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ABOUT COVER

Editor-in-Chief of *World Journal of Otorhinolaryngology*, Amr El-Shazly, MD, PhD, Professor, Department of Otorhinolaryngology, Liege University Hospital, Liege 4000, Belgium

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World Journal of Otorhinolaryngology (*World J Otorhinolaryngol*, *WJO*, online ISSN 2218-6247, DOI: 10.5319) is a peer-reviewed open access academic journal that aims to guide clinical practice and improve diagnostic and therapeutic skills of clinicians.

The *WJO* covers topics concerning endoscopy, rhinology, pharyngology, laryngology, tracheo-esophagology, otology, tracheology, cancer, nasal symptomatology, congenital nasal diseases, inflammatory diseases of the external nose, rhinitis, allergic rhinitis, nasal polyps, nasal septal diseases, nasal bleeding, nasal or sinus foreign bodies, sinusitis, rhinogenic complications, diagnostic imaging, evidence-based medicine, epidemiology and nursing. Priority publication will be given to articles concerning diagnosis and treatment of otorhinolaryngologic diseases. The following aspects are covered: Clinical diagnosis, laboratory diagnosis, differential diagnosis, imaging tests, pathological diagnosis, molecular biological diagnosis, immunological diagnosis, genetic diagnosis, functional diagnostics, and physical diagnosis; and comprehensive therapy, drug therapy, surgical therapy, interventional treatment, minimally invasive therapy, and robot-assisted therapy.

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EDITORIAL

CX3CR1 receptor as a potential therapeutic target in chronic rhinosinusitis and allergic rhinitis

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Abstract

Chronic rhinosinusitis and allergic rhinitis are chronic inflammatory diseases that affect the mucous membrane of the nose and paranasal sinuses. These diseases are characterized by recruitment of inflammatory cells to the upper airway. For this to take place a complex interaction between inflammatory cells and the cytokines/chemokines (ligand) liberated at the site of inflammation is involved in a process termed chemotaxis or directed cell migration against concentration gradient of the ligand. This entails signal transduction through the cell surface receptor resulting in cellular functional response and directed migration. In this editorial the novel role of CX3CR1 receptor in the immunopathology of chronic inflammation of the nose and paranasal sinuses will be explored with its potential role as therapeutic target in chronic nasal inflammation.

Key words: Nasal Inflammation; CX3CR1 receptor; Chronic rhinosinusitis; Allergic rhinitis; Therapeutic modalities

Core tip: In this editorial, we explore the role of CX3CR1 as therapeutic target in diseases, characterized by recruitment of inflammatory such as chronic rhinosinusitis and allergic rhinitis. Both diseases are chronic inflammatory diseases that affect the mucous membrane of the nose and paranasal sinuses. In this editorial, the novel role of CX3CR1 receptor in the immunopathology of chronic inflammation of the nose and paranasal sinuses will be explored with its potential role as therapeutic target in chronic nasal inflammation.

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INTRODUCTION

Chronic rhinosinusitis (CRS) is a disease characterized by chronic inflammation of the mucous membrane lining the nose and paranasal sinuses. The immunological response of CRS is biased towards T_{H1} immunological response and its cytokine profile. On the other hand, allergic rhinitis (AR) is another form of chronic inflammation of the mucous membrane of the nose. It is broadly classified as seasonal (intermittent) or perennial (persistent). Here the immunological response is biased towards T_{H2} and its cytokine profile. However, both diseases; CRS and AR, may coexists especially in poorly controlled AR that is complicated by sinuses. Both diseases demonstrate leukocytes infiltration at the mucosal level. This entails a complex interaction between the ligand (chemokine or cytokine) and its specific receptor in the cell surface resulting into specific signal transduction and hence chemotaxis or infiltration of the inflamed tissue by inflammatory leukocytes.

CX3CR1 is the receptor for its ligand CX3CL1 or fractalkine and also for eotaxin 3[1,2] that is involved in adhesion and migration of leukocytes[3]. CX3CR1 is expressed by lymphocytes, natural killer (NK) cells and monocytes. It is recently shown to be also expressed by human neutrophils in CRS and CRS associated with airways allergy[4]. This further highlights the importance of this receptor in recruiting important types of inflammatory cells to the inflamed nose in CRS and/or AR. In the lower airway, CX3CR1 receptor signaling promotes T_{H2} survival in the inflamed lungs[5] and promotes asthma inflammation.

CX3CR1 seems to be significantly upgraded in allergy of the airways. It was shown that allergen challenge upregulates the function of CX3CR1 in peripheral blood NK cells in AR patients and that NK cell infiltrated the epithelial layers of nasal tissue only in patients with CRS with allergy[2]. Likewise, infiltrating neutrophils to the sub-epithelial layer of nasal mucosa showed maximum expression of CX3CR1 in CRS patients with combined airway allergy; AR and asthma[4]. Interestingly, CX3CL1 or fractalkine was also shown to be expressed by inflammatory cells infiltrating the inflamed nasal tissue in CRS with AR. A maximum expression of fractalkine was seen in CRS patients with combined airway allergy; AR and asthma[2].

Although there are only few reports in the literature reporting on this novel receptor role in the nasal inflammation, it is clear from the existing evidences that CX3CL1/CX3CR1 axis in CRS and AR, play a pivotal role in promoting the chronicity of inflammation in CRS and AR patients, through inflammatory cells recruitment. It is interesting the predominance towards T_{H2} immunological response for the NK cells and neutrophils in this scenario. This makes this receptor a novel target for CRS with associated combined airways allergy. Therefore, it is mandatory to further explore the signaling of CX3CR1 in CRS and AR. Further studies are required to explore the role of this very important receptor in the pathophysiology of CRS and AR.

While Anti FKN (E6011; KANAb001) is under continuous evolution and evaluation as pharmacological target for CXCL1/CX3CR1 axis in rheumatoid arthritis patients[6], the author believes a similar approach in blocking CX3CR1 would be a promising therapeutic modality in treating inflammation seen in CRS and AR, especially the recalcitrant CRS and severe AR. Blocking of this receptor can attenuate the inflammatory cells influx in the epithelial and sub-epithelial layers of the nasal mucosa of NK cells and neutrophils, respectively.

