Spongiotic Gingival Hyperplasia in a Child with Asperger Syndrome: a Case Report

Aikaterini Aktypi-Bampouranou[1], Eleni-Marina Kalogirou[2], Ioannis Skamnakis[3], Dimitrios Vlachodimitropoulos[4], Konstantinos I. Tosios[2,5]

[1]Private practice in dentistry, Athens, Greece.
[2]Private practice in oral medicine, Athens, Greece.
[3]Private practice in pediatric dentistry, Athens, Greece.
[4]Department of Forensic Medicine and Toxicology, School of Medicine, University of Athens, Athens, Greece.
[5]Department of Oral Medicine, Pathology, and Hospital Dentistry, School of Dentistry, University of Athens, Athens, Greece.

Corresponding Author:
Konstantinos I. Tosios
15, Lambraki street, 16675, Athens
Greece
Phone: +302107461003
E-mail: ktosios@dent.uoa.gr

ABSTRACT

Background: Asperger syndrome is a type of autism spectrum disorder that may affect oral health and dental management. Spongiotic gingival hyperplasia is a rare lesion with unique clinicopathological features and unknown pathogenesis that has not been previously reported in a patient with autism spectrum disorder. The purpose of this case report is to present the first case of spongiotic gingival hyperplasia in a child with Asperger syndrome.
Methods: A 14-year-old boy with Asperger syndrome was referred for diagnosis and management of bright red granular overgrowths of the marginal gingiva and interdental papilla of the mandibular right incisors and marginal gingiva of the mandibular left incisor. A biopsy was performed on the interdental papilla between the mandibular right incisors.
Results: Microscopic examination and cytokeratin 19 immunopositivity confirmed the diagnosis of spongiotic gingival hyperplasia. The parents of the patient declined any further intervention, and four months later the gingival lesions, including the biopsied area, did not show any significant difference from the initial examination.
Conclusions: Patients with autism spectrum diseases, such as Asperger syndrome, cannot achieve a good level of oral hygiene. Thus, it is expected that the incidence of spongiotic gingival hyperplasia should be higher in this group of patients, in case oral microbiome participates in its pathogenesis. Management of such lesions is challenging, as such patients do not comply with a proper oral hygiene program and do not cooperate with surgical excision.

Keywords: Asperger syndrome; autism spectrum disorder; gingival diseases; gingival hyperplasia.

Accepted for publication: 29 September 2023
To cite this article:
Aktypi-Bampouranou A, Kalogirou EM, Skamnakis I, Vlachodimitropoulos D, Tosios KI.
Spongiotic Gingival Hyperplasia in a Child with Asperger Syndrome: a Case Report
J Oral Maxillofac Res 2023;14(3):e5
URL: http://www.ejomr.org/JOMR/archives/2023/3/e5/v14n3e5.pdf
doi: 10.5037/jomr.2023.14305

INTRODUCTION

Asperger syndrome (AS) is a subgroup of autism spectrum disorder (ASD) neurodevelopmental disabilities, characterized by impairment of social interaction, communication, and behaviour [1]. Patients with AS usually have a high level of intelligence quotient (IQ) and normal language development, but exhibit several unique characteristics, including abnormal and repetitive behaviours, difficulties in communication due to poor understanding of non-verbal languages, e.g., facial expressions and body language, and delay in motor development, e.g., clumsiness and uncoordinated motor movements [2,3]. AS prevalence in the general population is 10 to 26 cases per 10,000 and is predominantly diagnosed in males during childhood or adolescence [4].

Lack of manual dexterity in AS, combined with difficulty in following oral hygiene instructions [5] and dislike of the feeling of the toothbrush and the taste and texture of the toothpaste [6] result in inability in achieving a acceptable level of oral hygiene, and thus high susceptibility to periodontal disease [7]. In addition, dental management of those patients can be challenging due to poor communication, increased anxiety prior to dental visit and sensory hypersensitivity that can lead to aversion to the touch of the dentist, as well as the light and sound of the dental unit, and odor of the dental materials [8].

