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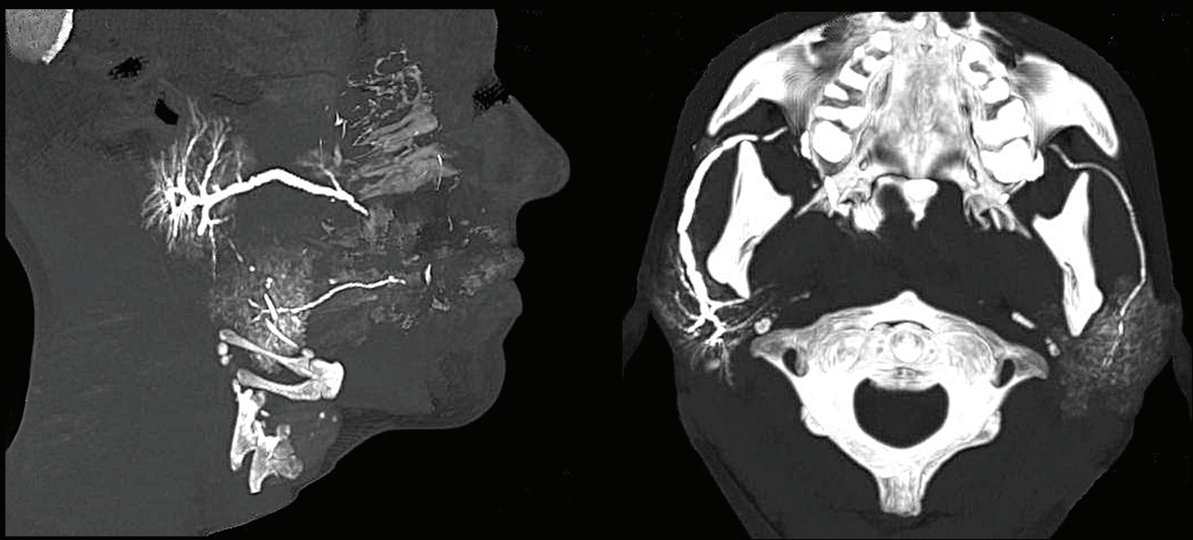

Technology

Comprehensive Reconstruction of Mandibular Defects With Free Fibula Flaps and Endosseous Implants

New Technique

Minimally Invasive Techniques for Management of Salivary Gland Pathology

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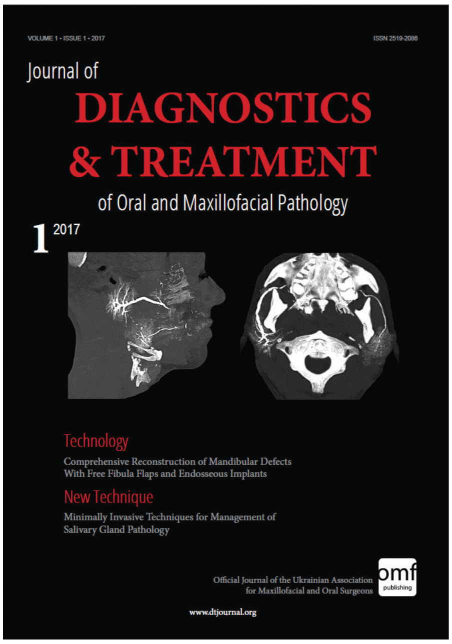

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New Horizons

Prof Oleksii O. Tymofieiev

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You are holding in your hands or reading online the first issue of a completely new journal. Treatment of any oral or maxillofacial pathology is not performed without modern diagnostics. CBCT, MDCT, MRI, ultrasound, 3D printing, 3D planning of surgeries and other technologies have become a part of the everyday practice of general practice dentists, periodontists, oral and maxillofacial surgeons, oncologists, and radiologists. Before us are opened up completely new horizons that we need to explore with the maximum benefit for our patients and ourselves.

The President's decision on the adoption of a year 2016 an a year of the English language in Ukraine is dictated by the requirements of the modernity. English opens the door to the whole world of science, conferences, places dating, it gives opportunity to demonstrate our results around the world, to develop joint projects and to increase the profitability of your practice. Therefore we continue that initiative in 2017 with *the Journal of Diagnostics and Treatment of Oral and Maxillofacial Pathology*, which became the first publication dedicated to maxillofacial pathology in post-Soviet states. The editorial board of the journal includes leading experts from all over the world. With this approach and cooperation with the world, we will all win.

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Comprehensive Reconstruction of Mandibular Defects With Free Fibula Flaps and Endosseous Implants

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ABSTRACT

Purpose.
The goal of this paper is to review the use of fibula free flaps in reconstruction of various mandibular defects, as well as illustrate that placement of dental implants into free fibula flaps is a viable option ensuring a superior functional outcome.
Patients and Methods.
Nine of patients with mandibular fibula free flap reconstruction who underwent dental implant placement were included in this study to demonstrate the versatility of this reconstructive technique.
Results.
In all nine patients, fibula flaps provided adequate bone stock for implant placement. All 30 implants were placed in bicortical fashion and none had issues with primary stability at the time of placement.
Conclusion.
Fibula free flap reconstruction is the treatment of choice for patients with various disease processes resulting in significant mandibular defects and can ultimately be restored with fixed dental prostheses.

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Introduction

The advent of microvascular surgery facilitated the development of novel techniques that provided superior esthetic and functional reconstruction of large maxillofacial defects. One of the major advantages of microvascular free tissue transfer is that it contains its own vascular pedicle, thus allowing for improved healing in wounds compromised by radiation and chronic infection [1]. The goal of any reconstructive surgery is to restore natural form and function. Mandibular defects resulting from ablative surgery due to malignant or benign pathology, osteonecrosis, or trauma often result in significant functional and esthetic compromise. The free fibula flap is considered the gold standard for mandibular reconstruction due to its versatility, outcome predictability, and suitability for dental implant placement [2]. Dental rehabilitation plays a pivotal role in improving patient's quality of life, since edentulism has been shown to result in significant psychological morbidity to patients [3].

The goal of this paper is to review the use of fibula free flaps in reconstruction of various mandibular defects, as well as illustrate that placement of dental implants into free fibula flaps is a viable option ensuring a superior functional outcome.

Patients and Methods

Retrospective review of patient charts treated from 2005-2015 was completed. Total of 116 patients with mandibular fibula free flap reconstruction were identified. Nine of these patients who underwent dental implant placement were included in this study to demonstrate the versatility of this reconstructive technique. Exclusion criteria were lack of dental implant placement, lack of adequate follow up, or incomplete and lacking records. Although dental implant placement can be recommended to everyone, the cost of dental implants is often prohibitive for a majority of our patients. Unfortunately, many medical and dental insurance companies do not offer 100% coverage for dental implant rehabilitation, even in cases of malignant disease. The selected patient group included 4 females, 5 males, age ranged from 20 to 72 years old with a mean

TABLE 1. Patient Data.

Patient Age, Gender	Diagnosis	Jewer Classification	Fibula Class	Timing of Implant Placement (months) after FFF
20M	GSW[a]	LCL	Class III	41
33M	Ameloblastoma	L	Class I	33
42F	Ameloblastoma	L	Class I	3
45M	Ameloblastoma	CL	Class II	9
49M	SCCA[b], post op XRT[c]	CL	Class II	Immediate at time of FFF[d] surgery
52M	Glandular odontogenic tumor	LCL	Class II	9
52M	SCCA, post op XRT	L	Class I	7
55F	SCCA, no XRT	C	Class II	8
72F	Bisphosphonate related osteonecrosis	L	Class I	12

[a] *Gunshot wound*
[b] *Squamous Cell Carcinoma*
[c] *Radiotherapy*
[d] *Fibula free flap*

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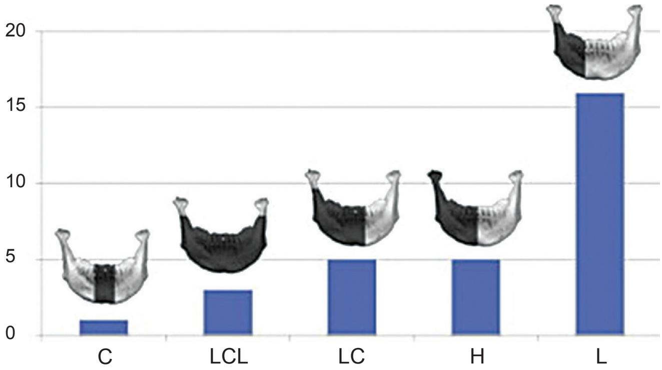


FIGURE 1. Location of mandibular defects according to the Jewer classification (C = central defect; LCL = bilateral defect; LC = central and lateral defect; H = hemimandibulectomy; L = lateral defect) [4].

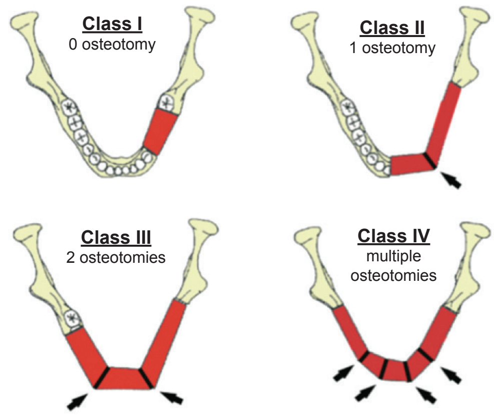


FIGURE 2. Fibula classes according to number of osteotomies [5].

age of 42. Follow up ranged from 1 year 5 months to 6 years 9 months, and median was approximately 4 years. The diagnoses included squamous cell carcinoma, ameloblastoma, glandular odontogenic tumor, and self-inflicted gunshot wound. The resulting mandibular defects and reconstruction were classified based on Jewer and fibula osteotomy classifications (Figs 1, 2). Detailed patient information, including timing of implant placement, is presented in Table 1.

Results

In all nine patients reviewed, single barrel free fibula flaps were utilized. No intra-operative or immediate post-operative complications were noted and no flap failure occurred. Two out of nine patients developed intra-oral dehiscences that healed uneventfully and required no additional operating room interventions. In all nine patients, fibula flaps provided adequate bone stock for implant placement. All 30 implants were placed in bicortical fashion and none had issues with primary stability at the time of placement. No implants required removal to date. Additional surgical procedures, such as vestibuloplasty, keratinized mucosa grafting, and flap debulking were completed in four out of nine patients (Table 2). One patient with diagnosis of medication related osteonecrosis of the jaw did not complete dental rehabilitation due to issues with recurrent infection, need for fibula hardware removal and delayed healing complicated with extra-oral fistula. Three patients had implant supported overdentures fabricated and reported satisfaction with the results. Two patients underwent fixed partial denture fabrication and were also happy with the functional and esthetic results. The remaining four patients were awaiting final prosthesis delivery at the time of study.

The most common complication encountered in our group was peri-implant tissue inflammation and infection. Peri-implantitis resolved with granulation tissue removal and meticulous oral hygiene without causing peri-implant bone loss in two patients. One of the patients developed bone loss adjacent to the terminal implant that at the time of evaluation did not appear to compromise stability of the implant. Although eventual loss of implant with compromised bone support is certainly possible, conservative measures to address peri-implantitis were undertaken to prolong the life-span of the existing prosthesis.

Discussion

Free fibula flap for reconstruction of mandibular defects was first introduced by Hidalgo in 1989 [6]. Numerous studies since then have demonstrated the effectiveness and predictability of free fibula flaps for mandibular reconstruction (Figs 3-6).

TABLE 2. Patient outcomes.

Patient Age, Gender and Diagnosis	Jewer Classification	Number of implants in fibula	Additional Implant Surgery	Prosthesis	Complications
20M, GSW	LCL	2	Flap debulking	Overdenture	None
33M, amelobasltoma	L	3	None	Fixed partial denture	Periimplantitis requiring granulation tissue debridement, oral hygiene
42F, ameloblastoma	L	3	None	In process of being fabricated [2]	None
45M, ameloblastoma	CL	4	Vestibuloplasty	In process of being fabricated	None
49M, SCCA	CL	3	None	In process of being fabricated [2]	None
52M Glandular odontogenic tumor	LCL	4	Vestibuloplasty	Overdenture	None
52M, SCCA	L	3	None	Fixed partial denture	Peri-implantitis, radiographic bone loss distal implant
55F, SCCA	C	4	Vestibuloplasty, flap debulking, palatal mucosa graft	Overdentures	Peri-implantitis requiring granulation tissue debridement, antibiotic treatment, oral hygiene
72F, BRONJ	L	4	None	None	Hardware infection requiring removal, extraoral fistula

In order to facilitate comprehensive orofacial rehabilitation, a flap has to satisfy several requirements. First, it must provide sufficient bone length to ensure adequate repair of the continuity defect. Up to 26cm of fibula can be harvested, which allows for reconstruction of mandibular defects spanning almost the entire length of mandible [7]. The long segment of bone can be osteotomized in multiple locations, thus allowing for esthetic reconstruction of patient's anatomy. Complex defects, requiring more than 2 osteotomies can be reconstructed with

FIGURE 3. Fibula harvesting.