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CASE REPORT

Effect of intranasal stents (AlaxoLito, AlaxoLito Plus and AlaxoLito Xtreme) on the nasal airway: A case report

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Abstract

BACKGROUND

The study of intranasal stents on the nasal airway is limited in the medical literature. The authors aim to provide objective measurements on their effects on the nasal airway. The aim is to study the feasibility of three novel intranasal stenting devices, AlaxoLito, AlaxoLito Plus, and AlaxoLito Xtreme, as treatment for nasal obstruction.

CASE SUMMARY

A 58- year-old man, who had right sided nasal obstruction, used stents during sporting activities intermittently for four years and subsequently in addition to intermittent sports use regularly for sleep for another two years. Magnetic resonance imaging (MRI) of the nasal passages and rhinomanometric measurements were taken with and without stents *in situ*. The stents tested are all braided from thin nitinol wires. The AlaxoLito Nasal Stent has a length of 35 mm. The AlaxoLito Plus and AlaxoLito Xtreme Nasal Stents have a length of 60 mm. Both have a diameter of about 10 mm in unloaded state and comprise a widened, ball-shaped section (which is positioned at the nasal alar) of about 11 and 14 mm, respectively. Rhinomanometric nasal airflow after application of the stents improved 1.11, 1.23, and 1.38 fold, respectively, with application of the AlaxoLito, AlaxoLito Plus and AlaxoLito Xtreme stents. MRI showed that after application of the stents, the nasal passage increased in diameter.

CONCLUSION

Intranasal stenting shows improvement in nasal airflow. Intermittent and regular longterm use had been shown to be safe, with no discomfort and no side effects.

Key words: Intranasal stent; Airway; Obstructive sleep apnoea; Rhinomanometry; Magnetic resonance imaging

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Core tip: Intranasal stents have a number of applications in the clinical setting, but no current study existed to determine objective measurements. This case shows objective data on rhinomanometry and imaging measurements, to determine long term effects of three types of intranasal stents on the nasal airway.

Citation: Zhang H, Kotecha B. Effect of intranasal stents (AlaxoLito, AlaxoLito Plus and AlaxoLito Xtreme) on the nasal airway: A case report. *World J Otorhinolaryngol* 2019; 8(1): 4-11
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INTRODUCTION

A variety of devices exist to treat nasal obstruction, ranging from nasal valve dilator devices (Max-Air Nose Cones) to external devices (Breathe Right nasal strip), and these have been shown to improve nasal airflow[1].

Nasopharyngeal stenting, which splints open the velopharynx, has been used to treat sleep disordered breathing[2], and plays a role in surgical planning for Obstructive Sleep Apnoea (OSA)[3,4]. Previous studies of nasopharyngeal stenting devices have shown reduced snoring and OSA as evidenced by an improvement in audiometric data and the apnoea-hypopnoea index during sleep[5]. These devices however, are not designed to splint the nasal airway and to improve the nasal breathing.

Fluid dynamics studies of nasal airflow have shown that a larger proportion of airflow is through the middle nasal passage, drawing conclusions that ensuring optimal airflow through this area may provide better laminar flow through the nasal passage[6,7]. This led to the development of intra-nasal stenting devices.

This article presents for the first time, a single case report with patient experience using intranasal stents (Figure 1), the AlaxoLito, AlaxoLito Plus and AlaxoLito Xtreme Nasal Stents. The authors describe symptomatic treatment including changes in objective measurements, imaging and rhinomanometric data.

MEDLINE, Scopus, and the Cochrane Library were searched from inception to September 2018, for treatment of nasal obstruction, rhinitis, snoring with a nasal stenting. Terms searched included the various combinations of nasal obstruction and nasal trumpet, stent, obturator, airway or tube. There were no studies in Medicine describing the use in nasal obstruction or rhinitis.

CASE PRESENTATION

A 58 years old healthy male volunteer enrolled into clinical testing for the use of three intranasal stents (AlaxoLito, AlaxoLito Plus and AlaxoLito Xtreme). The patient had a previous tonsillectomy, and no other significant past medical history. Symptomatically he had mild right sided nasal obstruction. On clinical examination, he had a mildly deviated nasal septum to the right. Anterior rhinomanometry (Flowhandy ZAN 100 USB, ZAN Meßgeräte GmbH, Oberthulba, Germany; flow at 150 mbar pressure difference) showed a baseline of 370L/min on the right and 380L/min on the left nasal passage.

FINAL DIAGNOSIS

Right nasal septal deviation associated with mild non-allergic rhinitis.

TREATMENT

The subject applied the intranasal stents for a total of 6 years, starting in 2012. All stents always have been inserted into the middle nasal passage. The application of these stents was performed by the subject themselves. The stents have an insertion tube which facilitates insertion, which is removed after the stents are in a comfortable

