TIG cleft fellows to substantive jobs in NHS trusts concerns about a bottle-neck effect have been exacerbated. A lack of success for OMFS trainees in TIG application processes is open knowledge via social media and other forums, with a suspicion that a less than meritocratic approach has been taken by some involved in the recruitment process. Disenchantment with the TIG process is also a significant problem in other subspecialty areas of OMFS, most notably head and neck oncology, where a number of leading units have actively withdrawn participation.

In my experience OMFS trainees are amongst the highest achievers overall and most dedicated in their training journey through necessity. It seems unlikely that they will be significantly assisted therefore by intensified interview preparation sessions, in contrast perhaps to trainees in sister specialties.

Ultimately, given that it is unlikely to prove feasible to reverse historical trends of centralisation of services, the best recourse would appear therefore to facilitate an increase of flow in to cleft and craniofacial training opportunities from OMFS. This could be achieved through reform of the current TIG process to make competitive selection more meritocratic and provide equal opportunities for candidates regardless of specialty of affiliation, as was originally intended. Alternatively, the creation of designated RCS approved fellowships in cleft and craniofacial around the UK would also serve to increase OMFS participation in this area, and consideration should be given to this idea at association level.

Conflict of interest

Not applicable.

Ethics statement/confirmation of patient's permission

Not applicable.

Reference

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Functional compensation of a hypertrophied sublingual gland and the absence of the ipsilateral submandibular gland

Sir,

The absence of the submandibular gland and the enlargement of the ipsilateral sublingual gland has been reported previously, and may occur unilaterally or bilaterally. The finding may be incidental, as a symptom associated with reduced saliva, or an enlarged gland, or it can manifest as a mass in the neck.[1–5] Although aplasia or atrophy of the submandibular gland is thought to cause hypertrophy of other glands, including the sublingual gland, compensation of function by the glands has not been shown so far. To the best of our knowledge, 4 cases of the bilateral absence of the submandibular gland and hypertrophy of the sublingual gland have been reported to date[1–4]; one had relatively poor oral hygiene and several cavities, and the others had no symptoms of salivary gland dysfunction. A 41-year-old woman was also diagnosed with aplasia of the submandibular gland and ipsilateral hypertrophy of the sublingual gland (with symptoms such as xerostomia and dysphagia), but showed no uptake on ^{99m}Tc-pertechnetate scintigraphy in the submandibular-sublingual area.[5] We think, therefore, that functional compensation may be independent of hypertrophy of the sublingual gland.

We report a patient who presented with enlargement of the sublingual gland that appeared to compensate for the atrophied ipsilateral submandibular gland. To the best of our knowledge, no reports on this condition have been published before.

A 66-year-old man visited the otorhinolaryngology department with a 7-day history of voice change. He was had no dryness of the mouth and his oral hygiene was good. Past diagnoses included Behçet's enteritis and uveitis, and ischaemic heart disease, but there was no history of cervicofacial surgery, radiotherapy, or salivary gland-related disease.

Physical examination showed no ulceration or scarring indicative of Behçet's disease in the oral cavity. Bimanual palpation, however, indicated an absence of the right submandibular gland, and no saliva was being expelled through the papilla. Laryngitis was diagnosed on endoscopy, then after informed consent was given, computed tomography (CT), magnetic resonance imaging (MRI), and sialoscintigraphy were also done. CT showed no abnormality except the absence of the right submandibular gland, and a $3.3 \times 1.3 \times 2.9$ cm enlargement of the ipsilateral sublingual gland. T2-weighted MRI showed complete replacement of the right submandibular gland with fatty tissue, with isointense and enlarged right sublingual and left submandibular glands (Fig. 1). The maximal uptake on ^{99m}Tc-pertechnetate scintigraphy occurred 30 min after the injection in the right submandiular-sublingual region, and was reduced in comparison with the left side (Fig. 2).

