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Adenomatoid odontogenic tumour – two case reports

Guz gruczolakowato-odontogenny – opisy dwóch przypadków

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Summary

Adenomatoid Odontogenic Tumour (AOT) is an uncommon, benign odontogenic tumour affecting predominantly young females and occurring typically in the anterior maxillary region. This paper aims to report two clinical cases of AOT, emphasizing the clinical, radiographic, and histopathological features of this tumour.

The first case involves a 24-year-old male who presented to the Oral Medicine and Oral Surgery Department with a hard, painless gingival swelling in the right maxillary canine area. The second case concerns a 30-year-old male patient with epilepsy, who was referred by his orthodontist following the incidental radiographic finding of an impacted canine associated with a mixed radiolucent-radiopaque lesion. Both patients underwent surgical enucleation of the lesions. The diagnosis of AOT was confirmed with histopathological examination in each case.

AOT is a slowly growing, often asymptomatic tumour, frequently associated with impacted teeth, especially maxillary canines. Management typically involves conservative surgical removal, with a low risk of recurrence.

These cases underscore the importance of early detection and accurate diagnosis, and emphasize the importance of clinical and radiographic evaluation to ensure effective management of this rare odontogenic neoplasm.

Streszczenie

Guz odontogenny gruczolakowaty (AOT) to rzadki, łagodny guz, który dotyka głównie młode kobiety i występuje zazwyczaj w przednim odcinku szczęki. Niniejsza praca ma na celu przedstawienie dwóch przypadków klinicznych AOT, ze szczególnym uwzględnieniem cech klinicznych, radiologicznych i histopatologicznych tego guza.

Pierwszy przypadek dotyczy 24-letniego mężczyzny, który zgłosił się do Kliniki Medycyny i Chirurgii Stomatologicznej z powodu twardego, niebolesnego obrzęku dziąsła w okolicy prawego kła szczęki. Drugi przypadek dotyczy 30-letniego pacjenta z padaczką, który został skierowany przez ortodontę po przypadkowym stwierdzeniu na zdjęciu radiologicznym zatrzymanego kła z towarzyszącą zmianą radiologicznie mieszaną – przezierną i nieprzezierną. U obu pacjentów wykonano chirurgiczne wyłuszczenie zmian. Rozpoznanie AOT zostało potwierdzone badaniem histopatologicznym w obu przypadkach.

AOT to wolno rosnący, zazwyczaj bezobjawowy guz, często związany z zębami zatrzymanymi, zwłaszcza kłami szczęki. Leczenie zazwyczaj polega na zachowawczym usunięciu chirurgicznym, z niskim ryzykiem nawrotu.

Przypadki te podkreślają wagę wczesnego wykrywania i skrupulatnej diagnostyki, a także znaczenie oceny klinicznej i radiologicznej dla zapewnienia skutecznego leczenia tego rzadkiego nowotworu odontogennego.

Introduction

Adenomatoid odontogenic tumour (AOT) is a rare benign neoplasm of odontogenic origin that was first described by Steensland in 1905. It was later recognized as a distinct pathological entity by Stafne in 1948. Over the years, AOT has been referred to by various names, including adenoameloblastoma, adamantinoma, epithelioma adamantinum and teratomatous odontoma.[1,2] AOT has undergone several redefinitions in the World Health Organization (WHO) classification over the years, reflecting the evolving understanding of its nature, histopathology and clinical behaviour. In the 1992 WHO classification, AOT was considered a mixed odontogenic tumour associated with odontogenic epithelium and ectomesenchyme, with or without the formation of dental hard tissues.[3] In 2005, the WHO reclassified AOT as a benign epithelial odontogenic tumour, describing it as composed exclusively of odontogenic epithelium without odontogenic ectomesenchyme.[4] In the most recent WHO classification published in 2022, AOT remains categorized as a benign epithelial odontogenic tumour. The WHO highlights its purely epithelial origin, nonaggressive and slow-growing nature, frequent encapsulation and excellent prognosis. Three well-recognized subtypes are described: the follicular type (associated with impacted teeth), the extrafollicular type (not associated with teeth), and the peripheral type (arising in the gingival soft tissues). Additionally, the 2022 WHO classification provides clearer distinctions between AOT and other odontogenic lesions, such as dentigerous cysts and ameloblastomas, to ensure more accurate diagnoses.[5,6] These findings underscore the importance of early recognition and accurate diagnosis, emphasizing the need for a thorough clinical evaluation to ensure the successful management of this rare odontogenic tumour.

