Cervical lymphocele: A rare complication following salvage surgery for ocular adnexal apocrine adenocarcinoma. A case report and review of the literature

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Background. An ocular adnexal apocrine adenocarcinoma (OAAA) is an extremely rare, but potentially aggressive and life-threatening tumor with ill-defined management based only on recommendations from a limited number of reported cases. The development of cervical lymphocele following neck dissection is a very rare complication, but one with well established methods for prevention and treatment. Here we describe a previously unreported case of salvage surgery including neck dissection for OAAA in addition to an emergence of cervical lymphocele. A literature review of current knowledge on both pathological conditions is included.

Methods and Results. A 58-year-old man suffering from OAAA, previously treated with multiple eye-sparing excisions and adjuvant proton therapy, underwent salvage surgery for locoregional recurrence of the tumor. A partial orbitectomy with orbital exenteration, primary reconstruction and left-sided neck dissection was performed. The procedure was complicated by a cervical lymphocele resolved after the surgical therapy. The patient remained disease-free during the one-year follow-up.

Conclusion. OAAA is a locally aggressive tumor with potential to local or distant metastatic spread. Whole-body staging, regular clinico-radiological follow-up and stage-dependent therapy with surgery as the first-choice treatment is required. A cervical lymphocele as a complication of especially left-sided neck dissection is managed with a conservative or surgical therapy according to the level of lymph leakage, extent and localization of lesions, presence of local or systemic disorders and the period from primary surgery.

Key words: cervical lymphocele, thoracic duct injury, neck dissection, adnexal tumor, apocrine adenocarcinoma, orbit, salvage surgery

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INTRODUCTION

Apocrine adenocarcinomas arise from apocrine sweat glands occurring in the skin of the axilla or anogenital area, in the areola and nipple of the breast and in the auditory canal[1,2]. Ocular adnexal apocrine adenocarcinoma (OAAA) is an extremely rare tumor emerging from modified apocrine glands of the eyelids (glands of Moll), which are situated at the base of eyelash follicles. A quite unique origin of OAAA from sweat glands of the lacrimal caruncle or lacrimal sac was also described[1,3,4]. OAAA was first reported by Stout and Cooley in 1951 (ref.[5]) and only 30 cases have been reported since[1,3,4,6]. Because of its sporadic occurrence, little is known about the biological behaviour of this tumor. OAAA may follow a relatively indolent and protracted clinical course, but some tumors have the potential for rapid growth with deep soft tissue or bone invasion, recurrence, and regional lymph-node and even distant metastases[2]. OAAA usually manifests as an eyelid mass with a clinical appearance similar to a chalazion, leading to misdiagnoses and delay in treatment[6]. The paucity of data about OAAA has not yet enabled the establishment of a management standard for this malignant disease. Owing to potential of the metastases and deep invasion in OAAA, adequate clinical staging (ideally the whole-body) using imaging methods should be performed. The mainstay of treatment is surgical therapy, meaning en bloc excision or resection aiming at the achievement of histologically negative surgical margins on the entire surface of the specimen[6]. The appropriate size of the clinical surgical margins is unclear. Patients with orbital extension require exenteration, accompanied with orbitectomy in selected cases. For patients in whom lymph node involvement is suspected, therapeutic neck dissection is necessary. Radiotherapy is used to improve locoregional control in patients with an aggressive course of the disease. The role of chemotherapy is still unknown[1]. Primary determinants of patients' survival include an oc-

currence of lymph node or distant metastases and local invasion into deep structures[1].

Thoracic duct injuries associated with neck dissections (especially those including levels IV and VI) are infrequent but potentially lethal complications, with an estimated incidence of 1-6% (ref.[7]). They may lead to a postoperative cervical lymphorrhea and development of chylous fistula, chylothorax or an exceptionally rare lymphocele[8]. The cervical lymphocele (chyloma) is defined as a circumscribed lymph collection in a neocavity lacking an epithelial lining, which is predominantly left-sided[9,10]. These lesions may also develop following other neck surgical procedures, trauma or spontaneously, probably due to the congenital or acquired lymphatic vessel wall weakness and degeneration or obstruction at the terminal end of the duct. Spontaneous lymphoceles compared to postoperative ones usually have a simple endothelial lining revealed by histopathological analysis[7]. Current recommendations for the management of cervical lymphoceles following neck dissections were adapted from experiences with the therapy of these lesions in other localizations in the body.

