Papillary ThyroidCarcinoma In AThyroglossalCystInAn AdultPatient

ABSTRACT:

AThyroglossalcystisamongstthemostcommonlyencounteredanteriorpainless neck swelling. Its usual presentation is seen in childhood and rarelypresents in adulthood. The management for Thyroglossal cyst is, the Sistrunk'sOperation.TheetiologyisexplainedbyembryonicdevelopmentoftheThyroglossalductanditsdeficiencytoobliterateafterbirth.Herewepresentacaseofa45yearoldgentlemanwhopresentedwithapainlessmidlineneckswelling since 6 years. Onset of the swelling was insidious and graduallyincreased insize overthe years.Patientdid notgiveany historysuggestive ofaninfectionorthyroidglanddysfunction.MRIoftheNeckrevealeda3.5x3x3.5cm well-definedlesioninthe anteriornecksuggestingaThyroglossal Cyst. Patientwasworkedupforasistrunk'sprocedure.Histopathologyrevealedawell-differentiatedPapillaryThyroidCarcinoma.ThyroglossalductCystinadulthoodis a rare entity as most of thecase usually presented at childhood. Presence ofaThyroglossal Cystinan adultshouldprompt thecaregiverto evaluateforamalignantchange.Sistrunksprocedureremainsthestandardtreatmentofchoice for all the age groups. Infection of the cyst remains the most commoncomplicationofThyrglossalcyst.

KEYWORDS-

ThyroglossalCyst,ThyroglossalDuct,Sistrunk'sProcedure,PapillaryThyroidCarcinoma,Malignancy.

INTRODUCTION

Thyroglossalductcyst(TDC)isoneofthemostcommonmidlineswellingof the neck, which arises from an unobliteratedthyroglossal duct which ispresent in the embryonic period[3]. It represents the embryonic pathway ofdescentofthethyroidgland.Asitisacongenitaldiseaseitpresentsmostcommonlyi nthechildhoodperiodandseldompresentsinadulthood.Hence,most of the data available on this subject focuses on management of thisconditioninthechildhoodperiod.

Sistrunk's operation is the procedure of choice forthis disease and isacceptedgloballyasthestandardofcare[4].Theoperationincludestheexcisionofthe cystalongwiththecentralpartofthehyoidboneandcoringofthetracttill the foramen cecum. This is driven by the knowledge of the embryonicdevelopment of the Thyroglossal duct and its intimate relation with the Hyoidbone.

Data available reveal satisfactory cure rates with Sistrunk's Operation forThyroglossalcystoccurringinchildhoodandveryfewcasesarereportedinadulta gegroupandhencelessdataisavailableforcomparison[6].

CASEREPORT

Herewepresentacaseofa45yearoldgentlemanwithnocomorbidities,whopresen tedwithapainlessmidlineneckswellingsince6years,theswellingwas first noticed by the patient 6 years ago when it was the size of a peagraduallyitprogressedtothecurrentsizeofalemon.Patientdidnotgiveanyhistor yofFever,Pusdischargefromswelling.Nohistorysuggestiveofthyroiddysfunction or difficulty in swelling/breathing/change of voice. Examinationrevealed a 4 cm x 4 cm midline neck swelling, moving with deglutition andprotrusionoftongue,smoothsurfacewithroundededges,notendernessor

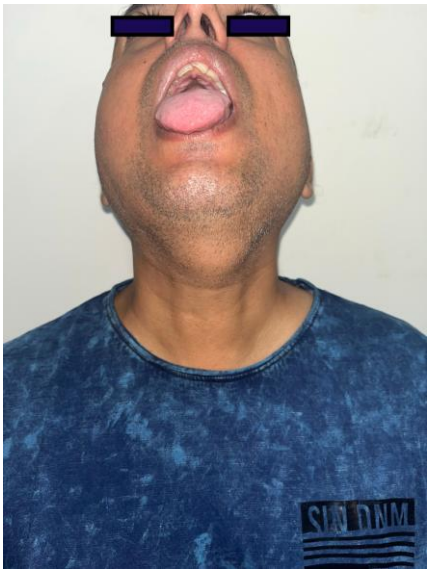

Fig1. Movementwithtongueprotrusion.