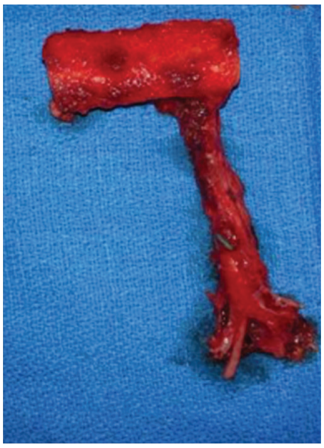


FIGURE 4. Fibula segment with pedicle.

computer assisted virtual surgical planning [7] (Fig 7A, B). Second, adequate bone and tissue stock has to be available for endosseous implant placement and provide satisfactory long-term implant survival rates. The dense cortical bone of the fibula, and its 1-3cm thickness, provide ample primary implant stability by allowing bicortical engagement of conventional 12-14mm implants [7]. Skin paddle size can reach up to 32cm x 14cm thus allowing reconstruction of significant intraoral and extraoral soft tissue defects as well [8]. Next, for a successful microvascular anastomosis, donor and recipient vessels must be of similar caliber. The flap is based on the peroneal artery, 1.5-2.5mm diameter, and two venae comitantes, 2-4mm in diameter which is similar to the diameter of commonly used recipient vessels in the head and neck [7-9]. In addition, donor site morbidity is minimal with anticipated return to normal ambulation in 4 weeks after surgery [8].

Several specific considerations must be taken into account when planning implant placement into fibula free flap. Optimal results can be achieved only when orofacial reconstruction is approached with the end result in mind. Thus input from the restorative dentist responsible for fabrication of the final prosthesis is essential in order to avoid unfavorable outcomes.

Optimal timing of implant placement has yet to be agreed upon to date. Eight out of nine patients included in this study underwent delayed implant placement with mean delay of 23 months, ranging from 3 to 41 months. In one case

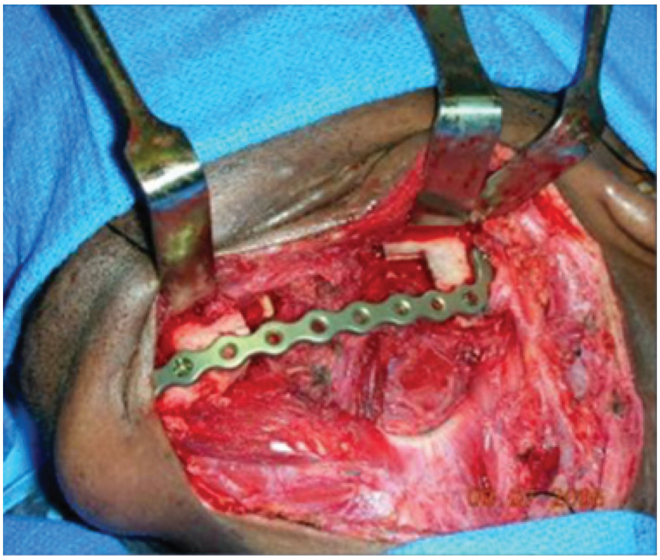


FIGURE 5. Recipient site.

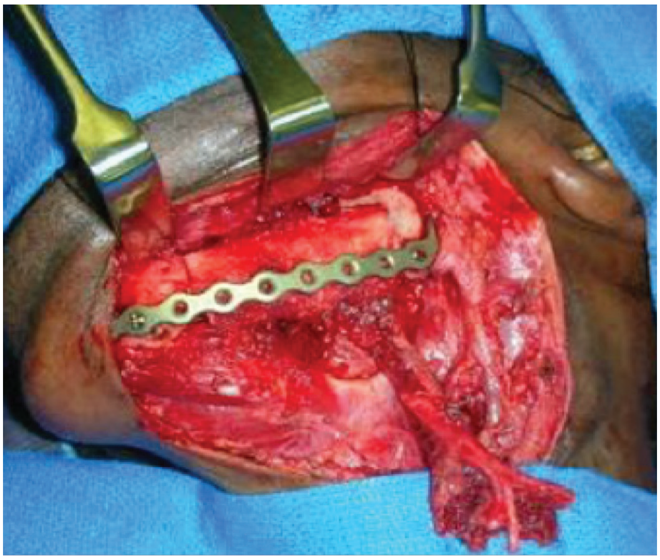
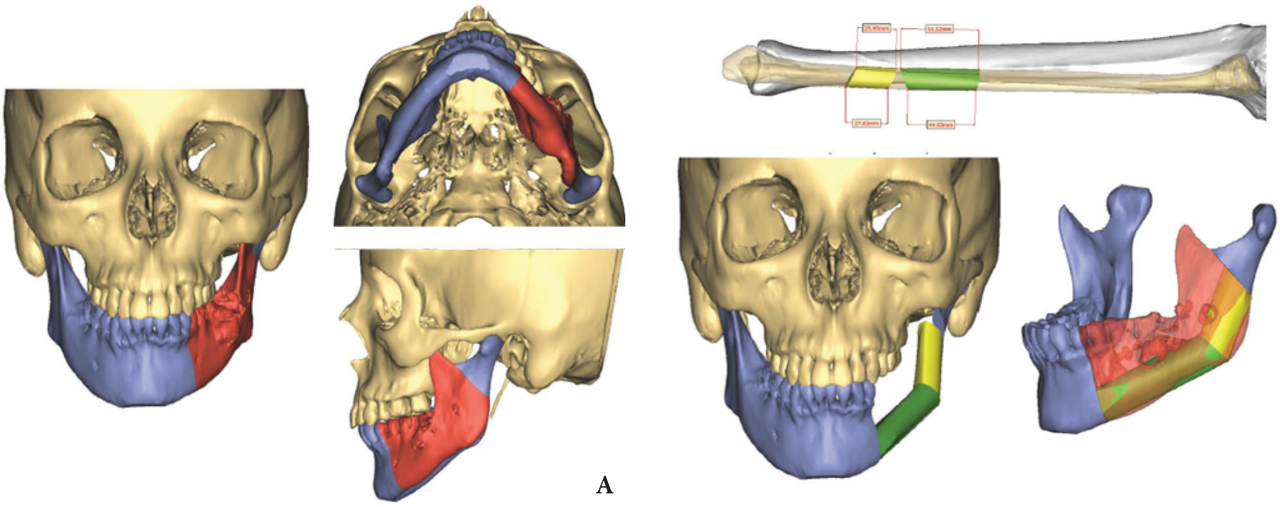


FIGURE 6. Inset fibula with vascular pedicle.



A

B

FIGURE 7. Virtually assisted surgical planning demonstrating resection margins (**A**). Virtually assisted surgical planning demonstrating free fibula flap reconstruction (**B**).

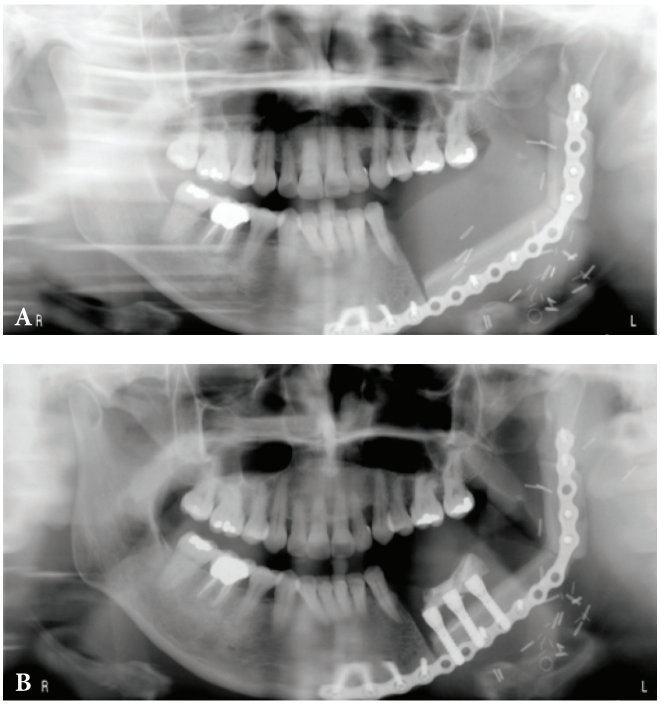


FIGURE 8. A – Immediate post-operative panoramic radiograph of free fibula flap reconstruction; **B** – 3 months post-reconstruction panoramic radiograph after dental implant placement and flap debulking.

immediate implant placement was performed at the time of reconstruction. Prolonged interval to implant placement was primarily a result of socioeconomic or personal issues rather than medical complications. The timing of implant placement did not appear to have an effect on the overall outcome in our patients. Some authors recommend at least a 6-month period of healing prior to implant surgery [10], while others advocate immediate implant insertion into fibula at the time of reconstructive surgery [11]. Delayed implant placement allows for sufficient bone remodeling and soft tissue healing thus allowing more precise implant placement (Fig 8A, B) [12]. In cases of malignant disease, a 6-12 month waiting period also allows monitoring for early disease recurrence, presence of which would discourage implant surgery due to poor overall prognosis. Disadvantages of this treatment option include need for additional surgery and prolonged period of time with suboptimal function due to delayed prosthetic rehabilitation. Immediate implant placement eliminates the need for additional surgery and its associated morbidity. However, it increases the risk of future implant disuse due to difficulty predicting final implant position once bone remodeling and soft tissue healing reach its final stages [13]. Since cone beam CTs became widely used in dental practices, computer assisted surgical planning has also gained wide acceptance and has been shown to provide more predictable results with optimal final restorations [14]. Nevertheless, desired implant placement may be impossible due to position of internal fixation screws necessary to secure the flap to native mandible. With delayed implant placement, simultaneous removal of fixation hardware is also possible, thus eliminating the risk of future hardware infection development. A total of 12 to 16 weeks are recommended for healing and osseointegration of implants prior to uncovering [8].

Discrepancy in the bone height between native mandible and fibula was implicated in creation of unfavorable crown to fixture ratio that may increase the risk of implant failure [10]. Several strategies were devised to circumvent this problem. Positioning of the fibula superior to the inferior border of the mandible improves the crown to implant ratio, but may result in evident facial deformity [9]. Placement of the reconstruction plate along the inferior border is often used to correct this issue [9]. Double-barrel fibula and vertical distraction osteogenesis are more technically challenging and demanding options available for fibula height correction [8]. From a restorative stand point, a milled bar framework may be used to help correct the height discrepancy, as well as facilitate distribution of masticatory forces (Fig 9) [8].

All of our patients were reconstructed with fibulas that were aligned with inferior border of the mandible and 8 out of 9 were satisfactorily restored with dental prosthesis, or are in the process of being restored, without the above-mentioned corrections. One patient with a history of medication related osteonecrosis of the jaw (MRONJ) did not have a prosthesis fabricated to date due to delay in healing that was complicated by hardware infection.

One of the disadvantages from the standpoint of dental rehabilitation is the excessive mobility and thickness of the fibula skin paddle. Reconstruction of intraoral soft tissue defects often results in vestibular obliteration and requires vestibuloplasty with tissue debulking to facilitate prosthesis fabrication and use [8]. Lack of attached mucosa adjacent to the implant abutments increases risk of irritation and inflammation of the hypermobile fibula skin paddle [10].

One of our patients required vestibuloplasty, flap debulking, and keratinized tissue graft. Two others required vestibuloplasty and one more patient required flap debulking. In total 4 out of 9 patients (i.e 44%) required additional procedures, which is consistent with reports in the literature [6].

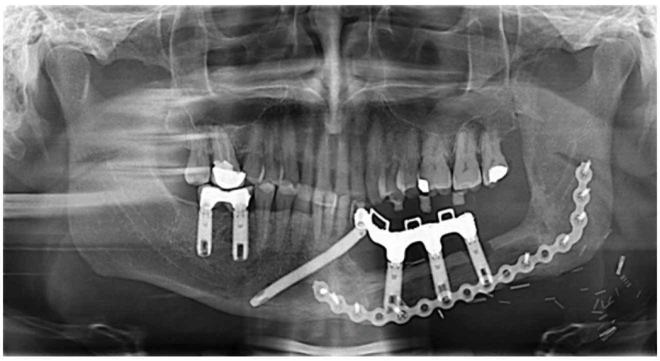


FIGURE 9. Milled bar framework for a fixed partial denture.

FIGURE 10. Implant supported prosthesis.

Conclusions

Implants placed in fibulas have high success rates comparable to native mandible, >95% [15]. Immediate or late implant failure is uncommon, and in our cohort, no implants were lost. Two out of nine patients developed peri-implantitis that had eventually resolved, although required invasive intervention. Peri-implant tissue inflammation is one of the most common complications reported in the literature [16]. Chronic peri-implantitis may result in peri-implant bone loss as was observed in one of our patients.

As illustrated by our selection of cases, fibula free flap reconstruction is the treatment of choice for patients with various disease processes resulting in significant mandibular defects and can ultimately be restored with fixed dental prostheses (Fig 10).

Since comprehensive orofacial rehabilitation is a multi-step complex process involving different healthcare specialists and multiple surgeries, patients' prognoses, interest and enthusiasm needs to be assessed. Multiple studies have shown that patient's quality of life is dramatically improved when these surgical techniques are used to restore patient's form and function [17].