Here we present a case report of salvage surgery including neck dissection for recurrent, metastatic OAAA with the further complication of a cervical lymphocele. A review was carried out of all published cases dealing with both pathological conditions to improve knowledge of current management of these lesions.

CASE REPORT

A 57-year-old man consulted an ophthalmologist due to a one-month history of a swelling affecting the left lower eyelid. On clinical examination the eyelid margin and skin showed no visible pathology, but a painless indurated mass was palpable in the medial region. Visual acuity was unimpaired. During eye elevation (sursumduction), ocular motility was limited in the extreme position in the left eye with binocular diplopia in this position (vertical image disparity). A mild protrusion of the eyeball caused by the pathological process was found. Both eye anterior and posterior segment examinations were normal and other findings were in the physiological range. The medical history of the patient was irrelevant. The MRI revealed a hypervascular pathological mass 25x20x23 mm in extent predominantly infiltrating the medial half of the left orbital space with extension into the lower eyelid. The cranial part of the nasolacrimal duct was completely

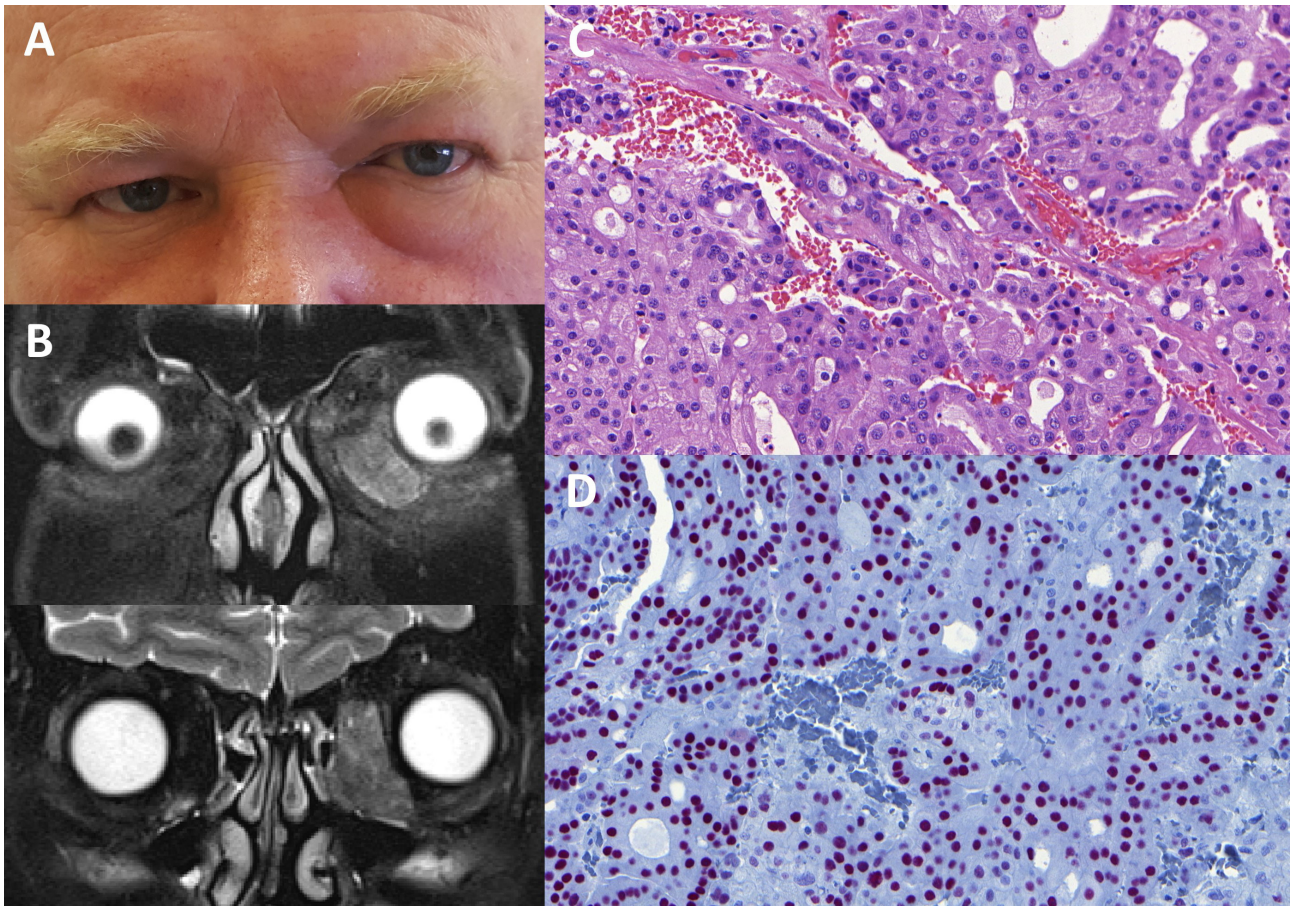

Fig. 1. (A) The clinical finding at the time of diagnosis, (B) T2-weighted coronal MRI scans (Image courtesy of Department of Imaging Methods, University Hospital and Faculty of Medicine in Pilsen, Charles University) and (C, D) histology and immunohistochemistry of ocular adnexal apocrine adenocarcinoma. (C) Hematoxylin-eosin stain: The tumor has a solid and tubular growth pattern and it is composed of polymorphic cells with abundant granular eosinophilic cytoplasm. (D) The cells reveal positive nuclear staining for androgen receptors (AR).

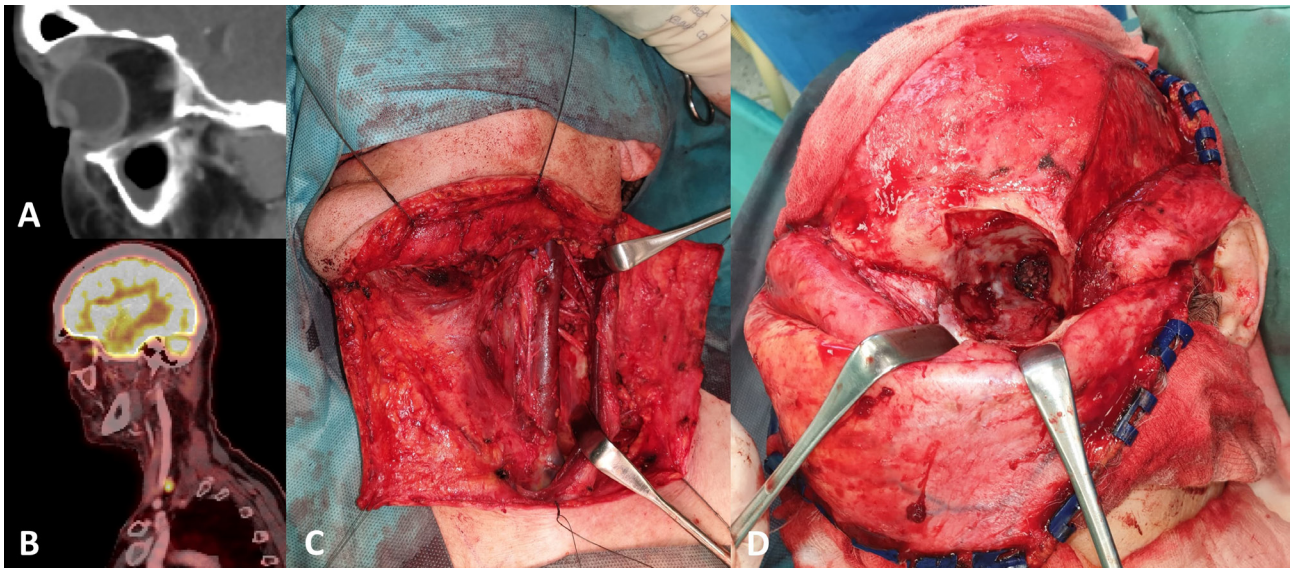

Fig. 2. (A) Sagittal CT and (B) 18F-FDG PET/CT scans showing the tumor recurrence and the lymph node suspicious for metastasis (Image courtesy of Department of Imaging Methods, University Hospital and Faculty of Medicine in Pilsen, Charles University). The state after the salvage surgery: (C) therapeutic neck dissection (ND L (I-V)) and (D) partial orbitectomy with the upper eyelid-sacrificing orbital exenteration. The temporalis muscle flap is prepared for orbital reconstruction.