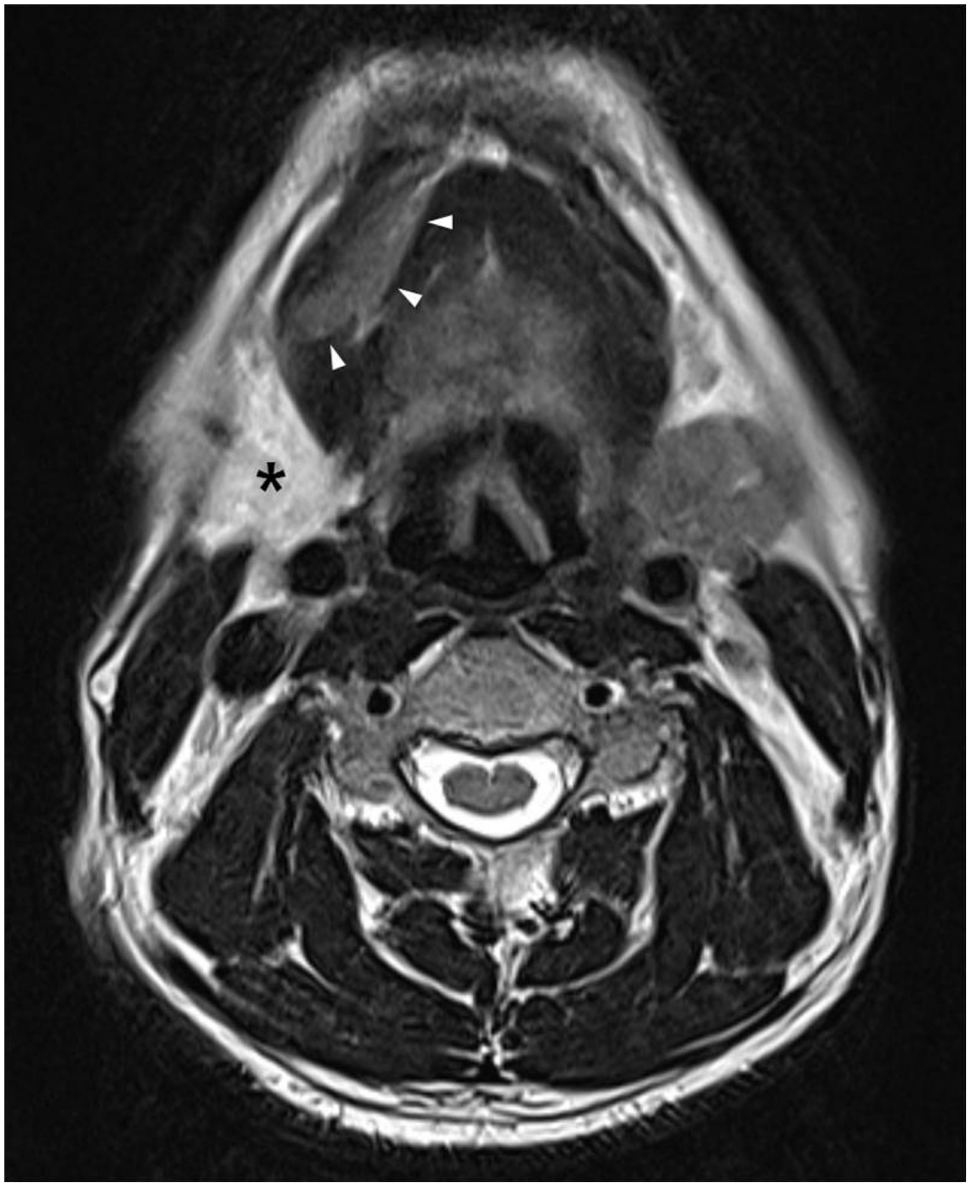

Fig. 1. Axial T2-weighted magnetic resonance image that shows total replacement of the right submandibular gland with fatty tissue (asterisk), the similar signal intensity of the hypertrophied, ipsilateral sublingual gland (arrowheads), and the left submandibular gland.