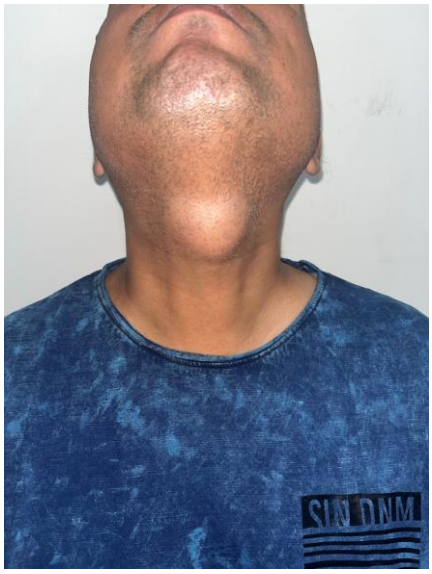

Fig2. Midlineneckswelling

riseoflocaltemperature.Thyroidglandnotpalpable.Nocervicallymphnodespalpated.

CECTofNeckrevealedawell-definedperipherallyenhancingcystic

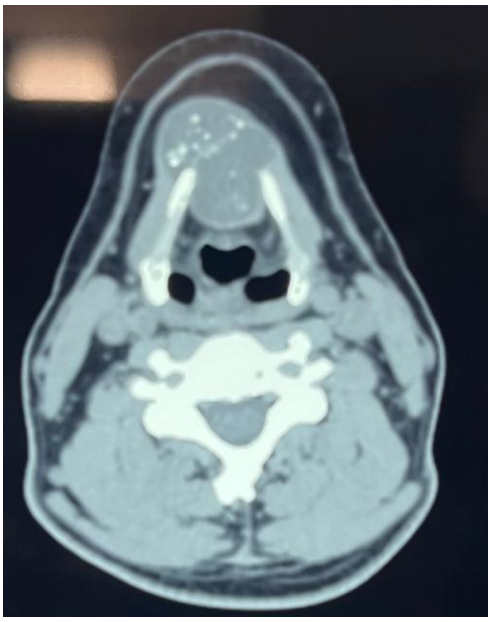

Fig3. ImageShowingcalcifications.

structureinmidlineupperneckinfrahyoidinlocationmeasuring3.6cmx2.9cmx3.5cmwithfociofcalcification.