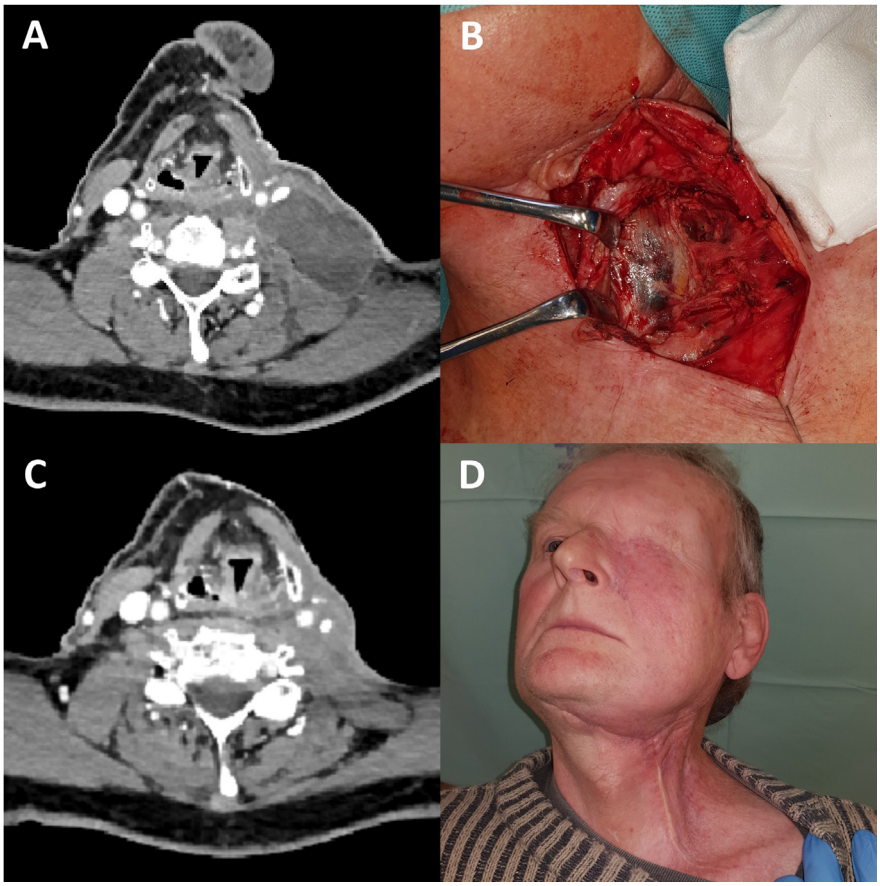

Fig. 3. (A) Axial CT scan showing the lymphocele in the left V neck level (Image courtesy of Department of Imaging Methods, University Hospital and Faculty of Medicine in Pilsen, Charles University) and (B) the intraoperative view of this lesion. (C) The axial CT scan (Image courtesy of Department of Imaging Methods, University Hospital and Faculty of Medicine in Pilsen, Charles University) and (D) clinical state after the lymphocele resolution.

Table 1. Cases of ocular adnexal apocrine adenocarcinoma reported in the literature.

Number of case	(Reference); Year of publication	Gender; Age of patient	Primary tumor localization	Lymph node metastases	Distant metastases	Treatment	Follow-up (mths)	Outcome
1	(5); 1951	F; 44	right lower eyelid, orbital invasion	No	Lung	3 non-radical excisions, adjuvant RT to the orbit, orbital exenteration	22	PD, DOD
2	(11); 1955	M; 55	right upper eyelid	No	No	2 non-radical excisions, orbital exenteration	7	NED
3	(12); 1970	M; 58	left upper eyelid	No	No	excision and radical re-excision	4	NED
4	(13); 1971	F; 59	right upper eyelid	ipsilateral preparotid LN	No	non radical excision, total eyelid resection, ipsilateral superficial parotidectomy, RND	84	NED
5	(14); 1984	M; 66	left lower eyelid	preauricular, submandibular LN on recurrent tumor	No	excision, orbital exenteration due to recurrence 1 year later, LN dissection	12	PD (intracranial involvement), DOD
6		M; 50	left lower eyelid, orbital invasion	preauricular, submandibular LN	No	RT to the orbit, orbital exenteration, subtotal maxillectomy, RND, adjuvant CHT	10	AWD (enlarged subclavicular LN)
7	(15); 1989	M; 66	right lower eyelid, orbital invasion	No	No	non-radical total eyelid resection, orbital exenteration	15	NED
8	(16); 1993	F; 85	left upper eyelid	No	No	excision	12	NED
9	(17); 1993	F; 36	right eyelid	cervical LN on recurrent tumor 3 mths later	No	excision	36	AWD (orbital recurrence)
10	(18); 1997	F; 47	lower eyelid	NA	NA	NA	NA	NA
11	(3); 2000	M; 34	left caruncle, orbital invasion	No	No	orbital exenteration	21	NED
12	(19); 2001	M; 80	right upper eyelid	No	No	excision	24	NED
13	(20); 2002	M; 57	left upper eyelid, orbital invasion	ipsilateral cervical LN	iliac bone, lumbar vertebra, sacrum	orbital exenteration, RND	6	locoregional NED
14	(21); 2005	NR	eyelid	NR	NR	NR	NR	NR
15	(22); 2006	M; 66	reccurent tumor on left lower eyelid (malignant transformation of papillary apocrine hidradenoma)	No	No	excision	108-180	NED
16		M; 53	right lower eyelid	No	No	excision	108-180	NED
17		M; 71	recurrent tumor on left lower eyelid	No	No	excision	108-180	NED
18	(23); 2008	M; 74	right lower eyelid, orbital invasion	No	No	excision and orbital exenteration, adjuvant RT to the orbit	22	NED

