Tooth abnormalities in congenital infiltrating lipomatosis of the face

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Objective. The aim of this study was to present a literature review and case series report of tooth abnormalities in congenital infiltrating lipomatosis of the face (CIL-F).

Methods. Four typical cases of CIL-F are presented. Tooth abnormalities in CIL-F documented in the English literature are also reviewed. The clinical and radiological features of tooth abnormalities are summarized.

Results. In total, 21 cases with tooth abnormalities in CIL-F were retrieved for analysis. Accelerated tooth formation and eruption (17 cases), macrodontia (9 cases), and root hypoplasia (8 cases) were observed in CIL-F.

Conclusion. Tooth abnormalities including accelerated tooth formation or eruption, macrodontia, and root hypoplasia are common in CIL-F. (Oral Surg Oral Med Oral Pathol Oral Radiol 2013;115:e52-e62)

Lipomatosis refers to a diffuse overgrowth or accumulation of mature adipose tissue, which can occur in various anatomical regions of the body including the trunk, extremities, head and neck, abdomen, pelvis, or intestinal tract.[1] Congenital infiltrating lipomatosis of the face (CIL-F) was first described by Slavin et al.[2] in 1983 with the following main characteristics: a nonencapsulated mass containing mature adipocytes; fat infiltration in muscles and adjacent soft tissue; absence of malignant characteristics; absence of lipoblasts; presence of fibrous elements and increased number of vessels and nerves; and adjacent bone hypertrophy or exophytic overgrowth.[3]

Facial structures from the inferior orbital rim to the angle of the mandible are regularly involved in this disease.[4] The parotid gland, tongue, cheek, lip, soft palate, masticatory muscles, and jaw bones tend to be frequently affected by diffuse fat tissue infiltration.[4-6] Oral mucosa neuromas also frequently occur in this disease.[4] Cosmetic deformities, oral malfunction, speech and sleep disorders, and subsequent psychological problems can occur.[4,6,7]

Although radiological manifestations of this disease have been documented in the literature, related tooth developmental disorders have not been systematically reviewed. Various tooth developmental abnormalities including accelerated tooth eruption, macrodontia, abnormal root shape, and early loss of deciduous or permanent teeth have been documented.[4-8] In this article, we report 4 additional typical cases and present a review of associated tooth developmental abnormalities in this disease.

Congenital unilateral enlargement of hemifacial soft and bone structures, together with dental malformations, has also been documented in hemifacial hyperplasia. The diagnostic distinction between these 2 congenital disorders has been imprecisely documented. Some authors have reported that hemifacial hyperplasia is related to increased fat cells in the soft tissue.[9,10] Therefore, we also present our opinions on these 2 disorders.

CASE REPORTS

Case 1

A girl with a congenital diffuse left facial mass was referred to our hospital at 5 years and 9 months of age. The patient had undergone a partial resection of the mass at 8 months of age through a preauricular and submandibular incision. Abundant subcutaneous fat tissue was found and pathologic examination showed the mass to be composed of mature fat cells. A diagnosis of diffuse lipoma was made at that time. The mass increased proportionally in size as the patient grew. Her parents reported that the permanent maxillary and mandibular left first molars erupted when she was 4 years old. Family history and blood tests revealed no unusual findings. Physical examination revealed a soft, nontender, noncompressible and ill-defined mass on the left face (Figure 1, A). Ipsilateral hemimacroglossia was detected and several small nodules were seen at the tip of the tongue (Figure 1, B). The left soft palate was also thickened with an irregular soft mass. Early eruption of numerous permanent teeth was noted, including the permanent maxillary left first premolar and first molar and the permanent mandibular left first molar. No hypoplasia of the enamel was noted in existing primary or

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Fig. 1. Clinical photograph shows hemifacial enlargement on the left side (A). Intraoral view of the patient shows hyperplasia of the left hemitongue (B).

Fig. 2. Panoramic radiograph shows generally accelerated root formation (23-27, 31-37) and eruption (24, 26, 31-37) on the left side. Early calcification of 28 could be observed, whereas 18 is not seen. Macrodontia could be observed in 26 and 36, in which the roots are elongated and the apices have almost completely formed. Age-appropriate primary dentition is observed on the right side.