References

1. Markiewicz MR, Bell RB, Bui TG, Dierks EJ, Ruiz R, Gelesko S, Pirgousis P, Fernandes R. Survival of microvascular free flaps in mandibular reconstruction: a systematic review and meta-analysis. Microsurgery 2015;35:576–87.
2. Boyd JB. The iliac crest and the radial forearm flap in vascularized oromandibular reconstruction. Am J Surg 1990;159(3):301–8.
3. Rogers SN, McNally D, Mahmoud M, et al. Psychologic response of the edentulous patient after primary surgery for oral cancer: a cross-sectional study. J Prostate Dent 1999;82:c317.
4. Adrian Ooi, Jiajun Feng, Hiang Khoon Tan, Yee Siang Ong. Primary treatment of mandibular ameloblastoma with segmental resection and free fibula reconstruction: achieving satisfactory outcomes with low implant-prosthetic rehabilitation uptake. Journal of Plastic, Reconstructive & Aesthetic Surgery 2014;67(4):498–505.
5. Iizuka T, Häfliger J, Seto I, Rahal A, Mericske-Stern R, Smolka K. Oral rehabilitation after mandibular reconstruction using an osteocutaneous fibula free flap with endosseous implants. Clinical Oral Implants Research 2005;16:69–79.
6. Hidalgo DA. Fibula free flap: a new method of mandible reconstruction. Plast Reconstr Surg 1989;84:71–9.
7. Wei F-C, Mardini S. Flaps and reconstructive surgery. Philadelphia: Saunders/Elsevier; 2009. 624 p.
8. Kademani D, Tiwana P. Atlas of oral and maxillofacial surgery. Saunders; 2015. 1520 p.
9. Su T, Fernandes R. Microvascular reconstruction of the mandible: an argument for the fibula osteocutaneous free flap. Revista Española de Cirugía Oral y Maxilofacial 2014;36(2):1–8.
10. Anne-Gaëlle B, et al. Dental implant placement after mandibular reconstruction by microvascular free fibula flap: current knowledge and remaining questions Oral Oncology 2011;47(12):1099–104.
11. Sclaroff A, Haugey B, Gay W, Paniello R. Immediate mandibular reconstruction and placement of dental implants at the time of ablative surgery. Oral Surg Oral Med Oral Pathol 1994;78(6):711–7.
12. Chiapasco M, Abati S, Ramundo G, Rossi A, Romeo E, Vogel G. Behaviour of implants in bone grafts or free flaps after tumor resection. Clin Oral Implants Res 2000;11:66–75.
13. Disa J, Hidalgo D, Cordeiro P, Winter R, Thaler H. Evaluation of bone height in osseous free flap mandible reconstruction: an indirect measure of bone mass. Plast Reconstr Surg 1999;103(5):1371–7.
14. Okay DJ, Buchbinder D, Urken M, Jacobson A, Lazarus C, Persky M. Computer-assisted implant rehabilitation of maxillomandibular defects reconstructed with vascularized bone free flaps. JAMA Otolaryngol Head Neck Surg 2013;139(4):371–81.
15. Kramer F-J, Dempf R, Bremer B. Efficacy of dental implants placed into fibula-free flaps for orofacial reconstruction. Clinical Oral Implants Research 2005;16:80–8.
16. Blake F, Bubenheim M, Heiland M, Pohlenz P, Schmelzle R, Gbara A Retrospective assessment of the peri-implant mucosa of implants inserted in reanastomosed or free bone grafts from the fibula or iliac crest. Int J Oral Maxillofac Implants 2008;23(6):1102–8.
17. Vu DD, Schmidt BL. Quality of life evaluation for patients receiving vascularised versus nonvascularized bone graft reconstruction of segmental mandibular defects. J Oral Maxillofac Surg 2008;66(9):1856–63.

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Періімплантит

РЕЗЮМЕ

Мета. Розглянути використання ВМК при реконструкції різних дефектів нижньої щелепи, а також проілюструвати, як розміщення зубних імплантатів в ВМК є життєздатним варіантом забезпечення чудового функціонального результату.

Пацієнти та методи. Дев'ять пацієнтів з реконструкцією нижньої щелепи ВМК, в які встановили імплантати були включені в це дослідження, щоб продемонструвати універсальність цього методу реконструкції.

Результати. Найбільш частим ускладненням в нашій групі був періімплантит та інфікування.

Висновки. Реконструкція ВМК є методом вибору при лікуванні хворих з різними патологічними процесами, в результаті яких утворюються значні дефекти нижньої щелепи і в кінцевому рахунку можуть бути відновлені фіксованими зубними протезами.

Комплексная реконструкция дефектов нижней челюсти свободными малоберцовыми лоскутами и внутрикостными имплантатами

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Циторедукция лоскута
Периимплантит

РЕЗЮМЕ

Цель. Рассмотреть использование СМЛ в реконструкции различных дефектов нижней челюсти, а так же проиллюстрировать как размещение зубных имплантатов в свободных малоберцовых лоскутах является жизнеспособным вариантом обеспечения превосходного функционального результата.

Пациенты и методы. Девять пациентов с реконструкцией нижней челюсти СМЛ, в которые установили имплантаты были включены в это исследование, чтобы продемонстрировать универсальность этого метода реконструкции.

Результаты. Наиболее частым осложнением в нашей группе был периимплантит и инфицирование.

Выводы. Реконструкция СМЛ является методом выбора при лечении больных с различными патологическими процессами, в результате которых образуются значительные дефекты нижней челюсти и в конечном счете могут быть восстановлены фиксированными зубными протезами.

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Minimally Invasive Techniques for Management of Salivary Gland Pathology

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ABOUT ARTICLE

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ABSTRACT

Purpose.
The goal of this article is to describe the technique used at our institution and highlight potential pitfalls during sialendoscopy.
Discussion.
Indication for sialendoscopy, sialendoscopy technique are discussed.
Results.
Despite the high reported success rates with sialendoscopy, the procedure is deemed to be technically challenging and correlation between success rates and operator experience has been shown.
Conclusion.
Sialendoscopy is a minimally invasive technique that is gradually replacing the classic open surgical approach to the treatment of obstructive salivary gland diseases as the standard of care.

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Introduction

Obstructive salivary gland disease is common and regardless of etiology historically it has been treated with gland excision. Although removal of the gland results in complete symptom resolution, the surgery itself carries inherent risks. Depending on which major salivary gland is involved the risks range from minor cosmetic defects to major complications such as facial nerve damage or airway compromise. These considerations encouraged the development of minimally invasive approaches in order to avoid surgical gland resection.

Sialendoscopy takes advantage of the naturally present salivary gland duct orifice thus obviating the need for surgical incisions. Small endoscopes may be utilized to examine and assess the pathologic process within the ductal system, i.e. diagnostic sialendoscopy, as well as intervene as indicated, i.e. therapeutic sialendoscopy. Use of endoscopes to remove a sialolith was first described by Katz in 1991 (Katz, 1991) [1]. Following the introduction of this technique numerous authors have reported success utilizing sialendoscopy for a wide range of indications. In addition to removal of sialoliths, it has been used for retrieval of foreign objects (Nahlieli, Nakar, Nazarian, & Turner, 2006) [2], breaking up of adhesions (Ardekian, Shamir, Trabelsi, & Peled, 2010) [3], treatment of juvenile recurrent parotitis (Singh & Gupta, 2014) [4], and radio-iodine induced and autoimmune sialadenitis (Atienza & Lopez-Cedrun, 2015) [5].

The purpose of this article is to describe the technique used at our institution and highlight potential pitfalls during sialendoscopy.

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Discussion

Sialendoscopy is indicated in any situation where there is a physical obstruction to the salivary outflow that eventually results in inflammatory or infectious processes within the gland parenchyma. Therefore, patients most commonly present with complaints of painful facial or submandibular swelling that may also be associated with overlying skin erythema and systemic fever, especially during or after meals. Complete assessment consists of history, physical exam, including an attempt to "milk" a salivary gland, and imaging. The imaging modalities used include plain radiographs, ultrasound, non-contrast CT (Fig 1), and MRI (Hitti, Salloum, & Mufarrij, 2014) [6]. Plain radiographs and US are adequate to assess a minimally symptomatic patient with no suspicion of retrotropharyngeal or parapharyngeal abscess formation. Non-contrast CT allows visualization of deep neck spaces as well as provides sufficient gland tissue detail, has 100% sialolith detection sensitivity (Hitti et al., 2014) [6] and can be quickly obtained, thus making it a good choice of imaging in

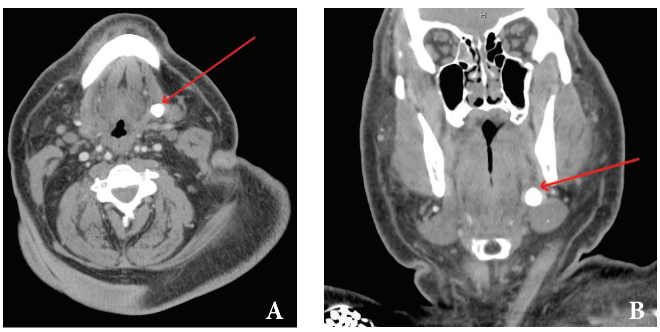


FIGURE 1. Non-contrast CT images (**A**, **B**) with *arrows* pointing to sialolith in the left submandibular gland duct (Wharton's duct).

patients who are suspected to have more extensive disease.

Several different types of etiologies lead to the same obstructive symptomology but may require different therapeutic approaches. Intraluminal obstruction of the salivary gland ductal system most frequently is due to sialolithiasis (Marchal et al., 2001) [7] with submandibular gland being affected 80% of the time and parotid gland involved in 20% (Zenk et al., 2012) [8]. Small size of the sialolith, oval shape, mobility and distal location within the duct were determined to be positive prognostic factors for simple removal (Luers, Grosheva, Stenner, & Beutner, 2011) [9]. Specifically, it has been shown that simple removal of 3mm stones from parotid gland and 4mm stones from submandibular gland can be easily achieved with wire baskets, mini graspers, or forceps (Marchal et al., 2001) [7]. Where a stones of larger diameter, up to 8-9mm, require fragmentation by either direct instrumentation or lithotripsy (Zenk et al., 2012) [8]. Ductal mucous plugs and less commonly foreign bodies such as tea leaves and hair were reported (Nahlieli et al., 2006) [2]. Ductal strictures have also been implicated as a cause of recurrent sialadenitis and their incidence was thought to be underestimated (Ardekian et al., 2010) [3]. Etiology of ductal strictures is not well defined, although it is assumed to be the result of epithelial healing after traumatic, infectious, or recurrent inflammatory insults (Ardekian et al., 2010) [3].

Sialendoscopy Technique

After appropriate patient selection and review of relevant pre-operative images, parotid and submandibular sialendoscopy can be performed either under general endotracheal anesthesia (GETA), intravenous sedation, or local anesthesia. It has been shown that patients with no co-morbidities and small sialoliths tolerate sialendoscopy under local anesthesia well (Luers, Stenner, Schinke, Helmstaedter, & Beutner, 2012) [10]. The authors prefer nasal GETA due to improved access, patient cooperation, and risk of airway compromise if inadvertent fluid extravasation into the floor of mouth during submandibular procedures.

The basic set up includes the following armamentarium:

- IV tubing and extension with 3-way stopcock
- 60cc syringe
- 500cc bag of 0.9% NaCl solution
- Endoscopy tower and monitor
- Salivary probes and dilators (Figure 2)
- COOK® Kolenda Introducer Set (Figure 3)
- Karl Storz® ALL-IN-ONE Sialendoscopes, ERLANGEN model (Figure 4):
    - All endoscopes are 0°angle
    - 0.8mm diameter for diagnostic sialendoscopy
    - 1.1mm and 1.6mm diameter for therapeutic sialendoscopy
- Karl Storz® Foreign Body Forceps
- Disposable Items:
    - Karl Storz® Stone Extractors (wire baskets)
        - 0.4mm or 0.6mm diameters
    - Karl Storz® Balloon Catheter
        - 0.7mm diameter
    - COOK® NGage Stone Extractor

The procedure, both for parotid and submandibular sialendoscopy, is initiated by serial dilatation of the salivary gland ducts. In case of difficulty with visualizing salivary gland papilla milking of the gland or use of methylene blue was suggested in the literature (Kent, Walvekar, & Schaitkin, 2016) [11]. Schaitkin salivary gland dilators can be used, or serial dilation with standard salivary gland dilators from size 0000 to 8. Marchal bougies can then be used to further dilate the ducts (Fig 2).

FIGURE 2: Sialendoscopy dilation set. Standard dilators No. 0000 to 8, conical bougie, Marchal dilators, papillotomy scissors, grasping forceps. (From top left to bottom left in clockwise orientation).

Once appropriate dilation has been performed, COOK® Kolenda Introducer Set can be placed in the duct to secure ductal access (Fig 3).

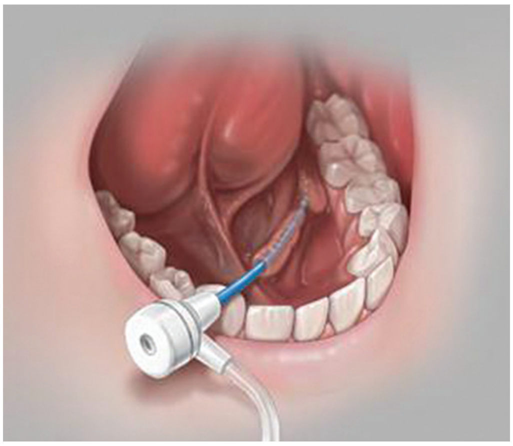


FIGURE 3. COOK® Kolenda Introducer Set.

