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Case Report / Приказ болесника

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Edema of the larynx – an emergency caused by angina Ludovici

Едем ларинкса – хитно стање узроковано Лудвиговом ангином

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SUMMARY

Introduction Laryngeal edema is a rare complication of Angina Ludovici. Infections of this region are mostly of dentogenic origin, less often caused by tonsillitis or other infections in the pharynx. We present a case of a patient with laryngeal edema and dyspnea caused by a lower jaw tooth infection and an ipsilateral submandibular abscess.

Case outline The clinical picture of our patient progressed rapidly. From toothache, painful swelling of the floor of the oral cavity, submandibular and submental regions, bilaterally, all the way to life-threatening dyspnea. A flexible nasopharyngolaryngoscopy was performed. Swelling of the base of the tongue on the left side was observed, along with pronounced edema of the aryepiglottic fold on the same side, which narrowed the breathing space. Since the breathing space was significantly reduced, the patient was urgently hospitalized. The surgical treatment was carried out in the form of an external incision and drainage of the abscess collection of the left submandibular region, with the use of oxygen support and parenteral therapy, in accordance with the recommendations from the available medical literature. Constant monitoring of saturation levels indicated a significant improvement after just a few hours of medicamentous therapy.

Conclusion The goal of our work is to point out a very rare but serious complication, laryngeal edema, which can lead to airway obstruction even in the first few days of the development of the infection, and endanger the life of the patient.

Keywords: edema of the larynx; stridor; submandibular abscess

САЖЕТАК

Увод Едем ларинкса је ретка компликација Лудовигове ангине. Инфекције ове регије најчешће су дентогеног порекла, ређе узроковане тонзилитисом или другим инфекцијама у фаринксу. Приказујемо случај болесника са едемом ларинкса и отежаним дисањем, настао као последица инфекције зуба доње вилице и ипсилатералног субмандибуларног апсцеса.

Приказ болесника Клиничка слика код нашег болесника је брзо напредовала. Од зубобоље, болног отока пода усне дупље, субмандибуларне и субменталне регије обострано, све до прогресивне и потенцијално животно угрожавајуће диспнеје. У току дијагностике учињена је флексибилна назофаринголарингоскопија. Уочен је оток базе језика лево, омега епиглотис, као и изразит едем ариепиглотисног набора са исте стране који је сужавао дисајни простор и потискивао ипсилатерални пириформни синус. Дисајни простор у моменту прегледа је био редукован те је болесник хитно хоспитализован. По хоспитализацији болесника, спроведен је хируршки третман, у виду спољашње инцизије и дренаже апсцесне колекције субмандибуларне регије са леве стране, уз примену кисеоничне потпоре и парентералне терапије, а у складу са препорукама из доступне медицинске литературе. Константни мониторинг и редовно праћење сатурације указали су на значајно побољшање већ након неколико сати од примене медикаментозне терапије.

Закључак Циљ нашег рада је указати на веома ретку али озбиљну компликацију, едем ларинкса, која може довести до опструкције дисајног пута, већ у првим данима развоја инфекције, и животно угрозити болесника.

Кључне речи: едем ларинкса; стридор; субмандибуларни апсцес

INTRODUCTION

Edema of the larynx, as a result of an infection of the submandibular region, is a rare condition, but nonetheless life-threatening. Infections of this region are most often of dentogenic origin. Wide communication with other deep spaces of the head and neck, as well as the mediastinum, represents an ideal route for the spread of the infection that can lead to sepsis and a fatal outcome [1]. In this paper, we will present a case of a 45-year-old man with laryngeal edema and difficulty breathing, caused by lower jaw tooth infection and an ipsilateral submandibular abscess.

