DOI: 10.7860/NJLM/2021/43269:2469

Original Article

Pathology Section

Unusual Presentations of Disseminated Cutaneous Rhinosporidiosis Masquerading as Tumour Like Lesions of Skin: A Clinicopathological Study

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ABSTRACT

Introduction: Rhinosporidiosis is a chronic granulomatous infective disease caused by Rhinosporidium seeberi which primarily affects the mucosa of the nasal cavity. Cutaneous lesions are infrequent. However, disseminated cutaneous lesions with varied presentations are very rare in routine pathology practice.

Aim: The study aimed to diagnose cutaneous lesions due to rhinosporidiosis on Fine-Needle Aspiration Cytology (FNAC) and to confirm the same by histology.

Materials and Methods: This was a cross-sectional study. A total of 8 cases of disseminated cutaneous Rhinosporidiosis out of 815 cutaneous lesions were observed over five years, from September 2013 to October 2018, in Department of Pathology, VIMSAR, Burla. FNAC and scrape cytology of the lesions were performed and studied after Diff-Quik and H&E stain. The lesions were excised and histopathological examination was done.

Results: It was observed that the overall incidence of cutaneous rhinosporidiosis diagnosed on FNAC was very low (less than 0.1%) They were mainly seen in middle-aged males from low socio-economic status and rural areas. The skin lesions were mostly subcutaneous nodules, few wart-like growth and ulcerative growth, some of which were initially diagnosed as neurofibroma and squamous cell carcinoma. FNAC and histopathological examination helped to confirm the diagnosis of rhinosporidiosis in all cases.

Conclusion: Disseminated cutaneous rhinosporidiosis has varied clinical presentations, mimicking other cutaneous lesions. Hence, proper diagnosis of the patient is necessary which can be achieved by FNAC and histopathology.

Keywords: Granulomatous infection, Nasal cavity, Rhinosporidium seeberi

INTRODUCTION

Rhinosporidiosis is a chronic granulomatous disease that mostly involves the nasal and nasopharynx, and is caused by Rhinisporidium seeberii. Painless friable polypoidal growth is the usual presentation, that may hang anteriorly into the nares and posteriorly into the pharynx [1,2]. It also affects the conjunctiva and the lacrimal sac. Cutaneous dissemination, although known, is quite rare, usually associated with nasal mucosal lesions [1]. Skin lesions typically start as friable papilloma that becomes pedunculated. This may also present as subcutaneous nodules, warty papules, and nodules with whitish spots, ulcerative growth, crusting, and bleeding on the surface [2].

The main objective of the study were to determine the epidemiology, incidence, sites and varied clinical presentations of cutaneous rhinosporidiosis.

MATERIALS AND METHODS

The present study was a cross-sectional study conducted over a period of five years from September 2013 to October 2018 in the Department of Pathology, VIMSAR, Burla. This study followed all Institutional Ethical Guidelines in place and also followed Helsinki declaratation during that time. All the patients presented to the Outpatient Department with subcutaneous nodules at different sites and suspected cutaneous rhinosporidiosis with mucosal lesions in nasal cavity were included in the study by consecutive sampling. Patients with no lesions in nasal cavity or pharynx were excluded from the study. Informed consent was obtained from all included patients. A detailed history was taken, and a clinical examination was carried out in all the patients. The FNAC was performed in all the patients with cutaneous lesions with a 22-gauge needle attached to a 10 mL syringe and scrape cytology from the ulcerative growth was done with the help of a glass slide. The aspirated material was smeared on the glass slides and stained with Diff-Quik and H&E stain.

Diff-Quik stain: Air dried smears were fixed in methanol solution. Fixed smears were dipped 5-12 times in stain-1containing buffered solution of Eosin G. Excess stain was drained into filter paper followed by 6-8 dips in stain-2 containing buffered thiazine dye. Excess stain was blotted into filter paper.Then slides were rinsed in running tap water,allowed to air dry, mounted with DPX and examined directly under microscope in low power and high power objective.

Hematoxylin and Eosin stain (H&E stain): Alcohol fixed smears were kept in Hematoxylin for 1-3 minutes, followed by rinsing in tap water for 1 min, again washed in scott's solution for bluing. After washing, the slides were placed in 95% ethanol followed by staining with Eosin dye for 45 seconds.Then stained slides were dehydrated in graded alcohol, consisting of one change in 95% ethanol & two changes in 100 % ethanol for 1 minute each,following which the slides were treated in two solutions of Xylene 2 minutes each. Then the stained slides were mounted with DPX and examined directly under microscope with low power and high power objective.