Observations

The study was conducted following the CARE Checklist. A written informed consent for patients' information and images to be published was provided by the concerned parties.

Case 1

A 24-year-old male was referred to the Department of Oral Medicine and Oral Surgery for evaluation of a swelling accompanied by displacement of the upper right canine and the first premolar (teeth 13 and 14). His medical, family and dental histories were unremarkable. Extraoral evaluation revealed mild facial asymmetry in the right maxillary region, with no changes observed in the overlying skin. Intraoral examination revealed an oval-shaped, painless swelling located on the attached gingiva of the maxilla. The lesion was firm on palpation and covered by clinically normal-appearing mucosa. It extended mesiodistally from the free marginal gingiva of teeth 12 and 13 to the distal surface of tooth 14. Notably, tooth 13 exhibited distal angulation, while tooth 14 showed mesial angulation (Fig. 1).

A panoramic radiograph and cone beam computed tomography (CBCT) were performed for comprehensive radiological assessment. The panoramic radiograph revealed a large, oval-shaped, well-defined unilocular radiolucent lesion with corticated borders. The lesion extended horizontally from tooth 13 to tooth 14, causing displacement of both teeth. Vertically, it reached toward the alveolar ridge, which remained intact. CBCT imaging confirmed the presence of a well-demarcated, homogeneous, radiolucent lesion measuring approximately 18 mm in height and 13 mm in mesiodistal width. There was no evidence of root resorption, and the sinus floor appeared intact, with no signs of sinus involvement. Displacement of teeth 13 and 14 was observed. Additionally, small

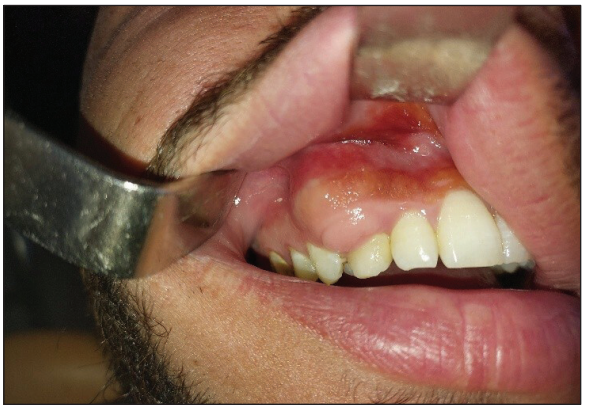

Fig. 1. Intraoral examination revealed an oval-shaped swelling, covered by clinically normal-appearing mucosa, localized on the maxillary attached gingiva. It extended mesiodistally from the free marginal gingiva of teeth 12 and 13 to the distal surface of tooth.

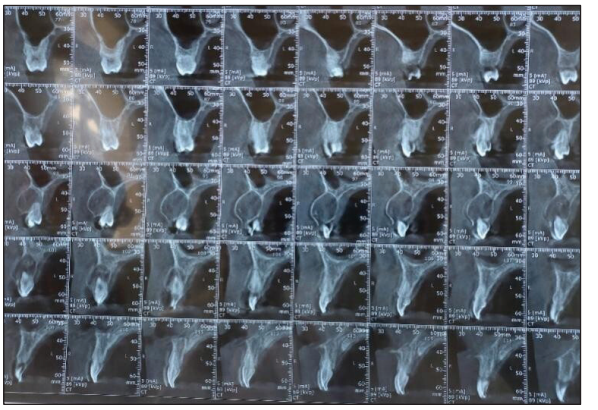

Fig. 2. CBCT scan of the right maxilla showing a well-circumscribed, homogeneous radiolucent lesion estimated to measure 18 mm in height and 13 mm in mesiodistal width, located between the roots of teeth 13 and 14. Note the displacement of both teeth, buccal cortical plate expansion, and the detection of small peripheral foci of calcification within the lesion.

