Madelung disease, also known as benign symmetrical lipomatosis, was first described in 1846 by Brodie and subsequently identified as a diffuse multilobular lipomatosis by Madelung in 1888. The disease usually affects men, with a sex ratio of 15:1 to 30:1, during the fifth and sixth decades of life and is characterized by symmetrical diffuse adipose tissue deposits in the neck, shoulders, and arms. The present report described the case of a 51-year-old man diagnosed with Madelung disease who presented with masses primarily in the neck. He had previously shown partial improvement after injection lipolysis and shoulder surgery. However, 4 years later, following corticosteroid administration for the treatment of acute deafness, cervical lipomatosis progressed to the extent that he was unable to fasten his shirt. The initial treatment plan involved bilateral surgical excision of the lipomatous masses of the neck and liposuction for those in the submental area. However, the lipomas were adherent to the surrounding tissue and were partially fibrosed, presumably due to the previous injection lipolysis; thus, liposuction was not possible, and all the masses around the neck were carefully excised using cervical lymph node dissection technique. Thirty-two months later, the patient showed good cosmetic results, with no recurrence of cervical lipomatosis. Radical resection of the lipomas using a cervical dissection technique is useful in the treatment of Madelung disease; however, clinicians must consider the potential for adhesions and plan a meticulous dissection in those who have undergone injection lipolysis before the surgery. (Plast Reconstr Surg Glob Open 2015;3:e375; doi: 10.1097/GOX.0000000000000337; Published online 15 April 2015.)

CASE REPORT

A 51-year-old man was presented to our facility with large subcutaneous tumors affecting the neck. He had previously undergone corticosteroid administration for the treatment of acute deafness, which resulted in the progression of cervical lipomatosis. He was unable to fasten his shirt due to the masses around the neck. The initial treatment plan involved bilateral surgical excision of the lipomatous masses of the neck and liposuction for those in the submental area. However, the lipomas were adherent to the surrounding tissue and were partially fibrosed, presumably due to the previous injection lipolysis; thus, liposuction was not possible, and all the masses around the neck were carefully excised using cervical lymph node dissection technique. Thirty-two months later, the patient showed good cosmetic results, with no recurrence of cervical lipomatosis. Radical resection of the lipomas using a cervical dissection technique is useful in the treatment of Madelung disease; however, clinicians must consider the potential for adhesions and plan a meticulous dissection in those who have undergone injection lipolysis before the surgery.

Disclosure: The authors have no financial interest to declare in relation to the content of this article. The Article Processing Charge was paid for by the authors.
entire neck. Eight years earlier, he was diagnosed with Madelung disease based on the characteristic presentation of symmetrical subcutaneous tender masses throughout the body. Pathological and magnetic resonance imaging (MRI) findings showed that all the masses were lipomatous. The patient had a history of chronic alcohol abuse. At 46 years old, he underwent a shoulder lipectomy and monthly injection lipolysis of the neck for 7 months, which resulted in moderate improvement of the diseases.

Four years later, he experienced sudden deafness and was administered 10 mg betamethasone (intravenous drip tapered over 8 days), followed by 5 mg prednisolone for 10 days. Following steroid administration, progression of the neck lipomatosis was noted, and the patient was unable to fasten his shirt buttons (Fig. 1). When he visited our facility, the circumferences of the mandible and suprasternal notch were 52 cm and 58 cm, respectively.

He had no history of smoking but had consumed 360 mL/d of sho-chu alcohol (25–35% vol/vol) for the past 20 years. MRI images showed a high-intensity, clearly marginated, widely dispersed, fat-intense mass in the posterior and bilateral neck that was located beneath the platysma and trapezius muscles (Fig. 2).

We planned to perform liposuction to the submental area and radical excision of the mass using the cervical lymph node dissection technique for the lateral and posterior aspects of the neck, where the tumor extended deeply.

An S-line incision was made anterior to the ear, and multiple lipomas were visible beneath the platysma. We attempted to remove the lipomas completely using cervical neck lymph node dissection to prevent recurrence from the residual fat tissue; however, the lipomas were strongly adherent to the surrounding tissues, and gentle manipulation was required to preserve major nerves and vessels.

We then attempted liposuction at the submental area, but suction was hampered because of lipoma fibrosis, and all the masses were eventually dissected (Fig. 3).

The excised lipomas weighed 250 g (bilateral submental area), 240 g (right posterior neck), and 130 g (left posterior neck) (Fig. 3).