Special stain (Periodic Acid Schiff (PAS) stain) was done in few selected cases for demonstration of endospore and sporangia. Following the FNAC, the excision of the lesions was done. After that, the tissue was fixed with 10% formalin, stained with H&E stain, and subjected to histopathological examination.

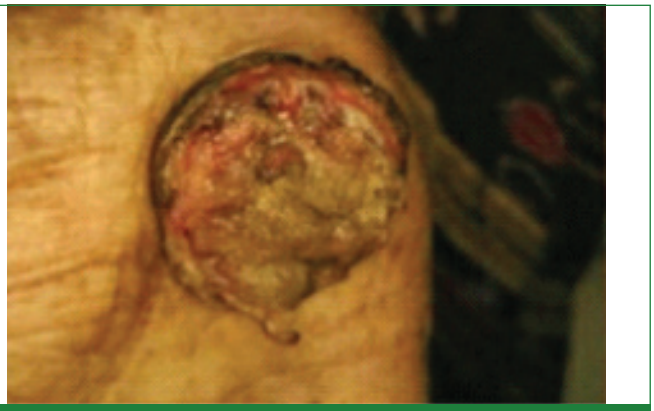
STATISTICAL ANALYSIS

Statistical analysis was done and data were presented as frequencies.

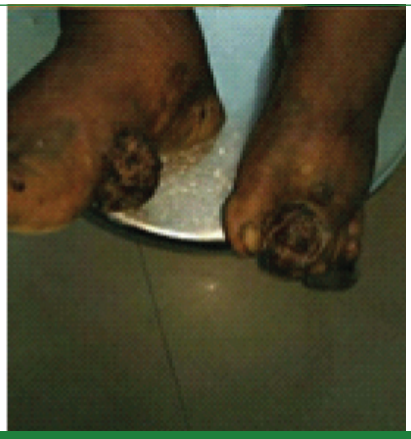
RESULTS

Eight cases of disseminated cutaneous rhinosporidiosis with different clinical presentations were reported here. The incidence of FNAC and histopathologically confirmed cases among all the cutaneous lesions were less than 0.1% (8 out of 815). The recurrence rate for nasal lesions was about 25% (2 out of 8 cases). Most of these cases were presented in the age group of 40 to 70 years, males being more commonly affected than females with a male: female ratio of 1.6:1. All the patients gave the history of bathing in stagnant water. Majority of the patients (seven out of eight) were immunocompetent with one having history of diabetes mellitus, and all the others had negative serology for HIV, hepatitis B and C.

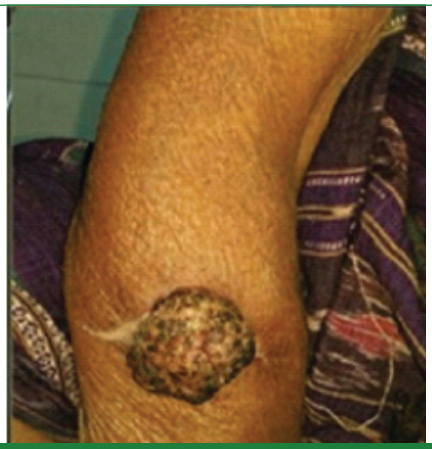
Most of the cases presented with non-pedunculated nodular growths, while one patient presented with ulcerative lesion [Table/Fig-1a] and another with tumour-like swellings on both the feet [Table/Fig-1b]. The majority of the skin lesions (6/8) were found in extremities [Table/Fig-1c], one on the forehead and one on the back [Table/Fig-1d]. All the patients presented with multiple lesions associated with the nasal or oropharyngeal growth [Table/Fig-1e,f]. The diagnostic accuracy of cytology was 88% (7/8).