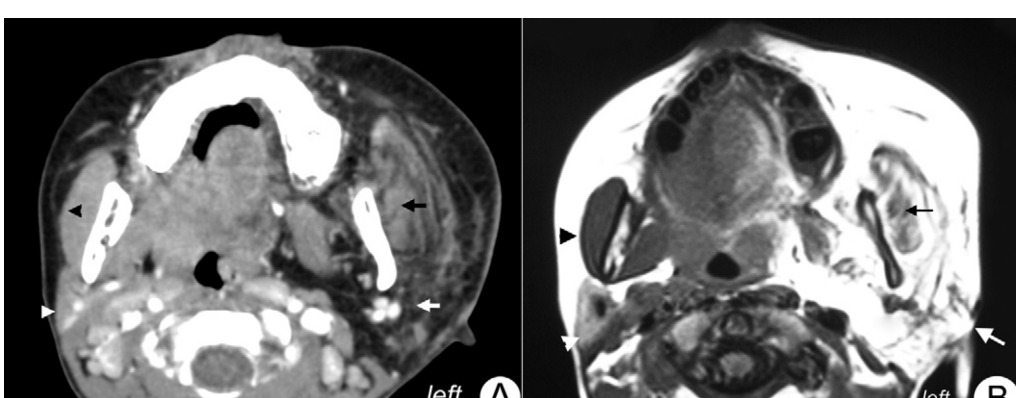

Fig. 3. Axial computed tomography (CT; A) and magnetic resonance imaging (MRI; B) show abundant fat tissue infiltrating the left masseter (black arrow), parotid gland (white arrow), pterygomandibular, and parapharyngeal spaces. Also note the normal appearance of the masseter (black arrowhead) and parotid gland (white arrowhead) on the right side. The fat tissue could be determined by its specific CT attenuation (−100 and −80 Hu before and after enhancement (A). Fat tissue infiltration observed on MRI shows high signal intensity on the T1-weighted image (B).

Fig. 4. Computed tomography volume rendering image show the asymmetry and hypertrophy of the left zygoma, maxilla, and mandible.

permanent teeth. A panoramic radiograph (Figure 2) showed that the formation of the roots of the permanent maxillary and mandibular teeth on the left side was generally accelerated compared with the right counterpart teeth. Especially notable are the permanent maxillary left canine and the permanent mandibular left first molar, in which the root apices had almost completely formed. Advanced crown formation of the permanent maxillary left third molar could also be observed, not seen on the contralateral side. Computed tomography (CT; Figure 3, A) showed that diffuse increased subcutaneous fat tissue accumulated in the facial and temporal region. Diffuse infiltration of fat tissue inside the left masseter and left temporalis could be seen between individual muscle fibers. The infiltrating fat tissue could also be confirmed on magnetic resonance imaging (MRI; Figure 3, B). The left zygoma, maxilla, and mandible were enlarged and deformed (Figure 4). This patient was kept under close follow-up. A series of surgical treatments was planned including pharyngoplasty, incision of the lingual nodules, cosmetic surgery, and possible facial bone plastic surgery.

Case 2

An 8-year-old girl with a congenital right facial mass was referred to our hospital. The patient had undergone a transoral reduction of the mass at 3 months of age. Pathologic examination revealed that the mass was composed of mature fat cells. Another partial resection of the mass took place at 5 years of age through a preauricular and submandibular incision. The mass increased proportionally with growth. Physical examination revealed a diffuse mass of the right cheek, which was soft and nontender on palpation (Figure 5). A scar from a facial incision was noted on her right face. The right upper lip was thickened and soft, but the tongue and soft palate were acceptably symmetric. No other clinical findings

Fig. 5. Clinical photograph shows the enlarged right cheek and the ptotic upper lip.

in the rest of the body were remarkable. A panoramic radiograph (Figure 6) showed generally accelerated formation and eruption of the right permanent teeth. Accelerated tooth and root formations were observed in the permanent maxillary right second and third molars, the permanent maxillary right second premolar, and the permanent mandibular right second molar and premolars. Short or stunted roots were prominent in the permanent maxillary right first molar, the permanent maxillary right first premolar, the canine, and the permanent mandibular right first molar. CT (Figure 7, A) showed diffuse infiltration of fat tissue into the right parotid gland, masseter, temporalis, and subcutaneous tissue. Fat tissue attenuation was observed in the involved parotid gland. Hypertrophy of the right zygoma and maxilla was also observed (Figure 7, B). Cheiloplasty was performed and pathologic examination of the excised specimen showed that abundant fat cells infiltrated the muscle fibers and inside the minor salivary gland acini (Figure 8).