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Table 1. *(Continued)*

Number of case	(Reference); Year of publication	Gender; Age of patient	Primary tumor localization	Lymph node metastases	Distant metastases	Treatment	Follow-up (mths)	Outcome
19	(24); 2008	M; 57	right lower and upper eyelids, orbital invasion	No	No	partial excision and RT for residual tumor	16	AWD
20	(25); 2011	M; 62	left lower eyelid, orbital invasion	cervical LN	No	CHT (cisplatinum, adriamycin, cyclophosphamide)	72	AWD (PD, terminal cancer)
21	(26); 2012	M; 78	recurrent tumor on right lower eyelid, orbital invasion	ipsilateral submandibular LN	No	orbital exenteration, partial orbitectomy, SNB, adjuvant RT to the orbit and ipsilateral parotid and neck areas	30	NED
22	(27); 2012	M; 63	left lower eyelid	No	No	excision	36	NED
23	(28); 2013	M; 87	right upper and lower eyelids, orbital invasion	No	No	RT to the orbit	24	NED
24		F; 56	left lower eyelid	No	No	excision	12	NED
25	(29); 2016	F; 59	recurrent tumor on left lower eyelid	No	No	excision	6	NED
26		M; 81	left upper eyelid	NR	NR	excision	NR	NR
27	(4); 2017	M; 62	right lacrimal sac, orbital invasion	bilateral submandibular LN	No	frontal craniectomy, subtotal maxillectomy, partial rhinectomy, orbital exenteration, ND, adjuvant CHRT (cisplatin, paclitaxel)	several months	NED
28	(30); 2018	M; 91	left upper eyelid	No	No	excision	12	NED
29	(6); 2019	F; 58	left upper eyelid	No	No	excision	4	NED
30	(1); 2019	M; 60	left caruncle, orbital invasion	No	No	orbital exenteration, adjuvant RT to the orbit	4	NED
31	present case	M; 57	left lower eyelid, orbital invasion	ipsilateral lower jugular LN on recurrent tumor	No	2 non-radical excisions and proton beam RT, recurrence after 17 mths - upper eyelid-sacrificing orbital exenteration, partial orbitectomy, ND L (I-V)	15	NED

NR, not reported; NA, not available; LN, lymph nodes.
Gender: F, female; M, male.
Treatment: RT, radiotherapy; CHT, chemotherapy; CHRT, chemoradiotherapy; SNB, sentinel lymph node biopsy; ND, neck dissection; RND, radical neck dissection; ND L (), left-sided neck dissection (levels).
Outcome: PD, progressive disease; DOD, dead of disease; AWD, alive with disease; NED, no evidence of disease.

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Table 2. Cases of cervical lymphoceles associated with neck dissections reported in the literature.