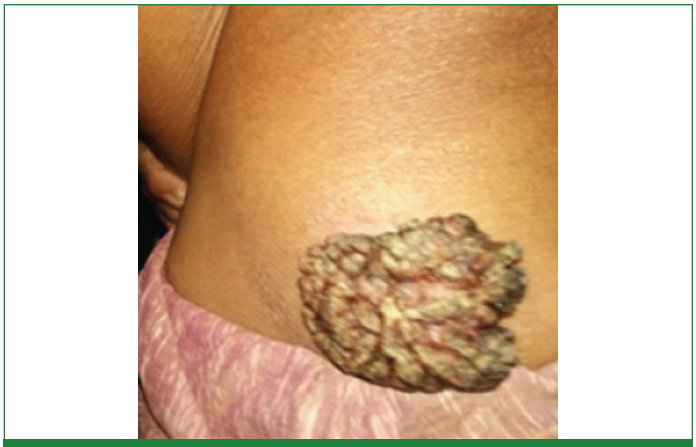
[Table/Fig-1a]: An ulcerative growth on the leg.



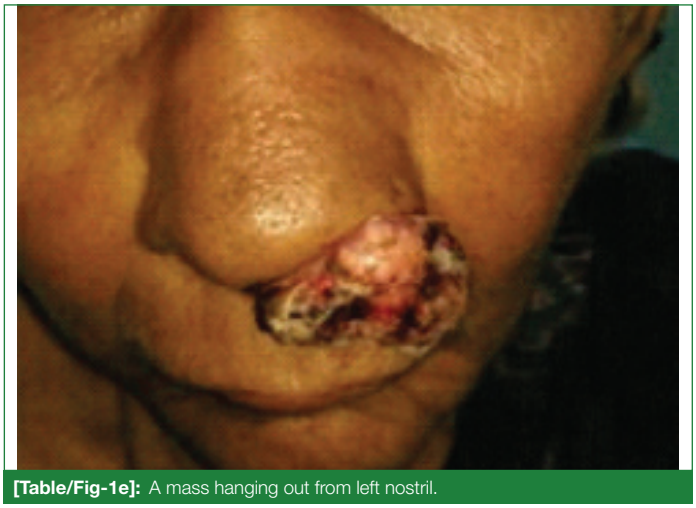
[Table/Fig-1b]: Tumour-like swellings over both the feet.



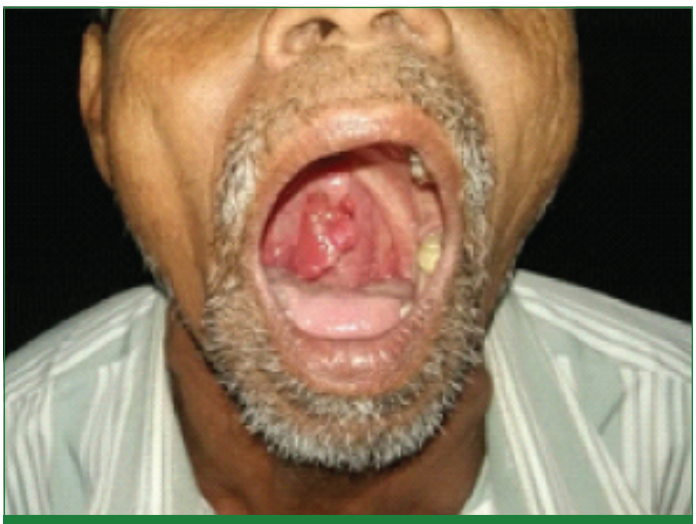
[Table/Fig-1c]: A nodular swelling over right arm.



[Table/Fig-1d]: A fungating growth on back.

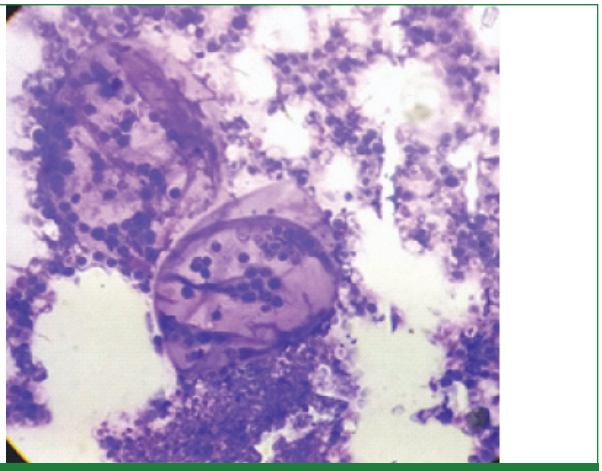


[Table/Fig-1e]: A mass hanging out from left nostril.