Case 3

A 16-year-old girl appeared at our unit with a congenital, painless left facial mass. Four surgical procedures (partial resection of the mass and bone contouring) had been performed when she was 1, 2, 3, and 6 years of age. The mass continued to grow slowly as she grew up. Physical examination showed that the mass was diffuse from the left cheek to the inferior orbital region (Figure 9). It was soft, ill-defined, and immobile. A facial scar caused by a skin incision was prominent. A thickened left cheek, lower lip, and left hemitongue were noted. Two small soft-tissue nodules were seen at the tip of the tongue. No

Fig. 6. Accelerated root apex formation could be observed in 15, 17, 42, 43, 44, and 47. Accelerated formation of 18 is remarkable. Stunting of the root formation could be observed in 16, 14, 13, and 46.

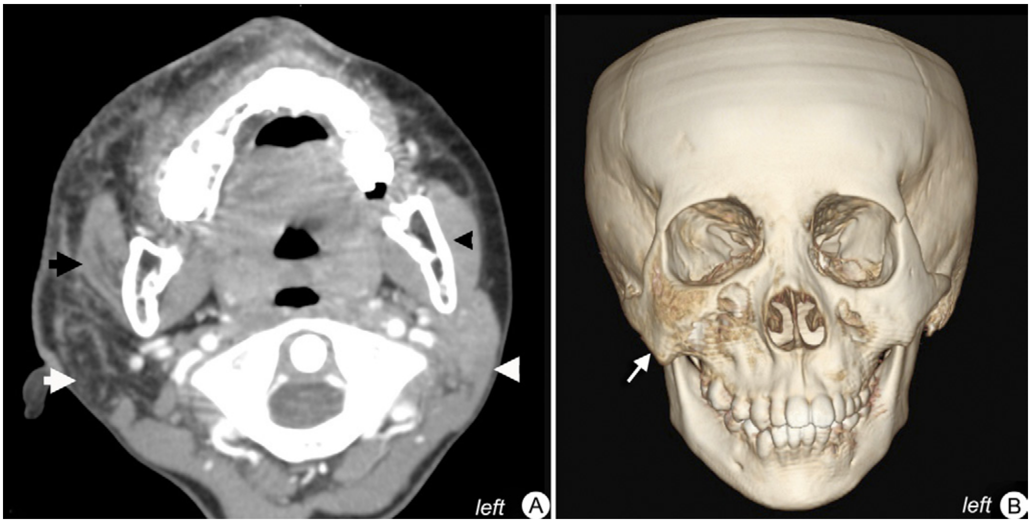

Fig. 7. Axial computed tomography (A) shows the fat infiltration in the right masseter (black arrow) and parotid gland (white arrow), compared with the normal counterparts on the left side (black and white arrowheads). Volume rendering image (B) shows the hypertrophy of the right zygoma and maxilla (white arrow). Note that the midline is deviated toward the left side because of the enlarged right maxilla.

other findings in the rest of the body were remarkable. Obstructive sleep apnea and hypoxia was confirmed by polysomnogram monitoring.

Numerous permanent maxillary and mandibular left teeth were missing, including the maxillary left canine and the first and second premolars. A panoramic radiograph (Figure 10) taken at 16 years of age showed general malformation of the remaining left permanent teeth, which were clearly displaced. Exophytic overgrowth of the left maxilla and zygoma were observed. The left mandible was also enlarged. A lateral oblique x-ray projection of the left mandible taken at 10 months of age (Figure 11) showed accelerated calcification of the permanent mandibular first molar and the primary mandibular second molar.

CT (Figure 12, A) and MRI (Figure 12, B) showed diffuse infiltration of fat tissue inside the left masseter, parotid gland, temporalis, tongue, and soft palate. The abundant fat tissue showed a high signal intensity on T1-weighted MRI and low signal intensity on fat-suppressed T2-weighted MRI. Exophytic overgrowth of the left zygoma and maxilla was prominent (Figure 13). The cortex of the zygoma became discontinuous with a deformed trabecular bone structure inside.

A biopsy was performed using an intraoral approach and a small piece of the zygoma was removed for pathologic analysis. Macroscopically, the specimen was brown and loose. Microscopic examination showed that the bone was made up of abundant mature fat cells intermingled with trabecular bone (Figure 14).