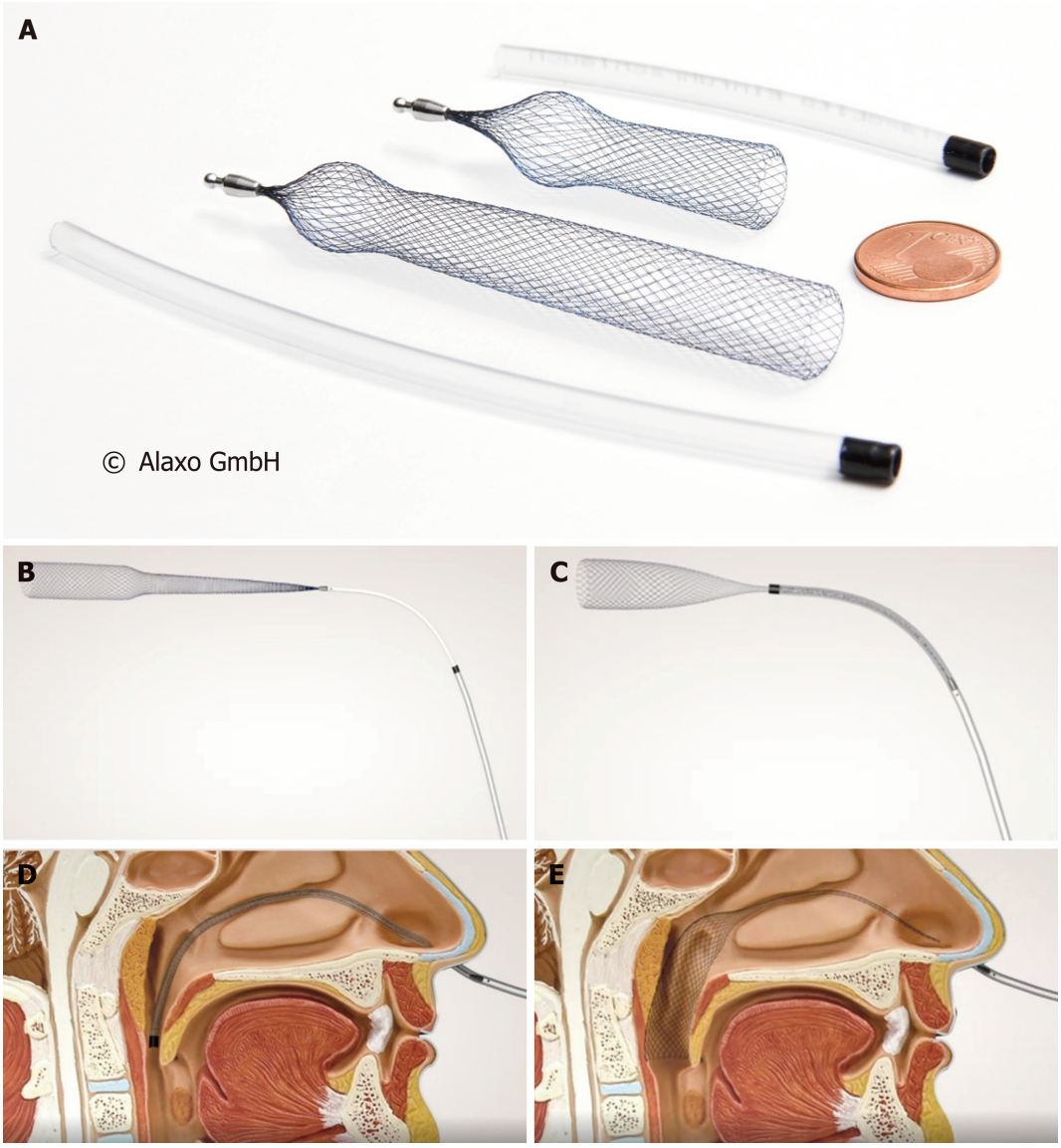

Figure 1 AlaxoLito (A) and AlaxoLito Plus (C-E) Nasal Stents with insertion tube. Insertion method: B and C: The stent is placed inside the insertion tube, which allows for easy insertion; D: Once the insertion tube is placed into the nasopharynx; E: It is withdrawn, allowing the stent to open inside the nasal cavity.

position. For the first four years, the subject applied the stents only during exercise and sporting activities, AlaxoLito Plus on the left nasal passage and AlaxoLito on the right nasal passage, due to the mild septal deviation. The AlaxoLito positioned just in front of the septum deviation. After regular nightly use of these stents for determination of the effects during sleep from May 2016 on, after two weeks the subject noticed he could insert the longer AlaxoLito Plus stent also on the right side, initially only in the morning, and after about two months around the clock, and without pain. Since then, the subject used the AlaxoLito Plus on both sides. In May 2018 the subject started using a new stronger variant of the AlaxoLito Plus (the AlaxoLito Xtreme) on both sides.

OUTCOME AND FOLLOW-UP

Effects on rhinomanometry

Subjectively, the patient experienced an improvement in nasal airflow with application of the stent, especially during physical activity, but also during sleep. He also reported an improvement in nasal airflow when applied during an episode of upper respiratory tract infection.

Nasal rhinomanometry was performed without and with the application of the three stent types. Mean measurements were taken to account for the nasal cycle. The results are shown in Figure 2. An increase in nasal airflow from 751 L/min to 832

L/min was observed with AlaxoLito stent. A further increase to 925 L/min and 1036 L/min, respectively, was measured with the application of the AlaxoLito Plus and AlaxoLito Xtreme stents. The subject reported that inspiration was hampered by the olive of the rhinomanometry system starting with the AlaxoLito Plus and the more for the AlaxoLito Xtreme, due to a reduced diameter of the possible air inflow into the nasal valve. Thus, the real values and factors of increase seem to be higher than measured.

Ten horizontal and vertical points of measurements of the nasal airway were taken in the coronal plane, and the mean of these calculated. Using this method, an increase in the diameter of the middle nasal airway of 1mm was seen from 2015 to 2017 without the nasal stents *in situ*, and of 2.3 mm with the AlaxoLito Plus *in situ*. The strongest widening occurred at the position of the septum deviation in the right nasal passage with 4.1 mm. With the AlaxoLito Xtreme additional widening and an almost round shape of the air canal was observed.

Imaging studies

Multiple magnetic resonance imaging (MRI) studies were performed. Figure 3 shows a comparison of coronal MRI of the nasal passages prior to regular nightly use of the stents (December 2015) and with about 1 year post regular AlaxoLito Plus stent application (June 2017). A clear improvement by widening of the nasal airway post regular stent usage is seen on the MRI, especially on the right side, as evidenced by the increase in the black area of the middle nasal passage representing the air canal provided by the stent (red arrows). A comparative MRI was performed with the stents *in situ*. The yellow arrows show a compression effect they have on the middle turbinates. Further, a certain reduction by rounding of the probably cartilaginous structure of the septum deviation is observable (red arrows).

Summary comparisons of the middle nasal airway without and with stents are shown in Figure 4. Without stents, the middle nasal airway has improved on both sides from December 2015 to June 2017, and again in September 2018. With the stents *in situ*, a clear improvement by increase of the cross-sectional area in the middle nasal airway is shown, with the shape having become more or less round due to the application of the AlaxoLito Xtreme.

In Figure 5 axial and sagittal planes in MRI are shown for September 2018 with the AlaxoLito Xtreme stents *in situ*. It is evident that the stents create a rather consistent airway of about 6.5 mm diameter.