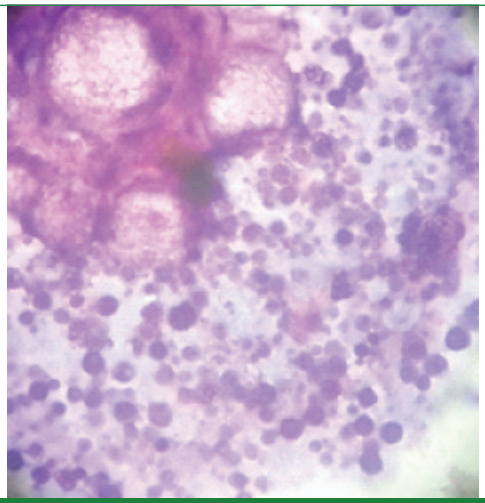


[Table/Fig-1f]: A hanging mass in oral cavity.

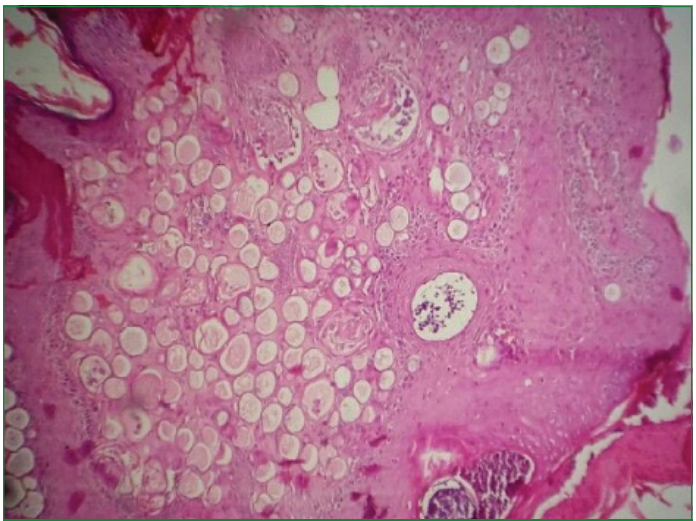
The FNAC taken from multiple sites in all the cases showed similar cytomorphology. The cytosmears showed many mature sporangium containing spores, a few empty sporangia along with plenty of endospores seen in the background. The scrape cytology smears taken from the ulcerated lesion showed ruptured matured sporangium and endospores [Table/Fig-2,3]. PAS stain showed positivity for the double layered chitinous wall along with sporangium and endospores. The impression was suggestive of rhinosporidiosis. The histopathological evaluation revealed hyperplastic stratified squamous epithelium. Many well-defined thick-walled mature sporangia along with immature and collapsed sporangia were seen within the epidermis and underlying upper dermis [Table/Fig-4]. The cytology, along with the histopathology, helped in the diagnosis of disseminated cutaneous rhinosporidiosis in all the cases.



[Table/Fig-2]: Cytology showing mature sporangium with endospores (x400) (Diff-Quik).



[Table/Fig-3]: Cytology showing PAS positive chitinous wall of sporangium (x 400) (PAS).



[Table/Fig-4]: Histopathology showing hyperplastic stratified squamous epithelium and underlying sporangium at different stages of maturation (X4100) (H&E).

DISCUSSION

Rhinosporidiosis is a chronic granulomatous disease caused by Rhinosporidium seeberi. Involvement of site other than naso-pharyngeal mucosa, especially skin, is very rare. Males acquire the disease more commonly compared to females and it is usually seen between the second and fourth decades of life [2]. Stagnant water exposure, bathing in water same as that used for washing cattle, and repeated trauma have been reported as the causative factors [3,4]. In the present study, five cases were males, and the other three were females, all in the age group of 40-70 years. All of them had a history of bathing in stagnant water.

Nasal and nasopharyngeal rhinosporidiosis usually affect males (70-90%), while the ocular infection is more prevalent in females [2]. The lesions are pink, or purple-red friable polyps stubbed with tiny white dots, sporangia-containing spores [2,5]. Nasal rhinosporidiosis is most commonly associated with nasal obstruction and bleeding as the presenting symptoms. Conjunctiva and lacrimal sac are involved in 15% of cases. Occasionally, it affects scalp, ear, uvula, palate, lips, maxillary antrum, epiglottis, larynx, trachea, bronchus, vulva, vagina, penis, rectum and very rarely the skin [5]. In the present study, all the cutaneous lesions were associated with involvement of nasal mucosa.