Case 4

A 23-year-old woman was referred to our hospital because of a facial deformity. The patient had left facial enlargement at birth, which grew proportionally with age. No pain or other discomfort was reported by the patient. She had undergone a

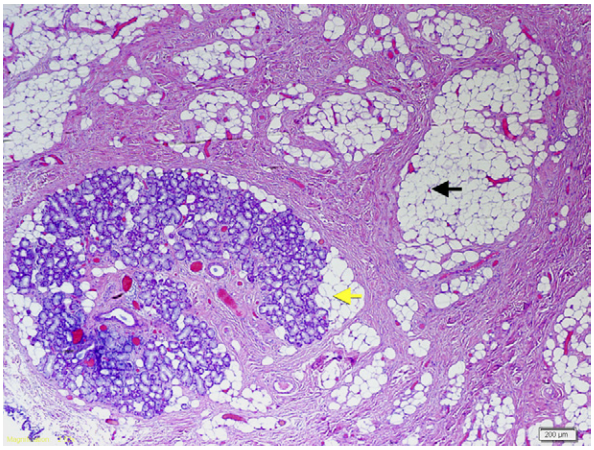

Fig. 8. Photomicrograph reveals abundant mature adipocytes infiltrating between lip muscle fibers (black arrow) and inside the minor salivary gland (yellow arrow). Hematoxylin–eosin stain, original magnification ×40.

partial reduction operation when she was 17 years old, but was still unsatisfied with the treatment outcome. Physical examination showed that the left facial area was diffusely enlarged from the inferior orbital region to the cheek (Figure 15), which was soft and nontender on palpation. A left preauricular scar was prominent as a result of the most recent operation. The left hemitongue, left maxillary, and mandibular alveolus were also significantly enlarged (Figure 16). The permanent maxillary and mandibular dentition was crowded and the occlusal plane was tilted. Panoramic radiograph (Figure 17) indicated short roots in several permanent maxillary and mandibular teeth (26, 27, 35-37). The root formation of the permanent maxillary and mandibular left third molars was observed. Two impacted permanent mandibular teeth were observed (38, 35). Enlargement of the left mandible, maxilla, and zygoma was also observed. Fat infiltration into the left masseter, parotid gland, and cheek could be observed on CT (Figure 18). Analysis of previous pathologic sections showed that abundant mature fat cells were present in the parotid gland (Figure 19).

DISCUSSION

A PubMed search from 1946 to 2012 identified 40 cases of CIL-F.[2,3,5,6,11-13] Tooth abnormalities did not receive the full attention they deserve in most of the previous studies. Seventeen cases were identified with tooth abnormalities in 6 English articles (Table I).[4-8,14] Padwa and Mulliken's study remained the largest case series of CIL-F, in which dental abnormalities occurred in 12 of 13 patients.[4] Poor formation of the roots (4 cases), missing permanent teeth (3 cases), macrodontia (3 cases), and dentigerous cyst (2 cases) were documented in this study.[4] Dentoskeletal enlargement and exophytic growth of facial bones were documented in several studies.[8,13]

In this study, we add to the knowledge that the stunting and acceleration of root formation could occur in very proximate teeth (case 2) and that tooth formation could be accelerated from the early intraosseous stage (case 3).

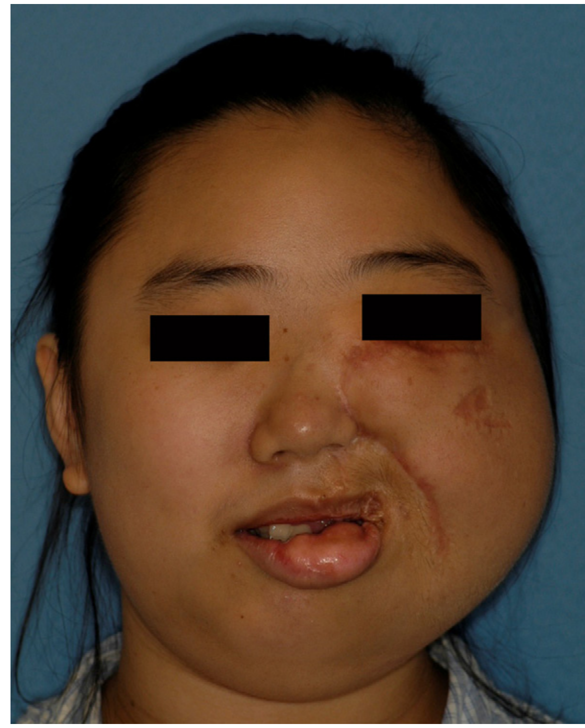

Fig. 9. Clinical photograph shows enlargement of the left cheek and the affected lower lip.

