

FIGURE 4. Intraoperative view showing the adhesion of cysts.

The surgery was realized through a vestibular route shifting the incision in the lip to avoid penetrating the cysts directly. A careful dissection of the horizontal then vertical fibers of the orbicularis oris lets the 2 cysts appear, showing the typical pearl-white aspect of dermoid cysts. Their thick envelopes were totally independent from each other. The adhesion area was found on the anterior surface, in contact with the muscle, toward the deep layer of the dermis (Fig. 4). The histologic examination revealed a typical aspect of dermoid cyst in both tumors. The surgical suites were simple with no recurrence at 1 year after surgery.

DISCUSSION

Dermoid cysts are slow-growing benign tumoral formations. They have 3 possible origins:

- Traumatic implantation of skin cells into deeper layers: they are similar to epidermal cysts. They represent 10% of dermoid cysts.²
- Congenital teratoma are composed of mesoderm, endoderm, and ectoderm derivatives³⁻⁵; they are principally located in the testes and ovaries but have been reported in the tongue.^{6,7}
- Congenital inclusion dermoid cysts form along the lines of embryologic closure. They contain both dermal and epidermal derivatives.

At the neck and the face level, New and Erich¹ distinguish 4 groups of possible location. The most frequent ones being the lateral ends of eyebrows (47%–70%) then the nasal dorsum (8%–12%)⁸⁻¹⁰ with a possible intracranial extension.^{11,12} The possibility of this intracranial location imposes the realization of a computed tomographic or MRI scan. The sublingual and neck regions are the 2 last locations described.¹ Bauer¹³ reported a case of dermoid cyst located on the base of the columella in relation with the maxillary sinus. In our patient, the attachment points were situated in the deep dermis.

The human face derives from 5 prominences of ectomesenchyme covered by surface epithelia. Union of the facial prominences occurs between the sixth and the eighth week of development.¹⁴ Fusion of the medial nasal and maxillary prominences provides for continuity of the upper jaw and lip and for separation of the nasal pits from the stomodeum. Medial nasal prominences form the columella, the tip of the nose, the philtrum of the upper lip, and the primary palate. This fusion requires the disintegration of contacting surface epithelia of prominences, allowing the contact of underlying mesenchymal cells. At this time, dermal inclusions may arise between frontonasal and medial nasal prominences, which manifest themselves more than 40 years later.

According to literature data, we present the first described case of bilateral dermoid cysts of the upper lip.

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Surgical Technique of the Transoral Approach to Remove a Lipoma of the Buccal Fat Pad

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Background: In 1727, Heister (Compendium anatomicum. Altdorf, Guille, Koleshii: editio tertia 1727: 134, table VIII and figs. 36–37) described the buccal fat pad (BFP) as an independent anatomic structure of the face; in 1801, Bichat (*Anatomie generale appliquee a la physiologie et a la medecine*. Paris, France: Brosson, Gabon et Cie Libraires, 1801:60) reported his fatty histologic finding. According to the literature, several pathologic tumorous conditions can

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Received March 28, 2011.

Accepted for publication July 26, 2011.

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The authors report no conflicts of interest.
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ISSN: 1049-2275

DOI: 10.1097/SCS.0b013e318231fe2b



FIGURE 1. A photograph demonstrating the echotomography.

arise from BFP, such as lipoma, lipoblastomatosis, liposarcoma, hemangioma, arteriovenous malformation, and nodular fasciitis; all of which are rare. After a revision of the English literature performed through PubMed between 1948 and 2008, we found 10 cases of lipomas arising from the BFP (7 cases are simple subtype, 2 are spindle cell lipoma, and 1 is fibrolipoma). The aims of this study were to introduce our clinical report of this rare pathologic entity, describe the surgical technique of the transoral approach, and discuss the potential pitfalls regarding the preoperative diagnosis and the close interrelation among the BFP, the facial buccal branches (FBBs), and the parotid duct (PD).

Clinical Report: A 43-year-old man was referred to the Maxillofacial Unit of the Novara Major Hospital with a 6-month history of a painless swelling in the right cheek. Clinical examination revealed a clearly visible, tender, slightly fluctuant mass, situated anterior to the masseter muscle and extended to the submandibular region. The patient underwent an ultrasound, a computed tomography, and a magnetic resonance imaging. Under general anesthesia with nasotracheal intubation, the patient underwent intraoral resection of BFP lipoma.