Number of case	(Reference); Year of publication	Gender; Age of patient	Primary tumor	Type of neck dissection (classification corresponding with published data)	Side of lymphocele	Treatment of lymphocele	Outcome of treatment
1	(33); 1989	F; 78	SCC of the skin (mandibular angle)	RND	left	surgery	healed
2	(34); 2004	F; 24	papillary thyroid carcinoma	MRND	left	surgery	healed
3	(35); 2008	M; 58	esophageal SCC	MRND	left	OK-432 sclerotherapy	healed
4		F; 59	papillary thyroid carcinoma	MRND, CND	left	OK-432 sclerotherapy	healed
5		M; 65	hypopharyngeal SCC	RND	right	OK-432 sclerotherapy	healed
6		M; 70	SCC, unknown site	MRND	right	OK-432 sclerotherapy	healed
7	(7); 2011	NA	SCC, site NR	NR	NR (3 separate lymphoceles)	NR	NR
8		F; 37	thyroid cancer	NR	bilateral	sclerotherapy	"good results"
9	(36); 2012	F; 37	papillary thyroid carcinoma	NR	left	surgery	healed
10	(37); 2012	M; 65	papillary thyroid carcinoma	MRND (IJV and CN XI sparing)	left	surgery	healed
11		M; 62	papillary thyroid carcinoma	MRND (IJV, SCM and CN XI sparing)	left	surgery	chyle leak persistence, death of sepsis
12	(9); 2013	F; 31	papillary thyroid carcinoma	ND L (IV, VI)	left	surgery	healed
13	(8); 2017	F; 64	oral SCC	RND	left	conservative	no change, death of cancer
14	present case	M; 58	OAAA	ND L (I-V)	left	surgery	healed

NR, not reported.
Gender: F, female; M, male.
Primary tumor: SCC, squamous cell carcinoma; OAAA, ocular adnexal apocrine adenocarcinoma.
Type of neck dissection: RND, radical neck dissection; MRND, modified radical neck dissection; CND, central neck dissection; IJV, internal jugular vein; CN XI, spinal accessory nerve; SCM, sternocleidomastoid muscle; ND L (), left-sided neck dissection (levels).

involved and there was suspicion of the medial orbital wall invasion by the tumor. The eyeball was mildly displaced laterally. A biopsy was performed, resulting in a confirmatory diagnosis of OAAA (Fig. 1). Two non-radical eye-sparing excisions of the tumor within two months were performed. The result of histopathological examination showed intermediate to high grade apocrine adenocarcinoma arising from a gland of Moll with lymphatic and perineural invasion. Assessment of proliferative activity using Ki-67/MIB-1 immunostaining revealed a low level of MIB-1 index, namely 10%. Immunohistochemistry on formalin fixed paraffin embedded sections revealed positivity for cytokeratin CK-7 and AE1/3, epithelial membrane antigen (EMA) and androgen receptors (AR) (Fig. 1D). Immunostains were negative for CK-14 and CK-20. The detection of p63, SOX10, HER2/neu and S-100 proteins was also negative. The patient subsequently underwent adjuvant proton beam radiation therapy to the orbit and ipsilateral regional lymph nodes. Complete remission was attained but severe adverse effects of radiotherapy occurred. Radiation keratopathy with mycotic corneal ulcer and a secondary glaucoma led to total left eye blindness.

After 17 months of clinico-radiological follow-up, a locoregional recurrence of the tumor was discovered. A tumor mass (a size of 13x12x7 mm) under the left orbital roof in the extent of the frontal sinus base and a lymph node suspected of metastasis between IV and Vb neck levels were detected by the whole-body ^{18}F-FDG PET/CT. The histopathological analysis of samples obtained from the core cut biopsy confirmed the tumor recurrence. Salvage surgery was performed. The procedure included an en bloc partial orbitectomy with the resection of the orbital roof corresponding with the floor of the frontal sinus, accompanied with upper eyelid-sacrificing orbital

exenteration. This was performed from the anterior and coronal approach. The perioperative biopsy showed tumor-free superficial surgical margins. The orbital defect was reconstructed primarily with the temporalis muscle flap and cheek advancement flap. Therapeutic left-sided neck dissection of levels I-V was performed, sparing all non-lymphatic structures, with the dissection and ligation of the thoracic duct in order to achieve an oncological radicality (Fig. 2). The histopathological examination confirmed the recurrence of high-grade apocrine adenocarcinoma of identical histological structure with a bone, peri- and intraneural invasion. The MIB-1 index was 35%. None of the fusion oncogenes were proven by molecular genetic testing using FusionPlex Solid Tumor, CTL and Sarcoma Kits (ArcherDX, Boulder, CO, USA). A positive surgical margin at one point was found (R1 resection), but in this area, which was considered as the closest margin during the procedure, a further bone resection was added. Two lymph node metastases were detected in the specimen from the suspected region on PET/CT imaging (between IV and Vb neck levels). The postoperative course was uneventful except for the fact that the fluid from active drainage of the neck contained chyle. The chylous exudation ceased spontaneously on the third day after surgery and drains were removed. The wounds healed by the primary intention and the patient was without any further problems.