Table 1 lists the diameters of the right nasal passage at ten distinct points along its entire length. The values measured demonstrate (1) a clear increase in width and height of the nasal passage over its entire length without stent *in situ* (comparison 2015-12 to 2017-05), (2) an additional clear increase in width and height of the nasal passage over its entire lengths when comparing without and with stent *in situ* (2017-05), and (3) that the narrow space at the position of the local septum deviation (positions number 6 to 8) has been significantly widened (2017-05) without the stent *in situ* so that a more uniform shape of the nasal passage results. Insertion of the AlaxoLito Plus leads to a rather consistent tubular shape of the nasal passage. A similar result in terms of widening of the nasal passage has been obtained for the left side (data not shown).

DISCUSSION

Although nasopharyngeal airway tubes have been used by both anaesthesiologists and otolaryngologists to treat upper airway obstruction at the level of the velopharynx, the treatment of nasal airway obstruction using stenting devices has limited prior research.

The AlaxoLito, AlaxoLito Plus and AlaxoLito Xtreme stents are braided from thin nitinol wires, a memory metal. For the patient in this case report, all stents provided a subjective improvement in nasal breathing, with stepwise increasing effect from AlaxoLito to AlaxoLito Plus to AlaxoLito Xtreme.

Rhinomanometric measurement results improved stepwise with application of AlaxoLito to AlaxoLito Plus to AlaxoLito Xtreme. The longer AlaxoLito Plus stent was initially not insertable on the right side, where the patient had mild obstruction secondary to septal deviation, as the 4 mm diameter insertion tube could not pass the narrow position at the septum deviation, instead causing pain on the septum mucosa. The intermittent and irregular use of the shorter AlaxoLito mainly during sports, each time for some hours, over four years did not change this situation. However, after regular nightly use of the AlaxoLito stent for two weeks, the patient was able to insert the longer AlaxoLito Plus on the right side without pain and discomfort, indicating

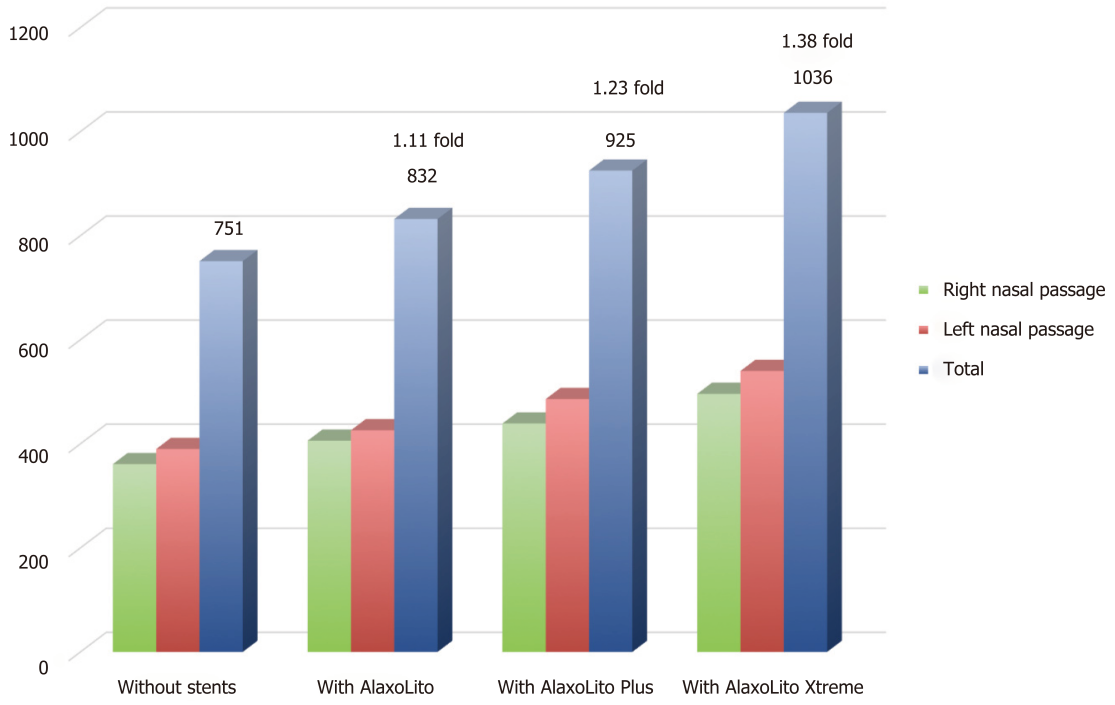

Figure 2 Rhinomanometric data without and with stents.

there may be cumulative effects on opening the nasal airway by regular nightly application for about 8 h per night. Initially this effect lasted for 2 to 3 h only. After about 2 mo this effect lasted around the clock so that the longer AlaxoLito Plus could be inserted also in the evening before sleep and was used regularly since then.

The use of the stronger version (the AlaxoLito Xtreme) again within 2 to 3 mo led to a further widening of the middle nasal airway at the position of the septum deviation. Although there was no resolution of the septal deviation, the nasal airway on the narrow side improved with measurements on MRI. This suggests that a period of about 2-3 mo of use is necessary to achieve the permanent widening effects which are visible in the MRI. All MRI studies have been performed in the evening, that means at least 10 h or more after removal of the stents from the nose in the morning.

An increase of the diameter of the middle nasal passage of 1 mm without stents and 2.3 mm with stents *in situ* was observed after one year of regular use of the AlaxoLito Plus stent spanning across this septum deviation position. 5.5 to 6.5 mm hydraulic diameter is considered to be the normal range of the nasal passage for good nasal breathing[8]. With the stents an almost ideal tube has been reached, so that the hydraulic tube diameter values should be directly applicable for comparison.

This is the first case report to examine the effects of intranasal stents in the treatment of nasal obstruction, using rhinomanometric data and MRI. Clear improvement in the nasal airway is shown on MRI, both with the stent *in situ*, and after regular nightly use of the stent for one year in the first step (AlaxoLito Plus) and 2-3 mo in the second step (AlaxoLito Xtreme). Further imaging and rhinomanometric data would be helpful after the patient ceases stent use, to analyse whether the stents have a longterm lasting effect on the nasal airway.

Although the patient reported subjective improvement in sleep quality, however more objective data would be needed to quantify this. Occasional cell phone app recordings (SnoreLab) indicated almost complete abolishment of occasional weak snoring finally with the AlaxoLito Xtreme. However, the authors did not systematically study the effect of these intranasal stents on snoring or sleep apnoea, and further studies would be warranted to study their effects on sleep.

