over the masseter muscle. The deep lobe of the gland extends through the stylomandibular tunnel and protrudes into the pre-styloid parapharyngeal space.

Histologically, the parotid gland is composed of parenchyma (the secretory unit and associated ducts) and mesenchymal stroma (the surrounding connective tissue including adipose tissue, nerves and blood vessels). Embryologically, the primordial parotid parenchyma develops from the invagination of stomodeum, which elongates to form a proliferative epithelial cord extending posteriorly to the preauricular and further to the retromandibular area.[1]

Disorder of this developmental progress gives rise to various malformations of the parotid gland,[1] including aplasia,[2] atresia, agenesis[3,4] and hypoplasia. Aplasia is defined in general as defective development or congenital absence of an organ or tissue. Atresia is a condition in which the orifice or duct is abnormally closed or absent. Agenesis refers to the failure of an organ to develop during embryonic growth and development due to the absence of primordial tissue. Hypoplasia is featured by underdevelopment or incomplete formation of a tissue or organ, which is similar to aplasia but less severe. Hypoplasia of the parotid gland may occur per se or associated with hypoplasia or aplasia of the submandibular gland and lacrimal gland.[5]

Imaging methods of ultrasonography, X-ray sialography, computed tomography (CT), magnetic resonance imaging (MRI) and scintigraphy could detect the absence of the parotid gland. Although the axial CT or MRI images could confirm the absence of the parotid gland by revealing the fat composition of the parotid space, the Stensen's duct needs to be visualized to resolve the confusion about the so-called "accessory" parotid gland overlying the masseter.[6,7] CT sialography provides as a potential effective method to distinguish the hypoplastic superficial lobe from the "accessory" parotid gland based on combined axial and three-dimensional observation.

We report two cases of hypoplasia of the parotid gland characterized by the absence of gland tissue in the retromandibular parotid space and the presence of hypoplastic superficial lobe with axial CT images combined with three-dimensional sialography.

CASE REPORT

Case 1

A 29-year-old male complaining of a preauricular mass was referred to our hospital. The patient accidentally discovered the mass a month before. No xerostomia, diet-related swelling or other remarkable salivary discomfort was noted.

Physical examination showed that a well-circumscribed, flat, non-tender and movable mass located in the right preauricular region and superficial to the masseter, which was approximately 3.0 cm × 3.0 cm × 2.0 cm in size. Intra-oral examination showed that the bilateral mucosa papillae of the parotid ducts were appreciably normal. The face was symmetric and no other prominent abnormality was noticed. The mass enlarged significantly after the contrast was injected via the right Stensen's duct during sialography (Figure 1).

CT examinations were performed using an 8-slice spiral CT machine of the GE Brightspeed Series (GE Healthcare, Pittsburgh, PA). The scans were carried out with the following

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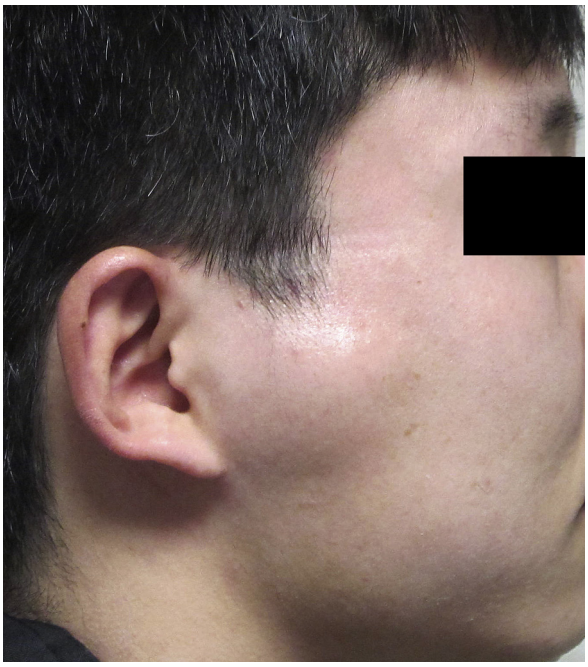

Fig. 1. Clinical photo of the case 1. The right preauricular mass enlarged after contrast media injection via the Stensen's duct.

parameters: 1.25 mm collimation; 1 s scanning time; 1:1.675 pitch; 120 kV and 200 mA. Axial images of 1.25 mm thickness were reconstructed with standard algorithm and the images were interpreted on the GE advantage workstation.