Three months later a cervical lymphocele (6x3x7 cm) localised in the V neck level on the left side was revealed on ^{18}F-FDG PET/CT. Only discreet swelling was clinically manifested in the relevant area of the neck. The lymphocele was first treated conservatively by repeated aspiration drainage and external compressions of the affected area for approximately one month. During the course of conservative management, the status was repeatedly monitored by a neck ultrasonography. This imaging showed a gradual mild progression of this lesion which became septated with minimal fluid content. Therefore, an extirpation of the lymphocele was performed. A lesion-feeding branch of the thoracic duct was identified and ligated. A small perforation detected in the main trunk was repaired by suture in combination with oxycellulose and the muscle flap from the dorsal edge of the sternocleidomastoid muscle. Histopathological examination revealed fibrofatty tissue with reparative changes, reactive fibroblastic proliferation and also necrosis, which supported the clinical diagnosis. The healing was uneventful (Fig. 3). During the one-year clinico-radiological follow-up *using t*he w*hole-body* ^{18}F-FDG PET/CT, the patient remained disease-free in terms of both tumor and lymphocele.

DISCUSSION

To date 27 cases of OAAA arising in eyelids, two cases originating from the lacrimal caruncle and one case from the lacrimal sac have been described in the literature (Table 1) (ref.[1,3-6,11-30]).

The male to female ratio of published cases was 2.8:1. The mean age of patients was 62.4 years (standard deviation 13.7 years, range 34-91 years, median=59.5 years). The lymph node metastases were detected in 32% (9/28) and distant metastases to the lung and bone in 7% (2/28) of cases. These data were available only in 30 and 28 patients, respectively, including the presented case. According to published data, primary surgery for OAA was performed in 90% (26/29) of cases. During the course of the disease, orbital exenteration was required in 41% (12/29) of patients and 21% (6/29) of patients had to undergo some type of neck dissection. The radiotherapy, especially as an adjuvant treatment, was indicated in 31% (9/29) of patients. These data were available only in 29 patients, including the presented case, which is the first reported case of salvage surgery including neck dissection due to multiple lymph node metastases. The surgery of OAAA performed in the previously irradiated field is reported only in two cases in the literature. Stout and Cooley described the salvage orbital exenteration without neck dissection following three non-radical tumor excisions and the adjuvant radiotherapy to the orbit[5]. The progressive course of this disease with the evidence of lung metastases resulted in patient's death. Ni et al. described the patient suffering from OAAA primarily treated with radiotherapy to the orbit[14]. The radiation exposure of 4000 roentgen in 29 days was used followed by the orbital exenteration and subtotal maxillectomy with the addition of neck dissection and the adjuvant chemotherapy in the further course of the disease[14]. But these surgical procedures cannot be called salvage surgery. This patient with the evidence of the enlarged subclavicular lymph node was lost to follow-up. Out of all published cases, 75% (21/28) of patients were disease free during the follow-up. But in most cases this follow-up involved an insufficient period for malignant disease: only in five patients did it exceed five years.

The thoracic or right lymphatic ducts have potential for numerous anatomical anomalies of their drainage to a cervical venous system in the lower neck. Multiple thoracic duct terminations in one or more veins are described in 10-45% of cases[9,31]. In addition, the walls of these lymphatic structures are extremely thin, fragile and difficult to identify in fatty tissue. The anatomy of this region may be further complicated by the presence of aberrant thoracic duct branch terminating on the right side. A real right-sided duct occurs with an incidence rate of about 4% (ref.[32]). For these reasons, peroperative injuries to the thoracic duct may be easily overlooked. In the presented case, a lymph leak occurred probably because of these facts despite the meticulous identification, dissection and ligation of the duct to achieve an oncological radicality in IV and Vb neck levels.