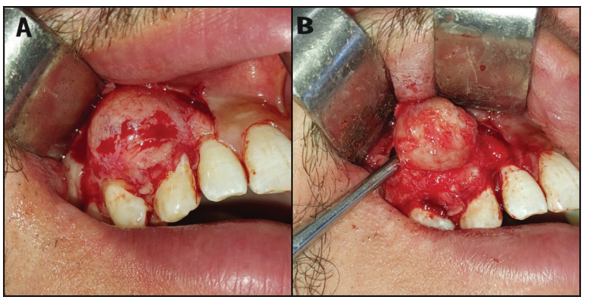

Fig. 3. Surgical procedure: (A) A vestibular mucoperiosteal flap was reflected to expose the lesion. (B) An osteotomy was performed, then, the lesion was enucleated surgically.

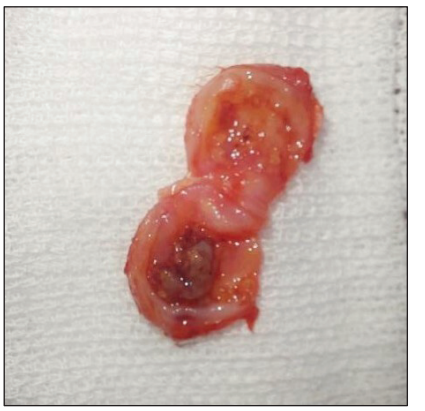

Fig. 4. Cross-sectional inspection revealed intraluminal vegetative growths, the largest of which measured 0.7 × 0.4 cm.

peripheral foci of calcifications were noted within the lesion (Fig. 2).

Under local anaesthesia, a vestibular flap was raised, followed by an osteotomy to access the lesion, which was subsequently surgically enucleated (Fig. 3).

Macroscopic examination revealed a cystic lesion measuring approximately 1.3 cm in diameter. Cross-sectional inspection showed intraluminal vegetative growths, the largest of which measured 0.7 × 0.4 cm (Fig. 4).

Histopathological examination revealed a thickened cystic wall containing anastomosing cords and irregular patches of odontogenic epithelium. The epithelial component was composed of basaloid cells, morphologically reminiscent of those observed in ameloblastoma. Areas of hyalinized fibrous stroma separated the epithelial proliferations. Multiple epithelial nodules were identified, composed of elongated cells that, in certain regions, exhibited rosette-like arrangements surrounding central luminal spaces. The stroma was partially dissociated

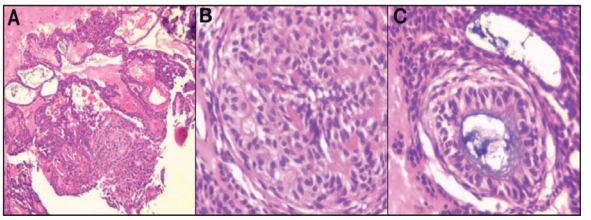

Fig. 5. Microscopic findings: Hematoxylin and eosin-stained sections at low power (A, ×200) show a partially cystic lesion with thick fibrous stroma and multiple epithelial nodules. Eosinophilic pseudo--amyloid material and calcifications are noted within epithelial islands,consistent with AOT. At higher magnification (B, C; ×400), epithelial nodules display rosettelike and duct-like arrangements lined by cuboidal to spindle-shaped cells with central lumina containing eosinophilic material and calcifications. No atypia or mitoses are observed.

by an eosinophilic, amyloid-like material, and numerous foci of dystrophic calcifications were interspersed throughout the lesion. These histological features are characteristic of an AOT (Fig. 5).

Correlation of clinical, radiological and histological findings confirmed the diagnosis of cystic AOT. The two-year follow-up revealed no postoperative complications or recurrence.

Case 2

A 30-year-old male with a previous history of epilepsy, currently managed with Depakene, was referred by his orthodontist to the Department of Oral Medicine and Oral Surgery following a chance finding on a panoramic radiograph of a mixed-density lesion involving the impacted left maxillary canine. The extraoral clinical examination revealed no significant findings. On intraoral examination, there was a slight, indurated, painless tumefaction in the vestibule opposite the maxillary deciduous left canine (63), with normal overlying mucosa. Additionally, a mild palatal swelling was noted opposite this tooth (63) with the appearance of the tip of the left maxillary permanent canine (23) (Fig. 6).