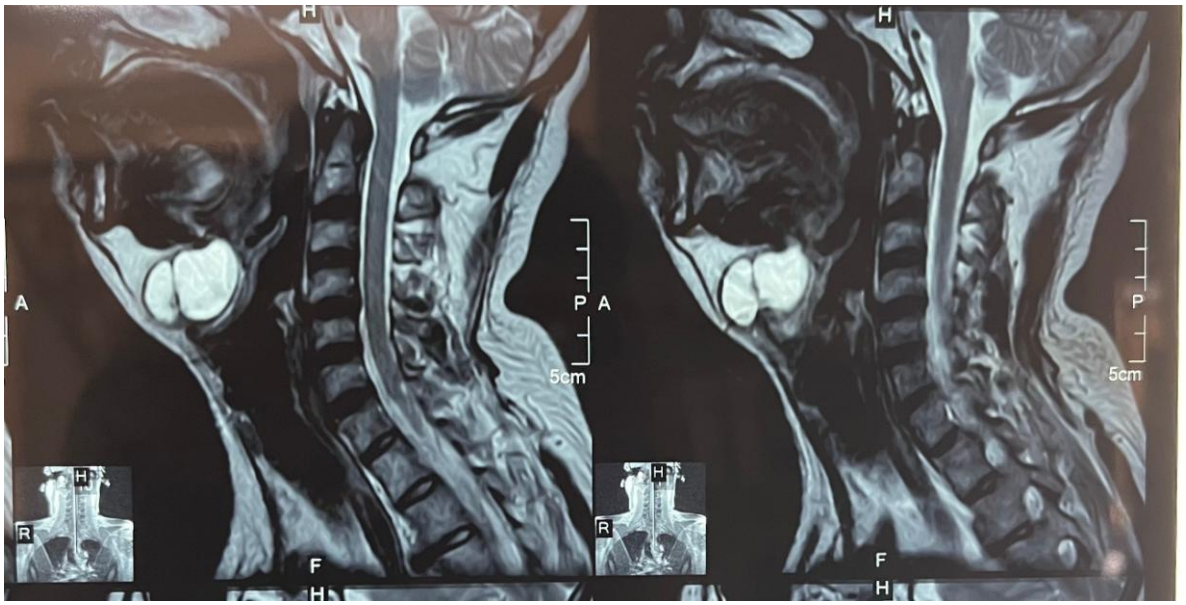

Fig4.MRINeckinsagittalview.

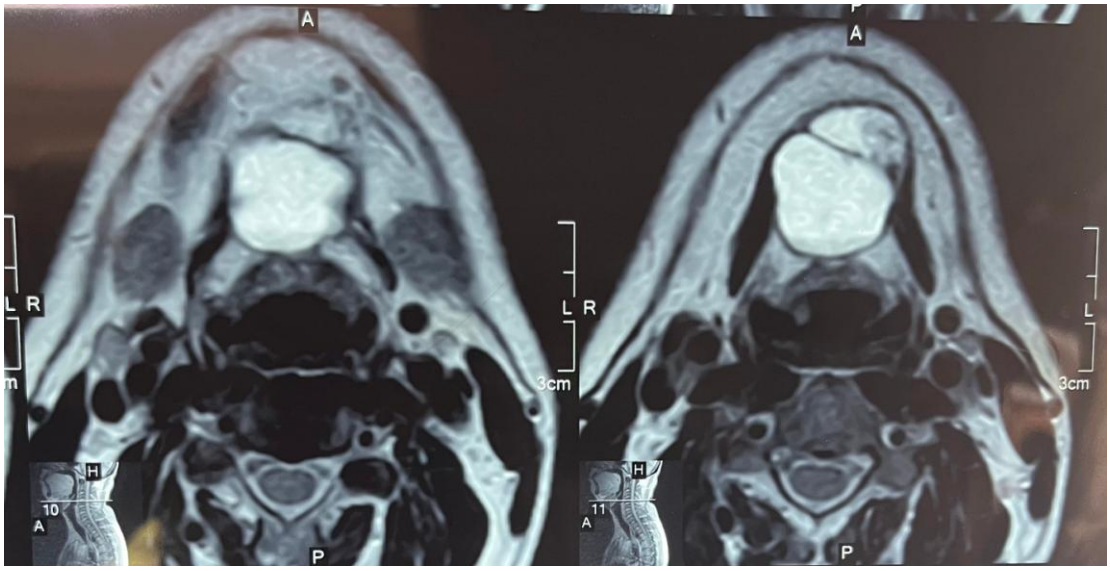

Fig5. Antero-posteriorviewshowingcloserelationthehyoidbone.

MRINeckrevealedwelldefinedT1hyperintenseswellingintheinfrahyoidregion withmultiplebloomingspotssuggestiveofthyroglossalcystwithcalcifications.Theth yroidglandwasnormalwithnodetectablecervicallymphnodes.

Patient was posted for Sistrunk's procedure. Patient, under generalanaesthesia,was taken in the supine position with neck extended and headsupportedwiththehelpofaring.Atransverseincisionwastakenovertheswellinga nddeepenedtilltheinvestinglayerofdeepcervicalfascia.Strapmuscles were retracted laterally andswelling was freed fromall adhesions.Hyoid bone was located superiorly to the swelling, off which, the muscleattachmentsfromthecentralpartwerereleased.Thehyoidbonewascutinthec entral part. The tract was then seen extending superiorly into floor of themouthandcoreduptothemylohyoidmuscle.Woundwasclosedinlayersafterachie ving homeostasis over a closed vacuum drain. Patient tolerated theprocedure wellandthespecimenwassentforaHistopathologicalExam.Postoperativeperiodwas uneventfulandthepatientwasdischargedofPOD-2withdrainin-situ.