For prevention of thoracic duct injury, a thorough knowledge of the normal and aberrant anatomy of the lymphatic system and painstaking dissection without undue traction of these structures is necessary. At the end of the operation, the surgical field should be systematically examined for chyle leakage, especially if thoracic duct breach is suspected. This requires lowering the patient's

head and performing the Valsalva maneuver[8,31]. If any breach with lymph leak is detected, a functional repair is enabled by suturing the perforation. It may be reinforced with local hemostatic agents (e.g. gelatin, oxycellulose, fibrin glue) alone, or in combination with adjacent fascia, local muscle flap or autologous fat graft[8]. If the suture is not feasible, a ligation of the duct may be another option. An active drain has to be positioned away from the site of lymphatic structures closure to avoid the direct aspiration of the lymph from this area[9]. The postoperative surveillance should include systematic observation of the drainage liquid quality and examination of the surgical area for any signs suggestive of lymph leakage (red and sensitive skin with induration caused by intense inflammatory reaction to lymph) (ref.[9]). An accumulation of lymph in the wound may cause fluid and electrolyte shifts, protein loss, as well as compromising local healing[8]. The postoperative lymphorrhea may sometimes resolve spontaneously, leads to chylous fistula, chylothorax or on rare occasions to later development of lymphocele. So far, only 16 cases of lymphoceles following neck dissections have been published (Table 2) (ref.[7-9,33-37]).

Our case was successfully treated surgically with no signs of recurrence during the follow-up. So far, only six cases of cervical lymphoceles following neck dissections managed surgically with the success rate of 83% (5/6) have been reported in the literature[9,33,34,36,37]. The lymphocele should be suspected when a patient develops a cervical mass in the neck dissection area. A fluctuation with overlying edema and erythema may be found out on physical examination[31]. Imaging methods such as ultrasound, CT and MRI are not contributive to diagnosis but may be helpful for determination the exact localization and extension of the lesion, its character (cystic or solid) and also relations to the surrounding structures[8]. Establishing the diagnosis of postoperative cervical lymphocele is based upon identifying the cyst fluid is chylous in origin. Therefore, an aspiration of the liquid is required for evaluation of its appearance and performing biochemical analysis. The pathognomonic hallmark is a creamy or milky fluid with increased cholesterol and triglyceride levels which are higher than in the blood[8,9].

Conservative management performed approximately for up to one month should be the first-line approach in the treatment of postoperative cervical lymphocele in cases of low-level lymph leakage (<100 mL/day) (ref.[9,38]). Diet modification has been shown to reduce the production of chyle. Enteric regimes include low-fat diet with medium-chain triglycerides which are directly absorbed by the portal venous system, bypassing the lymphatics[9,38]. Total parenteral nutrition eliminates any gastrointestinal absorption of fat and may be considered as an alternative. Somatostatin analogs (octreotide) are also used as a concomitant therapy for reduction of intestinal fat absorption and production of lymph[32,38]. Dietary modification was not applied in the presented case in order not to impair the nutritional status of the oncological patient. Other conservative measures comprise an ensuring elevation of the head, repeated aspiration drainage and applying external compression of the affected area[9,38]. In patients refractory to conservative management, more invasive procedures like the transcutaneously instillation of sclerosing agents such as OK-432, doxycycline, bleomycine, talc or povidone-iodine can be considered[8,9,31].

Surgery is indicated in cases of lymph leakage exceeding 600 mL/day or after failure of the conservative approach[9]. Revision surgery should be performed before local (a cutaneous involvement with wound healing disorders) or systemic complications arise (due to loss of electrolytes, liquids and proteins). Late indication for surgery is associated with elevated risk of injury to adjacent neuro-vascular structures because the preparation within the area of newly-formed granulation and fibrous tissue is challenging[9,31]. A preoperative administration of milk or cream may facilitate localization of the leak[9,31]. Surgical therapy includes extirpation of the lesion and above-mentioned procedures aimed at closing the thoracic duct or recreating the lymphovenous connection[38]. In patients who do not respond to this treatment, in cases of leakage exceeding 1000 mL/day and/or in lesions associated with chylothorax, ligature of the thoracic duct performed through an open thoracotomy or using less traumatic thoracoscopic approaches is required[9]. Other treatment options for lymphatic–chylous leaks such as a therapeutic lymphography, thoracic duct embolization or microsurgical derivative multiple lymphatic–venous anastomoses may be considered[38].

CONCLUSION

OAAA is a locally aggressive tumor with potential to local or distant metastatic spread. Whole-body staging, regular clinico-radiological follow-up and stage-dependent therapy with the surgery as the first-choice treatment is required. A cervical lymphocele as a complication of especially left-sided neck dissection is managed with a conservative or surgical therapy according to the level of lymph leakage, extent and localization of lesions, presence of local or systemic disorders and the period from primary surgery.

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