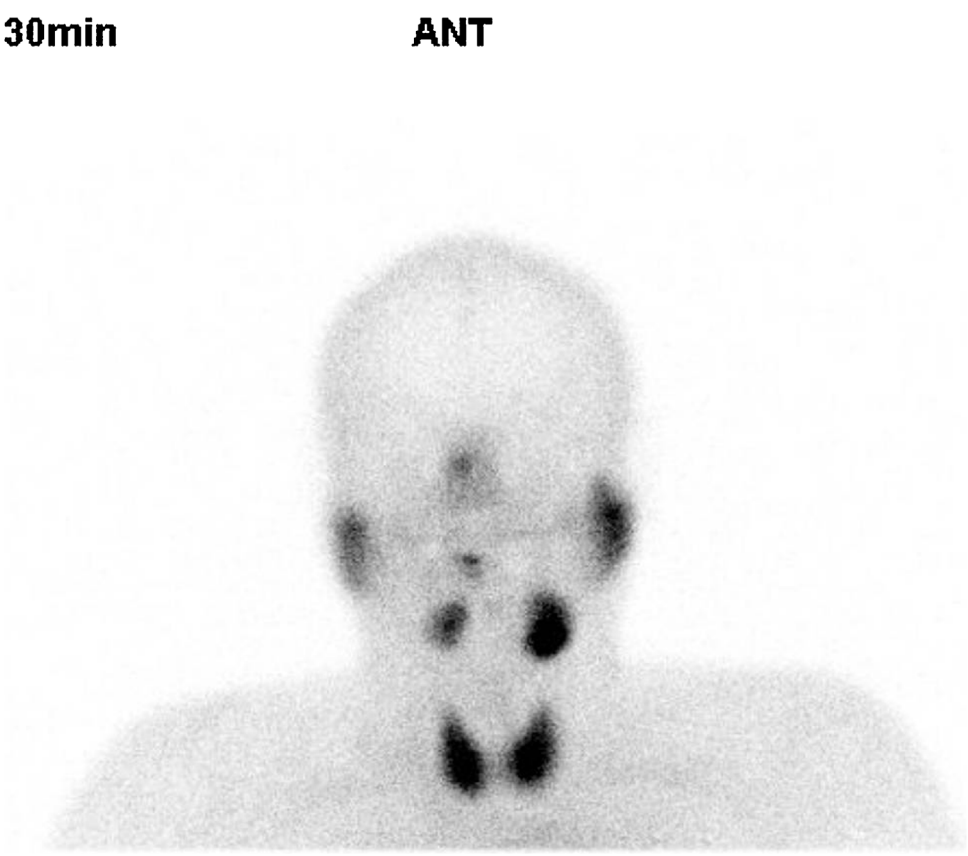

Fig. 2. Scintigraphic examination that suggests functional compensation of the right sublingual gland.

Although qualitative, the results of the scintigraphic examinations showed a distinct pattern of uptake in the submandibular-sublingual area, which were totally different from those reported previously. The change in the right submandibular gland seems to have been atrophic rather than aplastic, but the cause could not be established.

More research is needed to ascertain how the dysfunction of the submandibular gland leads to hypertrophy, and eventually, to understand the functional compensation of the ipsilateral sublingual gland. For patients having operations for cosmetic purposes, or other problems associated with enlarged sublingual gland, a functional evaluation should be done first to prevent the symptoms of dry mouth that may occur postoperatively.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patient's permission

This study was approved by the institutional review board of Veterans Health Service Daejeon Hospital. The authors have obtained the patient's informed consent.

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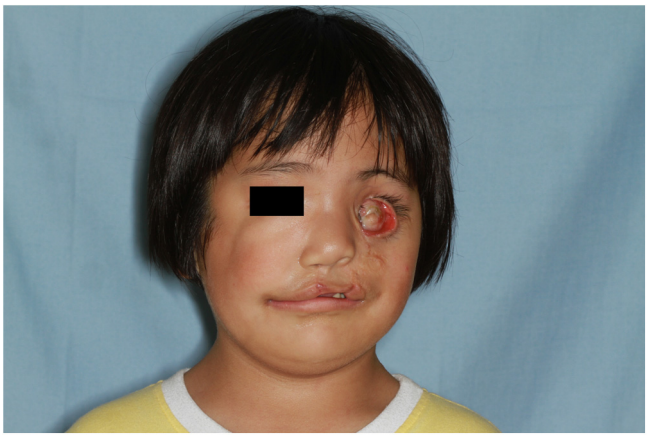

Fig. 1. Frontal view of the patient.