Local anesthesia (1% lidocaine with 1:100,000 epinephrine) is injected into the duct with an flexibile angiocatheter tip. Diagnostic sialendoscopy is then performed with the Karl Storz® ALL-IN-ONE Sialendoscopes (Fig 4), ERLANGEN model 0.8mm diameter endoscope.

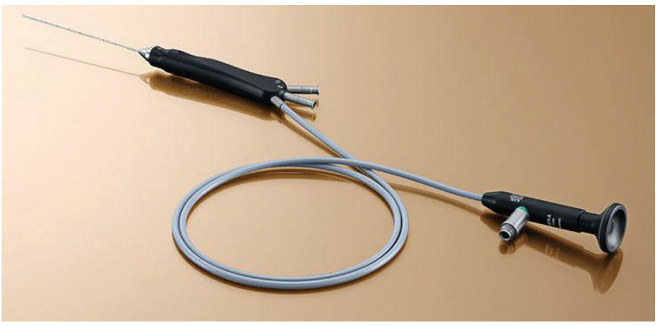
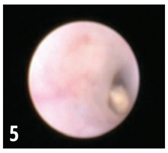
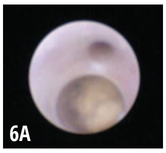
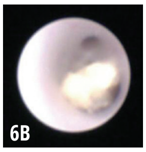


FIGURE 4. Karl Storz® ALL-IN-ONE Sialendoscopes, ERLANGEN model.

Following diagnostic sialendoscopy, therapeutic endoscopy can then be performed with either the 1.1mm or 1.6mm diameter endoscopes. If possible, the authors prefer to use the 1.6mm diameter scope for improved ability to perform stone extraction. It is important to maintain a constant and steady flow of fluid insufflation during the entire procedure to prevent ductal collapse and improve visualization (Figs 5-7). A three-way stopcock is attached to a 60cc syringe to allow the assistant to apply controlled insufflation during the procedure.







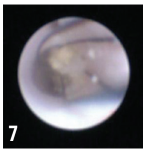


FIGURE 5. Sialolith visualiziation.
FIGURE 6A. Sialolith proximal to duct bifurcation.
FIGURE 6B. Sialolith distal to ductal bifurcation.
FIGURE 7. Wire basket applied to sialolith.

If performed this procedure for stone extraction, wire baskets are then used to attempt stone removal (Fig 7). They are introduced in the closed position through the working port of the endoscopy and passed beyond the stone. Once beyond the stone, the wire basket can be opened and rotated until it is engaging the stone. The basket and the endoscope are then retracted from the duct together, keeping visualization on the stone. For stones larger than 3mm or 4mm, parotid or submandibular respectively, papillotomy may need to be performed to allow for stone retrieval. For more advanced techniques and larger stones, laser lithotripsy can be performed to fragment the stone.

After stone extraction, 0.8mm endoscopy is used again to evaluate the salivary gland system to ensure that all stones have been removed and to irrigate all remaining debris and mucous plugs.

If performed for salivary duct strictures, similar initial procedures are performed with the exception of using wire baskets. Using the 1.6mm diameter endoscope, balloon catheters can be introduced through the working port in a closed position. Once the area or areas of strictures are encountered, the balloon is opened and held in place for roughly one to two minutes. The balloon catheter is then retracted and the stricture evaluated. If adequate dilation has not been achieved, catheter inflation can be performed again.

Once the procedure is completed, the endoscopes and COOK® Kolenda Introducer Set can be removed. Minor papillotomy can be performed at this time if needed. Compressive head dressing is then placed followed by patient emergence from general anesthesia and extubation.

Post-operative care includes instructing the patient to perform frequent warm compress massages over the involved salivary gland and appropriate hydration. Post-operative antibiotic use is not indicated, only the standard peri-operative prophylactic dose. In the literature, practice of prescribing antibiotics appears to be center or surgeon specific, although reported post-operative rates of glandular infection are around 2.5% (Nahlieli, Bar, Shacham, Eliav, & Hecht-Nakar, 2004) [12]. Local infection of papilla has been reported around 23%, thus suggesting that use of antiseptic mouth rinse, such as chlorhexidine, maybe warranted in the immediate post-operative period.

Pearls and Pitfalls

There are several technical problems and complications that can occur during sialendoscopy. Some technical errors are maceration of the papilla, which can be avoided by decreasing the amount of traction or force placed on the endoscope. Over insufflation or excessive pressure while irrigating can lead to significant edema, it is important to maintain a controlled level of pressure during irrigation to avoid this. This is monitored by the assistant using a 60cc syringe attached to a three-way stopcock, and only irrigating fluid with enough pressure to maintain duct patency for visualization. False passages and ductal perforation can also be created with using excessive force during dilation or blindly passive instruments through the working port. The most severe or life-threatening complication can occur during submandibular gland sialendoscopy, which is floor of mouth edema leading to airway compromise. The reported incident of upper airway obstruction occurred in the setting of irrigating solution extravasation after excessive pressures resulted in ductal tear (Baptista, Gimeno, Salvinelli, Rinaldi, & Casale, 2009) [13]. If this occurs, it is imperative to keep the patient intubated until the edema has subsided. The most common complication described in one study was failure of procedure due to peculiar duct anatomy, distal ductal stenosis or retained stone (Walvekar, Razfar, Carrau, & Schaitkin, 2008) [14]. Multiple other studies have validated sialendoscopy as a safe method with minor complications such as ductal tears, papillary infection, and facial swelling that usually self-resolve with minimal to now additional interventions (Marchal & Dulguerov, 2003) [15]. Possibility of lingual nerve damage exists, however it is seldom mentioned in the available reports.

Despite the high reported success rates with sialendoscopy, the procedure is deemed to be technically challenging and correlation between success rates and operator experience has been shown (Walvekar et al., 2008) [14]. In order to achieve the success rates of >90% as reported in literature, completion of 50 cases appears to be the benchmark (Steck, Stabenow, Volpi, & Vasconcelos, 2016) [16]. The most commonly cited difficulties surgeons new to sialendoscopy experience are difficulty canalizing the papilla, creation of false passage and duct lacerations (Steck et al., 2016) [16], (Farneti et al., 2015) [17]. Catheterization of the papilla is deemed the rate limiting step, since failure to achieve this step precludes completion of either diagnostic or therapeutic sialendoscopy (Farneti et al., 2015) [17]. Use of magnifying loops or even microscope, if available, may be beneficial in identifying and canalizing the papilla. There appears to be a consensus that surgeons experienced with endoscopic sinus surgery or dacryocystorhinostomy have no trouble with this aspect of the procedure. It has been suggested that practicing this skill on fresh cadavers of human or pig heads should be part of standardized training (Steck et al., 2016) [16] (Farneti et al., 2015) [17]. Laceration of the duct is undesirable due to potential of future ductal stenosis thus increasing patient's chance for recurrent obstructive symptoms and possible need for eventual gland removal. Moreover, false passage can be created through the laceration increasing the risk for irrigant extravasation and making completion of the procedure more arduous. Definitive papilla identification, clear visualization of the intraductal lumen, and gentle instrument manipulation and irrigation reduce the likelihood of duct laceration. Even though sialendoscopy can be performed under local anesthesia, general anesthesia is recommended until adequate level of comfort and confidence is achieved by the operator.

There is a steep learning curve when beginning the practice of sialendoscopy. In the authors' experience, several challenges have been encountered that have led to implementing changes in our technique. First, to prevent trauma to the ductal papilla by entering the duct repeatedly with the endoscopes, use of the COOK® Kolenda Introducer Set was implemented. This device allows one to maintain passage of the endoscopes and instruments into the duct without having to reenter the papilla. Second, use of the 0.8mm scope initially is essential. This allows you to visualize the ductal anatomy, locate sialoliths and mucous plugs, and measure the approximate depth or distance a sialolith is prior to using the therapeutic sialoscopes. Finally, when attempting to remove a large sialolith, if one encounters difficulty encircling the stone, do not hesitate to use instrumentation, i.e. Karl Storz® Foreign Body Forceps, to break the stone apart into smaller fragments. Attempting to force a wire basket around the stone can lead to inadvertent laceration of the salivary gland duct.

Conclusion

Sialendoscopy is a minimally invasive technique that is gradually replacing the classic open surgical approach to the treatment of obstructive salivary gland diseases as the standard of care. Although the initial challenges to the implementation of sialendoscopy into routine practice include high cost and need for specialized training, benefits have proven to be substantial. Ability to perform sialendoscopy in an outpatient setting and, in appropriate situations, under local anesthetic dramatically reduces patient's financial burden, post-operative morbidity, and recovery time. As cited in literature and per author's experience serious complications are rare and minor complications that do occur, frequently resolve on their own with no need for additional surgical intervention. In addition to providing an overall better experience for patients and allowing for quicker return to normal daily life, an overall reduction in patient care time per patient provides a surgeon with an opportunity to take care of greater number of patients. Thus, inclusion of sialendoscopy in surgeon's arsenal of practical skills is most definitely recommended.

References

1. Katz P. [Endoscopy of the salivary glands]. Ann Radiol 1991;34(1-2):110−113.
2. Nahlieli O, Nakar LH, Nazarian Y, et al.Sialoendoscopy: a new approach to salivary gland obstructive pathology. J Am Dent Assoc 2006;137(10):1394−400.
3. Ardekian L, Shamir D, Trabelsi M, et al. Chronic obstructive parotitis due to strictures of Stenson's duct − our treatment experience with sialoendoscopy. J Oral Maxillofac Surg 2010;68(1):83−7.
4. Singh PP, Gupta V. Sialendoscopy: introduction, indications and technique. Indian J Otolaryngol Head Neck Surg 2014;66(1):74−8.
5. Atienza G, Lopez-Cedrun JL. Management of obstructive salivary disorders by sialendoscopy: a systematic review. Br J Oral Maxillofac Surg 2015;53(6):507−19.
6. Hitti E, Salloum C, Mufarrij AJ. Imaging in acute obstructive sialadenitis. Emerg Med Australas 2014; 26(3):313−4.
7. Marchal F, Dulguerov P, Becker M, et al. Specificity of parotid sialendoscopy. Laryngoscope 2001;111(2):264−71.
8. Zenk J, Koch M, Klintworth N, et al. Sialendoscopy in the diagnosis and treatment of sialolithiasis: a study on more than 1000 patients. Otolaryngol Head Neck Surg 2012;147(5):858−63.
9. Luers JC, Grosheva M, Stenner M, et al. Sialoendoscopy: prognostic factors for endoscopic removal of salivary stones. Arch Otolaryngol Head Neck Surg 2011;137(4):325−9.
10. Luers JC, Stenner M, Schinke M, et al. Tolerability of sialendoscopy under local anesthesia. Ann Otol Rhinol Laryngol 2012;121(4):269−74.
11. Kent DT, Walvekar RR, Schaitkin BM. Sialendoscopy: getting started, how long does it take? Laryngoscope 2016;126(5):1083−5.
12. Nahlieli O, Bar T, Shacham R, et al. Management of chronic recurrent parotitis: current therapy. J Oral Maxillofac Surg 2004;62(9):1150−5.
13. Baptista P, Gimeno CV, Salvinelli F, et al. Acute upper airway obstruction caused by massive oedema of the tongue: unusual complication of sialoendoscopy. J Laryngol Otol 2009;123(12):1402−3.
14. Walvekar RR, Razfar A, Carrau RL, et al. Sialendoscopy and associated complications: a preliminary experience. Laryngoscope 2008;118(5):776−9.
15. Marchal F, Dulguerov P. Sialolithiasis management: the state of the art. Arch Otolaryngol Head Neck Surg 2003;129(9):951−6.
16. Steck JH, Stabenow E, Volpi EM, et al. The learning progression of diagnostic sialendoscopy. Braz J Otorhinolaryngol 2016;82(2):170−6.
17. Farneti P, Macri G, Gramellini G, et al. Learning curve in diagnostic and interventional sialendoscopy for obstructive salivary diseases. Acta Otorhinolaryngol Ital 2015;35(5):325−31.

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Мінімально інвазивні методики лікування патології слинних залоз

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Обладнання для сіалендоскопії
Розсічення під'язикового сосочку
Стеноз протоку
Внутрішньопротокові пробки із слизу

РЕЗЮМЕ

Мета. Мета данної роботи – описати метод, який використовується в нашому закладі та висвітлити потенційні труднощі під час сіалендоскопії.
Обговорення. Обговорюються показання до сіалендоскопії і методики її проведення
Результати. Незважаючи на зафіксований в світі високий рівень успішних сіалендоскопій, процедура вважається технічно сладною і частота успіху залежить від досвіду хірурга в данній методиці.
Висновки. Сіалендоскопія – це мінімально інвазивна методика, яка по поступово заміняє традиційний відкритий хірургічний доступ в лікуванні обструктивних захворювань слинних залоз, як стандарта лікування.