Spongiotic gingival hyperplasia (SGH) was first described in 2007 by Darling et al. [9] as "juvenile spongiotic gingivitis". The authors drew attention to the possibility of being erroneously diagnosed as puberty gingivitis. The initial nomenclature was replaced by the term "localized juvenile spongiotic gingival hyperplasia" proposed by Chang et al. [10], as most reported cases showed hyperplasia and spongiosis of the gingival epithelium and were solitary. However, cases of adult patients presenting with multifocal lesions were consequently reported [11-14] supporting use of the term SGH [11,12]. It clinically manifests as a painless, localized or multifocal, bright red gingival overgrowth with pebbly, papillary or velvety surface, usually measuring up to 1 cm in maximum dimension [9,10,15] Microscopically, SGH presents hyperplastic non-keratinized epithelium, exhibiting a slight papillary surface, spongiosis, as well as neutrophilic exocytosis [13,16] and CK19 immunoexpression that is considered diagnostic for SGH [9,11,13,14,17,18].

Herein, we present the first case of spongiotic gingival hyperplasia in a young boy with Asperger syndrome and discuss its purported aetiopathogenesis and management.

CASE DESCRIPTION AND RESULTS

A 14-year-old boy was referred to the private practice (Athens, Greece) of one of the authors (K.I.T.) on December 28, 2018, by his paediatric dentist for evaluation of multifocal gingival lesions noticed approximately a year before referral. The patient's medical history was significant for AS, while his parents denied any medication uptake. He was in regular three months dental recalls.

Intraoral examination revealed that the marginal gingiva and interdental papilla of the mandibular right incisors and the marginal gingiva of the mandibular left lateral incisor were enlarged, with bright red colour and granular surface (Figure 1). The lesions were painless and did not bleed on gentle rubbing. Additional findings were linear erythema on the marginal gingiva between the maxillary lateral incisors and an ulcer covered by pseudomembrane on the labial attached gingiva of the upper right lateral incisor (Figure 2). The patient's oral hygiene was poor, as evidenced by plaque and calculus deposits on his teeth, mostly on the mandibular incisors. The rest of the oral mucosa was within normal limits and extraoral examination did not reveal any abnormality.

The clinical diagnosis was multifocal SGH and traumatic ulcer, but his parents insisted for confirmation of the diagnosis with a biopsy. On the day of the biopsy, a week after the initial examination, the ulcer had healed completely.

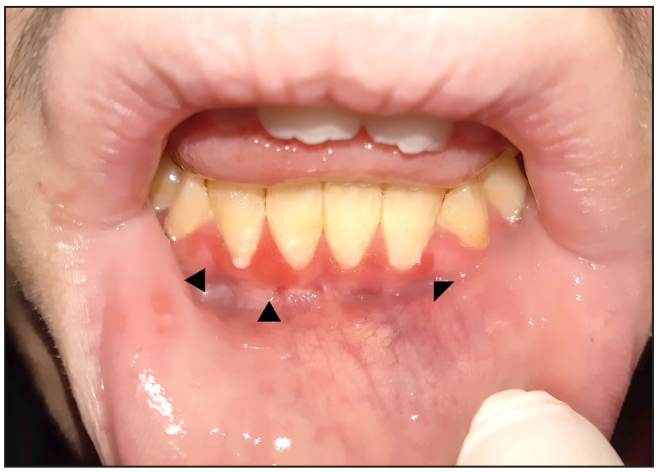

Figure 1. Clinical examination.
The marginal gingiva and interdental papilla of the mandibular right incisors and the marginal gingiva of the mandibular left lateral incisor are swollen and have bright red colon and granular surface (arrowheads).