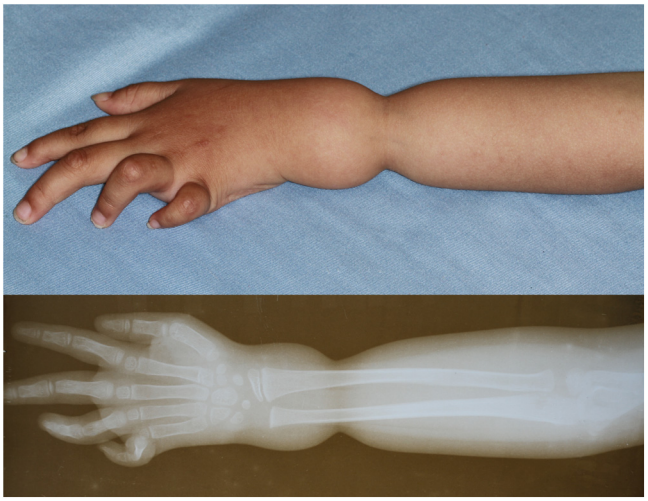

Fig. 2. Posterior view and radiograph of the left forearm.

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Amniotic band syndrome with Tessier number 4 and 7 clefts: a case report

Sir,

Our patient was a 7-year-old girl who had been abandoned at birth, so there was no prenatal history available, and information about her parents could not be accessed. Physical examination showed a constriction ring on her left forearm, and multiple types of craniofacial clefts.

The left-sided Tessier number 4 cleft passed through the high point of the Cupid's bow and extended laterally to the nasal alae up into the inferiomedial canthal region, and had been repaired when she was two. The bony alveolar cleft arose between the central and lateral incisors on both sides, and coursed laterally to the pyriform aperture, medial to the infraorbital foramen. The hard and soft clefts in the palate were complete on both sides. The left lower eyelid was cleft with involvement of the nasolacrymal duct, the left orbit was exposed, and there was slight macrostomia on the right side. A thorough physical examination showed no malformations other than the facial clefts and constriction ring.

At admission, her left eye was blind with severe injection and chemosis of the conjunctiva and subepithelial haze. Her left forearm had a constriction ring, but the bones were not involved. Her middle, ring, and little fingers had adopted a curled-up position when relaxed, but the hand functioned normally (Figs. 1 and 2).

A Tessier number 4 facial cleft is a rare, complex, and challenging craniofacial malformation, and when found in conjunction with amniotic band syndrome, is even more rare. Nivaldo Alonso et al[1] reported 21 cases of the cleft, in which four patients had amniotic bands in the limbs. The coexistence of clefts number 4 and 7 is extremely rare, however. Sigler et al[2] presented a case of a unilateral partial Tessier number 7 cleft accompanied by incomplete number 2 and 3 clefts. As far as we know, a combination of Tessier number 4 and 7 clefts has not yet been reported.

Two theories have been proposed to explain the pathogenesis of amniotic band syndrome. The intrinsic theory is based on a developmental anomaly of the embryonic germinal disc, which makes the amniotic band a by-product rather than the cause of the fetal anomalies. The extrinsic theory holds that an early rupture of the amniotic sac causes the formation of amniochorionic mesodermal bands that lead to the development of a constriction band. Early rupture may decrease amniotic fluid, which could cause the compressive consequences of early constraint such as scoliosis and clubfoot.[3]

Keller et al[4] observed similar anomalies in cousins affected by the syndrome, which suggested a genetic cause. Conversely, Torpin[5] reasoned that its malformations might be the result of external influences. He proposed several criteria such as: asymmetrical findings; syndactyly that starts distally with free proximal segments; no bony defects of the remaining parts; and lesions associated with fibrous bands that had originated from a denuded chorion.

Based on these criteria, we think that the extrinsic theory can better explain all abnormalities in this patient. According to previous publications and our own experiences, both intrinsic and extrinsic theories together explain a subset of cases of amniotic band syndrome more clearly, which suggests to us that it is aetiologically heterogeneous.

Conflict of interest

We have no conflicts of interest.