Discussion: The 2 major areas of discussion are the potential pitfall regarding the preoperative diagnosis and the close anatomic interrelation among the BFP, the FBB, and the PD. First, the spindle cell lipoma, one of the most common BFP lipoma variant, can be histologically and clinically similar to a well-differentiated liposarcoma, which can be recurrent and metastatic. This issue warrants that a careful workup of the tumorous mass of the buccal space and a BFP origin must be considered in every situation. Finally, according to the recent literature, the anatomic variations of the interrelation between the FBB and the BFP are classified into 2



FIGURE 2. A computed tomographic scan showing the tumor.

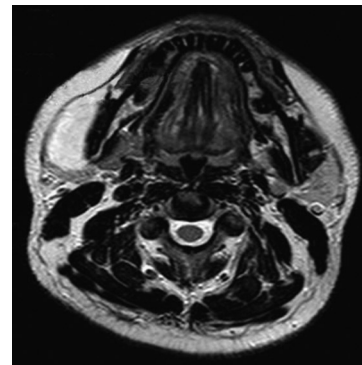


FIGURE 3. T2-weighted MRI scan demonstrating the hyperintensity of the lesion.

groups: (1) FBB passing lateral to the BFP and (2) branches crossing inside the BFP. The anatomic variations of the interrelation between the PD and the BFP are classified into 3 groups: (1) PD passing lateral to the BFP, (2) PD crossing deep to the BFP, and (3) PD running along the superior border of the BFP.

Key Words: Buccal fat pad, lipoma, liposarcoma, transoral approach, facial nerve, parotid duct

In 1727, Heister¹ described the buccal fat pad (BFP) as an independent anatomic structure of the face; in 1801, Bichat² reported his fatty histologic finding. According to the literature, several pathologic tumorous conditions can arise from BFP, such as lipoma,³ lipoblastomatosis,⁴ liposarcoma,⁵ hemangioma,⁶ arteriovenous malformation,⁷ and nodular fasciitis⁸; all of which are rare.

Moreover, pseudo herniation of the BFP can result in the formation of a mass. This condition is present either subcutaneously or intraorally. The former occurs in older individuals, and it is frequently associated with corticosteroids, trauma, or surgery due to loss of the ligamentous support of the fascia; the latter is usually seen in children, and it is associated with trauma.⁹

Although most of the BFP lipomas are of the simple subtype, other histologic variants, such as spindle cell lipoma and fibrolipoma, are also reported. After a revision of the English literature performed through PubMed between 1948 and 2008, we found 10 cases of lipomas arising from the BFP (7 cases are simple subtype, 2 are spindle cell lipoma, and 1 is fibrolipoma).¹⁰⁻¹⁴

The aims of this study were to introduce our clinical report of this rare pathologic entity, describe the surgical technique of



FIGURE 4. The mass clearly visible in the right cheek and extended to the submandibular region.



FIGURE 5. The planned incision.



FIGURE 6. The cut through the buccal mucosa revealing the buccinator muscle.



FIGURE 7. The intraoral removal of the tumor.



FIGURE 8. The closure of the wound.

the transoral approach, and discuss the potential pitfalls regarding the preoperative diagnosis and the close interrelation among the BFP, the facial buccal branches (FBBs), and the parotid duct (PD).

CLINICAL REPORT

A 43-year-old man was referred by his general physician to the Maxillofacial Unit of the Novara Major Hospital with a 6-month history of a painless swelling in the right cheek, around the body of the mandible; his medical history was unremarkable. He denied having other complaints, and there was no history of trauma. Clinical examination revealed a clearly visible, tender, slightly fluctuant mass, situated anterior to the masseter muscle and extended to the submandibular region.

The patient underwent an ultrasound, a computed tomography, and a magnetic resonance imaging (MRI; Figs. 1–3). These examinations showed a round, finely encapsulated fatty mass deformity with fibrotic changes anterior in the right cheek, lateral to the buccinator muscle, without infiltration of the surrounding structures (Fig. 4). This location led to the suspicion of a BFP lipoma, and a transoral approach to remove this tumor was planned (Fig. 5). Further preoperative investigations included a complete blood count, blood chemistry, electrocardiogram, and chest x-ray. Under general anesthesia with nasotracheal intubation, the patient underwent intraoral resection of BFP lipoma.

A bite block is inserted on the opposite side to operate on an adequately open mouth; infiltration of anesthetic and vasoconstrictor is performed to avoid bleeding, and steroid ointment is placed on the lips to avoid damage during the surgery. The incision is made with a no. 15 blade through the buccal mucosa and the buccinator muscle from just lingual to the external oblique ridge halfway up the mandibular ramus superiorly, to mesial of the second molar inferiorly (Fig. 6).