Accelerated tooth formation and early eruption are rare and have only been documented in a small number of congenital, systematic disorders or syndromes[15] such as cleft palate[16] and endocrine disorders.[17,18] In CIL-F, accelerated tooth formation and early eruption were confirmed in 17 cases,[4,6] which could have reasonably resulted from the accelerated root formation. Accelerated root development and apex formation could be either general in the hemimaxilla or mandible (case 1, this study) or sporadic (case 2, this study). Furthermore, it is remarkable that the accelerated crown formation could begin early in the intraosseous stage (case 3, this study), which again reinforces the congenital origin of CIL-F.

Hypoplasia of the roots in permanent teeth is also remarkable, and this was observed in 9 cases.[4,5,8] The degree of involvement may be variable. Short or club-like roots (7 cases) and absolute stunting of root development (case 2 in the present study) could be observed, which could reasonably result in early exfoliation of the permanent teeth. The hypoplasia of the root frequently involves hemimaxillary or mandibular permanent teeth. However, it is remarkable that stunting and accelerated apex formation of the root could occur in regional proximate permanent teeth in the same patient (case 2

Fig. 10. Hypertrophy of the left zygoma, maxilla, and mandible is prominent. Ipsilateral remaining permanent teeth (27-28, 34-37) are displaced with malformed roots; 23 to 26 are missing. Short roots could be observed in 27, 36, and 37.

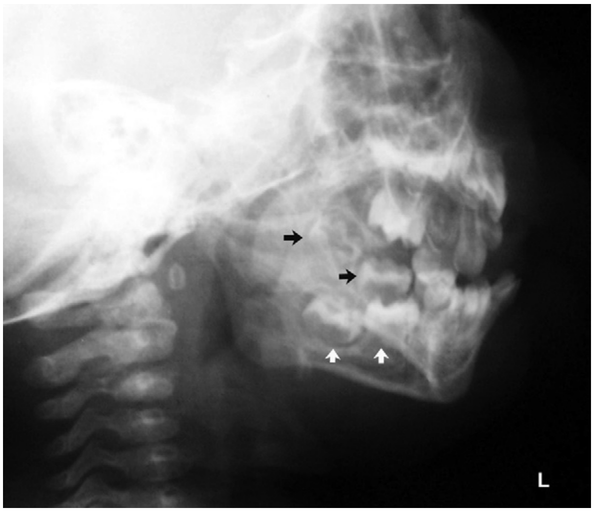

Fig. 11. A lateral oblique projection of the left mandible taken when the patient was 10 months old shows advanced calcification of the primary mandibular left second molar and the permanent mandibular left first molar (white arrows), compared with the right counterpart teeth (black arrows).

in the present study). This indicates that the developmental pathogenesis of CIL-F could either promote or hamper the root formation.

Macrodontia, which tends to be general in the affected jaws,[4,5,7,8,14] was observed in 9 cases. Enlargement of the crowns and elongated roots could be observed. Occurrence of macrodontia could possibly be related to accelerated dentoskeletal growth in CIL-F.[14] Macrodontia has also been documented in hemifacial hypertrophy,[9,19] which may present with a similar clinical appearance to CIL-F and is characterized by unilateral enlargement of facial soft and bony structures.

Diagnosis of CIL-F is not difficult based on the clinical and radiological features. Clinically, it presents as a congenital, diffuse, soft, and nontender facial mass, usually involving only the first and second branchial derivatives in the hemifacial area.[4,5,20,21] Diffuse infiltration of fat tissue inside various soft-tissue and skeletal organs could be specifically confirmed both on CT and on MRI.[20-22] Fat tissue could be determined by its low attenuation of approximately −100 Hounsfield on CT. On MRI, fat tissue presents with high signal intensity on T1- and T2-weighted images and with low signal intensity on fat-suppressed T2 images.