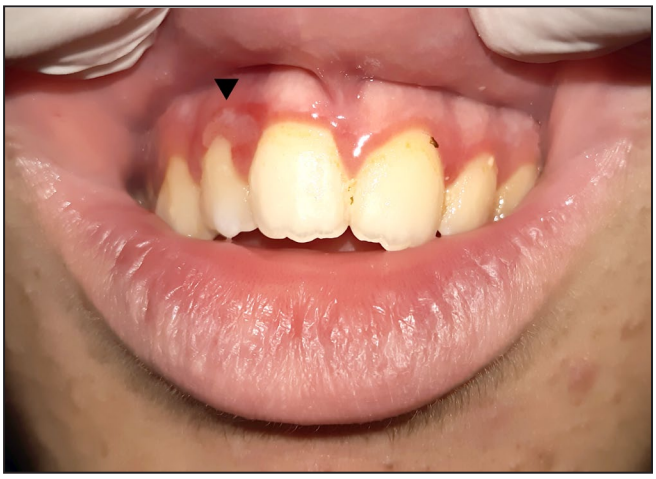

Figure 2. Clinical examination.
A linear erythema is present on the marginal gingiva between the maxillary lateral incisors and an ulcer on the labial attached gingiva of the upper right lateral incisor (arrowhead).

The patient was uncooperative, but after physical restraint by his mother, a specimen was excised under local anaesthesia from the interdental papilla between the mandibular right incisors. Histopathological examination of 5 μm thick formalin-fixed and paraffin-embedded tissue sections revealed a mucosal fragment covered by non-keratinized stratified squamous epithelium, showing papillary surface, acanthosis, spongiosis, focally elongated and interconnecting rete pegs, and neutrophilic exocytosis (Figure 3). The underlying vascular connective tissue showed dense, mixed type inflammatory infiltration. Immunohistochemistry with streptavidin-biotin-peroxidase method using the monoclonal mouse anti-human cytokeratin (CK) 19 (1 : 100 dilution, clone RCK108 - Dako; Glostrup, Denmark) showed intense cytoplasmic positivity in all cell layers of the lesional epithelium (Figure 4). The microscopic and immunohistochemical findings confirmed the diagnosis of SGH.

The parents declined any further excision for aesthetic reasons under sedation anaesthesia. The patient was asked to apply a topical 0.05% chlorhexidine gel on the anterior gingiva and brush and floss his teeth twice per day. Four months after the initial examination increased plaque and calculus deposits were noticed in most of his teeth, as he did not follow the oral hygiene instructions; the gingival lesions, including the biopsied area, did not show any significant difference from the initial examination.

DISCUSSION

The case presented herein is the first documented example of SGH in a patient with an ASD, i.e., AS. ASD patients commonly present periodontal disease [7] and drug-induced gingival hyperplasia [19]. Therefore, it may be hypothesized that cases of SGH may have been misdiagnosed as other types of periodontal disease, due to difficulty in cooperation with those patients [1] did not allow the establishment of diagnosis that can be achieved only through biopsy. Other intraoral findings in ASD patients include a 20% [20] to 52% [19] prevalence of dental trauma mostly on the anterior teeth due to patients' motor difficulties and hyperactivity; approximately 60% prevalence of bruxism [19,20]; 13.33% [20] to 44% [19] prevalence of angular cheilitis associated with saliva drooling; xerostomia, as a side effect of