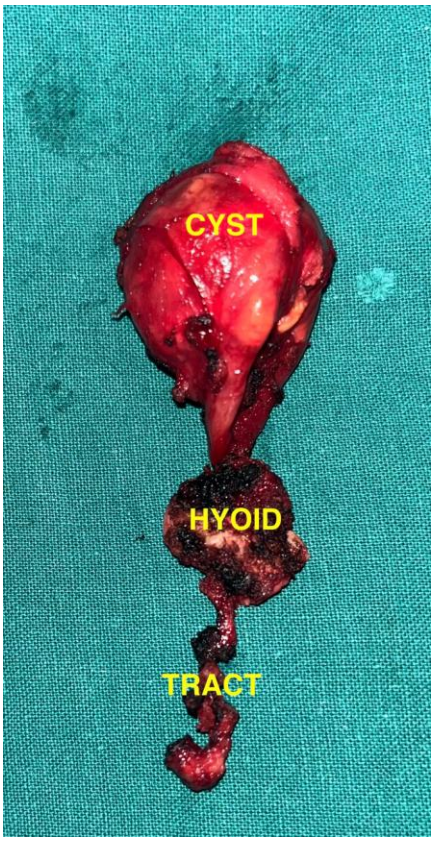

Fig6. Post operativespecimen.

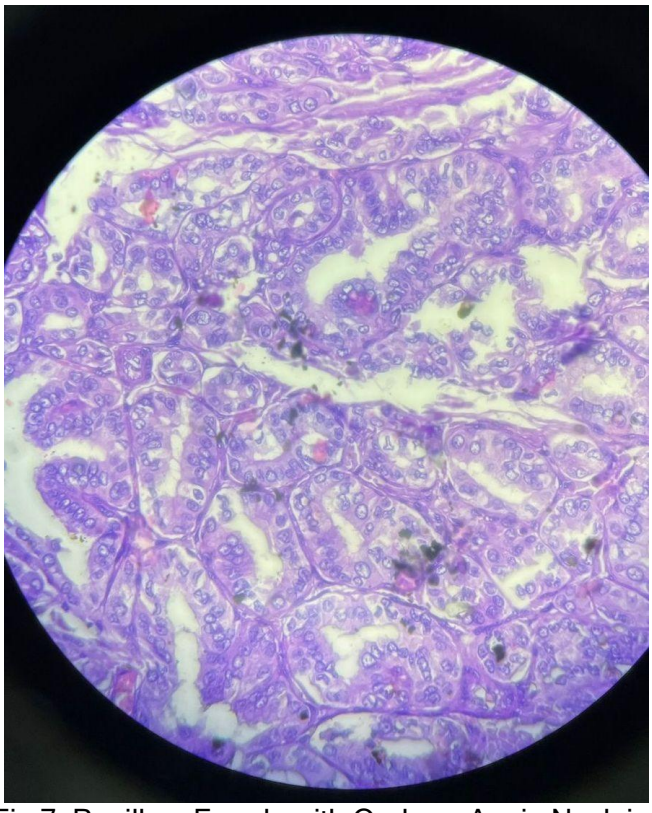

Fig 7. Papillary Fronds with Orphan- Annie Nuclei Noted.

Histopathologicalexamination,ongrossingreavealeda2.3x2x2cmovoidcysti cmasswithgraysurfacewithyellowishsolidandserousfluidcontainingcysticfluid,on microscopyawell-differentiatedPapillaryThyroidCarcinomainvolvingthe capsule with classical orphan Annie nuclei was noted.

DISCUSSION

Painlessanteriormidlineneckswellingisthemostcommonpresentationof Thyroglossal Cyst in adult patients. The presence of other symptoms such aspain,odynophagia,dysphagia,anddyspnoeaoftenindicatesthepresenceofcomplicationssuchasabscessformation.Locationofthyroglossalcystwithrespecttohyoidbonecanbevariable,whilemidlinepositionisthedominantlocationinbothchildrenandadults,lateraldeviationhasbeennotedinadultpresentation[3].With respecttothehyoidbone,cystscanbeabove,over,orbelowthehyoid,mostcommonlytheyareinfra-hyoid.Thesurgicalmanagementofthyroglossalcysthaschangedwithtime.EarlierIncision&Drainageorsimplecystexcisionwerepresentedwithunacceptablyhighlevelsofrecurrence,Schlangein1893,suggestedexcisionofthecystandmid-portionofthehyoidboneandleavingbehindtheproximaltract–atechniquewhichresulted in recurrence rates of 30 %. In 1920 Walter Ellis Sistrunk reported theclassicalSistrunkprocedurewhichsignificantlyimprovedpostoperativeoutcomes and has since remained the gold standard to date[2]. Recentadvancements such as robot-assisted, endoscope-assisted transoral, axillary &retro-auricularapproacheshavebeenattemptedforthyroglossalcystinadults.Theseproceduresarecosmeticallysuperiortothesistrunksoperationbutdataonefficacybeingthesameareinadequate.