The distinction between hemifacial hyperplasia and CIL-F is imprecisely documented. Hemifacial hyperplasia is used to describe a clinical condition involving half of the face, including the maxilla alone or with the mandible, or in concert with other parts of the body.[19] The involved jaw and facial bones have been documented to show unilateral enlargement, accelerated development, and premature loss of primary teeth.[19] Tongue and alveolar bone enlargement may be present on the involved side.[19]

Because CIL-F mostly involves only hemifacial structures and frequently leads to hemifacial enlargement of both soft and bone tissue,[4] its clinical, dental, and radiographic appearance is similar to that of hemifacial hypertrophy.[7,14] The inadequacy of CT, MRI, or pathologic evidence was common in most previous studies on hemifacial hypertrophy.[9,23,24] The pathologic nature of hemifacial hypertrophy is not clearly known.[19] Some documented cases of hemifacial hyperplasia that had been pathologically proved to be composed of mature fat cells should be reasonably diagnosed as CIL-F.[9] The hemifacial hypertrophy case reported by Oktay et al. also revealed that the hypertrophy was caused by abnormal fat tissue within the facial muscles and soft tissue spaces, which obviously also conforms to the diagnosis of CIL-F.[10] So striking are the similarities between these 2 disorders that the possibility that they represent the same disease has been

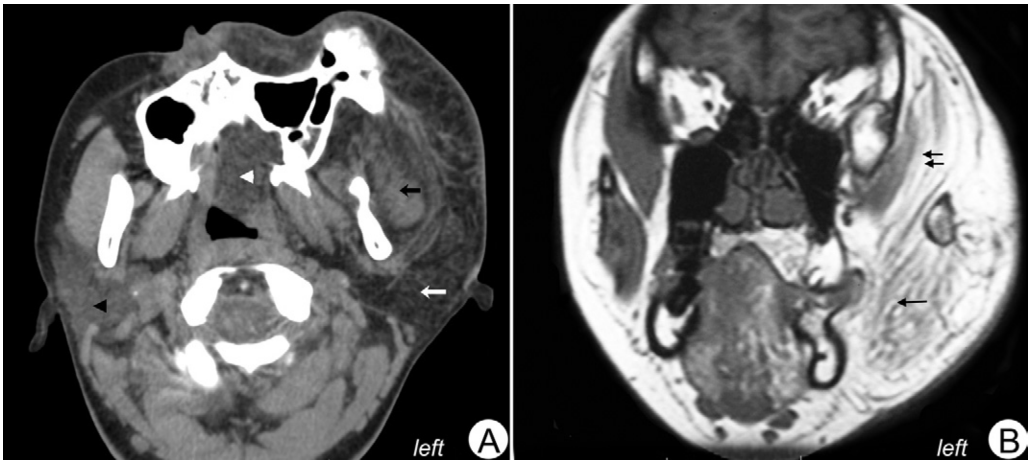

Fig. 12. Axial (A) computed tomography and coronal magnetic resonance (B) images show diffuse fat tissue accumulation in numerous hemifacial structures, including the infraorbital and cheek subcutaneous layer, masseter (black arrow), temporalis (double black arrows), parotid gland (white arrow), left hemitongue, and soft palate (white arrowhead). Note the normal parotid attenuation on the right side (black arrowhead).

Fig. 13. Volume rendering image shows hypertrophy and deformity of the ipsilateral zygoma, maxilla, and mandible.

suggested by some authors.[8] Because CIL-F could best illustrate the pathologic nature of this disease, we suggest distinguishing CIL-F from general hemifacial hypertrophy.

Hemimandibular hyperplasia is another frequently encountered deformity in dental practice, characterized by diffuse enlargement of the condyle, the condylar neck, and the ramus and body of the mandible.[25] The maxilla follows the downward growth of the mandible.[25] Hemimandibular hyperplasia clearly differs from hemifacial hyperplasia or CIL-F in that soft tissue overgrowth is consistently absent.[26]

