

Marked Clinical and Serologic Improvement After Lingual Tonsillectomy for Refractory PANDAS

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Background

- Pediatric Autoimmune Neuropsychiatric Disorder Associated with Streptococcal infection (PANDAS) involves abrupt-onset OCD, tics, and behavioral changes following Group A Streptococcus (GAS) exposure.
- **Pathogenesis:** post-streptococcal autoimmunity targeting basal ganglia circuitry via molecular mimicry.
- Palatine tonsillectomy has been proposed for refractory cases, but outcomes remain inconsistent.
- Lingual tonsillectomy has not been previously described as a therapeutic intervention for PANDAS.

Case Presentation

- 29-year-old male with adolescent-onset PANDAS (post-GAS infection at age 17).
- **Symptoms:** severe obsessive-compulsive behaviors, coprolalia, emotional lability, and choreiform movements.
- Previously underwent palatine tonsillectomy but remained refractory despite plasmapheresis, rituximab, and mycophenolate mofetil.
- ASO titers consistently paralleled symptom flares.
- Otolaryngology evaluation identified hypertrophied lingual tonsils, the only remaining lymphoid tissue of Waldeyer's ring.
- Underwent bilateral transoral robotic-assisted lingual tonsillectomy (TORS).

Pathologic Findings

- **Histopathology:** Dense lymphoid hyperplasia and reactive germinal centers (Figure 1).
- No evidence of acute infection or neoplasia.

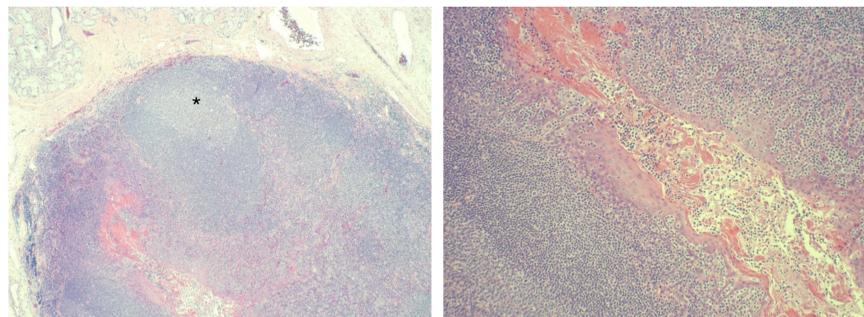


Figure 1. A) Low-power histology of lingual tonsil demonstrating follicular hyperplasia (*), crypt and adjacent minor salivary gland (H&E stain). B) High-power histology of lingual tonsil showing reactive germinal centers and lymphoid hyperplasia (H&E).

Postoperative Course

- **ASO titers:** Demonstrated perioperative stabilization, remaining in the ~200 U/mL range preoperatively (190–274 U/mL) and during early postoperative follow-up, with a value of 199.7 U/mL by postoperative day 18 (Table 1, Figure 2).
- Clinical outcome: Rapid resolution of tics, compulsions, and attentional deficits by postoperative day 11 (Table 1).
- Discontinued immunosuppressants and no recurrence of neuropsychiatric symptoms at follow-up.
- No surgical complications, returned to normal diet and activity.

Day (Pre/Post-op)	Event/Intervention	ASO Titer (IU/mL)	Outcome/Clinical Status
-24	Pre-op baseline	204.1	Stable with weekly PF, residual symptoms persist
-17	Continued PF	190.1	Mild clinical stability
-10	ASO Spike prior surgery	274.1	Increased neuropsychiatry symptoms
-3	Pre-op decline begins	230.5	ASO decrease likely related to ongoing weekly plasmapheresis; decision made to proceed with surgery
0	TORS Bilateral Lingual Tonsillectomy	-	Surgery completed without complication
+4	Post-op clinic visit	202.9	Marked behavioral improvement: no tics/compulsions; improved attention; mild dysphagia
+11	ASO trending down	203.8	Continued improvement; symptoms resolved; possible seizures being evaluated
+18	ASO normalizes	199.7	Symptom-free; stable

Table 1. Clinical Timeline: ASO Titers and Symptom Status Relative to Lingual Tonsillectomy (Day 0)

Discussion

- Supports hypothesis that persistent tonsillar immune reservoirs may perpetuate autoimmune activation in PANDAS.
- Prior reports focused on palatine and adenoid tissue. This case highlights the potential role of lingual tonsils.
- Removal of all lymphoid components of Waldeyer's ring may be warranted in select refractory cases.
- The correlation between lingual tonsillectomy, serologic normalization, and clinical remission suggests a possible causal link.

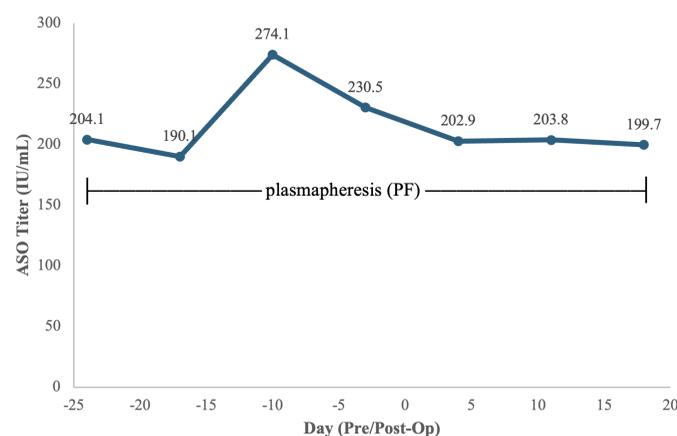


Figure 2. Line graph depicting anti-streptolysin O (ASO) titers (IU/mL) over time relative to transoral robotic surgery (TORS) lingual tonsillectomy, performed on Day 0. The patient continued weekly plasmapheresis throughout the pre- and postoperative period.

Conclusions

- First reported case of lingual tonsillectomy producing marked clinical and serologic improvement in refractory PANDAS.
- Suggests lingual tonsillar tissue may serve as a chronic immunologic reservoir sustaining disease activity.
- Comprehensive evaluation of Waldeyer's ring should be considered in refractory or recurrent cases.
- Further studies are needed to define the immunologic significance and therapeutic role of lingual tonsillectomy in PANDAS.

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