Serial axial CT images showed that the parotid gland was absent from the parotid tail to the isthmus and the deep lobe in the right retromandibular parotid space (Figure 2*A-C*). The CT attenuation of the right parotid space was approximately −100 HU (Hounsfield Unit). The retromandibular vein, external carotid artery and some interstitial strandings could be observed. The clinically palpated preauricular mass was well defined and located superficial to the right masseter. The attenuation and the enhancing appearance of the mass were identical with the normal parotid gland on the contralateral side (Figure 2*C*). Bilateral temporal bones, zygomatic arches, mandible and submandibular glands showed no remarkable abnormality.

Sialographic CT scan was initiated immediately after the contrast media was injected through the Stensen's duct (Iopamidol 2 mL, 37 g I/100 mL). CT sialography showed that the preauricular mass was significantly enhanced and the retromandibular parotid space was not enhanced (Figure 2*D*). Three-dimensional CT showed that the Stensen's duct was appreciably normal and connected directly to the mass (Figure 3*A*). The mass extended posteriorly to the posterior border of the ramus, superiorly to the zygomatic arch, inferior to below the sigmoid notch, which corresponded to the extending part of the superficial lobe. No parotid gland was seen in the retromandibular parotid space on CT sialography.

The preauricular mass was confirmed as a hypoplastic superficial lobe of the parotid gland. No surgical treatment was necessary. The patient had been followed up one year later and the mass showed no significant clinical change.

Case 2

A 32-year-old male was referred to our hospital for treatment of an oral mass. The patient had noticed a mass in the left posterior maxillary area for 2 years, which enlarged significantly in recent half year. No prominent pain or numbness was noticed. No xerostomia, diet-related facial pain, swelling or any other salivary related symptom was noticed.

Physical examination showed that a mass of approximately 2.0 × 2.0 × 2.0 cm located in the left maxillary tuberosity. The overlying mucosa was intact. Mouth opening function was normal. Another soft, non-tender and well-defined mass was palpated in the left preauricular region. The facial skin was normal. The bilateral papillae of the parotid duct were visually normal and secretion was clear. The contrast media (Iopamidol 2 mL, 37 g I/100 mL; Shanghai Bracco Sine Pharmaceutical Corp, China) was successfully injected through the left Stensen's duct and the mass enlarged significantly during sialography.

CT showed the bony destruction of the left maxillary tuberosity with soft-tissue occupying mass. The left retromandibular parotid space was composed of adipose tissue with the attenuation of approximately −100 HU except for the vessel structures (Figure 4*A-C*). Well-defined gland tissue was observed in the anterior portion of the superficial lobe and superficial to the masseter muscle (Figure 4*C*). The CT attenuations of the gland tissue before and after intravenous enhancement were 0 and 20 HU, which were identical with the right normal parotid gland (Figure 4*C*). Sialographic axial CT showed that the gland tissue was enhanced by the contrast media and the retromandibular parotid space was not enhanced (Figure 4*D*). The right Stensen's duct coursed directly from the gland tissue (Figure 3*B*). The gland extended posteriorly to the posterior border of the ramus, superiorly to the zygomatic arch, inferior to below the sigmoid notch. Due to its morphological similarity with the anterior extending part of the superficial lobe, the gland tissue should be considered as a hypoplastic parotid gland.

Partial maxillectomy was performed and the oral mass was excised under general anesthesia. Histopathological examination of the maxillary tumor proved to be chondrosarcoma. The hypoplastic parotid gland was an incidental radiological finding and did not necessitate any treatment.

DISCUSSION

Malformations of the parotid gland including aplasia and agenesis were documented in limited articles[8] and documentation of hypoplasia of the parotid gland was even rarer. In the present two cases, the absence of the gland tissue in the retromandibular parotid space with the presence of the Stensen's duct and the anterior extending part of the superficial lobe established the diagnosis of hypoplasia.

Malformations of the parotid gland frequently relate to several developmental syndromes, such as the first and second branchial syndromes, Klinefelter and Downs syndrome.[8-13] No remarkable abnormality of the eye, mandible, temporal and zygomatic bone was