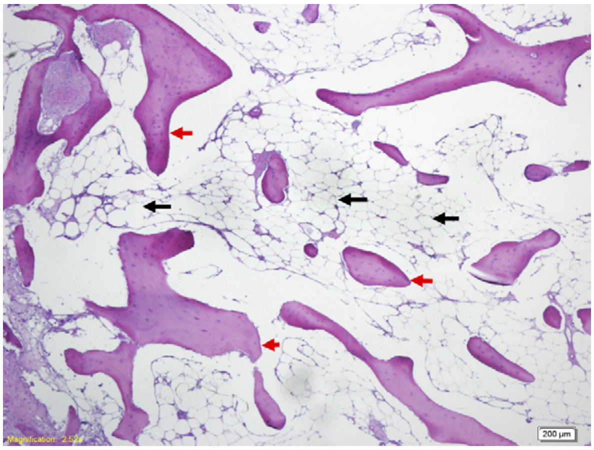

Fig. 14. Photomicrograph reveals abundant mature adipocytes (black arrows) infiltrating between trabecular bones (red arrows). Hematoxylin–eosin stain, original magnification ×40.

Intra- and intermuscular lipomas should be differentiated from CIL-F. Intramuscular lipomas are poorly defined but are usually restricted to single or numerous muscles. Intermuscular lipomas occur diffusely but are usually devoid of muscle infiltration. Lymphangioma, vascular malformation, and neurofibroma could also lead to the enlargement of jaw bones. But these disorders do not present with fat tissue characteristics on CT or MRI and could be easily differentiated from CIL-F clinically.

Pathologically, CIL-F characteristically manifests as lobules and sheets of mature adipocytes infiltrating among muscle fibers, trabecular bone architecture, or salivary acini. No malignancy or chromosome pleomorphism is observed.[2] CIL-F could be easily misdi-

Fig. 15. A 23-year-old woman with left facial lipomatosis who had undergone a partial reduction presented with hemifacial enlargement.

Fig. 16. Intraoral view shows the enlarged left hemitongue, upper and lower alveolus, and buccal mucosa.

agnosed as lipoma pathologically because they both manifest with mature fat cells microscopically. The pathogenesis is unclear because of its rarity. Neovascularization does not seem to play a role in the pathogenesis of this condition.[27]

No definitive management protocol exists for CIL-F. The long-term management goal should be to improve dental preservation, oral and sleep function, and psychological and cosmetic harmony. Because of high regrowth rates and the surgical risk to important anatomical structures, aggressive surgical excision should be delayed as long as possible.[20,21] Conservative surgical treatments, including cheiloplasty, liposuction, superficial parotidectomy, and bone plastic surgery, could be considered and should be performed at appropriate ages.

In conclusion, tooth abnormalities including accelerated tooth formation, premature eruption, and root malformations are common in CIL-F. CIL-F constitutes a distinct clinical entity that could manifest as hemifacial hypertrophy clinically.

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Fig. 17. Panoramic radiograph shows the enlarged maxilla, zygoma, and mandible. Short roots were observed in 26, 27, and 35 to 37. Also remarkable is the accelerated root formation of the left upper and lower third molars compared with the right counterparts.

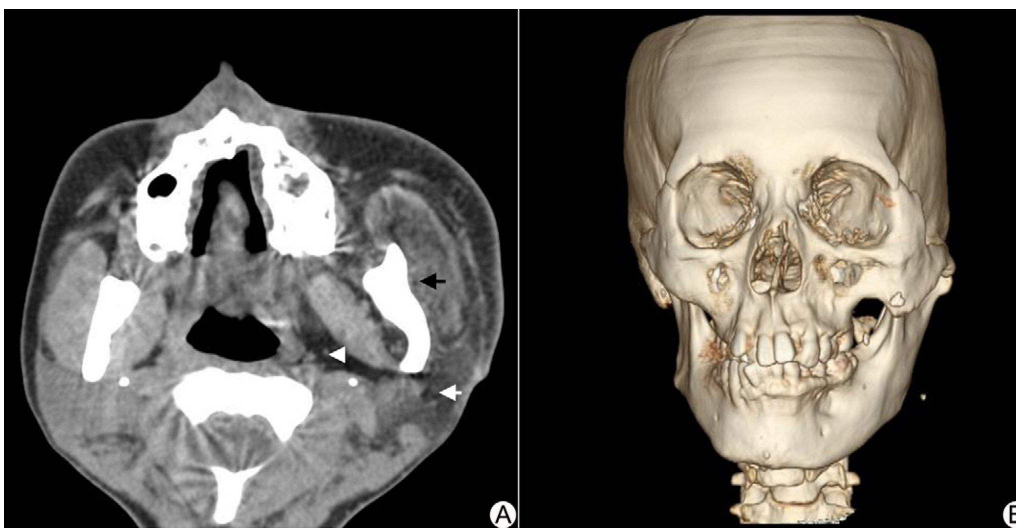

Fig. 18. Axial computed tomography (A) shows abundant fat tissue inside the left masseter (black arrow), parotid gland (white arrow), and parapharyngeal space (white arrowhead). (B) Three-dimensional view shows enlargement of the left zygoma, maxilla, and mandible.