Минимально инвазивные методики лечения патологии слюнных желез

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О СТАТЬЕ

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Сиалэндоскопия
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Оборудование для сиалэндоскопии
Рассечение подъязычного сосочка
Стеноз протока
Внутрипротоковые пробки из слизи

РЕЗЮМЕ

Цель. Цель данной работы – описать методику, которая используется в нашем учреждении и осветить потенциальные сложности во время сиалэндоскопии.
Обсуждение. Обсуждаются показания к сиалэндоскопии и методики ее проведения.
Результаты. Невзирая на зафиксированный в мире высокий уровень успешных сиалэндоскопий, процедура считается технически сложной и частота успеха зависит от опыта хирурга в данной технике.
Выводы. Сиалэндоскопия – это минимально инвазивная методика, которая по нарастающей замещает традиционный открытый хирургический доступ при лечении обструктивных заболеваний слюнных желез, как стандарта лечения.

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Features of Diagnostics, Clinical Course and Treatment of the Branchial Cleft Cysts

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ABOUT ARTICLE

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ABSTRACT

Purpose.
The aim of the present study was to determine the features of diagnostics, clinical course and treatment of the branchial cleft cysts.
Patients and methods.
The study composed of the branchial cleft cysts investigation and their complications in patients of different age groups, methods of diagnostics, anatomical features, surgical stages and pathomorphological study.
Results.
Diagnostic value of sonography, MDCT and MRI, pathomorphological study in verification of branchial cleft cysts and their complications have been proved. Surgical treatment technique is presented.
Conclusion.
Presented methods of diagnostics of the branchial cleft cysts and their complications, variants of clinical course and treatment can reduce the risk of failure at the pre-, intra- and post-operative stages.

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Introduction

Branchial cleft cyst (*synonyms:* lateral cyst of the neck, congenital lateral cyst of the neck, branchial cyst, lateral branchial cyst of the neck, lateral lymphoepithelial cyst) according to our data has been found in 25% of all cysts of the soft tissue in maxillofacial and neck area [1-15]. The branchial cleft fistulas are rarely detected.

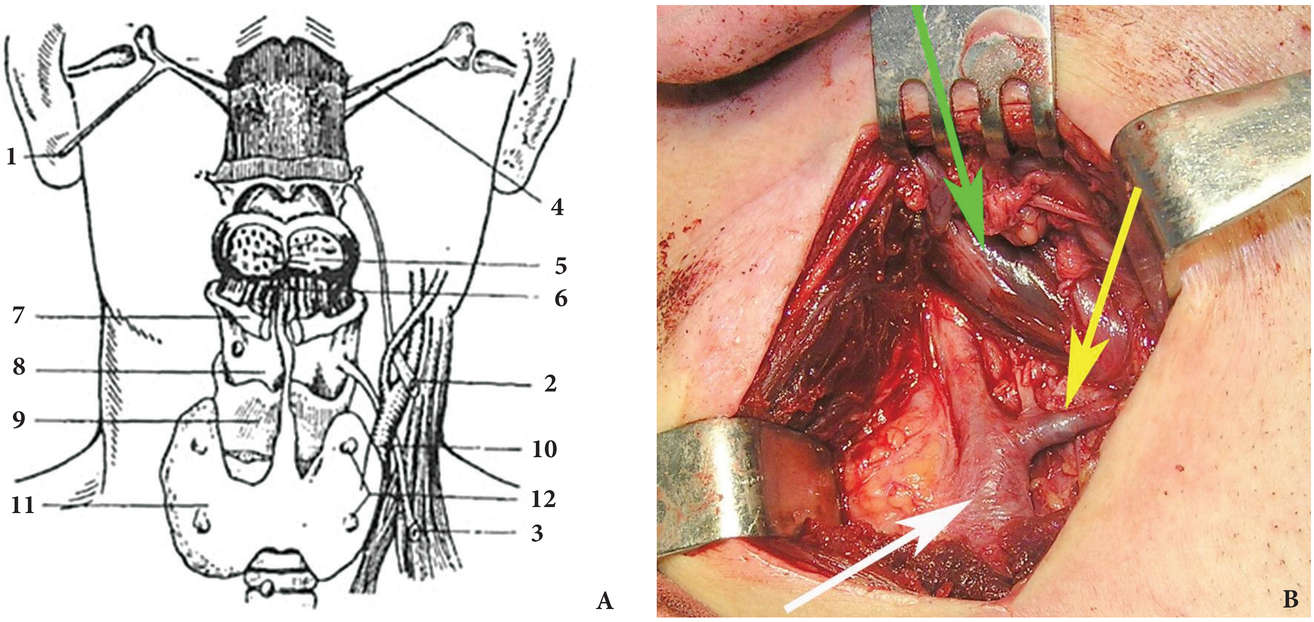

FIGURE 1. Location scheme of branchial neck fistulas (**A**): 1 – I branchial pocket; 2 – II branchial pocket; 3 – III branchial pocket; 4 – auditory tube; 5 – tongue; 6 – thyroglossal duct; 7 – the hyoid bone; 8 – thyroid-hyoid membrane; 9 – thyroid cartilage; 10 – common carotid artery; 11 – thyroid gland; 12 – parathyroid glands. Location of the inner pole (**B**) of BCC after its removal. Internal jugular vein is marked by *white arrow*, facial vein – *yellow arrow*, posterior belly of the digastric muscle – *green arrow*.

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Branchial cleft cysts (Greek, *branchia* gill) have dysontogenetic origin.

With regard to the pathogenesis of branchial cleft cysts (BCCs) and fistulas there is disagreement till present day. There are two theories of its origin. According to the **"thymus"** theory these cysts and fistulas are formed from the remnants of thymopharyngeal duct. **"Branchial"** theory links the origin of these lesions with abnormal development of branchial (pharyngeal) pockets. Anomalies of the 2nd or 3rd pair of pharyngeal (branchial) pockets are the source of the formation of the BCCs and fistulas. Internal branchial pockets are formed by endoderm and the external (or grooves) by ectodermal germ layers. BCCs can be both of endodermal and ectodermal origin (Fig 1A).

Cysts occur at any age, but are much more common in children and young adults (Fig 2). Their appearance is preceded (provoke) by the infections of the respiratory tracts (tonsillitis, flu, etc.). The sizes of the BCCs can be different (Fig 2). In contrary to dermoid (epidermoid) cysts the BCCs are often suppurate [1, 2, 3].

First, BCCs were classified according to their localization. Bailey H. (1929), divided them into 4 types [16]: **type 1** – deep to platysma, anterior to sternocleidomastoid (SCM); **type 2** – abutting internal carotid artery and adherent to internal jugular vein (most common); **type 3** – extending between internal and

FIGURE 2. Clinical view of the patients of different ages with BCCs (*arrows*) of various sizes (**A**, **B**, **C**, **D**). **(Fig 2 continued on the next page.)**

external carotid arteries; **type 4** – abutting pharyngeal wall and potentially extending superiorly to skull base.

CLINICAL PICTURE

BCCs are the round-shaped mass at the upper neck anteriorly to the sternocleidomastoid muscle (in carotid triangle). At the same time, they may be located in the middle and even lower parts of the neck. Typically, the BCCs localized in the upper or middle third of the neck adjacent to the anterior edge of the sternocleidomastoid muscle or partly comes under it. It is located between the 2nd and 3rd fascial leaf of the neck (between the superficial and deep fascia leaf of the own neck fascia) on the neurovascular bundle. The upper pole of the cyst is often found near or under the posterior edge of the digastric or stylohyoid muscles. Medially the cysts are adjacent to the internal jugular vein at the level of common carotid artery bifurcation. BCCs can be located in the upper, middle and lower parts of the neck. Along the length the cyst may extend down to the clavicle, and in the upper part of the neck reaches mastoid process (Fig 1B).

Visually, BCCs are showning as a painless limited rounded shape tumor-like lesion with a smooth surface. The skin above it is not changed in color. They are not soldered with surrounding tissues. A compulsory component of the

FIGURE 2 (cont'd). Clinical view of the patients of different ages with BCCs (*arrows*) of various sizes (**E**, **F**, **G**, **H**). **(Fig 2 continued on the next page.)**

FIGURE 2. (**cont'd**) Clinical view of female with the non-infected BCC (*arrow*) (**I**).

FIGURE 3. Clinical view of female with suppurated BCC.

cyst is a lymph node at the inferior pole. Upon swallowing the tumor-like mass does not move (as opposed to thyroglossal duct cysts). Consistency of the cysts are soft-elastic or elastically tense (elastically dense). A fluctuation may be determined. The BCCs does not cause respiratory and swallowing disorders. Systemic manifestations are not present. With secondary inflammation the cyst becomes dense, slow-moving, painful, can cause pain upon swallowing, and even talking. The systemic symptoms are (malaise, weakness, fever, etc.) appearing. Puncture of the cyst can get serous-mucous or muco-purulent transparent liquid light brown or dark brown (rare) color. Upon suppuration cyst fluid becomes turbid, pus appears. The skin over the cyst in case of its suppuration becomes hyperemated (Fig 3).

Desquamated epithelial cells, erythrocytes, lymphocytes, and cholesterol crystals can be detected microscopically in a punctate. Upon bacteriological examination a microflora in the content of uncomplicated cysts usually is not found. Only in rare cases low virulent *staphylococci* or *streptococci* are founded.

PATHOLOGY

Verification of the diagnosis is provided with pathomorphological investigation. Microscopically, the wall of the BCCs consists of a dense connective (fibrous) tissue that is lined with a stratified squamous non-cornified epithelium (ectodermal cysts), and multi-layered columnar epithelium (endodermal cyst). Some BCCs contain ciliated epithelium. Inside the wall (capsule) the lymphoid tissue, often forms the follicles (germinal centers) (Fig 4) [17]. Significant development of lymphoid tissue suggests that the BCCs originate from the branchial apparatus remnants. The inner surface of the cyst may be covered with warty growths of lymphoid tissue (crypts). In its wall, the formations like Hassel's corpuscles of thymus gland are identified. Upon suppuration of the cyst the epithelium can partially die and be replaced by the connective tissue, there is a thickening of the epithelial lining and its cornification. At the inferior pole of the BCCs the lymph node is often morphologically detected.

In front of the tragus, the preauricular (branchial) fistula can be found, which comes from I branchial pocket. The fistulous tract is lined by squamous epithelium. Preauricular (tragal) fistulas are often spread deep into the soft tissue to the parotid gland, and even penetrate into it. From these fistulas develop cysts localized in the parotid gland. The morphological difference between these fistulas is that the wall of the fistula, originating from the branchial I pocket has no lymphoid tissue, which is always present in BCCs or fistulas localized in the neck.

Diagnostics of BCCs is carried out between chronic **lymphadenitis** (non-specific and specific) [18-21], **dermoid** (**epidermoid**) **cysts** [22, 23], **tumors** and **tumor-like lesions** of soft tissues of the neck, blood vessels, nerves and thyroid gland, lymphangiomas (Fig 10) [24-28], metastases of malignant tumors, etc. For more accurate diagnosis the cyst- or fistulography with the administration of radiocontrast agents, CT, MRI, ultrasound can be performed (Fig 5).

ULTRASOUND

Upon **ultrasound diagnostics** are estimated: the location of the lesion, its size, wall thickness and the presence of septations, edges, borders, internal echogenicity, presence of acoustic enhancement artifact, fistula, vascularization at Doppler ultrasound. Compression of the mass by ultrasonic transducer confirms the true cystic nature helping in differential diagnosis. BCCs at ultrasonography are visualized as cystic mass of oval or round shape with smooth surface (Figs 6-8). Ahuja A.T. et al. (2000) [19] distinguish four echogenicity patterns of BCCs content: truly ***anehoic*** (41%), predominantly homogeneously ***hypoechoic*** but