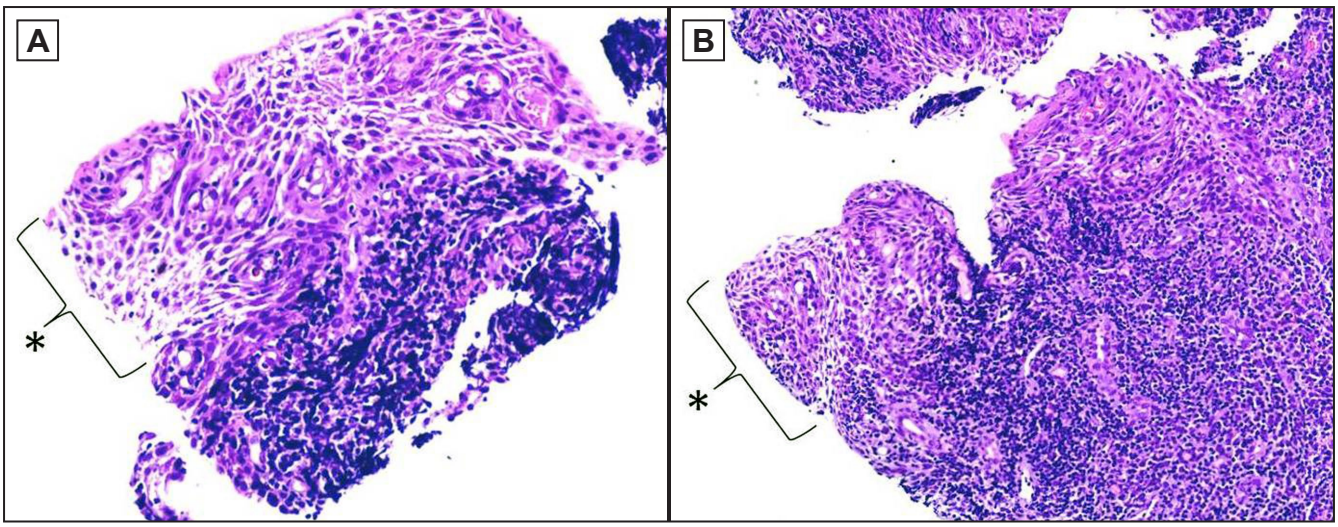

Figure 3. Microscopic examination: A = high (original magnification x100); B = low (original magnification x40). Non-keratinized stratified squamous epithelium, with papillary surface, acanthosis, spongiosis, focally elongated and interconnecting rete pegs, and neutrophilic exocytosis. The underlying vascular connective tissue shows dense inflammatory infiltration of mixed type.
(*) marks epithelium (haematoxylin and eosin stain).

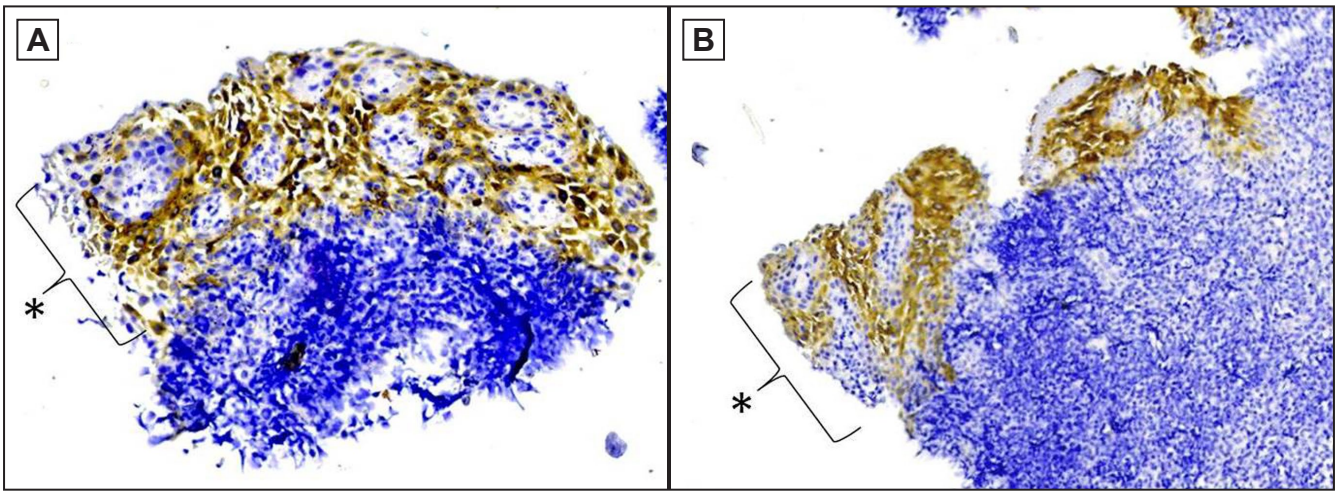

Figure 4. Immunohistochemistry: A = high (original magnification x100); B = low (original magnification x40). Intense cytoplasmic positivity for cytokeratin 19 is seen in all cell layers of the lesional epithelium.
(*) marks epithelium (streptavidin-biotin-peroxidase method).