Mode of spread of rhinosporidiosis can be mostly by three means [6,7]:

- Autoinoculation- This is the reason for the satellite lesions nearby the nasal lesions.
- Hematogenous spread- It causes distant skin lesions, as seen in our cases. The subcutaneous lesions all over the body, without breach of the overlying skin may be due to such haematogenous dissemination.
- Direct inoculation- This is another way of transmission of organisms through traumatised skin and is known as the primary cutaneous lesion.

The cutaneous lesion may present as pedunculated or sessile growth, furunculoid lesion, or ulceration [1,2,8]. Other clinically alike lesions are wart, verrucous tuberculosis, and granuloma pyogenicum [2]. In the present study, five of the eight cases presented with cutaneous lesions as sessile growth and one presented with both sessile and pedunculated growth, of which two were clinically diagnosed as possible neurofibroma. One of the cases presented with ulcerative growth mimicking squamous cell carcinoma and one with wart-like lesion.

The diagnosis can easily be clinched by performing a Giemsa stained imprint smear or scrape smear or FNAC smear from lesions [9]. The cytosmear exhibit many mature sporangia containing spores and few empty sporangia along with plenty of endospores. As the culture of the organism is not possible, histopathology is the gold standard for diagnosis [7]. Histopathology reveals an enormous number of sporangia in subepithelial connective tissue. These elements consist of sharply defined globular thick-walled cysts (sporangium), upto 0.5 mm in diameter, which contain numerous coated endospores, 6-7 micrometers in diameter. Immature and collapsed sporangia are also present [2]. FNAC and histopathological picture of all the cases in the present study were similar to the above findings and helped to reach a conclusive diagnosis of rhinosporidiosis.

The most common differential diagnosis is coccidioidomycoses, which can be differentiated by their size (<60 micrometers in diameter). Other differential diagnosis includes pyogenic granuloma and myospherulosis, an iatrogenic condition related to the application of oil-based ointment in the nose [2].

The life cycle of the parasite is intricate. Sporangia are the mature forms of the organism which contain multiple sporangiospores. After maturation, sporangiospores are released, which later develop into trophocytes. This immature form of Rhinosporidium seeberi is smaller and thinner than sporangia that do not contain endospores. Direct contact by humans with spores through dust, through infected clothing or fingers, and through swimming in stagnant water are the proposed methods of transmission in this disease [2].

The taxonomy of the causative organism is unclear. Malbran first described Rhinosporidium seeberi in 1892. Then later, it was classified as protozoan by Seeber in 1900. Ashworth thought it to be phycomycetes and proposed the name Rhinosporidium seeberi. It was also put under a new class of aquatic protistans called mesomycetozoea, along with other aquatic parasites that cause similar infections in amphibians and fish in some research [10]. However, recent studies by Ahluwalia KB hypothesised that the causative organism is a prokaryotic cyanobacterium, Microcystis

aeruginosa, as this organism was isolated from both the clinical specimen of patients and pond water samples where they bathed [11-13]. After analysis of 18s small subunit ribosomal DNA groups, Herr RA et al., proposed that R. Seeberi was related to a group of fish parasites referred to as the DRIP clade (Dermocystidium rosette agent, icthyophonus and psorospermium) [14]. Fredericks DN et al., also accepted the concept of a novel clade of aquatic protistan parasites named Ichtyosporea, pointing to the resemblance with other members of the DRIP clades [15].

The recurrence rate of nasal lesions is high, nearly 30% in some studies [16]. In the present study, the recurrence of nasal lesions was 25%. Dapsone, which is believed to arrest the maturation of sporangia and induce fibrosis in the stroma, may be used in medical management of cutaneous rhinosporidiosis [17]. But, surgical removal and electrodesiccation of the lesion remain the treatment of choice [6,18]. Also, it is noteworthy that many studies have suggested that the incidence of cutaneous rhinosporidiosis is very low [19].

Limitation(s)

Major limitation of present study was the small sample size due to the rare occurence of the disease.

CONCLUSION(S)

Disseminated cutaneous rhinosporidiosis may present with skin lesions masquerading as several common skin diseases, which leads to diagnostic dilemmas and social stigma for patients. A detailed enquiry about the history and clinical examination of the nasal or oral lesion is essential. FNAC and biopsy will further help in confirmation. The current study will succour the clinician for the proper diagnosis and treatment of the patient.