Table 1 Diameters of the right nasal passage at ten distinct points along its entire length (from anterior to posterior)

2015-12 without stent *in situ*			2017-05 without stent *in situ*			2017-05 with AlaxoLito Plus		
Width(mm)	Height(mm)	Sum(mm)	Width(mm)	Height(mm)	Sum(mm)	Width(mm)	Height(mm)	Sum(mm)
4.54	3.80	8.34	4.78	4.55	9.33	5.63	6.04	11.67
3.53	4.53	8.06	3.97	5.51	9.48	5.66	6.25	11.91
2.01	4.88	6.89	3.18	5.73	8.91	4.62	5.90	10.52
2.27	4.21	6.48	3.59	6.06	9.65	4.61	5.88	10.49
3.50	3.53	7.03	3.77	5.06	8.83	5.49	6.64	12.13
3.37	1.79	5.16	4.72	3.19	7.91	6.64	6.07	12.71
3.22	2.56	5.78	3.61	3.53	7.14	6.50	5.67	12.17
2.70	4.79	7.49	3.95	6.53	10.48	6.35	6.89	13.24
3.29	4.93	8.22	5.03	5.40	10.43	6.51	6.76	13.27
5.65	4.00	9.65	7.27	5.16	12.43	7.58	7.15	14.73

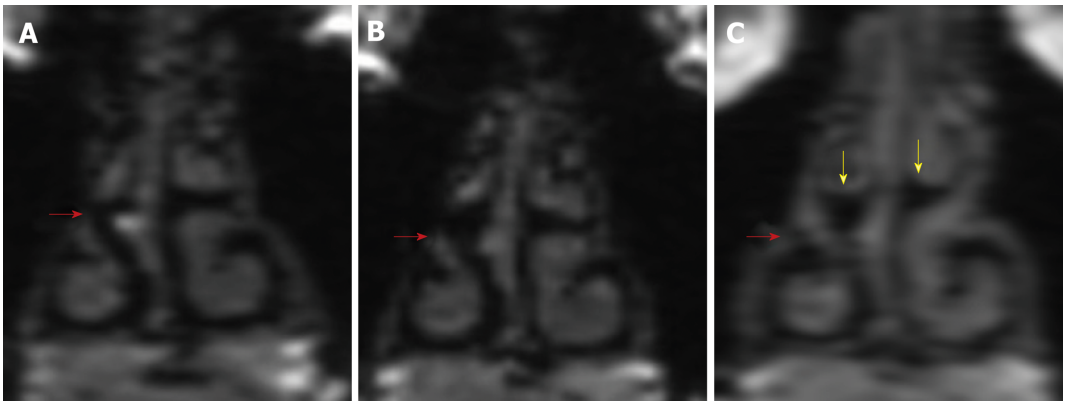

Figure 3 Comparison of coronal magnetic resonance imaging of nasal passages prior to and 1 year post regular stent use. A: Magnetic resonance imaging (MRI) from December 2015 prior to regular use of stents, without stents; B and C: MRI from June 2017 one year post regular use of AlaxoLito Plus stent (B: without stents *in situ*; C: with AlaxoLito Plus *in situ*).

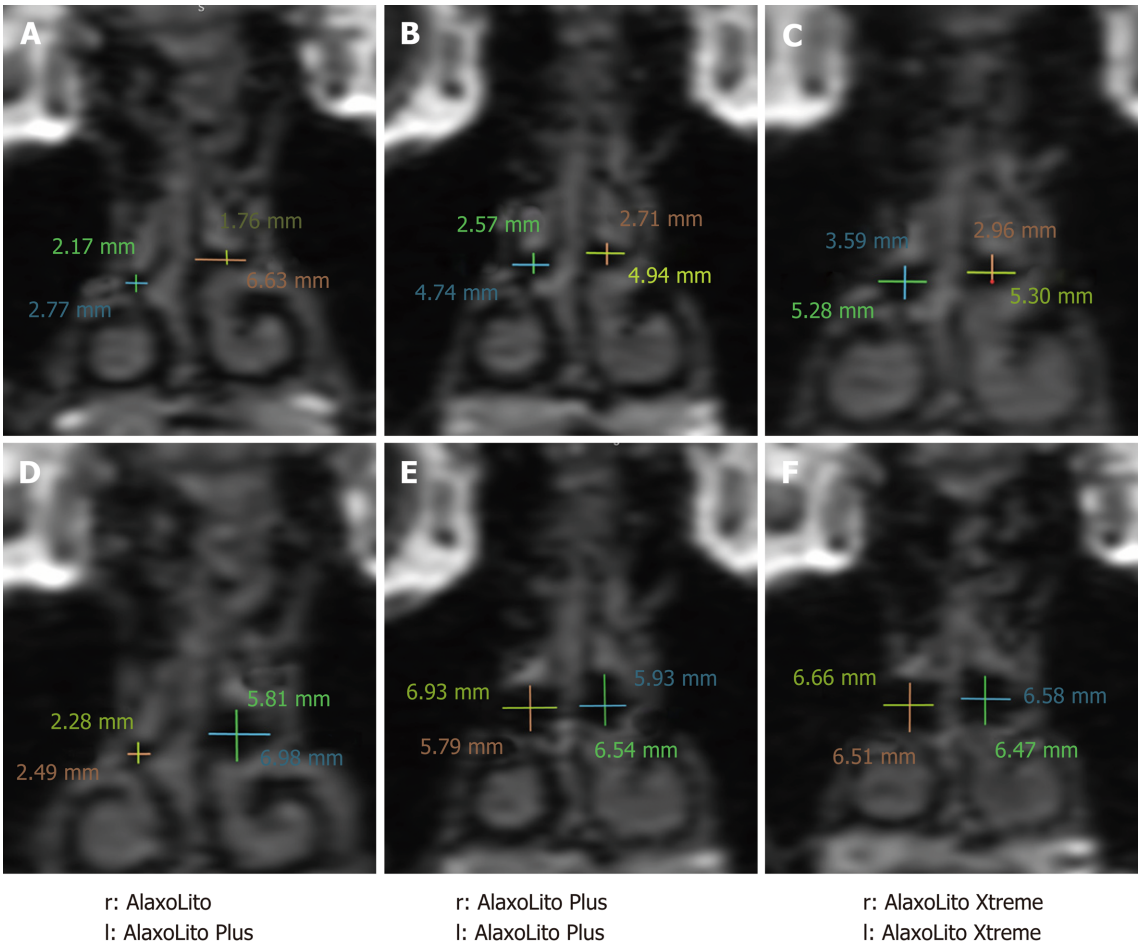

Figure 4 Coronal magnetic resonance imaging showing measurements of middle nasal passage without stents (A-C) and with stents *in situ* (D-F). A and D: Magnetic resonance imaging (MRI) from December 2015 prior to regular use of stents; B and E: MRI from June 2017 one year post regular use of AlaxoLito Plus; C and F: MRI from September 2018, 3.5 mo post regular use of AlaxoLito Xtreme.