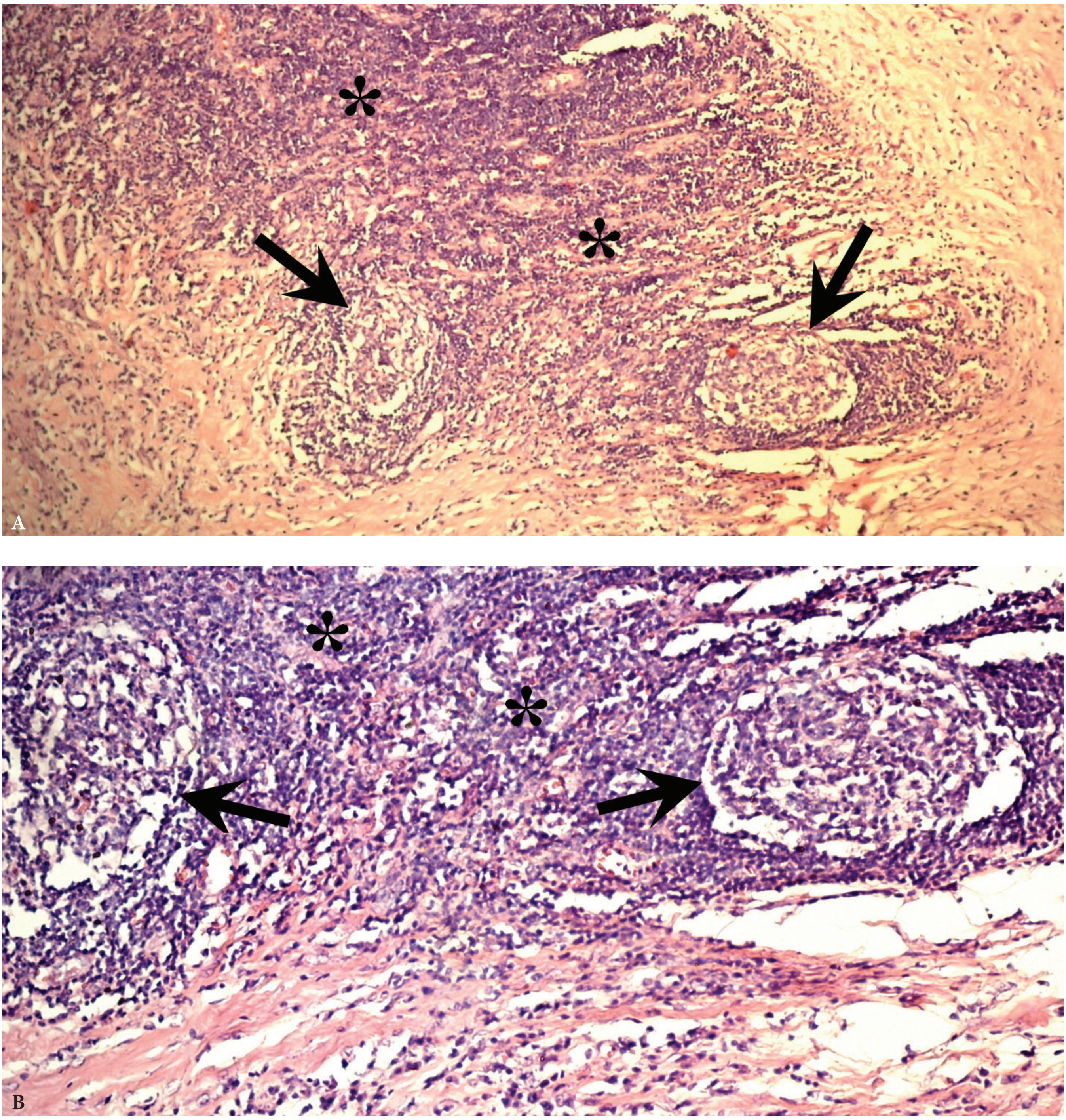

FIGURE 4. The histologic specimen of BCC of a 52-years-old female. Lymphoid formation (*asterisks*) in the thickness of the cystic wall with prominent bright breeding centers (germinal centers), which are marked by *arrows* (hematoxylin and eosin; magnification: **A** – x50, **B** – x100).

showing the presence of internal, low-amplitude, freely floating ***debris*** (diagnosed at 24%), ***hyperechoic*** with ***pseudosolid*** appearance (12%), ***heterogeneous*** internal echoes with internal debris and septa (23%). Echogenicity type is affected by the consistency of the content of the BCCs, which may vary depending on the presence of inflammatory processes (liquid-cystic, cystic-liquid with debris, pasty, suppurated). Pseudosolid appearance due to the presence of the protein content of the cysts produced by the epithelial lining [31]. On color and power Doppler ultrasound BCCs are avascular. Cyst wall appears as hyper- or isoehoic linear structure, often avascular at Doppler ultrasound. The thickness of the wall may vary in different parts of cysts and reach 1.0cm in recurrent inflammations [30,31], but also thinning is possible, i.e. the wall becomes non-differentiable [4].

CT & MRI

According to Weerakkody Y., Gaillard F. et al. on the **CT** and **MRI** images the BCCs have the following features.

For the patients with BCCs is recommended to perform magnetic resonance imaging in three modes. On **T1-weighted MR images** BCCs appears as a variable signal depending on the protein content. If their content is high – as high-intensity signal, low – as low intensity. On **T2-weighted MR images** BCCs are usually of high intensity. On **contrast-enhanced T1-weighted**

MRI, in uncomplicated cases, the BCCs have no enhancement.

On **contrast-enhanced multidetector CT images** BCCs are spherical or round shape, its walls are clearly distinguished from the surrounding tissue. The wall thickness varies from 0.1 to 1.0 cm. The cystic wall may penetrate between internal and external carotid arteries, in the region above the bifurcation of the common carotid artery (scraps symptom or beak tail) [6]. The density of the contents of the cavities (depending on the type of content and the presence of inflammation) ranges from 10 to +27,8 (± 6,0) HU, wall density is up to +102,0 (± 8,0) HU. **Hounsfield units** (HU) is a units of measure indicating the absorption of the X-rays by various tissues of the body. Remember that mostly absorbs x-rays the tooth enamel (3000 HU) and cortical bone (from 850 to 2000 HU), less of all – the blood (20-70 HU) and muscle (10-70 HU), adipose tissue (from −40 to −100 HU).

BCCs should be differentiated with esophageal diverticula. Esophageal diverticula is presented as a round shape lesion, which is located in front of sternocleidomastoid muscle. The lesion is soft or pastry to the touch, collapses on palpation and transmits peristaltic waves during swallowing. With eating it is filled, and increases in size. The pain is intensified with filling of diverticula after eating. Swallowing can be painful, especially during exacerbation of the inflammatory process.

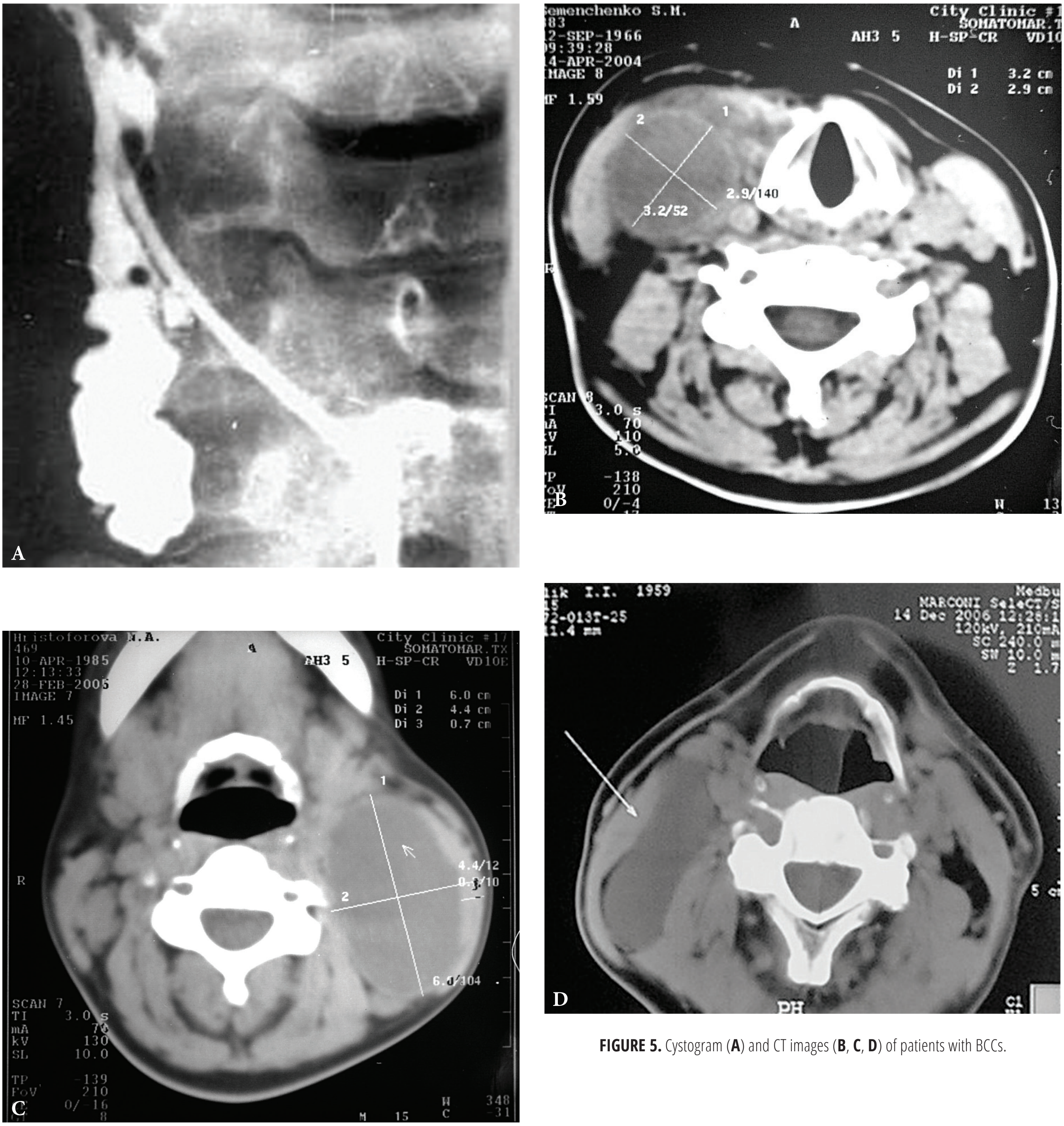

FIGURE 5. Cystogram (**A**) and CT images (**B**, **C**, **D**) of patients with BCCs.

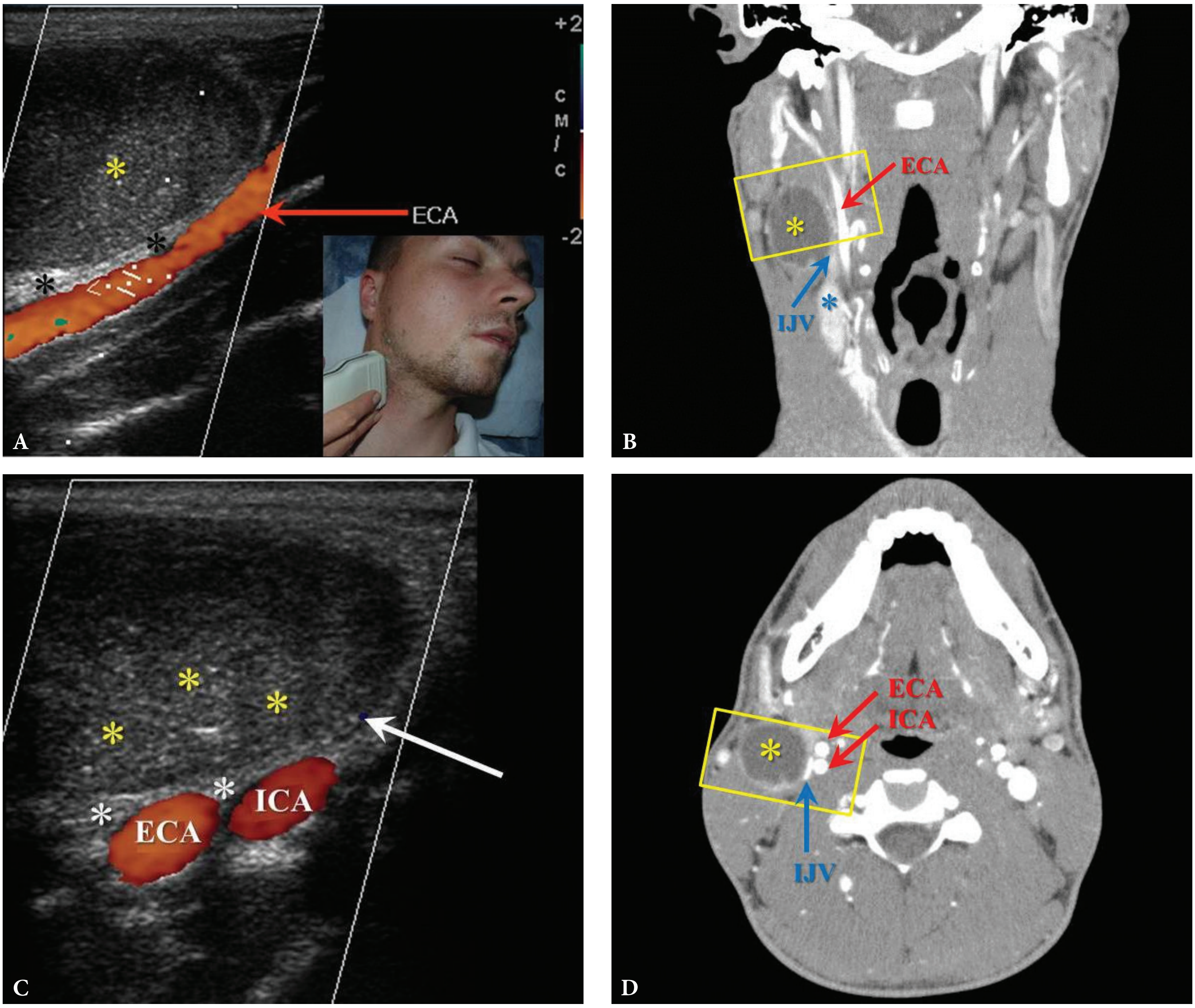

FIGURE 6. BCC in a 19-years-old patient. Longitudinal spectral Doppler sonogram (**A**) performed by linear transducer shows oval shaped cystic lesion with smooth margins, sharp edges, filled with a heterogeneous content (*yellow asterisks*) which simulate "pseudosolid" appearance. Note that cyst is adjacent to the external carotid artery (*ECA*) which is marked by a *red arrow*. Acoustic enhancement artifact (*black asterisks*) is distal to the cyst. Cyst is avascular. On contrast-enhanced CT (**B**) is confirmed the presence of cystic lesion (*yellow aterisk*) adjacent to the external carotid artery (*ECA*) and compression of the internal jugular vein (*IJV*). By *yellow frame* is marked the sonogram location performed at **Figure 6A**. Transverse spectral Doppler ultrasound (**C**) performed by linear transducer shows oval shaped cystic lesion with smooth contours, sharp edges, filled by heterogeneous content (*yellow asterisks*) which create "pseudosolid" appearance. The cyst is adjacent to the external (*ECA*) and internal carotid artery (*ICA*), and internal jugular vein (*IJV*), squeezing it (*white arrow*). Artifact of posterior acoustic enhancement (*white asterisks*) visualized distal to the cyst. Blood flow within the lesion and its wall is absent. On contrast CT image (**D**) confirmed the presence of cystic lesion (*yellow asterisk*) adjacent to the carotid arteries (*ECA, ICA*) and compression of the internal jugular vein (*IJV*). The density of the cyst content is +27,8 (± 6,0) HU. *Yellow frame* marks the position of a sonogram obtained at **Figure 6C**.