Panoramic imaging showed a clearly demarcated, oval-shaped, mixed radiolucent-radiopaque lesion, around 3 cm in size, surrounded by a radiolucent peripheral halo. The lesion was located between the roots of the maxillary left lateral incisor (22) and the impacted maxillary left canine (23) (Fig. 7).

CBCT image revealed an oval-shaped, mixed radiolucent-radiopaque lesion measuring approximately 35 mm in the mesiodistal dimension, 23 mm buccolingually, and 27 mm craniocaudally, related to the impacted canine (23). Both the buccal and palatal cortical plates exhibited expansion and thinning due to the lesion (Fig. 8).

Under local anaesthesia, both buccal and

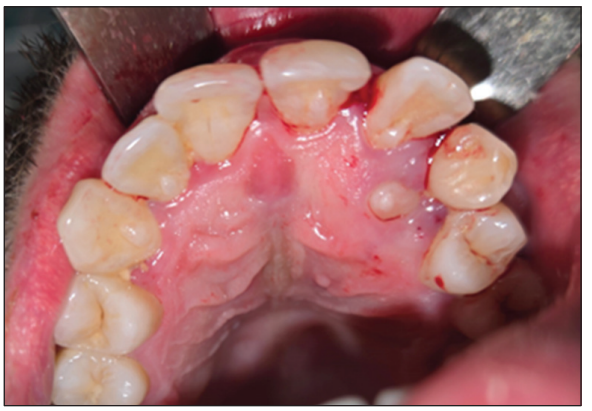

Fig. 6. Clinical intraoral evaluation demonstrated a mild palatal tumefaction opposite the left deciduous canine with the appearance of the tip of the left maxillary permanent canine.

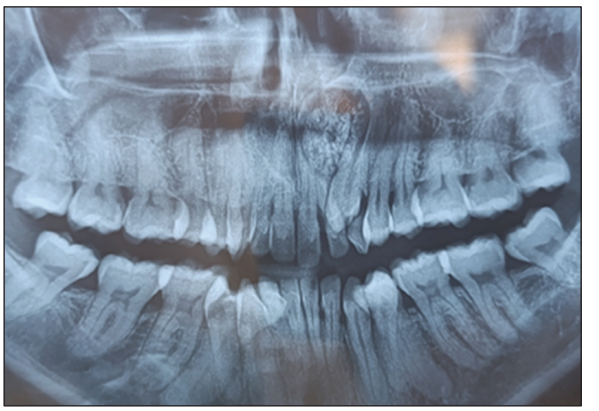

Fig. 7. Panoramic radiograph demonstrating the impacted left canine in the maxilla (23) associated with mixed radiolucent-radiopaque lesion surrounded by a radiolucent peripheral halo.

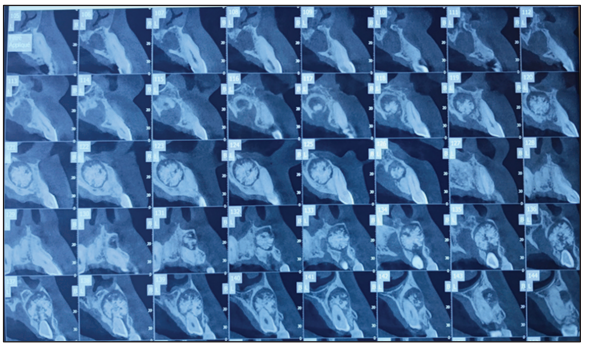

Fig. 8. CBCT image revealed an oval-shaped, mixed radiolucent-radiopaque lesion related to the impacted canine (23). Both the buccal and palatal cortical plates exhibited expansion and thinning due to the lesion.

palatal approaches were employed to access the lesion, which was completely enucleated. Simultaneously, the deciduous canine (63) was extracted. The impacted permanent left maxillary canine (23) was preserved, as the orthodontist had determined it could be successfully managed with orthodontic traction (Fig. 9).

Macroscopic examination revealed a well-circumscribed, round cystic specimen about 1.5 cm in diameter (Fig. 10).