Acknowledgement

The authors would like to thank all the patients, technical staff, and faculties of the Department of Pathology, VIMSAR, Burla.

REFERENCES

[1] Thappa DM, Venkatesan S, Sirka CS, Jaisankar TJ, Gopalkrishnan, Ratnakar C. Disseminated cutaneous rhinosporidiosis. J Dermatol. 1998;25:527-32.
[2] Kumari R, Nath AK, Rajalakshmi R, Adityan B, Thappa DM. Disseminated cutaneous Rhinosporidiosis: Varied morphological appearances on the skin. Indian J Dermatol Venereolleprol. 2009;75(1):68-71.
[3] Salim T, Komu F. Varied presentations of cutaneous rhinosporidiosis: A report of three cases. Indian J Dermatol. 2016;61:209-12.
[4] Ghorpade A. Polymorphic cutaneous rhinosporidiosis. Eur J Dermatol. 2006;16:190-92.
[5] Lupi O, Tyring SK, mcginnis MR. Tropical dermatology: Fungal tropical diseases. J Am Acad Dermatol. 2005;53:931-51.
[6] Kumari R, Laxmisha C, Thappa DM. Disseminated cutaneous rhinosporidiosis. Dermatol Online J. 2005;11:19.
[7] Nayak S, Acharya B, Devi B, Sahoo A, Singh N. Disseminated cutaneous rhinosporidiosis. Indian J Dermatol Venereol Leprol. 2007;73:185-87.
[8] Hadke NS, Baruah MC. Primary cutaneous rhinosporidiosis. Indian J Venereol Leprol. 1990;56:61-63.
[9] Kamal MM, Luley AS, Mundhada SG, Bohhate SK. Rhinosporidiosis: Diagnosis by scrape Cytology. Acta Cytol. 1995;39:931-35.
[10] Ghorpade A, Gurumurthy J, Banerjee PK, Banerjee AK, Bhalla M, Rabindranath M. Oculosporidiosis presenting as an under-eye swelling. Indian J Dermatol Venereal Leprol. 2007;3:196-97.
[11] Ahluwalia KB, Maheshwari N, Deka RC. Rhinosporidiosis: A study that resolves etiologic controversies. American J Rhinol. 1997;11(6):479-83.
[12] Ahluwalia KB. New interpretations in rhinosporidiosis enigmatic disease of the last nine decades. J Submicrosccytol Pathol. 1992;24(1):109-14.
[13] Ahluwalia KB. Culture of the organism that causes rhinosporidiosis. J Laryngol Otol. 1999;113(6):523-28.
[14] Herr RA, Ajello L, Taylor JW, Arsecularatne SN, Mendoza L. Phylogenetic analysis of Rhinosporidiumseeberi's 18S small- subunit Ribosomal DNA groups this pathogen among the members of the protoctistan mesomycetozoa clade. J Clin Microbiol. 1999;37:2750-54.
[15] Fredericks DN, Jolly JA, Lepp PW, Kosek JC, Relman DA. Rhinosporidium seeberi: A human pathogen from a novel group of aquatic protistan parasites. Emerg Infect Dis. 2000;6:273-82.
[16] Shariff MA. A clinicopathological study of rhinosporidiosis in a tertiary care hospital. Int J Otorhinolaryngol Head Neck Surg. 2018;4:981-85.
[17] Venkateswaran S, Date A, Job A Mathan M. Light and Electron microscopic findings in Rhinosporidiosis after dapsone therapy. Trop Med Int Health. 1997;2(12):981-85.
[18] Vijaikumar M, Thappa DM, Karthikeyan K, Jayanthi S. A verrucous lesion of the palm. Postgrad Med J. 2002;78:302,305-06.
[19] Sen S, Agrawal W, Das S, Nayak PS. Disseminated cutaneous rhinosporidiosis: Revisited. Indian J Dermatol. 2020;65(3):204-07.

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PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Jun 15, 2020
- Manual Googling: Oct 05, 2020
- iThenticate Software: Jan 21, 2021 (21%)

ETYMOLOGY: Author Origin

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was Ethics Committee Approval obtained for this study? No
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Jun 14, 2020**
Date of Peer Review: **Jul 16, 2020**
Date of Acceptance: **Oct 08, 2020**
Date of Publishing: **Apr 01, 2021**