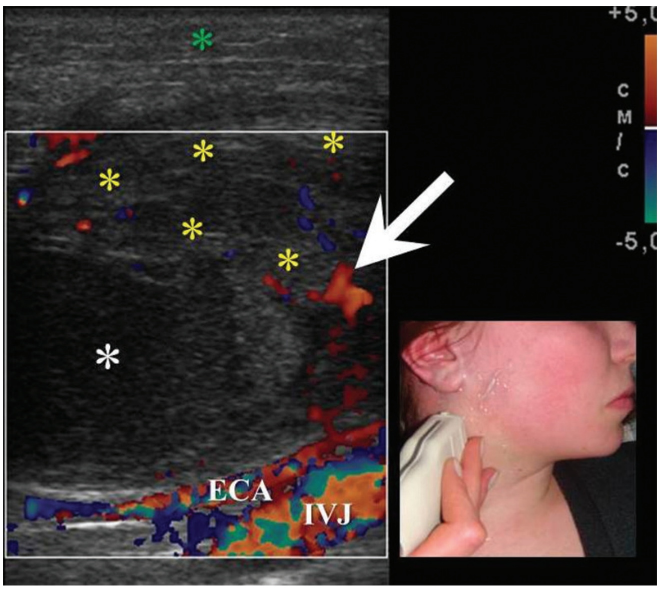

FIGURE 7. An infected BCC in a 18-years-old female. Longitudinal color Doppler ultrasound shows cystic oval shape lesion with hypoechoic content (*white asterisk*). Note inflammatory hyperemia of sternocleidomastoid muscle (*yellow asterisks*) in a form of its increased vascularity (*arrow*). Edema, decreased echogenicity of the surrounded tissues is marked by *green asterisk*. Lesion is avascular, adjacent to the neurovascular bundle of the neck. External carotid artery and internal jugular vein are marked by *ECA* and *IJV*.

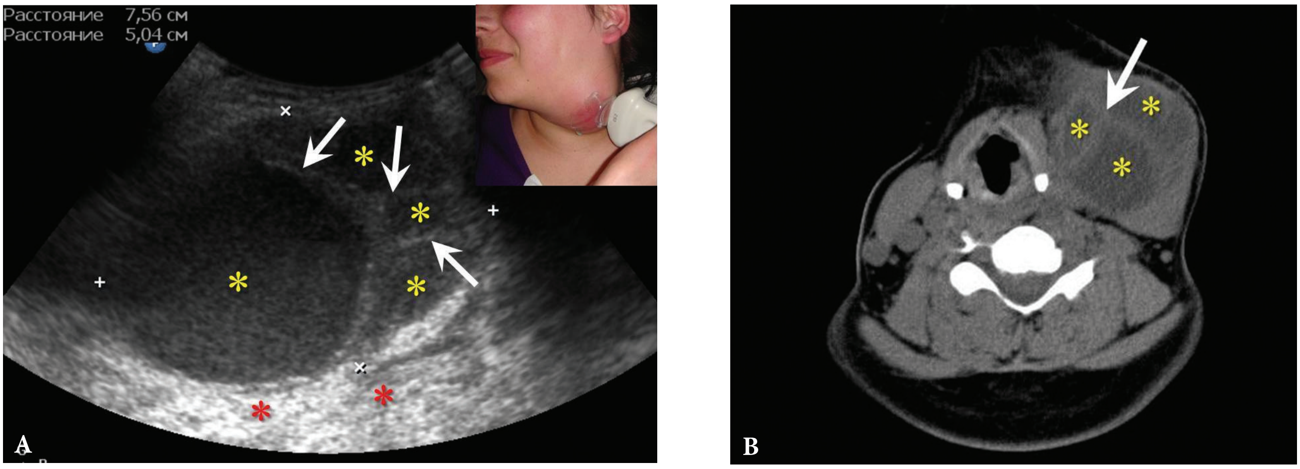

FIGURE 8. Suppurated multicameral BCC in a 33-years-old female. A sonogram (**A**) performed by convex transducer shows a cystic lesion (its size are marked with "+" and "x" are equal 7.5- x 5.0- cm) of the left neck with the presence of isoechoic septations (*arrows*). Anechoic cyst content in cameras are indicated by *yellow asterisks*, an artifact of acoustic enhancement – by *red asterisks*. An axial MDCT scan (**B**) confirms the presence of intracystic septations (*arrow*). The density of the cystic content is equal to +10, +15 HU. Cameras are marked by *asterisks*.

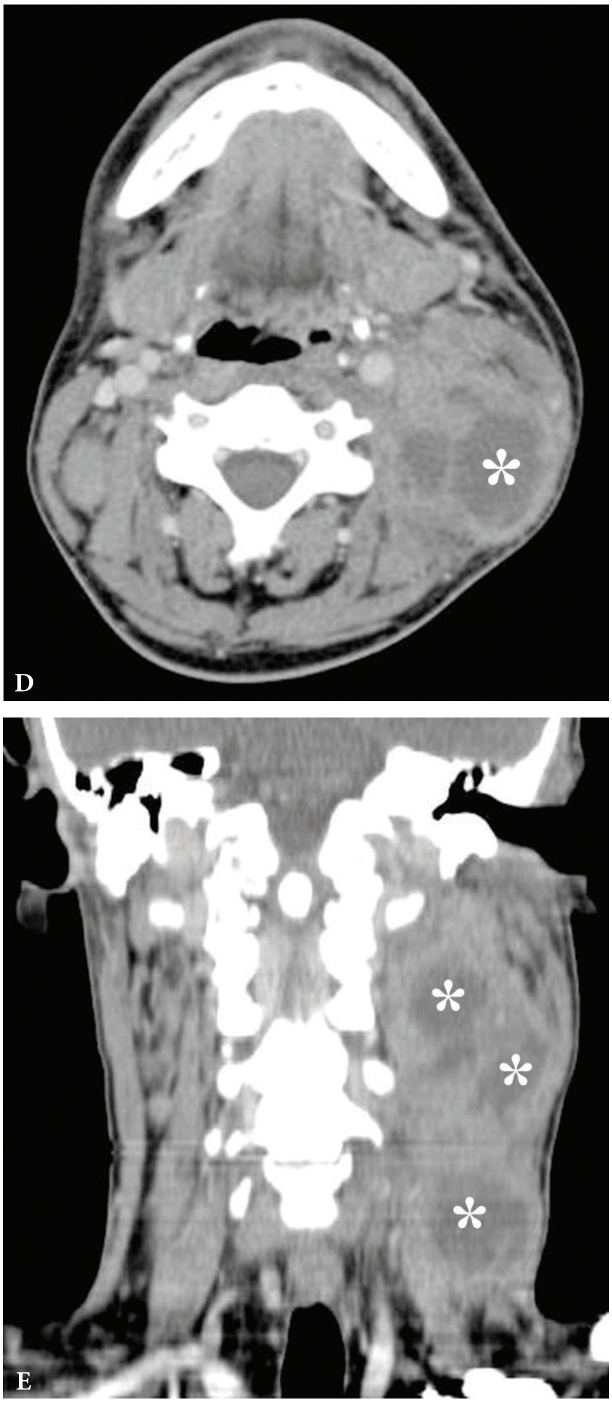

FIGURE 9. A 24-years-old man with cystic squamous cell carcinoma of the neck (poorly differentiated, which have the most aggressive behavior). Clinical photograph of the patient (**A**). Transverse gray scale ultrasound of the lower (**B**) and upper (**C**) neck shows multicameral lesion with anechoic cystic (*asterisks*) and heterogenous solid component, presented in the form of irregularly shaped intracystic growths (*arrows*). Acoustic enhancement artifact is presented. On contrast-enhanced CT images (**D**, **E**), the lesion on left side of the neck is multicameral (cameras are marked by *asterisks*) with solid component accumulating contrast. **(Fig 9 continued on the next page.)**

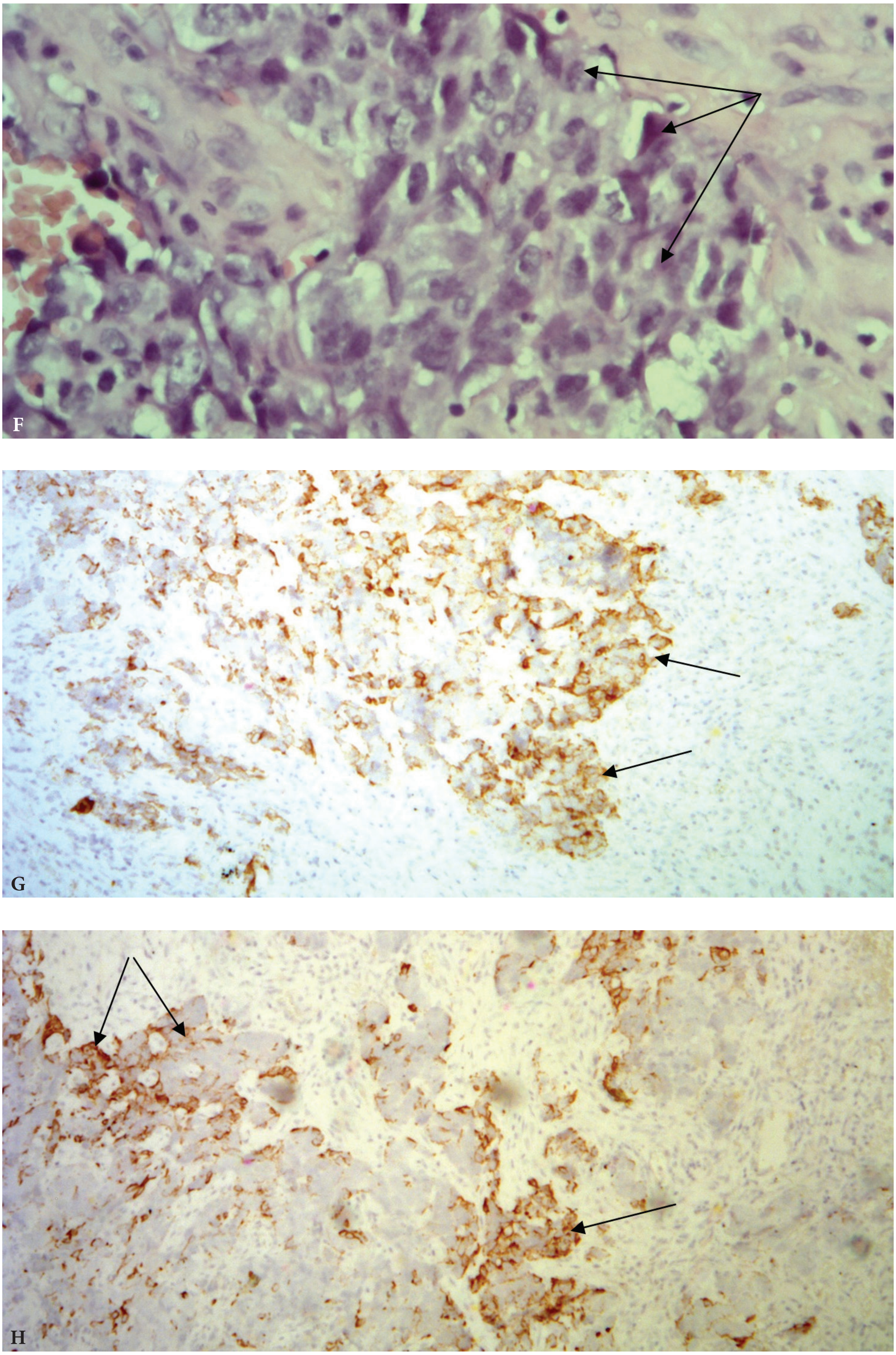

FIGURE 9 (cont'd). Trephine biopsy histology (**F**) shows, that in fragment of the lymph node tissue with presence of fibrous tissue, the growth of poorly differentiated malignant tumor (*arrows*) of epithelial nature with extensive necrosis in the tumor and lymph node tissue, hemorrhages is determined (hematoxylin and eosin; magnification x200). Immunohistochemistry (**G**, **H**) shows positive membrane reaction (*arrows*) with Cytokeratin AE1/AE3 and Cytokeratin 5/6. Reaction with CD45, S100 is negative (reaction with CD45- is determined in intact cells of the lymph node tissue). Thus, there is metastasis of poorly differentiated squamous cell carcinoma G_3 into lymphatic node tissue. (Histology and immunohistochemistry **Figure 9F**, **G**, **H** is courtesy of Dr. Antoniuk S.A., *Research Associate*, Dr. Petrenko L.I., *Junior Scientific Researcher*, Dr. Burtyn O.V., National Cancer Institute, Kyiv, Ukraine)