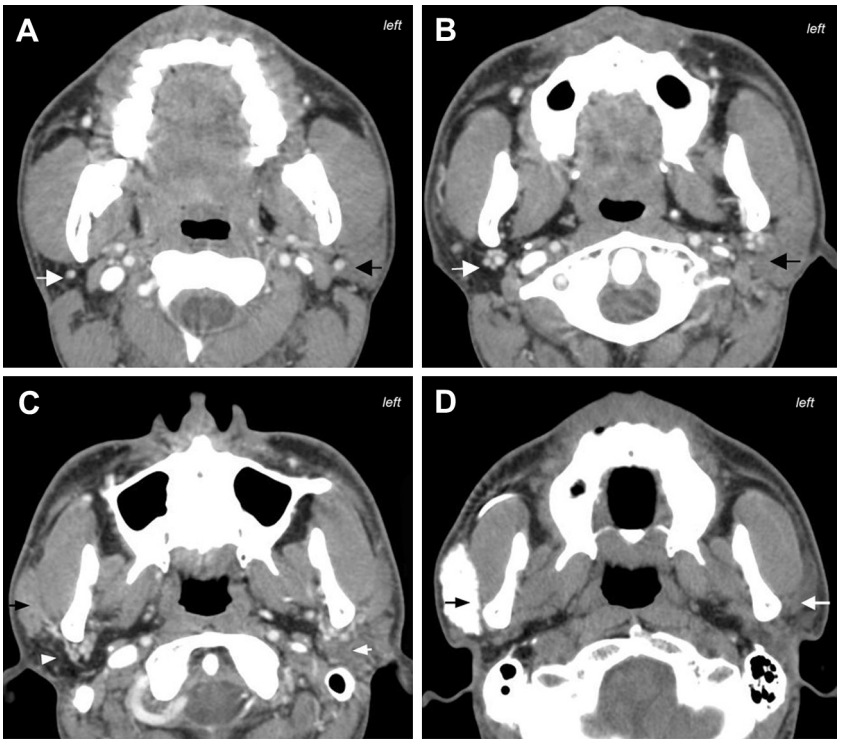

Fig. 2. Enhanced CT axial images in case 1. Axial images on the mandibular angle (**A**) and the mandibular foramen (**B**) levels showing the normal parotid gland on the left side (black arrows) and the absence of the parotid gland on the right side (white arrows). (**C**) Image on the condylar neck level shows the hypoplastic parotid gland (black arrow) overlying the masseter, which displays the identical attenuation with the normal left parotid gland (white arrow). Note the fat tissue in the retromandibular parotid space on the right side (white arrow head). (**D**) Axial image of the CT sialography shows that the hypoplastic parotid gland (black arrow) is enhanced with the contrast media and the retromandibular parotid space is not enhanced. Also note the normal parotid gland on the contralateral side (white arrow).

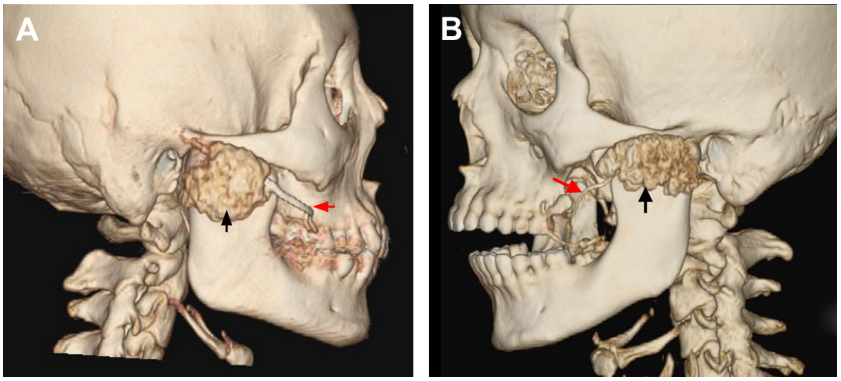

Fig. 3. Three-dimensional CT sialography images showing the hypoplastic parotid glands (black arrow) and the Stensen's ducts (red arrow) in two cases [(**A**) case 1; (**B**) case 2]. Note the absence of gland tissue in the retromandibular parotid space and the Stensen's ducts coursed directly from the gland. The gland takes the morphology of the anterior extending part of the superficial lobe and should be regarded as hypoplastic rather than accessory.

observed in the present two cases. Thus they should be non-syndromic malformation of the parotid gland.

Absence of all major salivary gland and severe decrease of the saliva may cause a profound xerostomia in children and result in several oral sequelae including dental caries, candidiasis, and disorders of smell, mastication and swallowing.[8] As only one parotid gland was involved, no xerostomia or other salivary related symptom was noted in the present two cases.