antidepressants or anticonvulsants, and phenytoin-induced gingival hyperplasia [19]. Patients with ASD do not present specific oral mucosal lesions, but oral habits such as tongue thrusting, lip and cheek biting, as well as picking at the gingiva that are common among them may lead to self-inflicted lesions [20]. The gingival ulcer seen in our patient is consistent with self-inflicted trauma.

As in our patient, SGH mostly occurs in the second decade of life, without gender preference [16]. It usually involves the anterior facial maxillary gingiva [16], while lesions on the anterior mandible have been reported in 3.6% [13] to 25% [9] of cases. There are, also, rare examples of SGH on the posterior maxillary [11,13,15] or mandibular gingiva [9,14]. The marginal gingiva are predominantly affected, but the attached or the interproximal gingiva may be also involved [10,13], as in the case reported herein.

The clinical appearance of painless, multifocal, bright red gingival overgrowths with pebbly, papillary, or velvety surface, usually measuring up to 1 cm in maximum diameter, is consistent with the diagnosis of SGH [9,10,15], as is the present case. The clinical differential diagnosis includes puberty gingivitis or plaque related gingivitis, plasma cell gingivitis, oral lichenoid reactions, linear gingival erythema, and strawberry gingivitis of Wegener granulomatosis [13,21]. Microscopic examination is imperative for the documentation of diagnosis in diagnostically problematic cases.

The aetiology of SGH is unknown. Due to the high prevalence of SGH in patients on the second decade of life the influence of hormones was initially hypothesized, but the absence of immunohistochemical expression of estrogen and progesterone receptors in those lesions [9], and the involvement of adults patients [10,13,14] contradict this assumption. Human papillomavirus (HPV) involvement was speculated based on the papillary surface of many cases, but although immunopositivity for p16INK4A was seen in 21 cases, polymerase chain reaction failed to identify the presence of HPV DNA [15]. The histopathological features of inflammation could be the result of a local reaction to environmental factors, but microscopically no foreign body or granulomas have been identified, while most lesions are localized and do not justify a hypersensitivity reaction [10]. Other potential aetiological factors include trauma, orthodontic appliances, and mouth breathing, but there is no adequate evidence to support a causal relationship [10,15]. The most prevailing theory attributes the origin of SGH to hyperplasia of the junctional epithelium that under the influence of yet unknown local environmental stimuli, such as minor trauma, tooth eruption, lip incompetence etc., “exteriorizes” and gradually acquires oral gingival epithelium-associated features in order to adjust to its new environment [10,17]. An odontogenic origin of SGH has been proposed based on the combined CK14 and CK19 immunopositivity [22].

The clinical similarity of SGH with bacterial plaque-related gingivitis implies that the oral microbiome may act as an environmental stimulus, but the absence of plaque and calculus deposits in some SGH cases [15] and the lack of involvement of the marginal gingiva in other cases [9], seem to contrast this theory. In line with this, Darling et al. [9] reported lack of response of SGH to oral hygiene improvement and surgical excision with scalpel or laser is currently considered as the treatment of choice, mostly for aesthetic reasons [9,10,15]. However, bacterial colonies may be evident microscopically [16],

while regression following dental plaque debridement and improvement of patient's oral hygiene have been documented [9,11,18]. Those observations, combined with the high frequency of SGH lesions on the marginal gingiva [11,13] where oral microbiome-induced gingivitis develops, raise the possibility that oral microbiome could participate in the development of SGH.