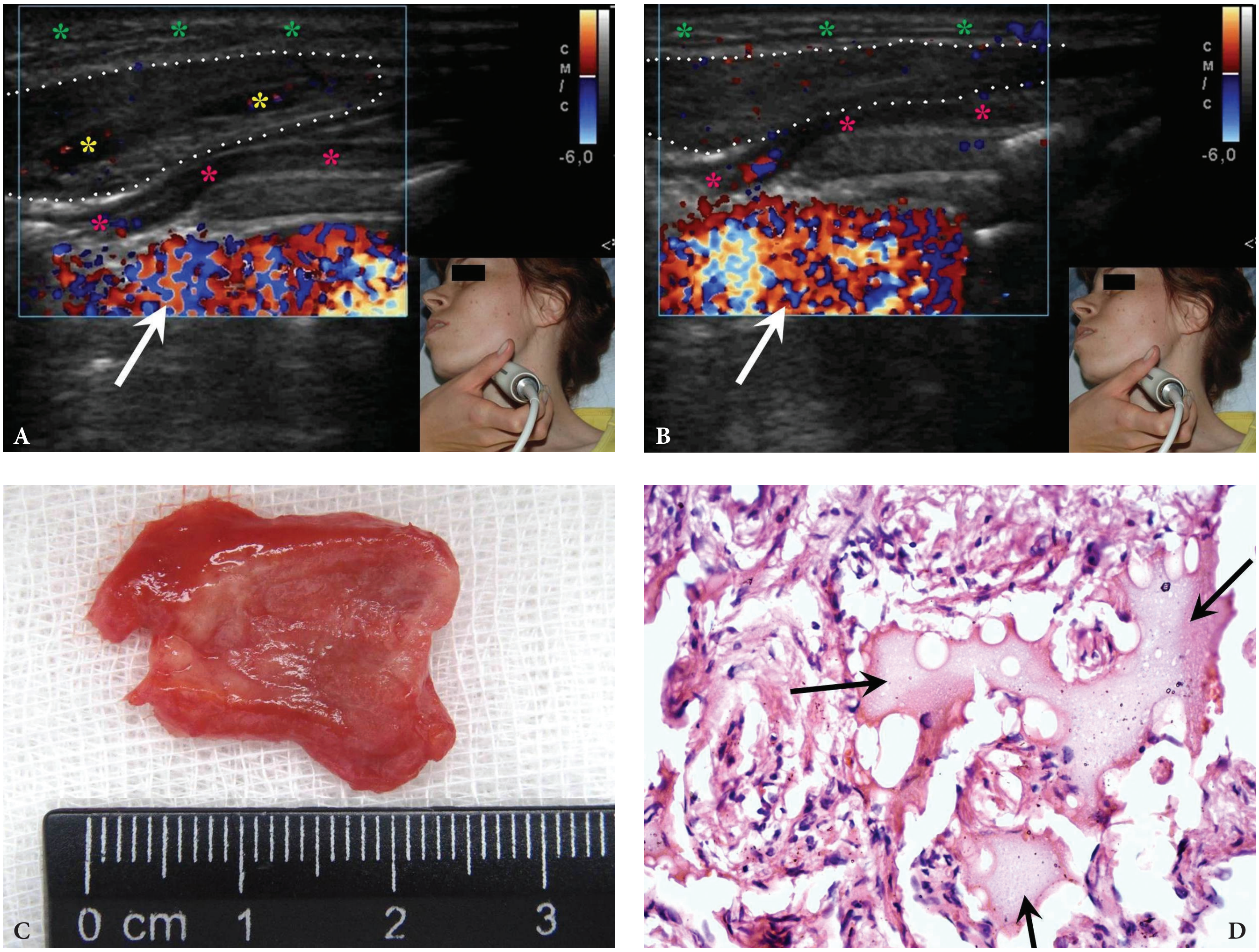

FIGURE 10. A 32-years-old female with cavernous lymphangioma of the upper neck. Oblique color Doppler ultrasound (**A**) shows well-demarcated lesion (marked by *white dots*) with a size of 3.0- × 1.0- cm elongated-oval form at the upper neck adjust to the omohyoid (*pink asterisks*) and anterior to the sternocleidomastoid muscles. Structure of the lesion is heterogeneous with presence of oval anechoic areas (cavities – *yellow asterisks*). The skin, subcutaneous tissue is marked by *green stars*. *White arrow* marks blooming artifact. Lesion have no arterial or venous blood flow. Oblique color Doppler ultrasound (**B**) with the compression by transducer (sonopalpation) noted the tumor shrinkage in 2-3 times, cavernous cameras are completely disappeared, indicating its spongy structure and confirmed after removal (lesion for the entire thickness was impregnated with light-gray liquid – lymph). Surgical specimen (**C**). Histology (**D**). In the lumen of the lymph vessels, lymph (*arrows*) is visualized. The inner layer is composed of endothelial cells of lymphatic vessels (oval form small inclusions with dark blue color). Hematoxylin and eosin; magnification x200.

TREATMENT

The treatment of the BCCs is only surgical (Figs 11, 12A). Surgery can be a difficult task due to the complex anatomical and topographical relations of the cysts with vessels and nerves of the neck. The surgery is performed under endotracheal anesthesia. The cut should be done on the anterior (medial) edge of the sternocleidomastoid muscle, or upper cervical crease. The first variant of incision is considered safer because in this area a large vessels and veins are located, and the second variant of cut is more cosmetic.

The surgeon may have difficulties in location place of internal pole of the BCC (Fig 1B), as in this place the internal jugular and facial veins are located. Especially need to be careful upon separating the BCC when its located nearby external and internal carotid arteries (Figs 12, 13). With the classical location of BCCs, it is more easy for the surgeon to navigate in the topographic anatomy of those vessels. However, often there are different variants for the location of the internal and external carotid arteries. This causes considerable difficulties in the intraoperative visualization of those vessels. Be especially careful in case of deep location of BCCs and the need to separate carotid arteries, i.e. topographic anatomy of the latter is not always classical (Fig 13).

COMPLICATIONS

Complications of BCCs the phlegmon of the and branchiogenic carcinoma are known. Phlegmon of the neck is more severe complication with severe intoxication in patient. Inflammatory processes can easily spread through the neurovascular bundle into the anterior mediastinum. Non-radical surgery may not only lead to recurrence, but also to the development of branchiogenic carcinoma (Figs 14, 15).

Branchiogenic carcinoma develops from the epithelium of the BCCs. Unlike cysts, tumor represents as a dense, tuberous, bad-movable (especially in the vertical direction) lesion, knitted with muscle and vascular bundle of the neck. Tumor (branchiogenic carcinoma) painless, relatively slow increases in size and can reach considerable size, quickly merges to the surrounding tissues. Tumor localization: from the submandibular region to the clavicle. Branchiogenic carcinoma merges to sternocleidomastoid muscle and vascular bundle of the neck. If the tumor has not merge into the vascular bundle, it can be separated from the vessel. Malignant tumors can merge not only in vascular bundle neck, soft tissue (muscles) of the side of the neck, but to the pharynx and larynx. Histologically branchiogenic carcinoma usually has a structure of squamous cell carcinoma (Fig 9F, G, H).

Development of branchiogenic carcinoma, according to the Maxillofacial Surgery Department of Shupyk NMAPE, is about 4.5% of patients with BCCs. A high percentage of branchiogenic cancer in these patients emphasizes the need for early and proper performed surgery (removal of the BCC). Treatment of patients with branchiogenic carcinoma is combined. Prognosis is poor, often recurs, metastases are rare.

Branchiogenic carcinoma should be differentiated from the **carotid chemodectoma** (Fig 16) and other tumors of the neck. Carotid chemodectoma *synonyms: carotid body paraganglioma, glomus tumor, endothelioma, perithelioma, pheochromocytoma, paraganglioma, potato tumor, receptoma, etc.* Chemodectoma [34, 35] develops from the carotid sinus (*synonyms*: chemoreceptoral glomus, glomus caroticum), located at the adventitia layer inwards from the bifurcation of the common carotid artery.

Carotid sinus (Greek, *karóō* to put to sleep and sinus) is an expanded portion of the common carotid artery at the site of its branching into the external and internal arteries.

In this glomus there is a cluster of chromaffin cells around capillary glomeruli, and there is lot amount of nerve endings (functions as a "chemoreceptor" – responds to changes in the level of oxygen in the blood). Making pressure on the vessel in the area of carotid glomus leads to a slowing of heart rate. There carotid sinus baroreceptors are also located, when they are stimulated the blood vessels dilate and blood pressure falls. Chemodectomas are located under the sternocleidomastoid muscle at the point of common carotid artery bifurcation.

The skin over the tumor is not changed. Tumor has spherical or elongated form, with dimensions of 3.0cm or more, smooth or slightly tuberous. *A characteristic feature of chemodectoma is its horizontal displaceability and absence of displacement in vertical direction, the inability to move aside tumor from the pulsating vessel and "transfer pulsation" over the tumor.*

Macroscopically carotid chemodectoma has a light gray or brownish-red color and is surrounded by a connective tissue capsule (Fig 16C). Treatment of chemodectoma is surgical. Postoperative mortality is very high, i.e. in the vast majority it is not possible to separate the tumor from the common or internal carotid artery.

PROGNOSIS

With timely and correct performed surgery, removing of the BCCs, the prognosis is favorable.

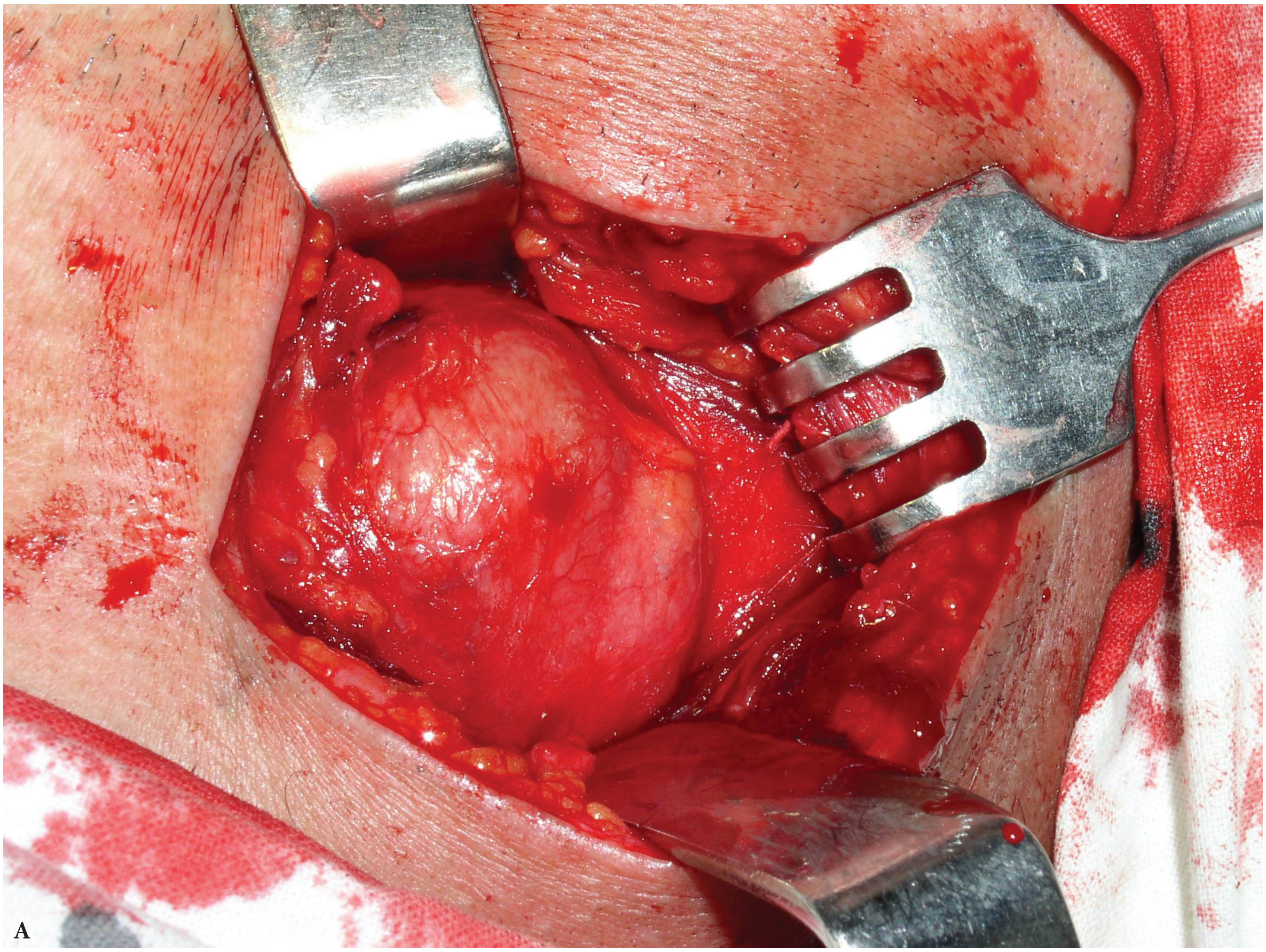

FIGURE 11. Surgical stage of BCC excision (**A**). **(Fig 11 continued on the next page.)**