Anatomopathological examination confirmed the diagnosis of AOT. The specimen revealed a well-circumscribed and encapsulated lesion composed predominantly of odontogenic epithelial cells. Histologically, the tumour showed characteristic single-layered cuboidal to columnar cells lined the duct-like structures, often arranged in a rosette-like or whorled arrangements. The epithelial cells exhibited uniform nuclei with minimal atypia and rare mitotic figures. Areas of eosinophilic, amorphous material were observed within the luminal spaces, along with scattered foci of calcifications. The absence of an ectomesenchymal component and the presence of these classic histological features supported the definitive diagnosis of AOT. The clinical course was favourable, with no

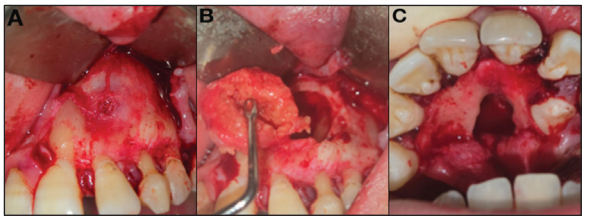

Fig. 9. Both buccal and palatal approaches were employed to access the lesion (A, C), which was completely enucleated (B).

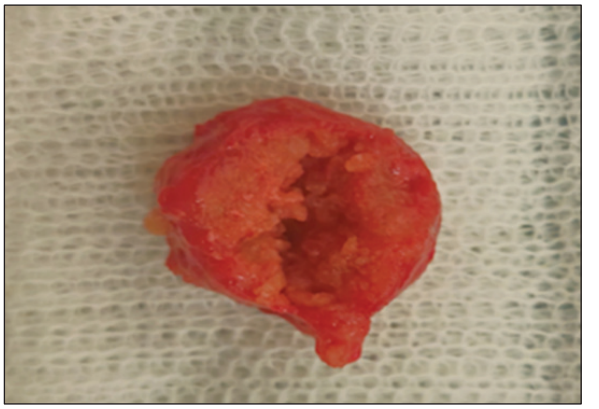

Fig. 10. Surgical specimen: a well-circumscribed, round cystic specimen about 1.5 cm in diameter.

signs of postoperative secondary infection at two weeks, and the patient remained free of recurrence after two years of follow-up.

Discussion

The AOT is considered a slow-growing benign tumour with a prevalence of 3 to 7% of all odontogenic tumours.[7,8] These lesions are more commonly encountered in the maxillary arch than in the mandibular one, in the anterior region more than in the posterior one with a particularly female predilection.[9,10] The location of the lesions in our two clinical cases was consistent with literature reports. Because nearly two-thirds of cases of AOT are observed in the maxilla, two-thirds affect young females, two-thirds are associated with impacted teeth, and two-thirds of the concerned teeth are canines, AOT is known as the "two-thirds tumour".[11] AOTs are generally diagnosed between 10 and 19 years of age, with

87.2% of cases seen between the ages of 10 and 30 years. The tumour typically measures between 1.5 and 3 cm along its longest axis. However, it can sometimes reach a larger size, causing, potentially, dental displacement as well as functional and/or aesthetic problems.[7] AOT is well recognized in two primary forms: central (intraosseous) and peripheral (extraosseous) variants. The central variant is further subdivided into two types based on radiographic characteristics:

Follicular type: this subtype accounts for approximately 70% of cases and involves a tumour surrounding the crown of an unerupted tooth, as observed in our second case.

Extrafollicular type: representing about 25% of cases, this subtype occurs independently of an unerupted tooth and is typically located between erupted teeth, as seen in our first case.

The peripheral (extraosseous) variant constitutes approximately 5% of AOT cases. It usually presents as a gingival swelling resembling an epulis.[7,8]