CONCLUSIONS

Patients with autism spectrum diseases, such as Asperger syndrome, cannot achieve a good level of oral hygiene. Thus, it is expected that the incidence of spongiotic gingival hyperplasia should be higher in this group of patients, in case oral microbiome participates in its pathogenesis. Management of such lesions is problematic, as such patients do not comply with a proper oral hygiene program and do not cooperate with surgical excision.

ACKNOWLEDGMENTS AND DISCLOSURE STATEMENTS

The authors report no conflicts of interest related to this study.

REFERENCES

1. Filipek PA, Accardo PJ, Baranek GT, Cook EH Jr, Dawson G, Gordon B, Gravel JS, Johnson CP, Kallen RJ, Levy SE, Minshew NJ, Ozonoff S, Prizant BM, Rapin I, Rogers SJ, Stone WL, Teplin S, Tuchman RF, Volkmar FR. The screening and diagnosis of autistic spectrum disorders. J Autism Dev Disord. 1999 Dec;29(6):439-84. [Medline: 10638459] [doi: 10.1023/A:1021943802493]
2. Fahlvik-Planefeldt C, Herrström P. Dental care of autistic children within the non-specialized Public Dental Service. Swed Dent J. 2001;25(3):113-8. [Medline: 11813447]
3. Klin A. Asperger syndrome: an update. Braz J Psychiatry. 2003 Jun;25(2):103-9. [Medline: 12975708] [doi: 10.1590/S1516-44462003000200011]
4. Gillberg IC, Gillberg C. Asperger syndrome--some epidemiological considerations: a research note. J Child Psychol Psychiatry. 1989 Jul;30(4):631-8. [Medline: 2670981] [doi: 10.1111/j.1469-7610.1989.tb00275.x]
5. Udhya J, Varadharaja MM, Parthiban J, Srinivasan I. Autism Disorder (AD): An Updated Review for Paediatric Dentists. J Clin Diagn Res. 2014 Feb;8(2):275-9. [Medline: 24701555] [PMC free article: 3972586] [doi: 10.7860/JCDR/2014/7938.4080]
6. Mansoor D, Al Halabi M, Khamis AH, Kowash M. Oral health challenges facing Dubai children with Autism Spectrum Disorder at home and in accessing oral health care. Eur J Paediatr Dent. 2018 Jun;19(2):127-133. [Medline: 29790776] [doi: 10.23804/ejpd.2018.19.02.06]
7. Shapira J, Mann J, Tamari I, Mester R, Knobler H, Yoeli Y, Newbrun E. Oral health status and dental needs of an autistic population of children and young adults. Spec Care Dentist. 1989 Mar-Apr;9(2):38-41. [Medline: 2533709] [doi: 10.1111/j.1754-4505.1989.tb01022.x]
8. Klein U, Nowak AJ. Autistic disorder: a review for the pediatric dentist. Pediatr Dent. 1998 Sep-Oct;20(5):312-7. [Medline: 9803429]
9. Darling MR, Daley TD, Wilson A, Wysocki GP. Juvenile spongiotic gingivitis. J Periodontol. 2007 Jul;78(7):1235-40. [Medline: 17608578] [doi: 10.1902/jop.2007.060502]
10. Chang JY, Kessler HP, Wright JM. Localized juvenile spongiotic gingival hyperplasia. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2008 Sep;106(3):411-8. [Medline: 18602289] [doi: 10.1016/j.tripleo.2008.04.024]
11. Siamantas I, Kalogirou EM, Tosios KI, Fourmousis I, Sklavounou A. Spongiotic Gingival Hyperplasia Synchronously Involving Multiple Sites: Case Report and Review of the Literature. Head Neck Pathol. 2018 Dec;12(4):517-521. [Medline: 29512024] [PMC free article: 6232213] [doi: 10.1007/s12105-018-0903-9]
12. Solomon LW, Trahan WR, Snow JE. Localized juvenile spongiotic gingival hyperplasia: a report of 3 cases. Pediatr Dent. 2013 Jul-Aug;35(4):360-3.
13. Vargo RJ, Bilodeau EA. Reappraising localized juvenile spongiotic gingival hyperplasia. J Am Dent Assoc. 2019 Feb;150(2):147-153.e2. [Medline: 30528764] [doi: 10.1016/j.adaj.2018.10.001]
14. Wang MZ, Jordan RC. Localized juvenile spongiotic gingival hyperplasia: A report of 27 cases. J Cutan Pathol. 2019 Nov;46(11):839-843. [Medline: 31322765] [doi: 10.1111/cup.13549]
15. Argyris PP, Nelson AC, Papanakou S, Merkourea S, Tosios KI, Koutlas IG. Localized juvenile spongiotic gingival hyperplasia featuring unusual p16INK4A labeling and negative human papillomavirus status by polymerase chain reaction. J Oral Pathol Med. 2015 Jan;44(1):37-44. [Medline: 25065444] [doi: 10.1111/jop.12214]
16. Theofilou VI, Pettas E, Georgaki M, Daskalopoulos A, Nikitakis NG. Localized juvenile spongiotic gingival hyperplasia: Microscopic variations and proposed change to nomenclature. Oral Surg Oral Med Oral Pathol Oral Radiol. 2021 Mar;131(3):329-338. [Medline: 33309266] [doi: 10.1016/j.oooo.2020.10.024]