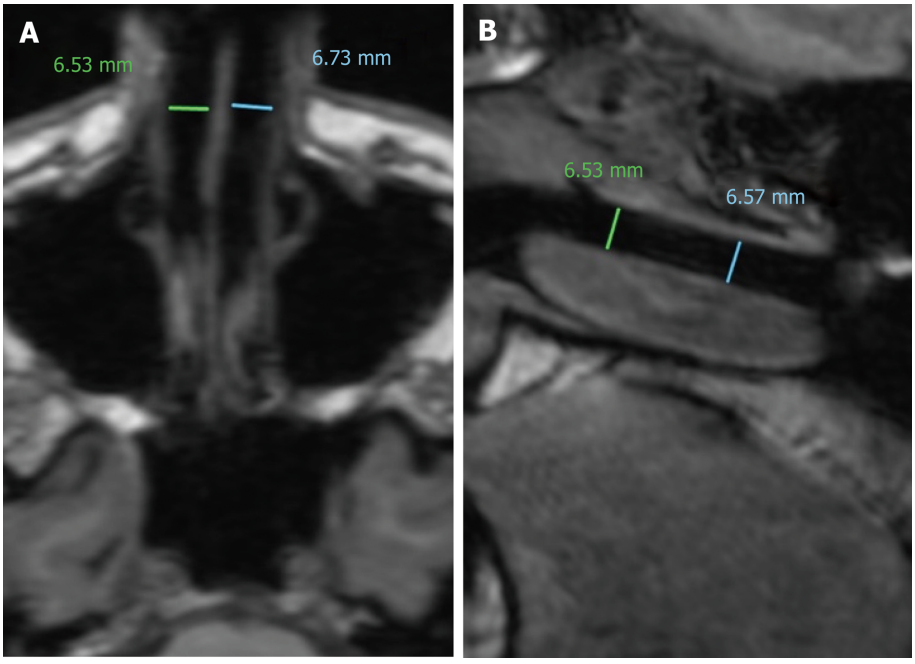

Figure 5 Axial (A) and sagittal (B) magnetic resonance imaging (from September 2018) showing measurements of middle nasal passage with AlaxoLito Xtreme stents *in situ*.

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CASE REPORT

Synovial osteochondromatosis of the temporomandibular joint: A case report

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Author contributions: Romero N drafted the initial manuscript, reviewed and revised the manuscript and participated in the ancillary care of the patient as a clinical assistant; Mulcahy CF reviewed the and contributed to the manuscript; Barak S was the patient's clinical pathologist and as such analyzed and interpreted the pathologic process of the disease and reviewed the manuscript; Shand MF reviewed, revised and formatted the manuscript; Badger CD contributed to, reviewed, and revised the manuscript; Joshi AS was the patient's head and neck surgeon who developed the treatment plan, cared for the patient in the follow up period, reviewed and revised the manuscript; All authors approved the final manuscript for submission.

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Abstract

BACKGROUND

Synovial osteochondromatosis is a rare but benign condition that can result in significant impairment of joint functionality. This case report documents an uncommon presentation of this disorder occurring within the temporomandibular joint, causing the patient significant pain, trismus, and difficulty with daily activities such as eating and speaking. A review of the literature including disease mechanisms and previously documented cases is included to provide comprehensive background for clinical decision-making.

CASE SUMMARY

A 48-year-old male patient presented with a 3-mo history of trismus, crepitus with jaw movement and significant pain while chewing. Physical examination revealed a firm mass and tenderness to palpation at the right temporomandibular joint. Further workup revealed a bilobed mass extending into the joint space as well as significant bony erosion of the glenoid fossa. The patient underwent mass excision with joint reconstruction and pathology revealed synovial osteochondromatosis. The patient reported significant improvement in his symptoms postoperatively.

CONCLUSION

This report outlines the investigative approach and treatment course of synovial osteochondromatosis. The positive outcome following surgical intervention in this case emphasizes the importance of interdisciplinary collaboration and the potential for improvement in quality of life of this patient population.

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Core tip: Synovial osteochondromatosis is a rare condition that arises from metaplasia and proliferation of synovial cells lining a joint space, which can impair joint function. It can be easily overlooked when working up non-specific symptoms such as preauricular pain and swelling. We present the case of a 48-year-old man with a history of significant unilateral temporomandibular joint (TMJ) pain, trismus, and dysphagia who was found to have synovial osteochondromatosis of the TMJ with erosion of the glenoid fossa. The patient underwent resection of the mass and TMJ reconstruction with a rib cartilage graft, resulting in alleviation of symptoms.

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INTRODUCTION

Synovial osteochondromatosis is a condition in which the synovial cells lining a joint space undergo metaplasia and proliferation, resulting in nodular masses, which may ossify within the joint capsule. Ultimately several loose or pedicled bodies form within the joint and cause significant symptoms and impairment of joint functionality. Affected regions most commonly occur within the axial skeleton, including the knee or shoulder capsule[1]. The temporomandibular joint (TMJ) is rarely affected and has only been documented in about 100 cases over the last 20 years[2,3]. Etiology of this condition is unknown, but it is hypothesized to involve abnormal expression of specific growth factors and cytokines[4,5]. This disease is generally monoarticular and tends to present in adults. Involvement of the TMJ is more common in women than men, most often occurring between the ages of 30 and 60[6]. Although considered a benign condition, synovial osteochondromatosis carries a risk of aggressive and progressive bone erosion which can extend intracranially and even invade the dura[3]. There is also a small incidence of malignant transformation to chondrosarcoma[7].