CASE REPORT

A 45-year-old man was urgently hospitalized at the Department of Otorhinolaryngology with maxillofacial surgery due to painful neck swelling and stridorous breathing. As the patient stated, the first symptom was a toothache in the left side area of the lower jaw, which intensified significantly by the time of hospitalization. Progressive dyspnea lasted for three days. The clinical examination revealed palpable, painful swelling of the submandibular region on both sides as well as submentally; it was of a harder consistency, without the phenomenon of fluctuation, followed by hyperemia and tightness of the skin of the above-mentioned regions. Oropharyngoscopy was difficult to perform due to the presence of trismus, edema of the tongue and the tissues of the sublingual region. Since it was impossible to view the distal structures, a flexible nasopharyngolaryngoscopy was performed. During this procedure a few findings were noted: a swelling of the base of the tongue on the left side together with omega epiglottis, as well as a massive edema of the aryepiglottic fold on the same side, which narrowed the breathing space and suppressed the ipsilateral pyriform sinus (which did not open during the examination). The partially visible structures of the glottis were in order, both halves of the larynx were mobile (Figure 1). The breathing space was reduced yet sufficient at the time of the examination, while the measured oxygen saturation was around 92%.

Laboratory analysis revealed elevated parameters of inflammation, CRP 234 mg/L, Le $20 \times 10^9/L$, Ly 5.1%, Gr 93.8%, while other values were within the reference limits. Upon admission to the department, parenteral therapy was administered (ceftriaxone, metronidazole, and levofloxacin, along with corticosteroid and rehydration therapy), as well as oxygen support through a nasal cannula, with oxygen flow values of about 5 L/min. During hospitalization, a computed tomography (CT) scan of the neck was performed, the findings of which indicated the existence of a liquid collection of dense content, about 15 mm in diameter, at the level of the tooth root in the left ramus of the mandible. The submandibular left abscess formation with a transverse diameter of 55x35mm propagated posteriorly towards the parapharyngeal and paraglottic space with a mass effect on the oropharynx and supraglottis, obliterating the left pyriform sinus (Figure 2, 3).

Other findings, such as chest X-ray and abdominal ultrasound, did not show any pathological changes. Upon arrival of the diagnostic imaging findings (CT of the neck), an incision of the abscess collection of the left submandibular region was made externally, during which a large amount of purulent-hemorrhagic content was obtained, after which passive drainage was placed. The patient was observed carefully, with regular monitoring of the

saturation levels. After 4 hours of applied corticosteroid therapy, the patient reported a subjective improvement in breathing, while after 24 hours of applied therapy, a significant reduction of laryngeal edema was observed by fiberoptic examination. After the stabilization of the general condition, a maxillofacial surgeon was consulted, who performed the extraction of tooth 36 under local anesthesia, which had caused the dentogenic infection.

Appropriate surgical therapy (timely incision and drainage of the abscess) as well as medical therapy, regular bandaging and removal of the cause of the infection (tooth extraction) resulted in regression of the laryngeal edema and healing of the infection, along with stabilization of laboratory parameters of inflammation, which were within reference values at discharge.

All procedures performed were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki Declaration and its later amendments and comparable ethical standards. Written consent to publish all shown material was obtained from the patient.

DISCUSSION

Obstruction of the respiratory tract caused by edema of the mucous membrane of the larynx, although an uncommon symptom, is one of the numerous complications that can occur as a result of a submandibular abscess. Depending on the location and the size of the edema, as well as the patient's age, it can manifest itself in the form of minor breathing difficulties, up to a potentially life-threatening condition. Etiological factors for the development of laryngeal edema can be various. To start with, they can be infectious, caused by viruses, bacteria, or fungi. Additionally, there are allergic ones in the form of angioedema, or as part of an anaphylactic reaction to the use of certain medications or food. Other potential causes of airway obstructions should also be noted, such as trauma to the larynx, foreign bodies, autoimmune diseases, and tumors of the larynx [1, 2, 3].