Our experience showed that the hypoplastic parotid superficial lobe might present as a preauricular mass clinically or be discovered as an incidental radiologic

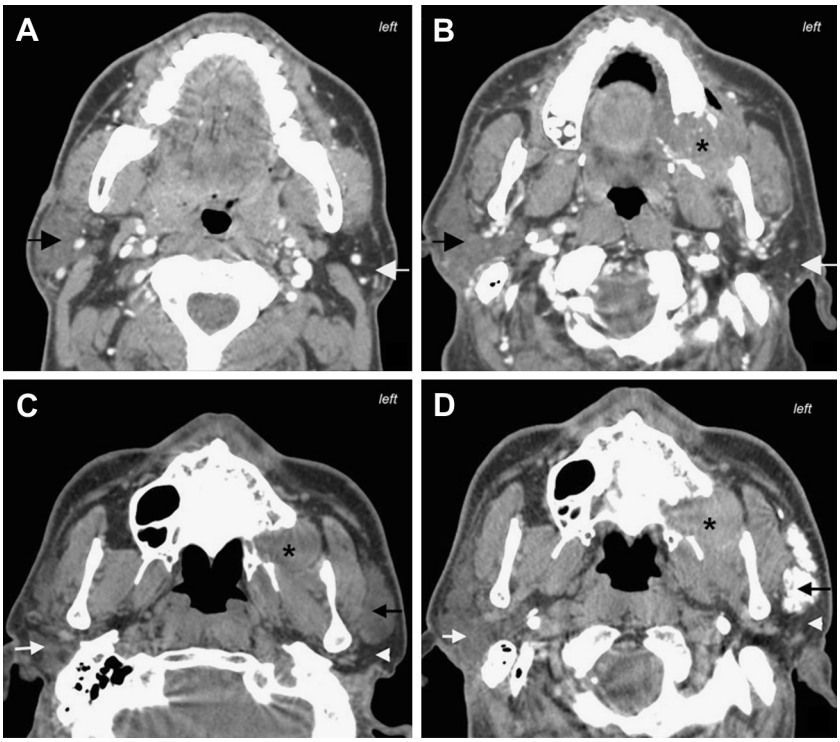

Fig. 4. Enhanced CT axial images in case 2. Axial images on the mandibular angle (**A**) and the mandibular foramen levels (**B**) showing the normal parotid gland on the right side (black arrows) and the absence of the parotid gland on the left side (white arrows). (**C** and **D**) Image on the condylar neck level shows the hypoplastic parotid gland (black arrows) overlying the masseter and the absence of the gland in the retromandibular parotid space (white arrow heads), compared with the normal parotid gland on the contralateral side (white arrows). The hypoplastic parotid gland (black arrows) could be enhanced during sialography (**D**). Also note the left maxillary mass (*).

finding, which should be carefully differentiated from a parotid tumor. Due to the absence of the parotid parenchyma and the replacement of adipose tissue in the parotid space, the retromandibular parotid area appeared to be relatively concave and much soft on palpation in contrast with the hypoplastic parotid gland, which made the hypoplastic gland mimic a parotid tumor.

During the literature review, we found out that several similar reports used "accessory" to describe the gland tissue overlying the masseter.[6,7] As the accessory parotid gland is anterior to and separate from the main parotid gland, adjacent to Stensen's duct as it passes over the anterior border of the masseter,[14,15] "hypoplasia" could better describe the embryological nature of the observed gland tissue overlying the masseter. Inadequacy of the axial CT or MRI images to show the three-dimensional spatial relationship of the Stensen's duct and the gland tissue might have led to this confusion. X-ray sialography could help to differentiate accessory parotid gland from the extending part of the superficial lobe. CT sialography could best show the holistic view of the gland and give an accurate diagnosis. Ultrasonography and scintigraphy could detect the absence of the gland parenchyma and loss of uptake function but could not show the holistic view of the gland.

CT and MRI are both sensitive in revealing the adipose component changes of the parotid gland. Normally the adipose tissue distributes homogeneously throughout the gland.[16] When the proportion of adipose tissue increases with the body mass index and age, the CT attenuation of the parotid gland decreases toward but could hardly approach that of adipose tissue (−100 HU). The attenuations of the parotid glands are homogeneously distributed within one gland and identical between bilateral glands of one individual.[16]

In accordance with previous reports, we found out that the parotid space without the gland parenchyma could be specifically characterized by fat tissue composition.[6,7,14,17,18] We could observe that the CT attenuations of the retromandibular parotid spaces devoid of the gland parenchyma were approximately −100 HU and significantly lower than that of the normal control side. In contrast, the hypoplastic parotid gland showed identical CT attenuations with the normal control side, which helps to differentiate from a real parotid tumor.

The treatment of the developmental abnormalities of parotid gland is highly dependent on the symptoms. In the present two cases xerostomia did not occur due to the presence of the other major salivary glands. Only very minor esthetic asymmetry of bilateral preauricular region could be remarkable, so we do not suggest any surgical treatment to these patients.

CONCLUSION

In conclusion, the hypoplastic parotid gland may clinically present as a preauricular mass or an incidental radiologic finding. CT sialography is effective in the diagnosis of the parotid gland hypoplasia. The parotid space could be mainly composed of adipose tissue when the gland parenchyma is underdeveloped.

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