Patients with synovial osteochondromatosis of the TMJ often present with unilateral pain, malocclusion, and swelling[8-10]. Evaluation of these patients should include comprehensive history, physical, imaging studies, and cytological studies of the joint capsule tissue or fluid. Definitive treatment for synovial osteochondromatosis is surgical removal of the nodules or loose bodies from the joint capsule and often partial synovectomy[11]. Removal of the growths relieves symptoms as well as decreases the risk of possible progression to chondrosarcoma and further degeneration of the joint. In cases with concurrent bone erosion, surgical reconstruction of the mandible or temporal bones may also be required[3].

CASE PRESENTATION

Chief complaints

A 48-year-old male patient was referred for otolaryngology evaluation by his dentist after complaining of trismus, crepitus with jaw movement, significant pain with chewing, and development of a right-sided facial mass.

History and physical exam

The patient's symptoms had started about three months ago and he had seen several physicians previously who provided reassurance and conservatively managed his symptoms. The pain was localized to the right side and aggravated by jaw movements, particularly opening the mouth. He had no constitutional symptoms. Clinical examination revealed a firm mass in the pre-auricular region with tenderness to palpation at the right temporomandibular joint. No obvious facial asymmetry was

present at rest. All cranial nerves appeared intact.

Imaging examinations

Several imaging studies were obtained. A CT scan of the face demonstrated a bilobed mass extending into the joint space on either side of the mandibular condyle measuring 1.7 cm by 1.2 cm medially and 2.6 cm by 1.6 cm laterally (Figure 1). Additional features included significant bony erosion of the middle fossa floor and glenoid fossa, as well as several cysts present in the subchondral bone on the temporal side of the TMJ. T1 and STIR magnetic resonance imaging (MRI) sequences also indicated marked joint capsule distention and joint effusion (Figure 2). The mass appeared to be well circumscribed and did not appear to invade adjacent soft tissue structures.

Laboratory examinations

A fine needle aspiration biopsy was performed, which revealed a matrix-producing lesion with osteoclast-like giant cells and cellular atypia of undetermined significance.

FINAL DIAGNOSIS

The final diagnosis of the presented case is synovial osteochondromatosis of the temporomandibular joint with significant erosion of the glenoid fossa.

TREATMENT

Surgical intervention was indicated for this patient given his failed response to medical management and aggressive nature of bony erosion displayed on imaging. The patient underwent surgical removal of the mass and reconstruction of the joint. A pre-auricular incision was made and a pedicled temporoparietal flap was used to fully expose the condylar mass and joint capsule. The TMJ capsule was opened and revealed well-formed pieces of cartilage which were removed. Circumferential subperiosteal resection of condylar neck was performed to completely remove the adherent mass. A freer elevator was utilized to circumferentially dissect the mass off of the condyle. Sharp excision was required to free the mass from surrounding soft tissue. Care was taken to preserve the external carotid artery. The mass was delivered in a complete, but piecemeal fashion. It became apparent that the mass had filled the superior aspect of the infratemporal fossa and extended into the parapharyngeal space. Following the resection, a rib cartilage graft was used to resurface the superior aspect of the glenoid fossa. The temporoparietal flap was wrapped around the cartilage graft for vascular support and the graft was inset into the joint defect. The condyle itself was preserved adequately preserved during excision of the mass so that condyle reconstruction was not necessary. Specimens acquired from within the TMJ capsule (Figure 3), the mandibular condyle, and surrounding soft tissue were sent for intraoperative frozen-section pathology analysis which showed no evidence of malignancy.

OUTCOME AND FOLLOW-UP

The postoperative course of this patient was uncomplicated. At his first post-operative visit two weeks after surgery, he reported significant improvement in symptoms, and his facial movement was symmetric. He was tolerating a soft diet with only moderate soreness. Physical exam revealed residual swelling and a well-healing facial incision. He was referred for physical therapy for restoration of joint motility. Final pathology of surgical specimens showed lobules of cartilaginous tissue, lacking cytologic atypia, hypercellularity or mitosis, consistent with synovial osteochondromatosis (Figures 4, 5). Unfortunately, after his first post-operative visit, the patient was lost to follow up.

DISCUSSION

Synovial osteochondromatosis is a non-neoplastic proliferation of joint cartilage, which results in the formation of nodular and sometimes ossified bodies within the joint space. Dysregulated tissue growth factors and cytokines such as TGF-B3, FGFR-3 and IL-6 are involved in the proposed mechanism of over-stimulating active

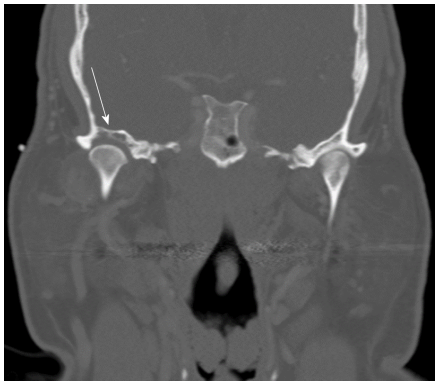

Figure 1 Coronal cut of computed tomography image (bone window) highlighting the erosion of the right glenoid fossa (arrow). A soft tissue mass can faintly be observed on either side of the mandibular condyle, which is better delineated in a soft tissue window (not pictured).

chondrocytes to proliferate and undergo metaplasia[4,5]. The disease process can be divided into several stages of growth: (1) Proliferation of the synovial lining resulting in nodular pedicled growths; (2) Release of the nodules into the joint space, detachment from the synovium; and (3) Ossification of the nodules within the joint space, with concurrent decline in synovial proliferation[12]. Symptoms most often present in the second and third phase of growth when the bodies become loose within the joint, ossify and cause impairment of joint functionality. Impairment of cranial nerve function generally indicates most advanced disease[6,8,10].