Cellulitis of the floor of the oral cavity was first described in literature in 1836 by a German military doctor Wilhelm Frederick von Ludwig, after whom this condition was named. It involves inflammation of the submandibular, sublingual, and submental regions, with a tendency to spread rapidly along the fascia to adjacent regions [4]. The infection usually starts from the infection of the roots of the lower premolars and molars, in cases where the purulent exudate spreads through the inflamed pulp to the periodontium and bone breaks through the

cortical part of the lower jaw below the attachment of the mylohyoid muscle [1]. The swelling increases rapidly and tends to spread, generally does not fluctuate and is extremely painful on palpation. The patient has an elevated body temperature, a headache, difficulty in the mouth opening, hypersalivation is present, and saliva can be seen leaking from the mouth due to difficult and painful swallowing. The tongue and the floor of the oral cavity are raised, which makes breathing difficult, and larynx edema can also occur. Subjective disturbances and clinical findings worsen rapidly. This condition can lead to asphyxia within 24 hours [1, 5, 6, 7].

In about 70% of cases before the "antibiotic era", tonsillitis was the most common cause of Ludwig's angina. However, the work of Ismi O. et al. indicates that there are more and more cases where the spread of infection has dentogenic causes [8]. In developing countries, given the economic conditions and insufficient education of the population, dentogenic infections are the primary cause of these conditions in more than 80% of examined patients, while it is less often the result of inflammation of the tongue (e.g., piercing), submandibular gland, tonsils or pharynx. Publications available on Medline support the fact that laryngeal edema as a complication of Ludwig's angina is a rare occurrence [5, 9].

Other possible factors are obesity, tobacco and alcohol abuse, as well as malnutrition [10]. Furthermore, chronic diseases such as diabetes mellitus, hypertension or immunocompromised conditions can additionally contribute to the development of a more severe clinical picture and complications. This anatomically significant region of the neck is bounded medially by the hyoglossus muscle, anteriorly and posteriorly by the digastric muscles, and externally by the platysma and skin. Since the submandibular gland and neck lymph nodes are also located in this area, this should also be taken into account in the differential diagnosis of inflammation and swelling of this area [5, 6].

The spread of infection through the so-called "dangerous" areas of the neck along the fascia can lead to the development of numerous complications, the most common of all is mediastinitis, which can further lead to the development of empyema, pneumonia, and then pericarditis. Also Lemierre syndrome if there is involvement of the carotid artery and the occurrence of septic thrombophlebitis, and finally sepsis. Rupture of the carotid artery is not a rare occurrence either. Death occurs due to suffocation, sepsis, mediastinitis or aspiration pneumonia [1, 11].

From the study by Almutairi et al., published in 2020, we see that the largest number of patients complained of neck pain (59.6% of them), followed by dysphagia (43.7%), and

toothache (42.6%) [12]. Also, a study by Bottin et al. showed similar results. Namely, 71.1% of patients complained of neck pain, odynophagia was present in 54.2% of cases, and dysphagia in 51.8% [13]. Our patient was no exception. Neck swelling was preceded by a toothache, followed by tongue swelling, trismus and difficulty breathing, which is not a common symptom and can be considered a complication. In the mentioned studies, swelling of the neck as the main symptom was accompanied by fever in almost 50% of cases and trismus in about a quarter of the monitored patients [12]. In our case, the patient did not have a fever.

These dentogenic infections are basically polymicrobial, with the unwritten rule being that the deeper the infection is localized, the more dominant the anaerobic agent is [1]. In the case of our patient, the causative agent of the infection was not isolated.

Immediate hospitalization of such patients is mandatory. Hospital treatment recommends continuous use of broad-spectrum antibiotics and corticosteroids, securing the airway (diagnostic endoscopy, intubation, or tracheotomy in case of stridor), as well as immediate surgical treatment of the abscess, in terms of incision and drainage. Prevention of the spread of infection, as well as prevention of its recurrence, will be best ensured by timely and energetic therapy that includes the extraction of the tooth causing the infection [12]. In the case of our patient, we were guided by the principles of good medical practice, which resulted in a positive outcome.

Infections of dentogenic origin are unfairly underestimated, even though in a certain number of cases they represent potentially life-threatening conditions. Although in most cases they are resolved during routine dental interventions, insufficient experience of the doctor, non-cooperation of the patient, along with the other listed risk factors can lead to serious complications.