Recurrence is the most important post-op outcome following the Sistrunkprocedure, with a recurrence noted in 3%-6% of cases. This is often attributedtotechnicalshortcomingsviz.incompleteexcisionoftheductorthepresenc

e

oframificationofducts,whichremainunrecognizedatthetimeofsurgery.Mostrecurrences occur in the first 6 months[6]. Another important possiblecomplicationisdamagetothe12thCranialnerve.MeticulousdissectionofthecentralpartofhyoidboneandpreservationoftheſuperiorHornofHyoidbonecanhelpsignificantlyinpreventinginjuriestotheHypoglossalnerve.

IncidenceofThyroglossalcystintheadultpopulationisabout7%. Thyroglossal Cyst present with a <1% chance of harbouring a malignant foci inadult population[7]. There are only around 300 cases of Papillary thyroidcarcinoma reported in TDC since its first reporting by Brentano in 1911[7]. Mostcommonlyonhistopathologywefindpapillarythyroidcarcinoma(90%)[5].Manytreatmentoptionssuchas TotalThyroidectomywithBilateralneckdissection,radioactiveiodine,thyroidsuppressionhavebeensuggested.Sistrunksoperationisusuallyconsideredsufficientandneedforfurtherinterventionisdecidedonthebasisonthefollowingfindingsa.)Thyroidnodule(cold)pickeduponthyroidscanb.)cervicallymphnodesdetectedclinicallyoronimagingc.)prior history of irradiation to the neck. Calcification is the hallmark of papillarycarcinoma in a thyroglossal duct cyst[5]. Pre-operative role of FNAC remainsuncertain but should be recommended to pick up lesions early and to plan adefinitive treatment[6].In ourpatient, sincerole ofFNAC inThyroglossal cystisdebated & since there was nothing to suggest a malignancy pre-op, an FNACwasnotdone.Also,nofurthertreatmentintheformofthyroidectomyorcentral neck dissection was not performed as it did not meet the above mentionedcriterion.

CONCLUSION

Therareoccurrenceofthyroglossalcystinadultsisthereasonforthepaucityofdataintheadultpopulation.Themajorityofpatientsaremales.Reportsinliterature,however,donotcometoaconsensuswithsomereports

suggesting equal distribution, whilst somesuggest male or femalepreponderance.Afterlingualthyroid,theThyroglossalcystinthesecondmost-commonsiteforectopicthyroidtissue.Incidenceofwhichisapproximately60%.Inmost ofthecasesthethyroidglandisnormalandthepatientsareofteneuthyroidorhypothyroidatpresentation.Hence,pre-operativeevaluationwithathyroidfunctiontestandthyroidscanshouldbeundertaken, priortoitsexcision,soastoconfirmwetherthe tissueinthecystisnottheonlyfunctioningthyroidtissueinthebody.

Ethical Approval:
As per international standard or university standards written ethical approval has been collected and preserved by the author(s).
Consent
As per international standards or university standards, patient(s) written consent has been collected and preserved by the author(s).

FOLLOW-UP PROTOCOL

Inourcase,thepatienthasbeenadvisedayearlyfollowupwiththyroidfunctiontestsandUSGofneckwiththyrogllobuinlevelsashedoesnotmeetthe criteria for warranting additional intervention for the papillary thyroidcarcinoma.

CONFLICTOFINTEREST

Theauthorsdeclarethatthereisnoconflictofinteresttodeclarewithregardstothepublicationofthisresearchmaterial.

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