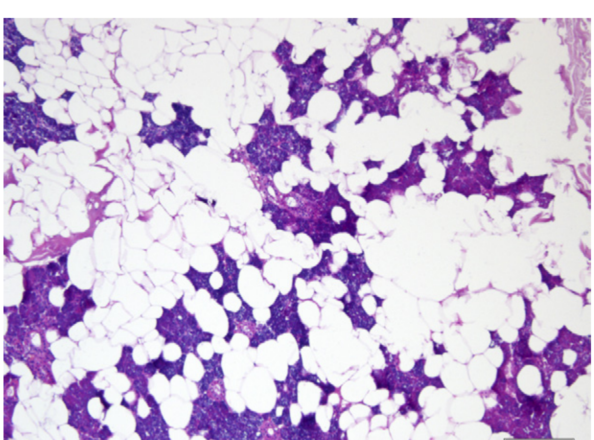

Fig. 19. Photomicrograph reveals abundant mature adipocytes infiltrating between the parotid acini (hematoxylin–eosin stain, original magnification × 80).

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Table I. Tooth abnormalities in congenital infiltrating lipomatosis of the face

Authors	*Cases*	*Sex*	*Age (years)*	*Side*	*Accelerated tooth development*	*Macrodontia*	*Root hypoplasia*	*Missing permanent teeth*	*Other tooth abnormalities*	*Enlarged facial bones*	*Soft tissue enlargement*
MacMillan et al.[8]	1 case	F	36	L	NS	Yes (26, 27)	5 left maxillary teeth	9 teeth (14, 15, 23, 24, 33, 34, 38, 44, 48)	Dentigerous cyst of impacted tooth	Left maxilla and mandible	Left facial from midline to preauricular region
Kang et al.[7]	1 case	M	25	R	NS	Yes (enlargement of right teeth, not detailed)	NS	NS		Right maxilla, zygoma	Right lower half of face and hemitongue
Bouletreau et al.[14]	1 case (case 1 in Kamal et al.[5])	F	16	L	NS	Yes (24-27)	Yes (22-27)	Yes (36, 38)	NS	Left mandible, zygoma	Left masseter, medial pterygoid muscle, hemitongue, cheek
Padwa and Mulliken[4]	13 cases	M (8), F (5)	1.1-22.9 (average age 12.4)	No predilection for affected side	Present in 12 cases	Present in 3 cases	Present in 4 cases	Present in 3 cases	Dentigerous cysts in 2 cases	Ipsilateral hypertrophy of the underlying facial skeleton (9 of 13)	Cheek (12), hemitongue (8), lip (12); mucosal neuroma (6)
Kamal et al.[5]	Case 3 of 3 cases	M	7	L	NS	Yes, in the left maxilla	NS	NS	NS	Left maxilla, mandible	Left cheek, parotid gland
Kim et al.[6]	1 case	F	3.5	L	Premature eruption of secondary teeth	Enlargement of left teeth	NS	NS	NS	Left mandible	Left ear, cheek, tongue, neck, parotid and submandibular gland
Sun (current study)	Current case 1	F	5	L	23-28, 31-38	16, 36	No	No	No	Left maxilla, mandible, and zygoma	Left cheek, hemitongue, soft palate, masseter, parotid gland
	Current case 2	F	8	R	18, 17, 15, 42, 43, 44, 45, 47	Not remarkable	16, 14, 13, 46	No	No	Right zygoma and maxilla	Right cheek, upper lip, masseter, parotid gland
	Current case 3	F	16	L	Primary mandibular left second molar and 36	Not remarkable	27, 36, 37	23-26	No	Left zygoma, maxilla, mandible	Left cheek, parotid gland, and masseter
	Current case 4	F	23	L	28	Not remarkable	26, 35-37	No	Impacted 35, 38	Left mandible, maxilla, and zygoma	Left cheek, parotid gland, masseter, and lower lip

F, female; *L*, left; *M*, male; *NS*, not specified; *R*, right.

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