17. Allon I, Lammert KM, Iwase R, Spears R, Wright JM, Naidu A. Localized juvenile spongiotic gingival hyperplasia possibly originates from the junctional gingival epithelium-an immunohistochemical study. Histopathology. 2016 Mar;68(4):549-55. [Medline: 26154781] [doi: 10.1111/his.12774]
18. Kalogirou EM, Chatzidimitriou K, Tosios KI, Piperi EP, Sklavounou A. Localized Juvenile Spongiotic Gingival Hyperplasia: Report of Two Cases. J Clin Pediatr Dent. 2017;41(3):228-231. [Medline: 28422590] [doi: 10.17796/1053-4628-41.3.228]
19. Al-Yassiri Ali MH, Abdul-ZahraaMahdi K. The presence of oral manifestations in autistic disorders among group of children samples in Babil and Najaf cities, Iraq. J Pharm Biomed Sci. 2015;(6):487-90. [URL: https://oaji.net/articles/2015/2317-1439515513.pdf]
20. Orellana LM, Silvestre FJ, Martínez-Sanchis S, Martínez-Mihi V, Bautista D. Oral manifestations in a group of adults with autism spectrum disorder. Med Oral Patol Oral Cir Bucal. 2012 May 1;17(3):e415-9. [Medline: 22143726] [PMC free article: 3476085] [doi: 10.4317/medoral.17573]
21. de Freitas Silva BS, Silva Sant'Ana SS, Watanabe S, Vêncio EF, Roriz VM, Yamamoto-Silva FP. Multifocal red bands of the marginal gingiva. Oral Surg Oral Med Oral Pathol Oral Radiol. 2015 Jan;119(1):3-7. [Medline: 25204611] [doi: 10.1016/j.oooo.2014.07.005]
22. Lafuente-Ibáñez de Mendoza I, Alberdi-Navarro J, Marichalar-Mendia X, Mosqueda-Taylor A, Aguirre-Urizar JM. Characterization of juvenile spongiotic gingival hyperplasia as an entity of odontogenic origin. J Periodontol. 2019 Dec;90(12):1490-1495. [Medline: 31294824] [doi: 10.1002/JPER.19-0022]

To cite this article:
Aktypi-Bampouranou A, Kalogirou EM, Skamnakis I, Vlachodimitropoulos D, Tosios KI.
Spongiotic Gingival Hyperplasia in a Child with Asperger Syndrome: a Case Report
J Oral Maxillofac Res 2023;14(3):e5
URL: http://www.ejomr.org/JOMR/archives/2023/3/e5/v14n3e5.pdf
doi: 10.5037/jomr.2023.14305