The first association of every doctor is that this condition can cause severe complications such as mediastinitis and sepsis. However, what we wish to emphasize is that the respiratory tract should be considered even at the very beginning, since it can become compromised already in the first days of the development of the infection. Quick reaction of the doctor, immediate hospitalization, and observation, even in situations where the airway obstruction still has not developed as much, are the key steps in the treatment of such conditions. If some of the recommended steps are skipped, a seemingly insignificant dentogenic infection can lead to a fatal outcome. Finally, we would like to emphasize that health education, care for physical and mental health, and especially oral hygiene are not only a matter of individual culture, but also of modern society to which we strive.

Conflict of interest: None declared.

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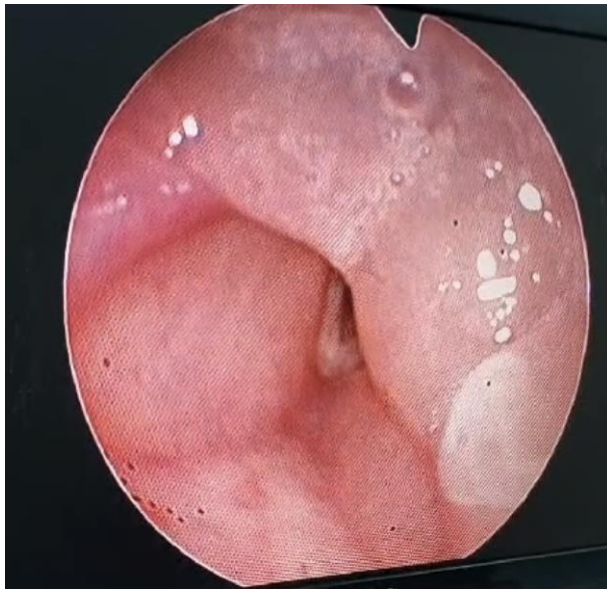


Figure 1. Massive edema of the left aryepiglottic fold with narrowing of the breathing space and suppressing the ipsilateral pyriform sinus



Figure 2. The finding corresponds to a dental abscess with parapharyngeal propagation to the left

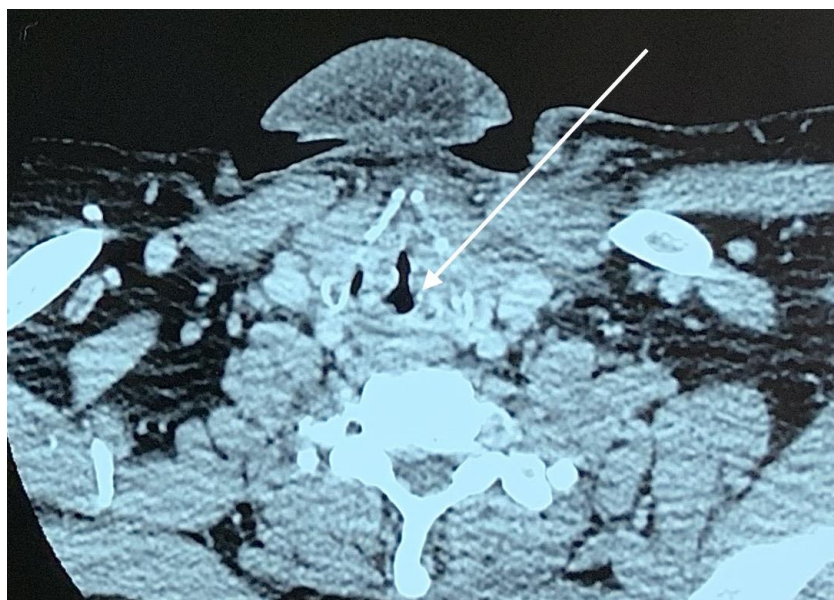


Figure 3. CT image of the massive edema of the left aryepiglottic fold with reduced breathing space