On histopathological examination, the majority of lipomas were composed of variable-sized adipocytes; others were composed of necrotic adipocytes and fibrous tissues that may have developed as a result of previous injection lipolysis (Fig. 3).

The patient did not experience any postoperative complications such as hematoma or seroma, and he was discharged from the hospital on the 10th postoperative day.

At 1 year postoperatively, the circumferences of the mandible and suprasternal notch had decreased to 39 cm and 43 cm, respectively. The patient complained of desensitization to the side of the neck, which persisted but was slowly recovering.

At the 32nd postoperative month, despite our recommendation to abstain from alcohol consump-
tion, the patient continued to drink. However, he had good cosmetic results, with no recurrence of the cervical lipomatosis, and he was able to fasten his shirt buttons (Fig. 4). An MRI 32 months postoperatively showed no recurrence of the lipomatosis beneath the platysma or trapezius muscles (Fig. 2).

**DISCUSSION**

Madelung disease is characterized by a significant cosmetic deformity resulting from symmetrical diffuse adipose tissue accumulation in the neck, auricles, and upper extremities.1 The disease etiology remains uncertain; however, a history of alcohol abuse is observed in up to 60–90%.4 Several reports have observed an association with mitochondrial dysfunction, various metabolic disorders affecting liver cytochrome p450 lipid metabolism and carbohydrate metabolism, and steroid consumption.5,6

The condition is resistant to lifestyle changes such as following a medical diet or abstaining from alcohol consumption, and surgical intervention is the first treatment strategy.4 However, several other treatment options such as injection lipolysis and liposuction have shown efficacy.5,6 Injection lipolysis is performed by injection of polyenylphosphatidylcholine-desoxycholic acid into the subcutaneous fatty tissue that reduces the extent of lipomatosis by destroying the cell wall.7

This treatment reduces the fat volume and is minimally invasive; however, it requires multiple applications, and its efficacy has been reported as moderate and temporary.7

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**Fig. 2.** Preoperative and postoperative T1-weighted MRI images. A, Preoperative image. B, Postoperative image 32 months after the surgery. Preoperative image shows a high-intensity, clearly marginated, fat-intense mass in the posterior bilateral neck, which was identified as a lipoma. The mass extends widely beneath the platysma muscle and does not compress the trachea. Postoperative image shows no lipoma masses beneath the platysma.

**Fig. 3.** Intraoperative findings and pathological images. A, Postexcision image. The resected masses weighed 250 g (right posterior) and 240 g (right anterior). B, Histopathological image of the surgical specimens (hematoxylin and eosin stain, ×40). Necrosed adipocytes (arrow) and fibrous tissue (arrowheads) were likely caused by the previous injection lipolysis.
By contrast, liposuction provides good aesthetic outcomes because lipomas are usually not encapsulated and they tend to expand diffusely. However, to avoid damage to adjacent vessels and nerves, a cautious approach should be taken, and serious hematomas or sensory deficits have been reported in 3% of treated patients.

In comparison, surgical intervention is far more reliable and is performed when rapid excision is required in patients experiencing functional disorders. However, surgery is more invasive compared with liposuction, and in some cases, a specific operative skill is required to remove masses located in the deep tissue layers.

Nevertheless, recurrence occurs when masses are not completely removed or when the patient cannot abstain from alcohol consumption, and it is difficult to accomplish complete removal of the tumor with liposuction and lipolysis; therefore, complete surgical resection is essential to avoid recurrence.

In the present case, the patient showed partial improvement in the lipomatosis after injection lipolysis; however, because of residual lipomatosis, the condition progressed after steroid administration.

We attempted to remove the lipomas using liposuction and surgical excision. However, liposuction was difficult, and surgical excision with careful exfoliation was the only choice for complete removal of the masses, which we accomplished by conducting cervical lymph node dissection technique.

Continual cautious observation of the patient is required; however, radical surgical excision might be necessary for the successful treatment of this disease.

CONCLUSIONS

In the present report, a patient diagnosed with Madelung disease underwent injection lipolysis and initially showed partial improvement; however, his condition worsened after receiving steroid treatment.

Liposuction could not be performed because of the presence of adherent fibrotic lipomas caused by previous injection lipolysis; therefore, cervical lymph node dissection was used.

During surgical planning, clinicians should consider the potential for adhesions and must exercise extensive intraoperative care for those who undergo injection lipolysis before surgery.

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PATIENT CONSENT

The patient provided written consent for the use of his image.

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