Although synovial osteochondromatosis rarely occurs in the facial bones, the TMJ is an important region of occurrence, which requires special consideration when evaluating patients with a pre-auricular mass or symptoms of malocclusion, crepitus and jaw pain. In many cases, patients experience symptoms for years before correct diagnosis can be made and surgical intervention performed[9]. Presenting complaints are often vague or non-specific, and late diagnosis occurs frequently due to symptom overlap with more common conditions such as temporomandibular joint disorder. Physical findings may include swelling and tenderness to palpation in the preauricular area, as well as decreased range of motion. Collaboration among colleagues in primary care, dentistry and surgery is therefore crucial to the timely diagnosis and proper management of these patients. Degenerative arthritis is a common complication due to the repeated injury of the joint[13,14]. Delay in diagnosis has the potential to cause significant complications and morbidity for the patient due to the possibility of local or intracranial invasion as well as permanent loss of joint function[8,11]. Invasion into the cranium can also result in permanent cranial nerve injuries, particularly facial nerve deficits[15]. The differential diagnosis of a mass within the TMJ includes many types of neoplastic growths and it is therefore important to rule out malignancy when evaluating these patients.

Diagnosis of synovial osteochondromatosis of the TMJ requires multiple imaging modalities, typically including CT and MRI. Common findings include expansion and effusion within the joint space as well as cartilaginous nodules or loose bodies[15]. MRI may be necessary for diagnosis of early disease in which the nodules have not yet ossified or calcified to better characterize joint effusion, capsular swelling and possible disruption of soft tissues[9,16]. Surrounding bony structures should be carefully evaluated to identify the likelihood of intracranial erosion and dural involvement[11,17]. Other degenerative changes such as subchondral cysts may also be evident within the joint, as in this case. Preoperative biopsy for tissue analysis to assess for malignancy is beneficial[3]. Histologic findings can confirm loose bodies within the joint space or pedunculated masses attached to the joint capsule lining[18].

Despite its classification as a benign condition, synovial osteochondromatosis occurring in the TMJ can lead to local erosion and complications from intracranial invasion[11]. In addition, the disorder carries a small risk of progression to malignant chondrosarcoma, and therefore prompt diagnosis and treatment is crucial[7,8]. Surgical intervention remains the most appropriate treatment due to the lack of effective medical management and the potentially aggressive nature of the disease process. Both arthroscopic and open technique surgeries have been utilized in the process of mass removal. Arthroscopic approaches are less morbid and may be useful for diagnosis, however an open arthrotomy may be more appropriate if the masses are large or numerous[19,20]. Cases with intracranial invasion typically require open surgical excision and reconstruction[3,15]. In this case as well as others, the extent and

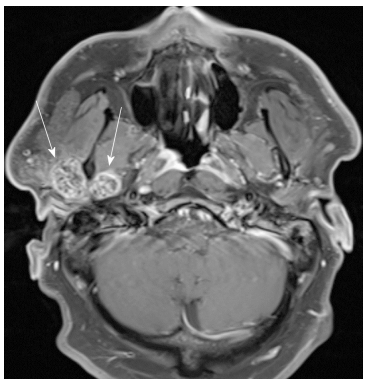

Figure 2 Axial cut of magnetic resonance imaging T1 weighted post-contrast image depicting the bilobed mass within the temporomandibular joint space on either side of the mandibular condyle (arrows) with associated joint capsule distention. On magnetic resonance imaging STIR sequence images (not pictured), significant joint effusion is noted.

stage of the disease may require condylectomy and reinforcement of the glenoid fossa to restore proper structural support[3].

Outcomes following surgical intervention reported in the literature have been overwhelmingly positive. Patients tend to report significant improvement in symptoms and reduction in pain[8,15]. Patients benefit from postoperative physical therapy to improve joint motility and help correct any previous malocclusion. Regular follow up should be encouraged for these patients due to the reported recurrence rate of approximately 11%[12]. Recurring disease should increase clinical suspicion for malignant process and careful re-evaluation should be performed.

CONCLUSION

Synovial osteochondromatosis is a rare condition and is easily overlooked when working up non-specific symptoms such as preauricular pain and swelling. The lack of response to medical management and the positive outcomes following surgical intervention reinforce the potential benefit to patients suffering from this treatable disorder. Treatment not only provides symptomatic relief and improved quality of life, but also decreases likelihood of progressive joint injury, intracranial erosion, and malignant transformation.

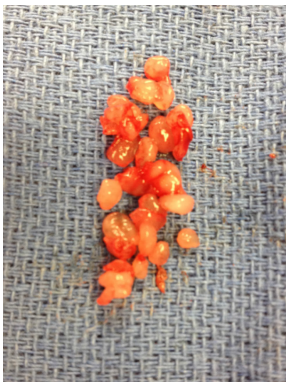

Figure 3 Multiple, irregular firm cartilaginous fragments removed from the temporomandibular joint space.

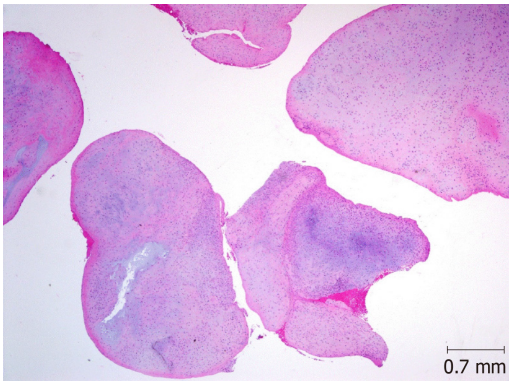

Figure 4 Multiple separate nodules of cartilaginous tissue (loose bodies) (HE, × 20).

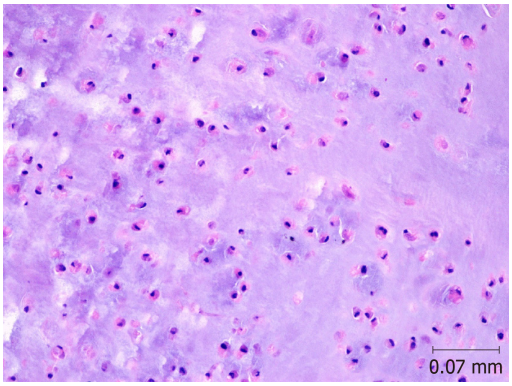

Figure 5 Lobules of hyaline cartilage. No atypia, hypercellularity or mitosis are noted (HE, × 200).

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