The cut is performed in a straight line with 5 mm of non-keratinized mucosa left buccally at the lower end of the incision for ease of suturing later; insufficient nonkeratinized soft tissue on the alveolar aspect can cause difficulty with suturing later and may lead to wound dehiscence. An incision too far lingually may damage the lingual nerve, causing lingual nerve anesthesia of the tongue; an incision too far buccally may cause excessive hemorrhage from the vessels in the masseter muscle. An incision too high may cause excessive hemorrhage from the vessels in the temporalis and/or masseter muscle.

The masseter muscle is left attached to the bone because tearing of periosteum and/or excessive stripping of the masseter muscle may cause hemorrhage, hematoma formation, and excessive postoperative swelling. Blunt dissection is performed to gain direct access to the anterior masticatory space. The tumor was extirpated entirely, and the wound was closed with slowly resorbing interrupted sutures

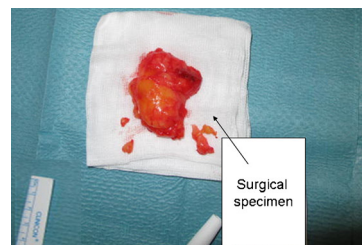


FIGURE 9. The surgical specimen.

(Figs. 7–9). The lesion was sent for pathologic analysis that confirmed the diagnosis of BFP lipoma: microscopically mature fatty tissue was seen separated by a fine network of connective tissue. No rich cellularity or lipoblasts were seen, and thus, the diagnosis of lipoma was confirmed. The postoperative course was uneventful; the patient was discharged 4 days after the surgical procedure, and a good mucosal healing was observed after 20 days, without any injuries of the FBB and the PD.

DISCUSSION

Despite the fact that several scientific speculations could arise from this case, we focus on 2 major areas of discussion: the potential pitfall regarding the preoperative diagnosis and the close anatomic interrelation among the BFP, the FBB, and the PD.

Lipoma of the BFP is a rare and underestimated pathologic condition; BFP lipomas show characteristics that can lead to confusion with other diseases. Extensive preoperative imagings are essential to understand the tumor extension and the potential infiltration. The spindle cell lipoma, one of the most common BFP lipoma variant, can be histologically and clinically similar to a well-differentiated liposarcoma, which can be recurrent and metastatic. This issue warrants that a careful workup of the tumorous mass of the buccal space and a BFP origin must be considered in every situation.^{5,15}

Liposarcoma of the BFP has been reported, constituting 10% to 15% of all the sarcomas; liposarcoma is the second most common soft-tissue sarcoma in adults, very rare in the head and neck region. Enzinger and Weiss¹⁶ described an incidence of 5.6%, and Cawson et al¹⁷ reported that the BFP is the most frequent head and neck location for liposarcoma. Moreover, if the clinical and histologic image of a well-differentiated liposarcoma can be similar to a lipoma, then it seems likely that more liposarcomas arise from the BFP than it is reported in the scientific literature.¹⁸

We think that a preoperative ultrasound and an MRI must be always performed to determine the potential origin from the BFP, visualize the extent in the various extensions, and exclude a liposarcoma; Gaskin and Helms¹⁹ stated that an MRI is 100% specific in the diagnosis of simple lipoma and has a 100% sensitivity with 83% specificity in distinguishing a liposarcoma. Moreover, MRI could be used preoperatively to perform an MRI sialography leading to a better visualization of any potential duct obstruction before the surgical resection.²⁰

These statements guide the surgical approach; whether an intraoral access could be sufficient in suspected simple lipoma or if a more radical cutaneous cervicofacial approach must be evaluated. Every resected BFP lipoma should be sent for pathologic analysis to determine the histologic feature because the clinical image is not always reliable.

Usually, a lipoma of the BFP is present as a mass in the cheek, underneath the superficial musculoaponeurotic system and lateral to the buccinator muscle. The clinical history is that of a slow-growing lesion with no history of trauma. On palpation, the overlying skin and oral mucosa are intact, and the tumor is tender. It does not adhere to the subcutis, and the mucosa can be moved freely over it.²¹

Finally, according to the recent literature, the anatomic variations of the interrelation between the FBB and the BFP are classified into 2 groups: (1) FBB passing lateral to the BFP and (2) branches crossing inside the BFP. The anatomic variations of the interrelation between the PD and the BFP are classified into 3 groups: (1) PD passing lateral to the BFP, (2) PD crossing deep to the BFP, and (3) PD running along the superior border of the BFP.²²

The results of these findings are that, during the procedure of removal of BFP by an intraoral approach, even with careful manipulation, some injuries to the PD can be inevitable and that some functions of facial muscles can be hampered; therefore, we recommend the use of a facial nerve monitoring during the surgical dissection, reducing the risk of any injuries.

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