The origin of AOT is still controversial. Several researchers hypothesize that it arises from the odontogenic epithelium associated with a dentigerous cyst, whereas others suggest it originates from epithelial remnants of the dental lamina. This controversy is further reflected in the developmental theory, which proposes that the lesion may develop within, or adjacent to, a nearby dental follicle.[1] Several hypotheses have been proposed regarding the development of AOT. It is believed that the tumour may originate from the enamel organ, the epithelial lining of a dentigerous cyst, the epithelial rests of Malassez associated with primary or permanent teeth, or remnants of the dental lamina. Nevertheless, none of these theories has been definitively confirmed or universally accepted. A notable clinical observation is that most cases of AOT are associated with successional teeth during the period of active dental development.[9] AOT is a non-aggressive lesion characterized by indolent but progressive enlargement.[3,7] Although it is usually asymptomatic, patients may occasionally present with pain, swelling or an unerupted tooth.[10] The tumour is often discovered incidentally during routine dental examinations, radiographic evaluations conducted for orthodontic purposes, or investigations of delayed tooth eruption, as seen in our second clinical case. In some instances, the appearance of a slowly enlarging and typically painless bony swelling may prompt the patient to seek medical attention. Tooth displacement, mobility of adjacent teeth, or delayed eruption of permanent teeth, as noted in our first clinical case, are some dental signs that can accompany the bony swelling.[9] The radiographic appearance of the intraosseous AOT is generally characterized by the presence of a unilocular osteolytic lesion with well-defined margins which may contain discrete calcified foci. More prominent radiopaque areas may be seen in more advanced stages as was the case with our second patient. It is rare to observe extraosseous, peripheral or gingival forms of AOT. However, a mild resorption of the alveolar bone plate beneath them can be noticed. Moreover, the radiographic existence of calcifications may lead to the exclusion of the diagnosis of a dentigerous cyst. Also, it must be noted that intralesional calcifications may not be visible in the panoramic radiographs. In many cases, the lesion appears entirely radiolucent on panoramic images, which may lead to a misdiagnosis of a simple cystic lesion. Therefore, a CBCT scan is recommended to achieve a more accurate evaluation and to better characterize the lesion.[11] The tumour may exhibit a partly cystic architecture, with solid tumour nodules present within the wall of a large cystic cavity, as observed in our second case.[7,12] Histopathological examination remains essential for an accurate diagnosis.[13,14] AOT is typically encapsulated by a well-formed

connective tissue capsule and may appear as a solid tumour, a large solitary cystic cavity, or multiple small cystic compartments.[1] Histologically, it is formed of spindle-shaped or polygonal epithelial cells arranged in sheets and concentric (whorled) formations within a sparse connective tissue stroma. Characteristically, amorphous eosinophilic material is observed both between the epithelial cells and centrally within rosette-like arrangements. The tumour is particularly characterized by the presence of tubule-like configurations surrounded by a lined layer of epithelial cells.[15] The differential diagnosis of AOT includes several odontogenic lesions with overlapping clinical and radiographic features, such as dentigerous cysts, odontogenic keratocysts, calcifying odontogenic cysts, and calcifying epithelial odontogenic tumours.[13] The treatment of choice for AOT is complete enucleation of the lesion, along with the extraction of any associated impacted teeth.[2,14,16] In our second case, enucleation of the lesion combined with curettage was performed, with preservation of the implicated canine for planned orthodontic traction. A thorough review of the literature revealed that cases in which impacted teeth linked to AOT are conserved and followed by orthodontic treatment are rare. The prognosis following treatment is excellent, with recurrence being exceptional and no reports of malignant transformation.[11]

Strengths and Limitations

These clinical case reports contribute to the existing literature by documenting two distinct clinical presentations of AOT, highlighting the importance of accurate radiographic and histopathological examination in the diagnosis of odontogenic lesions. One of the strengths of this report is the detailed description of the clinical, radiographic, surgical, and histological findings, which may assist clinicians in differentiating AOT from other odontogenic lesions such as dentigerous cysts or ameloblastomas. Moreover, the inclusion of both follicular and extrafollicular variants adds educational value, as these subtypes are rarely documented together in the same report.

However, this study has some limitations. First, as a case report, it inherently lacks generalizability and statistical analysis. Second, long-term follow-up data are limited, preventing a comprehensive assessment of recurrence or delayed complications. Lastly, molecular or immunohistochemical investigations were not performed, which could have provided deeper insight into the pathogenesis and differential diagnosis of the tumour.

Conclusion

The rarity of AOT contributes to the incomplete understanding of its biological behaviour and origin. Like many odontogenic pathologies, AOT can exhibit a wide range of clinical and radiographic presentations, varying in size, location and patient demographics, including age and sex predilection. This variability can pose significant diagnostic challenges and complicate treatment planning. Therefore, a comprehensive clinical evaluation, supported by appropriate imaging and histopathological evaluation, is crucial for achieving a precise diagnosis and ensuring optimal treatment. Continued documentation and study of such rare lesions are crucial for advancing our understanding and improving patient outcomes.

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